Resurfacing hemiarthroplasty of the shoulder for patients with juvenile idiopathic arthritis

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Background: This study reports the outcome of resurfacing hemiarthroplasty (RHA) in a cohort of patients with juvenile idiopathic arthritis (JIA) affecting the shoulder joint.

Methods: Fourteen uncemented RHA procedures were performed for 11 consecutive patients who required surgery because of JIA. Mean age at surgery was 36.4 years. Mean clinical follow-up was 10.4 years (range, 5.8-13.9 years). A significant humeral head defect (up to 40% surface area) was found in 5 shoulders and filled with autograft from the distal clavicle or femoral head allograft.

Results: At latest follow-up, no patient required revision. There was excellent relief from pain. The mean Oxford Shoulder Score and Constant-Murley Score improved significantly. No shoulder had a poor outcome, and 6 had a very good or excellent outcome. Worse outcome was associated with an intraoperative finding of significant humeral head erosion. Two shoulders required early arthroscopic subacromial decompression, but there were no other reoperations. There were no instances of radiographic implant loosening or proximal migration. Painless glenoid erosion was seen in 5 shoulders but was not associated with worse outcome.

Conclusions: The midterm results of RHA for JIA are at least comparable to those for stemmed hemiarthroplasty, with the added benefit of bone conservation.

Level of evidence: Level IV; Case Series; Treatment Study

Keywords: Juvenile idiopathic arthritis; juvenile rheumatoid arthritis; shoulder resurfacing; shoulder arthroplasty; shoulder replacement; resurfacing hemiarthroplasty

Juvenile idiopathic arthritis (JIA) is a chronic inflammatory arthropathy of unknown etiology that begins before the age of 16 years, persists for at least 6 weeks, and cannot be explained by any other cause. Patients with aggressive polyarticular disease often require arthroplasty in young adulthood, which is well established for the hip, knee, and elbow joints.\textsuperscript{1,2,6,10,12,19,21} The shoulder joint is estimated to be affected in 21\% of children within 5 years of diagnosis.\textsuperscript{9}

Arthroplasty options include stemmed or resurfacing hemiarthroplasty or total shoulder arthroplasty. In the adult rheumatoid literature, arthroplasty results in significant pain relief regardless of prosthesis choice,\textsuperscript{22} and some series have included very small numbers of patients with JIA.\textsuperscript{7,8} However, only 2 articles and 1 abstract have been published that are...
devoted to the outcome in patients with JIA, and all used stemmed humeral prostheses as part of a hemiarthroplasty or total shoulder arthroplasty procedure. Surface replacement of the humeral head represents a bone-sparing alternative for these young patients. This study reports the outcome of resurfacing hemiarthroplasty (RHA) in a cohort of patients with JIA affecting the glenohumeral joint.

Materials and methods

We retrospectively reviewed data gathered through routine clinical interaction with patients who underwent RHA of the glenohumeral joint because of JIA at our institution. Patients were identified through an arthroplasty database. Patients undergoing surgery for any other indication were excluded, as were patients in whom surgery had been performed less than 5 years earlier. Patients were referred to the senior author (M.T.) from a specialist rheumatology department after exhaustive medical management, including disease-modifying antirheumatic medication, physical therapy, and intra-articular corticosteroid injections. The indications for surgery were pain and loss of function with evidence of joint destruction on plain radiographs in patients whom nonsurgical management had not adequately controlled symptoms.

Operative technique

All procedures were performed by the senior author (M.T.) with the patient under general anesthesia with interscalene block and in the beach chair position. The Copeland uncemented Mark-3 prosthesis (Biomet Orthopedics, Warsaw, IN, USA) was used exclusively. Preoperative radiographs were templated, and a custom-made extrathin prosthetic was ordered in advance if thought necessary.

After a standard deltopectoral approach, the condition of the rotator cuff was noted. Division of the subscapularis tendon and anterior capsule allowed the joint to be carefully dislocated, and the humeral head was examined for erosion. In cases of humeral head erosion, the incision was extended proximally and laterally so that the distal clavicle could be excised and used as bone graft if quality allowed. A femoral head allograft was prepared for larger defects of up to 40% of the humeral head surface area.

A central guidewire was placed, and a pilot hole was created with a spade cutter and typically undersized to ensure maximal press-fit of the prosthetic tapered central post in the eroded, osteopenic bone. The head was sized and reamed, with the reamings retained as a graft. Before implant insertion, humeral head defects were treated with allograft or morselized reamings, or both, and distal clavicle bone with impaction grafting using the appropriately sized trial component. The graft was added to the undersurface of the prosthesis before it was impacted into place. The head was then gently retracted to allow inspection of the glenoid cavity, which was microfractured in the presence of significant patchy eburnation but never replaced.

The long head of biceps tendon was cut and tenodesed in the bicipital groove if diseased. The subscapularis tendon and anterior capsule were extensively released. The upper third of the pectoralis major tendon was released if felt likely to limit external rotation.

Postoperatively, the patient was typically immobilized in a broad-arm sling for 4 weeks before starting graduated mobilization exercises, dependent on the state of the soft tissue repair.

Data collection

Patient notes were consulted to obtain basic demographic information in addition to age at JIA diagnosis, disease subtype, the presence or absence of rheumatoid factor in the patient’s blood, and the number of prior and subsequent upper and lower limb arthroplasty procedures. Pain assessment using a 10-point visual analog scale score, active range of motion, complications, and Oxford Shoulder Score (OSS) are routinely recorded at our institution preoperatively by a specialist nurse and at yearly postoperative visits by the reviewing clinician (typically an assistant surgeon). We also record a Constant-Murley Score (CMS), with measurements for range of motion and strength taken in accordance with the originally described method. To assess the strength component, patients were asked to abduct their shoulder to 90° in the scapular plane against a spring balance attached to the forearm in pronation and anchored to the floor on 3 separate occasions. Patients who were unable to abduct their arm to 90° scored 0 for this component.

Preoperative anteroposterior, axillary, and scapular Y-view radiographs were assessed for severity of arthritis according to the classification described by Larsen et al. The most recent postoperative radiograph was assessed for evidence of glenoid erosion, prosthesis subluxation, and evidence of prosthesis loosening on all views by the first (E.F.I.) and senior (M.T.) authors.

Statistical analysis

Continuous outcome data sets were tested for normality using the Shapiro-Wilk W test and compared using the Student t test if parametric. Nonparametric and ordinal data were compared using the Mann-Whitney U test. Nominal data were compared using the Fisher’s exact test. A P value of <.05 was considered statistically significant.

Results

Since 2002, 17 RHA procedures have been performed at our institution for 13 consecutive patients because of JIA. We excluded 3 shoulders (2 patients) because follow-up was less than 5 years since the operation, leaving a study group of 14 shoulders in 11 patients (9 women, 2 men). The mean age at surgery was 36.4 years (range, 19-49 years), and mean age at diagnosis of JIA was 6.2 years (range, 2-15 years). Nine patients (11 shoulders) had been diagnosed with the systemic polyarticular form of the disease, 1 patient (2 shoulders) with the extended oligoarticular form, and 1 patient (1 shoulder) with the psoriatic form. Only 1 patient was positive for rheumatoid factor.

No patient had undergone a prior surgical procedure for their operated-on shoulder. At the time of the latest follow-up, patients had undergone a median of 4 primary and revision arthroplasties (1 to 9 arthroplasties) of other major joints in their lifetime, but only 1 patient had undergone ipsilateral upper limb arthroplasty (total elbow replacement; Fig. 1, a). The same patient had also undergone a contralateral stemmed shoulder hemiarthroplasty, before the current study period, and contralateral total elbow replacement (Fig. 1, b).
Follow-up

No patient had been lost to follow-up at the time of review. Mean postoperative clinical follow-up was 10.4 years (range, 5.8-13.9 years). Not all patients had been exposed to radiographs at last appointment, so radiographic follow-up was slightly shorter, with a mean 9.7 years (range, 5.8-13.5 years).

Operative details

The Copeland uncemented hydroxyapatite coated Mark-3 prosthesis was used. The rotator cuff was intact in all shoulders but thinned due to disease and disuse. Intraoperative findings, implant sizes, and additional procedures performed at the time of index operation are detailed in Table I.

Table I  Operative details for the 11 patients*

<table>
<thead>
<tr>
<th>Patient</th>
<th>Sex</th>
<th>Side</th>
<th>Operative difficulty</th>
<th>Implant size</th>
<th>Bone graft</th>
<th>Additional procedures</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>F</td>
<td>R</td>
<td>–</td>
<td>1</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>F</td>
<td>R</td>
<td>Severe posterior head erosion</td>
<td>1</td>
<td>Allograft</td>
<td>PMj release</td>
</tr>
<tr>
<td></td>
<td></td>
<td>L</td>
<td>Severe posterior head erosion</td>
<td>1</td>
<td>Distal clavicle autograft</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>M</td>
<td>L</td>
<td>–</td>
<td>3</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>L</td>
<td>Moderate posterior head erosion</td>
<td>1</td>
<td>Distal clavicle autograft</td>
<td>LHB tenodesis</td>
</tr>
<tr>
<td></td>
<td>R</td>
<td>Moderate posterior head erosion</td>
<td>1</td>
<td>Distal clavicle autograft</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>F</td>
<td>R</td>
<td>Severe posterior head erosion</td>
<td>Custom extra small</td>
<td>Allograft</td>
<td>Glenoid microfracture</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Bare glenoid, multiple adhesions</td>
<td></td>
<td>Distal clavicle autograft</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>F</td>
<td>L</td>
<td>Mild posterior head erosion</td>
<td>3</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Coracoid overgrowth</td>
<td></td>
<td>–</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>F</td>
<td>L</td>
<td>Moderate posterior erosion</td>
<td>3</td>
<td>Custom extra small</td>
<td>LHB tenodesis Glenoid microfracture</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>GT overgrowth, bare glenoid</td>
<td></td>
<td>Distal clavicle autograft</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>M</td>
<td>L</td>
<td>–</td>
<td>3</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>F</td>
<td>L</td>
<td>Moderate posterior and superior head erosion</td>
<td>2</td>
<td>Distal clavicle autograft</td>
<td>PMj release</td>
</tr>
<tr>
<td>10</td>
<td>F</td>
<td>L</td>
<td>–</td>
<td>1</td>
<td>–</td>
<td>PMj release LHB tenodesis</td>
</tr>
<tr>
<td>11</td>
<td>F</td>
<td>R</td>
<td>–</td>
<td>2</td>
<td>–</td>
<td>LHB tenodesis</td>
</tr>
</tbody>
</table>

* Excision of the acromioclavicular joint and mobilization of the subscapularis before repair was performed for all patients.

F, female; R, right; L, left; M, male; PMj, pectoralis major; LHB, long head of biceps tendon; GT, greater tuberosity.
Complications

There were no intraoperative or early postoperative complications such as infection. No implant has required revision to date. Symptoms suggestive of impingement syndrome developed in 2 patients at 6 months. Both failed to respond to nonsurgical treatment but improved after arthroscopic subacromial decompression at 1 year. At the time of arthroscopy, the rotator cuff was intact in both patients, with no evidence of progressive glenoid erosion since the index procedure. No complications have occurred to date in the other 9 patients.

Pain

All patients reported their shoulder was extremely painful before surgery. The mean preoperative pain score was 9.0 of 10 (range, 7-10 points). At the latest follow-up, 8 of 14 shoulders were pain free, and the remainder experienced only mild pain (mean, 0.64 points; range, 0-3 points; \( P < .001 \)).

Movement

Mean active range of motion in forward flexion, external rotation, and internal rotation had improved significantly at the latest follow-up (Table II). Range of motion in all 3 directions was improved in 11 of 14 shoulders. Only 1 of the remaining 3 shoulders did not improve in any direction.

Table II  Comparison of preoperative and postoperative mean values for range of motion and outcome scores

<table>
<thead>
<tr>
<th>Variable</th>
<th>Range of motion</th>
<th>Outcome score</th>
</tr>
</thead>
<tbody>
<tr>
<td>Preoperative</td>
<td>FF,°</td>
<td>ER,°</td>
</tr>
<tr>
<td>FF</td>
<td>69 (30-110)</td>
<td>12 (0-40)</td>
</tr>
<tr>
<td>ER</td>
<td>10 (0-60)</td>
<td>2.5 (0-4)</td>
</tr>
<tr>
<td>IR</td>
<td>&lt;.001</td>
<td>&lt;.001</td>
</tr>
</tbody>
</table>

FF, forward flexion; ER, external rotation; IR, internal rotation; CMS, Constant-Murley Score; OSS, Oxford Shoulder Score.

Data are presented as mean (range) except for IR, which is median (range).

* IR was measured according to predefined levels of thumb tip position achieved when attempting to reach behind the back: 0 = lateral thigh, 1 = buttock, 2 = sacroiliac joint, 3 = midlumbar spine, 4 = 12th thoracic vertebra, 5 = 6th thoracic vertebra.

Table III  Comparison of preoperative, intraoperative, and postoperative characteristics for patients who achieved an excellent or very good postoperative Constant-Murley Score vs. a good or fair result

<table>
<thead>
<tr>
<th>CMS post-op result</th>
<th>No.</th>
<th>Preoperative</th>
<th>Intraoperative</th>
<th>Postoperative</th>
</tr>
</thead>
<tbody>
<tr>
<td>CMS</td>
<td>OSS</td>
<td>Larsen grade V</td>
<td>Significant humeral head erosion</td>
<td>Follow-up, y</td>
</tr>
<tr>
<td>Excellent/very good</td>
<td>6</td>
<td>17.5</td>
<td>17.0</td>
<td>2/6</td>
</tr>
<tr>
<td>Good/fair</td>
<td>8</td>
<td>13.5</td>
<td>7.9</td>
<td>2/8</td>
</tr>
<tr>
<td>( P ) value</td>
<td>.39</td>
<td>.072</td>
<td>1.00</td>
<td>*.0047</td>
</tr>
</tbody>
</table>

CMS, Constant-Murley Score; OSS, Oxford Shoulder Score.

* Statistically significant (\( P < .05 \)).

Radiographs

Ten shoulders were assessed as Larsen grade IV, and 5 shoulders were Larsen grade V on the immediate preoperative radiograph (Fig. 2). Glenoid erosion could be appreciated on the latest postoperative radiographs of 5 shoulders (Fig. 3) but could not be measured accurately due to a lack of standardization of view. However, the amount of erosion was not...
estimated to be greater than 5 mm in any. Preoperative Larsen grade V or the presence of postoperative glenoid erosion was not associated with a poorer clinical outcome (Table III). Progressive superior migration of the humeral head, periprosthetic fracture, or implant loosening has not been encountered to date.

Discussion

This study is, by a small margin, the largest series with the longest follow-up published as a full manuscript and the first to report the results of RHA in patients with this condition. Levine et al presented a larger historical series, but it was published as an abstract in the proceedings of a meeting.

Thomas et al were the first to report the results of shoulder arthroplasty in this group, specifically using a stemmed hemiarthroplasty prosthesis in 9 shoulders. Compared with the present study, baseline patient characteristics were very similar, but mean follow-up was shorter (6 years vs. 10.4 years). Significant improvements in pain and CMS were noted, and a comparable proportion of patients achieved a very good or excellent outcome (4 of 9 shoulders), but 1 patient had a poor outcome. No prosthesis had required revision at last follow-up, but some stems were noted to be in varus, and the authors make clear their concern regarding lack of humeral bone stock should revision be required in the future for these young patients. These concerns would be mitigated by the use of resurfacing prosthesis.

Jolles et al subsequently published a larger series of stemmed prostheses (including 1 total shoulder replacement) in 13 shoulders, with a mean follow-up (9 years) approaching that of the present study. Significant improvements in postoperative pain score and range of motion were similarly seen. No shoulder had required implant revision or reoperation. Outcome scores were modest, but there was no preoperative comparison.

We corroborate the descriptions by Jolles et al and Thomas et al of the unique technical difficulties associated with shoulder arthroplasty in patients with end-stage JIA. Persistent arthritis results in a dysplastic proximal humerus and glenoid cavity with secondary degenerative change. Adduction and internal rotation contractures develop, resulting in painful dysfunction of the shoulder that affects basic daily activities such as axillary and perineal care. Significant soft tissue release is required for joint exposure and dislocation, and extensive subscapularis mobilization is usually necessary before repair to avoid limiting external rotation. The acromioclavicular joint is often approaching ankylosis and therefore likely to limit scapulothoracic motion unless addressed at the index.
operation. Jolles et al\textsuperscript{13} also report performing an open acromioplasty in 73% of shoulders at the time of prosthesis insertion. Although early symptoms did develop in 2 shoulders (14%) in this series that responded to arthroscopic acromioplasty, we do not feel this is necessary at the index procedure.

Unusual bony anatomy can be expected in most patients because of abnormal growth in addition to the disease process itself. These present specific challenges for stemmed hemiarthroplasty and total shoulder replacement. Glenoid replacement is precluded in most patients because of poor glenoid bone stock, the restrictive soft tissue envelope that would result in joint overstuffing, and concerns over longevity in these young patients. Cross-sectional imaging would be useful to determine those patients in whom glenoid bone stock might be sufficient to safely implant a glenoid component but was not used in this study. However, symptomatic progressive glenoid erosion has not been seen in any of the patients in JIA series to date.

Thomas et al\textsuperscript{14} noted difficulties with stem position and sizing due to a bowed metaphysis and proximal diaphysis as well as an often underappreciated mismatch between sagittal and coronal plane canal diameters. Jolles et al\textsuperscript{13} observed the high prevalence of significant osteopenia and cortical thinning. Accordingly, they did detect an intraoperative periprosthetic calcic fracture in 3 shoulders, although this did not change management or affect outcome. Multiple authors have raised concerns over the “vacant segment” stress riser between the tips of ipsilateral stemmed shoulder and elbow prostheses in patients with inflammatory arthritis (Fig. 1, b).\textsuperscript{11,17,20,22} A fracture here would be very difficult to treat and is therefore a perceived benefit of RHA.

We did not use RHA for patients with JIA at our institution before 2002 because a wide range of smaller sizes of the Copeland prosthesis were not available. RHA obviates concerns with diaphyseal shape, size, and bone stock but does present a different set of challenges. The presence of moderate or severe humeral head erosion was associated with worse outcome in the present study. Although none of the shoulders in this study exceeded the suggested 40% limit of humeral head destruction for use of the Copeland resurfacing prosthesis,\textsuperscript{16} the presence of significant erosion does make the procedure more complicated and necessitates an allograft in 3 shoulders.

The association of worse outcome with erosion may well reflect more advanced disease, because we did note a trend toward poorer preoperative OSS. Certainly, no radiographic evidence of failure of implant integration or progressive loosening was observed in these shoulders. We feel that undersizing of the central pilot hole is important in these patients because of the need to achieve good initial press-fit fixation of the tapered central peg. Bony ingrowth onto the hydroxyapatite coated undersurface occurs later. We recommend that an allograft be available for shoulders where preoperative imaging suggests significant erosion and that a stemmed prosthesis is always available in the operating theater as a backup option.

This study is limited by the small number of patients and lack of a control group with unoperated-on shoulders. The methodologic design is retrospective and therefore prone to bias, but the outcome data were collected at the time of patient consultation by observers independent of the operating surgeon. The same prosthesis was used throughout, and validated scoring methods were applied. Cross-sectional imaging was not performed before surgery for any of our patients because it did not change our surgical management. We do acknowledge that with the advent of stemless prostheses, some patients may be suitable for glenoid implantation without the need for a stemmed humeral implant. Another indication may be to assess humeral head erosion if a stemmed prosthesis is not available on the shelf. Cross-sectional imaging would also be useful in the postoperative period to assess glenoid erosion but was not available in this study. The lack of standardization of plain radiographs and, therefore, lack of precision in measurement of glenoid erosion is a weakness of the radiographic follow-up. However, the immediate postoperative and latest postoperative radiographs were at least comparable, allowing a broad assessment of glenoid erosion to be made.

**Conclusion**

Copeland surface RHA is a safe and effective intervention that significantly improves pain, range of motion, and function in the midterm for patients with end-stage arthritis of the shoulder due to JIA. The outcome is at least equivalent to that of stemmed hemiarthroplasty, with the added benefits of bone conservation, easier revision, and mitigation of periprosthetic fracture. Surgeons and patients should be prepared for the particular technical difficulty and poorer outcome associated with significant humeral head erosion.

**Disclaimer**

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**References**