Complex shoulder arthroplasty in patients with skeletal dysplasia can decrease pain and improve function

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**Background:** Patients with skeletal dysplasia are prone to the development of degenerative shoulder disease requiring shoulder arthroplasty at a younger age than in the general population. To date there have been no published reports on the complexities or outcome of shoulder arthroplasty in this unique patient group.

**Methods:** This is a review of 13 shoulder arthroplasties in 10 patients with skeletal dysplasia with mean follow-up of 7 years (2-17.6 years). There were 4 men and 6 women with a mean age of 53.1 years (23-76 years), mean height of 148 cm (122-177 cm), and mean weight of 60 kg (27-80 kg).

**Results:** The mean Oxford Shoulder Score increased from 13 (5-20) preoperatively to 28 (18-38) at final follow-up. Patients improved significantly in 2 of 8 Short Form 36 health-related quality of life domains: physical function (\(P = .04\)) and bodily pain (\(P = .04\)). Function was better in those who underwent non-constrained total shoulder arthroplasty as opposed to hemiarthroplasty. Four (31\%) required reoperation: 1 excision of heterotopic ossification, 1 relocation for anterior instability, and 2 revisions for periprosthetic fracture and glenoid erosion.

**Conclusion:** Shoulder arthroplasty is effective at relieving pain, optimizing movement, and improving function for patients with skeletal dysplasia; however, compared with the general population, there is a higher complication rate and function is not as good. Furthermore, this procedure is less effective at restoring health-related quality of life than total hip arthroplasty or total shoulder arthroplasty performed for osteoarthritis in the general population. Custom implants may be required to compensate for short stature and rotator cuff and glenoid deficiency.

**Level of evidence:** Level IV, Case Series, Treatment Study.

The skeletal dysplasias are a large, heterogeneous group of genetic disorders characterized by abnormal growth, development, and remodeling of the bones and cartilage that compose the human skeleton.\(^{20,36}\) The presence of multiple joint pathologic processes, abnormal alignment, generalized hypotonia, and ligament laxity predispose to degenerative joint disease at a young age.\(^5\) Hip and knee arthritis predominate; however, there is a significant incidence of shoulder disease, which tends to be bilateral and affects young patients.\(^{18,19}\) Thirty percent of patients with multiple epiphyseal dysplasia are affected by shoulder disease.\(^{18}\)
Early joint-preserving procedures include soft tissue repair, releases, débridement, decompensation, and corrective osteotomy to improve joint congruity, rotator cuff function, and stability. However, when painful osteoarthritis develops, shoulder arthroplasty may be indicated. Shoulder arthroplasty in these patients may be complicated by significant humeral and glenoid articular and metaphyseal angular deformity, bone deficiency, soft tissue and capsular contracture, hypotonia, and ligament laxity. There have been no reports on the status of the rotator cuff in patients with skeletal dysplasia.

Total hip arthroplasty (THA) and total knee arthroplasty have been shown to reduce pain and to improve function for patients with skeletal dysplasia; however, there have been no reports on the outcome of shoulder arthroplasty for such patients. We therefore determined function and health-related quality of life (HRQoL), radiographic findings, survival, and complications after shoulder arthroplasty for patients with skeletal dysplasia and compared these observations with those for shoulder arthroplasty in the general population.

**Methods**

Between 1996 and 2011, 10 patients with skeletal dysplasia and glenohumeral osteoarthritis were treated with either total shoulder arthroplasty (TSA) or hemiarthroplasty (HA). There were 4 men and 6 women with a mean age of 53.1 years (23-76 years), mean height of 148 cm (122-177 cm), and mean weight of 60 kg (27-80 kg) at the time of surgery (Table I). Three patients had staged bilateral procedures, leaving 13 shoulders available for review at a mean of 7 years (range, 2-17.6). Indications for arthroplasty were painful end-stage shoulder osteoarthritis affecting quality of life refractory to nonoperative treatment. Contraindications to the procedure included combined deltoid and rotator cuff insufficiency, infection, and patients with severe medical comorbidities that prevented general anesthesia. All patients had been referred to our institution with a mean duration of symptoms of 6.5 years (2-15 years) before surgery. This was due to prolonged nonoperative management at referring institutions.

We used the 2010 revision of the International Nosology and Classification of Genetic Skeletal Disorders to define skeletal dysplasia on the basis of clinical, radiographic, and molecular studies. Five patients had undergone previous THA; 5, total knee arthroplasty; 4, thoracolumbar surgery; 5, cervical surgery; and 1, elbow arthroplasty. One patient had undergone multiple previous shoulder operations for instability including anterior stabilization, débridement, decompensation, and contracture release before arthroplasty. The remaining patients had no history of shoulder surgery. Three patients had involvement of the ipsilateral elbow at the time of surgery.

 Patients were asked to subjectively evaluate their shoulder function. Responses were recorded as very satisfied, satisfied, not sure, or dissatisfied. Functional scoring was performed with the Short Form 36 (SF-36) HRQoL index and Oxford Shoulder Score (OSS). All remaining data were collected from clinic reviews, medical records, and radiographs. The SF-36 is a patient-completed health status measure designed for use in a broad range of patient populations and healthy subjects. It evaluates 8 dimensions of physical and mental health: physical functioning, physical role functioning, bodily pain, general health, vitality, social functioning, emotional role functioning, and mental health. Scores are coded, summed, and transformed onto a scale from 0 (worst possible health status) to 100 (best possible health status). Two summary scales, the physical and mental component summaries, are calculated. These are based on T-score transformations with a normative population mean of 50 and standard deviation of 10, based on American and European standards. The OSS is a patient-reported outcome measure specific to the shoulder that comprises 12 questions, each with 5 responses, to give a score between 0 (worst possible shoulder) and 48 (best possible shoulder). A higher score reflects better outcome.

Plain anteroposterior, axillary, and scapular Y-view radiographs were reviewed preoperatively, at 3 months, at 6 months, and annually thereafter. The most recent radiographs were evaluated by 2 independent reviewers and a consensus was reached. Radiographs of patients who had HA were reviewed to determine the presence or absence of glenohumeral subluxation, periarticular radiolucency, subsidence or a shift in the position of the humeral component, and glenoid erosion. Glenoid erosion was graded by the system of Sperling et al in relation to the anterior subchondral plate: mild (<5 mm), moderate (5-10 mm), or severe (>10 mm). Radiographs of patients who had TSA were reviewed for the presence of glenohumeral subluxation, periarticular radiolucency, and subsidence and migration or tilt of the glenoid component. Periarticular radiolucency was graded according to the system of Sperling et al; grade 0, no radiolucent line; grade 1, incomplete line ≤1 mm; grade 2, 1-mm complete line; grade 3, incomplete 1.5-mm line; grade 4, complete 1.5-mm line; and grade 5, 2-mm complete line. If radiolucent lines were present, all previous radiographs were compared for evidence of progression. Glenohumeral subluxation was graded as described by Sperling et al, evaluating the amount of humeral head translation relative to the center of the glenoid: none, mild (<25% translation), moderate (25%-50% translation), or severe (>50% translation).

**Statistical analysis**

This was performed with Stata/IC version 12.1 (StataCorp, College Station, TX, USA). Scores of the OSS were compared between groups by the unpaired t test for parametrically distributed data. The paired t test was used to compare SF-36 scores before and after surgery. A P value < .05 was considered significant.

**Surgical technique**

Preoperative cervical flexion and extension radiographs necessitated endoscopic intubation in several cases for cervical instability. All operations were performed by the senior authors (D.H., M.F., S.L., I.B.) through an extended deltopectoral approach in the reclining position under general anesthesia with interscalene regional block. The incision could be extended superiorly when access was difficult. The axillary nerve had a constant course in all cases, and was sought deep to the coracobrachialis and dissected to the quadrilateral space, where it was identified with a nerve stimulator. Inferior capsular release, progressing to circumferential capsulotomy, was an essential step to permit optimal visualization of the glenoid. The posterosuperior rotator
<table>
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<tr>
<th>Patient</th>
<th>Diagnosis</th>
<th>Side</th>
<th>Age (y)</th>
<th>Weight (kg)/height (cm)</th>
<th>Type of shoulder arthroplasty</th>
<th>Rotator cuff</th>
<th>Preoperative FF/Ab/ER</th>
<th>Postoperative FF/Ab/ER</th>
<th>Preoperative/postoperative OSS</th>
<th>Complications</th>
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<tr>
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<td>54</td>
<td>72/147</td>
<td>Unconstrained stemmed TSA</td>
<td>Intact</td>
<td>80/40/5</td>
<td>160/160/60</td>
<td>20/38</td>
<td>Superficial wound infection</td>
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<td></td>
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<td>55</td>
<td></td>
<td>Unconstrained stemmed TSA</td>
<td>Intact</td>
<td>80/50/5</td>
<td>170/160/60</td>
<td>16/38</td>
<td>RCT—SSp and ISp Glenoid erosion</td>
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<tr>
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<td>65</td>
<td>58/147</td>
<td>Stemmed hemi</td>
<td>Intact</td>
<td>70/70/10</td>
<td>70/65/10</td>
<td>11/24</td>
<td>Long head of biceps tendinitis</td>
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<td></td>
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<td>Right</td>
<td>55</td>
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<td>Linked custom TSA</td>
<td>SSp, ISp, and SSc tears</td>
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<td>Unconstrained resurfacing TSA</td>
<td>Fixed anterior dislocation with deficient SSc, SSp, and ISp</td>
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<td>70/70/40</td>
<td>10/24</td>
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<td>47/123</td>
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<td>105/90/10</td>
<td>16/35</td>
<td>TIA and UTI</td>
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<td>23</td>
<td>55/177</td>
<td>Linked custom TSA</td>
<td>Deficient SSc, SSp, and ISp</td>
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<td>45/40/0</td>
<td>10/29</td>
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<td>Fixed anterior dislocation with deficient SSc, SSp, and ISp</td>
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<td>20/10/0</td>
<td>7/25</td>
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<td>9</td>
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<td>80/153</td>
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<td>Intact</td>
<td>30/25/0</td>
<td>130/90/40</td>
<td>11/33</td>
<td>Anterior subluxation requiring EUA and Botox</td>
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<tr>
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<td>54/144</td>
<td>Resurfacing hemi</td>
<td>Intact</td>
<td>80/70/5</td>
<td>45/45/10</td>
<td>15/18</td>
<td>RCT and glenoid erosion Revision when medically fit</td>
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<tr>
<td></td>
<td></td>
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<td>72</td>
<td></td>
<td>Resurfacing hemi</td>
<td>Intact</td>
<td>70/70/10</td>
<td>45/45/20</td>
<td>18/24</td>
<td>Revision for RCT and glenoid erosion</td>
</tr>
</tbody>
</table>

ASL, aseptic loosening; EUA, examination under anesthesia; FF/Ab/ER, forward flexion/abduction/external rotation; ISp, infraspinatus; OSS, Oxford Shoulder Score; SSc, subscapularis; SSp, supraspinatus; TIA, transient ischemic attack; TSA, total shoulder arthroplasty; RCT, rotator cuff tear; UTI, urinary tract infection.
cuff was deficient in 4 shoulders, including both patients with fixed anterior dislocations; all tears were considered irreparable and non-reconstructable. Six humeri had a varus metaphyseal-diaphyseal angular deformity. The glenoid was dysplastic in all shoulders. Eight shoulders were judged to have clinically relevant glenoid rim bone loss in addition to the dysplasia. Mean glenoid rim bone loss was 16% (0%-60%). This was judged intraoperatively by the surgeon’s calculating percentage glenoid rim deficiency relative to glenoid circumference. Four shoulders had a glenoid vault depth of <15 mm as defined by Edwards et al. on the basis of preoperative computed tomography scan or radiograph (Fig. 1). When access was difficult, sequential partial pectoralis major tenotomy, coracoplasty (7 shoulders), and partial deltoid release were performed. Subscapularis tenotomy gained access to the shoulder and was repaired in 2 layers whenever possible with Ethibond. Wounds were closed in layers, and a drain was inserted.

Five unconstrained TSAs, 4 linked (constrained) TSAs, and 4 HAs (2 stemmed and 2 resurfacing implants) were implanted. The 5 unconstrained TSAs consisted of 3 resurfacing (Epoca; DePuy Synthes, Solothurn, Switzerland) and 2 stemmed implants (BioMet Merck, Bridgend, Wales, UK). The 4 linked (constrained) TSAs consisted of 3 custom and 1 noncustom reverse-type device (Stanmore Implants Ltd, Elstree, Stanmore, UK). Nine humeral components were uncemented; 4 were cemented. Indications for a resurfacing TSA or HA were (1) humeral head and glenoid bone stock of sufficient volume and quality for primary implantation of a standard prosthesis and (2) anatomically intact rotator cuff (Fig. 2). Indications for a stemmed unconstrained TSA or HA were (1) inadequate humeral epi-metaphyseal bone to support a proximal loading implant, (2) adequate glenoid bone to support an implant, and (3) intact cuff.

The indication for a linked TSA was a deficient rotator cuff. A reverse-type prosthesis (noncustom linked TSA) was used when there was adequate glenoid bone to support an implant. A custom linked device was used in the presence of rotator cuff deficiency and inadequate glenoid bone (defined as erosion medial to the coracoid with a vault depth <15 mm and >25% glenoid rim bone loss) to enable implantation of a standard glenosphere or custom inset component without augmentation with autologous or allograft bone (Fig. 3). The custom glenoid component consisted of a titanium hydroxyapatite-coated shell that is captured between the undersurface of the acromion, the deep surface of the coracoid, and the glenoid. Four screws (3.5- or 4.5-mm cortical; DePuy Synthes, Solothurn, Switzerland) achieve primary fixation into the lateral column of the scapula, base of coracoid, and body and spine of scapula. Secondary fixation relies on bone or stable fibrous ingrowth to the hydroxyapatite-coated shell. A polyethylene liner is cemented into the glenoid shell that provides a semi-constrained articulation with the humeral head.

Postoperative management

Prophylactic intravenous administration of cefuroxime was given at induction and for 2 postoperative doses. The operative side was placed in a polysling for 6 weeks. For those with linked prostheses, eccentric deltoid, active-assisted external rotation, abduction, and forward elevation to shoulder level were commenced on day 1. For those with nonconstrained devices, external rotation was restricted for 6 weeks.

Results

Function

All patients reported an improvement in pain and function at final follow-up (Table I). Subjectively, 5 patients were very satisfied, 4 were satisfied, and 1 was dissatisfied. The dissatisfied patient had HA with pain from glenoid erosion. Subjective improvements in activities of daily living are shown in Table II. Mean OSS improved from 13 (5-20) preoperatively to 28 (18-38) at final follow-up ($P < .001$). Mean range of active forward elevation and abduction improved from $54^\circ$ ($5^\circ$-$110^\circ$) and $43^\circ$ ($5^\circ$-$90^\circ$) to $84^\circ$ ($20^\circ$-$170^\circ$) and $76^\circ$ ($10^\circ$-$160^\circ$), respectively. Mean external rotation improved from $7^\circ$ ($0^\circ$-$30^\circ$) to $24^\circ$ ($0^\circ$-$60^\circ$). Subgroup analysis demonstrated mean postoperative OSS in patients who underwent TSA and HA of 30 (18-38) and 24 (18-29), respectively. Patients who underwent unconstrained TSA (5 patients; mean OSS, 34) had clinically and statistically significant better function (mean difference in OSS between groups, 10; $P = .027$) than those who underwent HA (4 patients; mean OSS, 24).

HRQoL

Figure 4 shows the SF-36 scores before and after shoulder arthroplasty. Of the 8 SF-36 dimensions, patients had statistically significant improvements in 2 domains: physical functioning ($P = .04$) and bodily pain ($P = .04$).

Radiographic analysis

Total shoulder arthroplasty

A nonconstrained TSA was implanted in 5 shoulders. Glenohumeral subluxation was present in 2 (1 mild posterosuperior and 1 mild anterior subluxation). There was a grade 1 incomplete radiolucent line adjacent to the medial aspect of a stemmed, cemented humeral component in 1 shoulder. There was a grade 2 complete 1-mm radiolucent line around a metal-backed glenoid component. There was no shift or subsidence of any component.

A constrained TSA was implanted in 4 shoulders. A grade 5 complete 2-mm radiolucent line was seen around a custom glenoid component in 1 patient who sustained a periprosthetic scapula fracture associated with component subsidence. This patient required revision. No radiolucent lines were present around the humeral components.

Hemiarthroplasty

HA was performed in 4 shoulders. All developed glenohumeral subluxation (2 moderate posterosuperior, 1 moderate superior, and 1 mild superior subluxation). Glenoid erosion was also present in all 4 (2 severe, 2 moderate). There were 2 grade 1 incomplete radiolucent lines adjacent to 2 humeral components, 1 adjacent to a stemmed,
cemented humeral component with medial stress shielding and 1 beneath the superolateral region of an uncemented resurfacing replacement. Both were nonprogressive and not associated with component subsidence.

Revisions and complications

Reoperation rate was 31%. There were 2 revisions (15%) at a mean time of 55 months (18-93 months). One patient with a resurfacing HA required revision to a custom linked TSA for pain secondary to severe glenoid erosion. The other patient required revision at 18 months for aseptic loosening of a custom glenoid component caused by a persisting scapula spine periprosthetic fracture nonunion. This patient has subsequently gained pain-free daily function at waist level. One patient required excision of heterotopic ossification at 27 months for stiffness. One patient, who underwent an unconstrained TSA for instability arthropathy, developed atraumatic anterior prosthetic subluxation at 12 months. The patient underwent examination under anesthesia and botulinum neurotoxin type A injection into an overactive pectoralis major muscle. Further intervention is not planned.

There were 3 posterosuperior rotator cuff tears, all in shoulders with HA and glenoid erosion; 1 patient underwent revision, 1 is awaiting revision when medically fit, and the other patient has acceptable function (OSS 24) and pain and is being managed nonoperatively. One patient developed long head of biceps tendinitis that resolved with image-guided steroid injection. There was 1 superficial wound and 1 urinary tract infection; both resolved with oral antibiotics. One patient had a transient ischemic attack in the early postoperative period; the patient recovered with no residual neurologic deficit.

Discussion

This study has shown that shoulder arthroplasty in patients with skeletal dysplasia can decrease pain, improve function, and 2 of 8 SF-36 HRQoL domains. The improvement in shoulder function and pain as measured by the OSS was
both clinically and statistically significant. The minimal clinically important difference for the OSS has been suggested to be 4.5 points, which is approximately 10% of the total score.

Compared with patients undergoing shoulder arthroplasty in other diagnostic groups (osteoarthritis, rheumatoid arthritis, acute fracture, and fracture sequelae), baseline preoperative scores of the OSS were lower in our study population. Final postoperative scores were also lower than in osteoarthritis or rheumatoid arthritis but comparable to those after fracture sequelae. Patients with fracture sequelae tend to do worse than other diagnostic groups as these shoulders are often very badly damaged with respect to both the joint and rotator cuff. Multiple joint pathologic processes, abnormal glenoid morphology, and rotator cuff dysfunction were features of patients in this study, which likely explains the relatively low preoperative and postoperative scores of the OSS. However, the relative improvement in OSS after shoulder arthroplasty (mean, 15) was similar to other diagnostic groups.

We found significant HRQoL improvements in 2 domains (physical functioning, bodily pain). There was no improvement in the other 6 domains: physical role functioning, general health, vitality, social functioning, emotional role functioning, and mental health. The general health perception score diminished by 3 points postoperatively. This apparent paradox has also been reported in the THA literature. Some of this effect is due to the fact that for control populations, general health perception scores diminish steadily with age for patients aged 50 to 70 years (0.8 per year for women and 0.5 per year for men).

Figure 3 Features of a patient with multiple epiphyseal dysplasia in whom a custom TSA was indicated. Preoperative anteroposterior (A) and axillary (B) radiographs and axial computed tomography scan (C) of the shoulder demonstrate diaphyseal bone deformity, glenoid erosion medial to the coracoid, and glenohumeral osteoarthritis. Postoperative anteroposterior (D) and lateral (E) radiographs show satisfactory position of a custom TSA. The glenoid shell articulates with the acromion, deep surface of coracoid, and glenoid. Four screws achieve primary fixation into the lateral column, base of coracoid, and body and spine of scapula. A polyethylene liner is cemented into the glenoid shell and provides a semiconstrained articulation with the humeral head.

<table>
<thead>
<tr>
<th>Activity of daily living</th>
<th>Ability to perform task preoperatively (n = 10)</th>
<th>Ability to perform task postoperatively (n = 10)</th>
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<tbody>
<tr>
<td>Eating</td>
<td>3</td>
<td>8</td>
</tr>
<tr>
<td>Hair brushing</td>
<td>0</td>
<td>7</td>
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<tr>
<td>Dressing</td>
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<td>8</td>
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<td>Shopping</td>
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<td>Personal hygiene activities</td>
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<td>Overhead activities</td>
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</table>
Complex shoulder arthroplasty in dysplastic patients

The management of glenoid bone deficiency for patients with severe dysplasia is a significant challenge. In this study, 8 shoulders had clinically relevant glenoid rim bone deficiency (mean loss of 16% judged intraoperatively), and 4 shoulders had a glenoid vault depth of <15 mm.9 Reconstructive options include bone grafting of the deficient region in conjunction with glenoid component placement, glenoid osteotomy or bone grafting in conjunction with HA, and accommodation of the deficiency with a glenoid component that substitutes metal for deficient bone. Historically, severe glenoid bone deficiencies were treated with glenoplasty, bulk bone grafts, and standard glenoid components18,34; however, this was associated with a high rate of unsatisfactory results (47%) and revision (29%).17 Many surgeons prefer to avoid structural bone grafts and perform HA instead of glenoid resurfacing. Recent reports have shown worse functional outcome with HA than with TSA in patients with glenoid bone deficiency and a nonconcentric glenoid and cautioned against performing HA in this setting.12,21,34

Custom glenoid devices have been used for patients with deficient glenoid bone.11,13 Gunther and Lynch13 reported good functional results and no complications in 7 patients at 4.3 years with use of a custom-inset glenoid implant. There is no consensus on the optimal treatment for deficient glenoid bone in TSA; multiple reports would suggest that TSA without version correction can provide good to excellent results in patients with congenital deficiency so long as there is sufficient glenoid vault to support the keel or pegs of a prosthesis.9,15,17,26 This was not always the case with patients in our study. When bone was eroded medial to the coracoid with >25% glenoid rim bone loss and a vault depth <15 mm, we thought the glenoid would not support a standard glenoid component. In this situation, we implanted a custom glenoid shell that articulated with the remaining glenoid and acromion, supported by screw fixation into the scapula and reliant on secondary bone ingrowth. This provided reliable pain relief and satisfactory function; however, long-term durability is not known. Historically, linked devices have been associated with high early complication (range, 8%-100%) and revision (range, 4%-54%) rates.2,38 A reverse shoulder arthroplasty combined with structural glenoid bone grafting is an alternative option to manage combined cuff and glenoid deficiency.25,27,29 Neyton et al27 described 9 patients who underwent, in 1 or 2 stages, reverse arthroplasty and glenoid bone grafting. Most patients were satisfied with their result because of pain relief. This remains a viable option to address combined cuff and glenoid deficiency.

We found that function after unconstrained TSA (5 patients; mean OSS, 34) was superior to that after HA (4 patients; mean OSS, 24) in those with an intact cuff and glenoid (mean difference in OSS between groups, 10; \( P = .027 \)). We preferentially implant a resurfacing TSA when bone stock permits. This has the benefit of preserving bone, facilitating revision, and reducing risk of humeral shaft fracture or stem perforation when there is angular deformity in the humerus (present in 6 humeri in this study). We found that management of humeral angular deformity is best achieved by a metaphyseal loading or resurfacing implant. When the metaphysis is not supportive, we recommend a custom device that accommodates the deformity on the basis of its success in the knee for patients.

Compared with age- and sex-matched controls, the preoperative self-assessed health status of patients with skeletal dysplasia undergoing shoulder arthroplasty was less in several domains (physical functioning, social functioning, physical role functioning, mental health, bodily pain, general health).28 With the exception of general health and physical role functioning, these improved after surgery; however, they did not reach the same levels found in the general population. Furthermore, TSA in patients with skeletal dysplasia did not restore the health status to the same levels seen after THA or TSA performed for osteoarthritis in the general population.1,4,16 Thus TSA in patients with skeletal dysplasia is less effective at improving HRQoL than are other types of joint replacement (total hip replacement, TSA) for the general population. Patients in this study are likely to be compromised by arthritis in multiple joints (3 had ipsilateral elbow involvement); thus, despite procedural benefits, overall health status is incompletely restored.

The management of glenoid bone deficiency for patients with congenital deformity is best achieved by a metaphyseal loading or resurfacing implant. When the metaphysis is not supportive, we recommend a custom device that accommodates the deformity on the basis of its success in the knee for patients.

Figure 4 Preoperative and postoperative SF-36 scores for the 10 patients in the study with 95% confidence intervals. There was a significant improvement in physical functioning and bodily pain. BP, bodily pain; GH, general health; MCS, mental component summary; MH, mental health; PCS, physical component summary; PF, physical functioning; RE, role—emotional; RP, role—physical; SF, social functioning; VT, vitality.
with skeletal dysplasia\textsuperscript{30} and experience from other scenarios in complex shoulder arthroplasty.\textsuperscript{32}

In this study, there was a higher rate of complications compared with shoulder arthroplasty in the general population, which usually has complication rates ranging from 10\% to 16\%.\textsuperscript{2} The revision rate was 15\% at 7 years, 1 for glenoid erosion in a patient with a resurfacing HA and 1 for glenoid component loosening secondary to a periprosthetic scapula spine fracture. One additional patient with a resurfacing HA is awaiting revision for glenoid erosion when medically fit. This rate is higher than would be expected for shoulder arthroplasty in the general population but comparable to that for patients undergoing shoulder arthroplasty for primary glenoid dysplasia.\textsuperscript{17,34}

There are limitations to this study: it is retrospective with small patient numbers and no randomization, making it prone to confounding and bias. Heterogeneity in prostheses used, temporal effects related to the long study period, and variable length of follow-up will introduce measurement and recall bias. Confounding from multiple joint pathologic processes and medical comorbidities will affect functional outcome. Given the paucity of published information on shoulder arthroplasty for patients with skeletal dysplasia, the study methodology is reasonable.

**Conclusion**

We have shown that shoulder arthroplasty can improve pain, function, and 2 of 8 SF-36 quality of life domains. Compared with the general population, function is not as good and revision rates are higher. Furthermore, this procedure is less effective at restoring health status than THA or TSA performed for osteoarthritis in the general population. In those with an intact cuff and glenoid, functional outcome appears superior when nonconstrained TSA is used over HA, although low patient numbers prevent accurate comparison. Linked devices are required for cuff deficiency. In those with a deficient cuff and glenoid, custom devices or reverse prostheses with structural glenoid bone grafting would seem appropriate. We conclude that shoulder arthroplasty is a viable treatment option for glenohumeral osteoarthritis in patients with skeletal dysplasia.

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