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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.

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Keywords: Quality of life; shoulder arthroplasty; dysplasia

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

No IRB/ethical approval was required for this study.

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Early joint-preserving procedures include soft tissue repair, releases, débridement, decompression, and corrective osteotomy to improve joint congruity, rotator cuff function, and stability.³³ 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.^{7,18,22,35} 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^{6,14,24,30,31}; 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, decompression, 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 index²⁴ and Oxford Shoulder Score (OSS).⁸ 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, periprosthetic 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 al³⁴ 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, periprosthetic radiolucency, and subsidence and migration or tilt of the glenoid component. Periprosthetic radiolucency was graded according to the system of Sperling et al³⁴: 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,³⁴ 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 shoulders 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

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