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Case report

REVERSE SHOULDER ARTHROPLASTY IN YOUNG PATIENT WITH ACHONDROPLASIA - TEN YEAR FOLLOW UP: CASE REPORT

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Abstract

Skeletal dysplasia in Achondroplasia can affect all body joints - including the glenohumeral joint- and is prone to develop to degenerative osteoarthritis (OA). This may cause pain and mobility problems at young age. Surgical treatment is challenging due to the dysplastic anatomy of the shoulder joint - with a dysplastic deformed short humerus, a small, hypoplastic medialized glenoid and lateralized acromion - andthe long life expectancy of these patients. The indications for Reverse Shoulder Arthroplasty (RSA) evolved during years with rotator cuff tears and rotator cuff arthropathy in combination with or

withoutglenohumeral OA as the main indicator, with good short to mid-term results.Long term results of RSA are rarely found in literature, especially in young patients.The use of a RSA in glenohumeral OA with an intact rotator cuff has rarely been reported. In this case report we present the ten year clinical and radiographic results of a RSA for the treatment of degenerative OA with glenohumeral dysplasia in a young patient with achondroplasia.

KEYWORDS: achondroplasia, dysplasia, reverse shoulder arthroplasty

CASE REPORT

A33-year-oldwomanwas diagnosed with Achondroplasia at young age. She did light manual work, practiced no sport and underwent previous prosthetic surgery for knee and hip OA because of destructive joint disease. She was complaining of right shoulder pain for 5 years and was treated conservatively by pain medication and steroidal injections for many years. Due to the increasing pain -limiting her daily activities-, decreased range of motion and failing of the conservative treatment, the decision to perform a shoulder arthroplasty was made. On clinical examination, palpation and mobilization of the glenohumeral joint was painful and range of motion was decreased. Preoperative, a Constant score of 30 and an Oxford shoulder score of 48 were reported. Passive elevation was 90 degrees and active anterior and lateral elevation 60 and 40 degrees respectively. Passive and active external rotation at all levels were zero degrees. There were no signs of rotator cuff deficiency or impingement. A normal neurovascular examination was reported. Preoperative radiographs (Fig 1) and Computed Tomography (CT) scan (Fig2) demonstrated a dysplastic deformed short humerus and a small, hypoplasticmedialized glenoid leading to degenerative glenohumeral OA.The humeral head was absent and the medialcalcar was projecting more medial and far below the inferior rim of the glenoid. The acromion showed a lateralized position. On CT scan, the subscapularis and supraspinatus muscle showed grade 3 fatty infiltration.

Due to this destructive glenoid and the low humeral position in relationship to the glenoid in combination withimportant fatty infiltration of the rotator cuff, a standard reverse shoulder prosthesis (TESS, Biomet Inc, Warshaw, IN) was implanted using a deltopectoral approach

under general anesthesia and interscalene block. At the time of surgery, no bio-RSA or lateralized implants were available, which could have been appropriate for this indication.

The humeral components consist of a standard 6*100mm uncemented humeral stem and reversed Corolla size zero. The glenoid was replaced by a standard size zero glenoid base plate, fixated with 2 screws (35mm and 40mm)with a reversed size zero, 36 mmdiameter head. A size 0 – 36 mm diameter – 6mm thick reversed polyethylene insert was used.Peroperatively, the supraspinatus and subscapularis tendon showed a degree of atrophy without signs of a tear. Atenodese for the long biceps tendon was performed. Subscapular opening was transtendinous and could easily be closed with tendon to tendon sutures. The supraspinatus tendon was resected. No peroperative complications were reported and the patient could be discharged from the hospital after 3 days without any problem. Postoperative, a shoulder sling with abduction pillow was applied for 6 weeks. Immediately after surgery, passive mobilization and Codman pendulum exercises were assessed for the duration of 3 weeks, followed by progressive active mobilization.

One year postoperative, the Constant score increased to 59 points and Oxford score decreased to 21 points. The patient was very satisfied of the clinical result. Passive elevation was increased to 130 degrees and active anterior and lateral elevation increased to 110 and 60 degrees respectively. Passive and active external rotation at all levels were still zero degrees. Radiographs one year postoperative were unchanged and showed excellent component position without signs of loosening. (Fig 3)

At ten year follow up Constant score and Oxford score even improved to respectively 64 and 16 points. Passive and active anterior elevation remained the same at respectively 130 and 110. However, lateral elevation improved to 90 degrees. Passive and active external rotationsat all levels were improved as well to 40 degrees. After ten years, radiographs

showed excellent component position without any sign of notching. (Fig 4) A small zone of radiolucency could be seen on the medial calcar of the humerus and around the humeral stem.This could be caused by slight osteolysis due to polyethylene wear. A bony spur could be seen on the inferior part of the glenoid, without clinical reference. An additional CT scan showed excellent glenoid and stem position. At ten years follow-up, the patient was still very satisfied with the clinical result and she would have chosen the operation again.Table 1 gives an illustration of the preoperative and postoperative Constant score and Oxford score and the mobility.

DISCUSSION

Achondroplasia is a common form of skeletal dysplasia. It is also known as *short limb dysplasia* because patients with this condition have an average sized torso (chest and abdomen) but smaller limbs.About one in 20000 babies are affected. On radiograph, almost all bones and joints of the skeleton are affected, including the scapula and the humerus. The latter is particularly shortened and can show metaphyseal flaring. [1]Patients with skeletal dysplasia are prone to the development of degenerative osteoarthritis requiring arthroplasty at younger ages than general.[2] Recently, Sewell et al published good clinical and radiographic results usingTotal Shoulder Arthroplasties and Hemiarthroplasties in ten patients (mean age 53.1 year) affected with skeletal dysplasia with a mean follow-up of 7 years.[3]However, the use of a RSA in the treatment of skeletal dysplasia is not reported in literature so far.

The results of implanting a RSA in primary glenohumeral OA in patients due to a dysplastic biconcave glenoid (Type B2)with a normal and functional rotator cuff werereported by Mizuno et al. He showed excellent results using a RSA in these indications at a mean follow-up of 54 months. [4] Increased Constant scores, improvement of mobility and low complication rate were reported in patients with a mean age of 74.1 year.

In general, the ideal surgical management of glenohumeral OA, resistant to non surgical treatment in patients younger than 55 is still not well defined[5], although a Total Shoulder Arthroplasty is recommended over hemiarthroplasty[6], but most authors do not recommend implanting a RSA in patients less than 65 years, due to the possible complications[7] and high revision rate. [8]

In this case,non surgical treatment was insufficient. Due to the destructed glenoid, the dysplastic anatomy of the glenohumeral joint with low position of the humerus according to the glenoid and moderate fatty infiltration of the rotator cuff, a RSA was the option of choice. Although the fact that the results of RSA in glenohumeral osteoarthritis with intact rotator cuff have rarely been reported, the RSA has shown its profit in several other indications as rotator cuff tears and rotator cuff arthropathy in short to mid-term follow up.[7,9-12]

At long term follow up, a large long term study, Favard et al found survivorship free of revision of 89% at 10 years. [13] But after 8 years the functional results started to decline. This trend was seen by Gurey's results as well, with a deterioration in function and increasing pain 6 years after RSA. [14] Deltoid fatigue is one of the bigger concerns in reversed shoulder arthroplasty at longer-term follow- up. [13]

However, in this case, nodeterioration of function or strength was observed at ten years follow up, despite the veryyoung age at the time of implantation.In addition, external rotation increased from 0 degrees preoperatively to 40 degrees at one year follow up and this was maintained for ten years. This improvement can be explained by the fact that the external rotation was passively and actively very limited preoperatively due to stiffness and disuse.Post surgical exercises and physiotherapy increased the total range of motion of the shoulder, including external rotation.Grafting of the medialized glenoid to lateralize the glenoid can increase external rotation, as well as the use of lateralized implants. However, grafting wasn't performed and lateralized implants were not available at time of surgery.

Improvements in active external rotation with the use of TSA were also found in the study of Sewell et al. He reported an increase in external rotation with 17 degrees. [3] Flurin et al demonstrated a significant improvement of external rotation postoperatively in TSA and RSA, respectively with 36.1 and 28.0 degrees.[15] Similarly, Latif et al found greater postoperative external rotation after TSA compared with RSA, respectively 43 and 12 degrees. [16]

The radiolucencies at the the humeral metaphyseal region should be closely followed as they may be an early radiographic sign of polyethylene wear.A long term follow-up study reported polyethylene wear as the cause or radiolucencies at the level of the humeral stem. The progression of notching was variable. [17]

TABLE

Table 1. Preoperative and postoperative mobility.

		Preoperative	Oneyearpostoperative	Ten yearspostoperative
	Constant Score (100 p)	30	59	64
	Oxford Score (60 p)	48	21	16
Passive	elevation	90	130	130
	externalrotation	0	0	40
Active	elevationanterior	60	110	110
	elevationlateral	40	60	90
	externalrotation at side	0	0	40
	externalrotation at 90°	0	0	40

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CONFLICT OF INTEREST DICLOSURE STATEMENT

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FIGURES LEGENDS

Fig 1. Preoperative radiographs show dysplastic deformation of glenohumeral joint with low and medial position of the humeral calcar. There was a lateralized acromion.

Fig 2. Preoperative CT scan shows dysplastic deformation of glenohumeral joint.

Fig 3. Radiographs, one year postoperative show excellent component position without signs of loosening.

Fig 4.Radiographs, ten years postoperative still show excellent component position without signs of notching. There was some radiolucency to be seen around the humeral component (white arrows). A bony spur could be seen on the inferior part of the glenoid (black arrow).