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Case report

## Anatomically-designed shoulder hemiarthroplasty made after 3-D models and implanted in a child with rheumatoid arthritis. A case report

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**SUMMARY.** *Introduction.* Shoulder arthroplasty is unusual at an early age. Diseases such as juvenile rheumatoid arthritis, however, warrant this kind of treatment because the shoulder pain and limited range of motion significantly compromise function. *Case report.* Male patient, 14 years old, diagnosed as having juvenile rheumatoid arthritis, with a deforming and disabling history, and broad glenohumeral destruction confirmed by X-rays. A prosthesis was designed after the natural anatomy of the patient according to preoperative planning using a 3-D anatomical model based on CT slices. Left shoulder hemiarthroplasty was performed with functional and pain improvement according to the UCLA score.

**Key words:** arthroplasty, shoulder, child, rheumatoid arthritis.

**RESUMEN.** *Introducción.* La artroplastía del hombro suele ser rara a temprana edad, sin embargo, patologías como la artritis reumatoide juvenil, en que el dolor del hombro y la limitación de movimiento comprometen importantemente la función, posibilitan la práctica de esta forma de tratamiento. *Reporte de caso:* Paciente masculino de 14 años con diagnóstico de artritis reumatoide juvenil, quien contaba con una evolución deformante e incapacitante, con amplia destrucción glenohumeral corroborada en la imagen radiográfica. Se diseñó una prótesis a la medida del paciente de acuerdo a planeación preoperatoria con apoyo de un modelo tridimensional anatómico basado en cortes tomográficos. Se practicó hemiarthroplastía de hombro izquierdo con mejoría funcional y del dolor según la escala de UCLA.

**Palabras clave:** artroplastía, hombro, niño, artritis reumatoide.

### Introduction

The first known report of shoulder arthroplasty is that by Pean,<sup>1</sup> a French surgeon who replaced a joint destroyed by tuberculosis with a prosthesis made of platinum and plastics. In 1951, and mostly in 1974, Neer became the strongest advocate of this practice by designing his own

systems. In the 1990s, knowledge on the kinematics and recognizing the significance of adjacent soft tissues involved in shoulder function, led to technique improvements having a significant impact on the outcome.

Shoulder arthroplasty is unusual at an early age. Diseases such as juvenile rheumatoid arthritis (JRA), however, warrant this treatment modality as shoulder pain and a limited range of motion significantly compromise function.<sup>2</sup> The main problem is that this disease leads to bone loss that usually occurs<sup>3</sup> together with soft tissue incompetence and muscular atrophy.<sup>4-9</sup>

The purpose of this paper is to present the case of a patient who underwent shoulder hemiarthroplasty due to JRA sequelae, and to review the literature about it.

### A case report

Male, 14-year-old patient, with a 6-year history of juvenile rheumatoid arthritis (JRA), who received several

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therapies beginning with anti-inflammatory agents and followed by multiple infiltrations. Multiple musculoskeletal conditions occurred due to the disease, including bilateral hip and wrist lesions, which had previously undergone arthrodesis. Both shoulders showed severe functional limitation with 15° of abduction, 10° of flexion, 10° of extension, and 15° of external rotation. According to the UCLA classification score (Table 1), the patient

had a score of 7, indicating poor function. The pain visual analog scale (VAS) indicated 9/10 (severe pain). X-rays revealed a severe panarticular destruction pattern and evidenced closure of the physis (Figure 1), which was confirmed by MRI.

The decision was made to place a hemiprosthesis in the left shoulder. The prosthesis was designed after the natural anatomy of the patient under the supervision of the bioengineering department of the orthopedics group in our hospital. A three-dimensional model was used for the surgery planning and optimum prosthesis manufacturing.

**Planning and manufacturing the prosthesis:** a functional anatomical tracking was made to detect bone and muscle deficiencies, which are very common in patients with JRA. Special care was given to the assessment of the glenoid cavity, as this structure is prognostic of proper joint performance. Patients with JRA usually have central erosions of the coracoid base and glenohumeral instability. Anteroposterior, lateral and true AP X-rays were therefore taken, showing the mismatch between the humeral head and the glenoid cavity. The use of computed tomography with slices at 3 mm intervals provided a more specific assessment of the bone quality of the joint. This allowed for the creation of a three-dimensional model made with clay to evaluate the need for bone grafts and to effectively plan the surgery. MRI was useful in evaluating the soft structures of the shoulder, especially the integrity of the rotator cuff, the deltoid muscle, and the presence of avascular necrosis of the humeral head.

Table 1. Functional and patient satisfaction evaluation score by UCLA	
<b>Pain</b>	
1. Always present and unbearable. It is usually strong even with medication 2. Always present but bearable with medication; occasionally strong 4. No or little pain at rest; present with light activities, and patient often takes salicylates 6. Frequent pain only with heavy activities 8. Occasional and mild pain 10. No pain	
<b>Function</b>	
1. Inability to use the limb 2. Function with light activities 4. Capable of performing light housework or activities of daily living 6. Housework, shopping, and driving, if possible; capable of grooming his/her hair, getting dressed and undressed, including wearing a bra 8. Only slight restriction. Capable of working at a level above the shoulder 10. Normal activities	
<b>Ventral active flexion</b>	
5. More than 150 degrees 4. 120 to 150 degrees 3. 90 to 120 degrees 2. 45 to 90 degrees 1. 30 to 45 degrees 0. Less than 30 degrees	
<b>Strength with active flexion</b>	
5. Normal or grade 5 4. Good or grade 4 3. Average or grade 3 2. Poor or grade 2 1. Perceptible muscle contracture 0. No contraction is perceived	
<b>Patient satisfaction</b>	
5. Satisfied and better 0. Dissatisfied and worse than before the surgery	
<b>Outcome evaluation</b>	
Excellent: 34 to 35 points Good: 28 to 33 points Average: 21 to 27 points Poor: 0 to 20 points	



**Figure 1.** Anteroposterior shoulder X-ray showing the significant involvement of the glenohumeral joint under Bombelli's osteoarthritis criteria.

The anatomical model was designed by the Dicom software system, capable of scanning topographic slices at 3 mm intervals and turning them into 3-D anatomical models made with sandy clay at a 1:1 scale with a minimum error margin (0.005) (*Figure 2*). With the data from this software system and the 3-D anatomical model, the necessary measures were taken to manufacture the prosthesis designed after the patient's natural anatomy, following the Neer II humeral component bases, with a titanium, aluminum and vanadium alloy.

The surgical technique described by Neer<sup>10-13</sup> was used to place the prosthesis. Notice that due to their prognostic relevance, muscles that compose the rotator cuff were in good status, allowing for the implantation of the hemiprosthesis.<sup>5,6,14,15</sup>

The patient has done quite favorably. At six months, the procedure has a 0 rating on the pain Visual Analog Scale. The UCLA functional score shows a 24-point improvement, which constitutes average function especially in terms of the pain and satisfaction parameters (*Table 2*).

The shoulder improved its ranges of motion achieving a 15° external rotation, 50° of abduction, 75° of flexion, and 75° of internal rotation. At each monthly visit we have seen good glenohumeral stability evidenced by dislocating maneuvers. This allows learning about the appropriate prosthetic length and soft tissue preservation. Postoperative control X-rays show an appropriate stem-cement-bone relationship with no evidence of loosening (*Figure 3*).

## Discussion

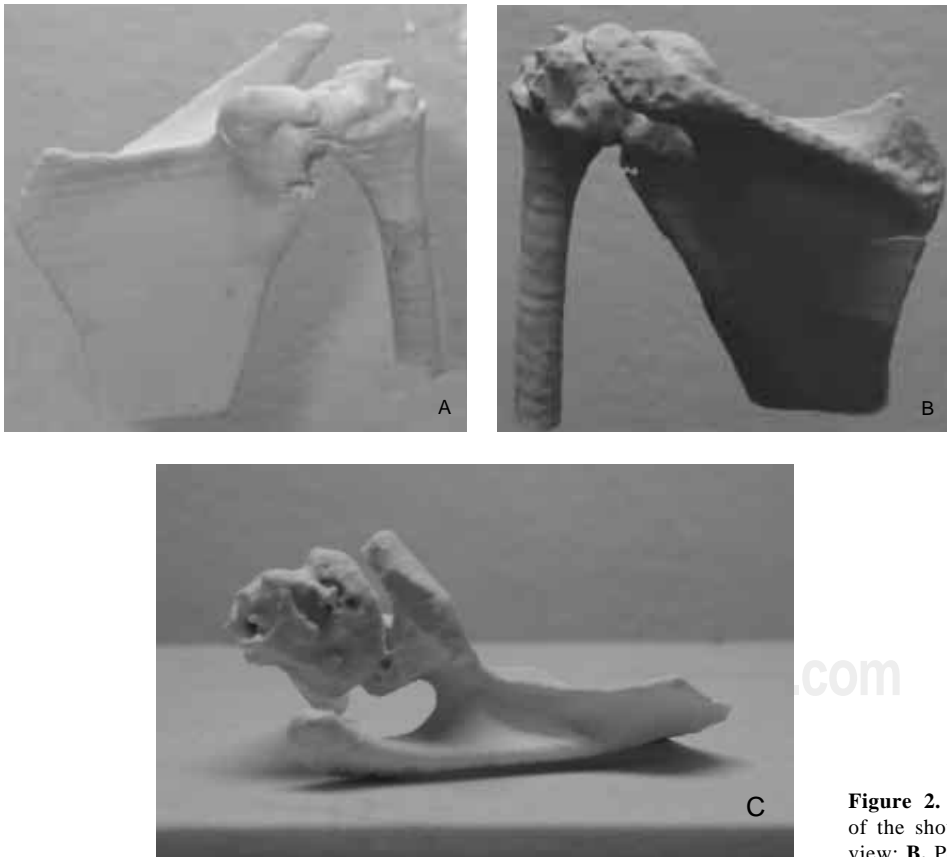
Rheumatoid arthritis is characterized by repetitive synovitis. This is reflected on the difficulty to treat and relieve pain. Initially, conservative methods are usually effective<sup>16</sup> but as the disease progresses, these methods begin to fail<sup>2,4,17</sup> leading to palliative pain management.

Reports on shoulder arthroplasty in patients with rheumatoid arthritis (RA) go back to 1979, when Neer practiced it with unsatisfactory results because of rotator cuff injury leading to later instability with dislocation and migration of the prosthesis.<sup>2,6,15,16,18-20</sup>

In 1990, Sneppen et al.<sup>6</sup> reported a prospective study in 62 patients with RA Larsen's Grade IV-V. These patients were treated with the Neer II prosthesis. The researchers

**Table 2. Functional evaluation based on the UCLA score.**

UCLA score	Preoperative	Postoperative
Pain	2	10
Function	2	4
Ventral anterior flexion	2	2
Strength with anterior flexion	1	3
Satisfaction	0	5



**Figure 2.** Sample of the 3-D anatomical model of the shoulder made of sandy clay. **A.** Anterior view; **B.** Posterior view; **C.** Bottom view.



**Figure 3.** Anteroposterior postoperative X-ray of the left shoulder.

found that within an average of 92 months, proximal migration of the humeral component occurred in 55% of patients; 40% experienced loosening as evidenced by radiographic findings and displacement of the glenoid component, together with loosening of the humeral component. The authors thus concluded that cementless hemiarthroplasty is better in patients with rheumatoid arthritis. In 1995, Cofield et al.<sup>7</sup> reported 35 shoulders with osteoarthritis and 32 with RA who underwent hemiarthroplasty. Of the latter, 18 shoulders required revision surgery and placement of the glenoid component due to erosion, which led us to questioning Sneppen's claim. We feel it is not possible to treat all patients with the same procedure, as some require total arthroplasty while others will do well with hemiarthroplasty alone.

The good results of shoulder prostheses essentially depend on the rotator cuff. We therefore believe that the benefits of muscle balance are responsible for centering the humeral head. Accordingly, shoulder prosthetic surgery is different from all other major joints, essentially in that most of the surgical procedure is soft-tissue surgery geared primarily to maintaining good muscle balance. Patients with a functional rotator cuff at the time of arthroplasty are handled with a total shoulder prosthesis.<sup>21</sup> The use of the glenoid component in patients with rheumatoid arthritis involves much more destruction of the rotator cuff and greater migration of the humeral component over time. Likewise, reaming of the glenoid cavity leads to loosening of the glenoid component.<sup>20</sup>

In 1991, Cofield et al.<sup>22</sup> reported a comparison between shoulder hemiprosthesis and total arthroplasty in 22 patients with rheumatoid arthritis and no rotator cuff. Patients treated with a hemiprosthesis reported worsening of pain. Those treated with total shoulder arthroplasty (TSA) had problems of a loosened glenoid component.

The loosened glenoid component was also reported by Matsen et al.<sup>7</sup> and Brems et al.<sup>23</sup> In 1992, Bigliani et al.<sup>24</sup> reported 30 patients with osteoarthritis and rotator cuff impairment. They were treated with hemiarthroplasty and TSA and the authors concluded that after a 40-month follow-up the hemiarthroplasty showed 95% improvement on pain. In 1996, William and Rockwood reported 21 shoulders in 20 patients with glenohumeral arthritis and impaired rotator cuff. After an average 4-year follow-up according to the Neer criteria, 86% of patients achieved satisfactory results including: absence of pain; improvement of external rotation from 27° before the surgery to 45° after the surgery; and improvement in flexion from 70° to 120°. None of the patients showed postoperative instability. These studies drove us to choose hemiarthroplasty for our patient.

Watson published a review of 14 bipolar prostheses of rough design with, nonetheless, very acceptable results considering they were indicated as a last resource. The mean assessment under the HSS scale improved from 5.3 before surgery to 18.9 after surgery. Watson stated that long-term results are not available and presumes progressive erosion of the glenoid surface leading to recurrence of pain and loss of motion.<sup>25-27</sup> We feel that short- and long-term residual pain essentially results from excessive tension or contracture of the soft tissues surrounding the joint because this increases tissue degradation and, consequently, its possibility of generating painful stimuli.

Stem cementing is desirable in rheumatoid arthritis, revision surgery, and fractures. It is generally recommended in cases in which a good rotational stability cannot be achieved. Cement must be applied to a stem of the immediately lower size than that of the last burr used.<sup>12,18,28</sup> We suggest not applying pressure on the cement to prevent it from leaking through the nutrient foramen of the shaft, located close to the radial nerve tract.

The size of the implant head should correspond to the one removed. When in doubt, it is preferable to use a smaller size. In joint disease due to rotator cuff tear, a larger size should be used and the retroversion maneuver should be overdone.<sup>16,23,24</sup>

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