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Bilateral reverse shoulder prosthesis in a patient with severe syringomyelia

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Syringomyelia refers to a cystic degeneration of the spinal cord. This cyst, called a syrinx, expands and elongates over time, causing damage to the spinal cord. This can result in pain, weakness, or stiffness, as well as conditions consistent with arthropathy. We report the first case of bilateral total shoulder arthroplasty with an inverted total shoulder prosthesis in a patient with post-traumatic syringomyelia. A satisfactory functional result and resolution of pain were obtained in both shoulders.

CASE REPORT

A 62-year-old woman presented to our clinic with a history of incomplete paraplegia resulting from spinal trauma with a fracture of the first lumbar vertebra in 1980. Over the subsequent years, permanent neurologic symptoms and cervicothoracic and lumbar syringomyelia developed with advanced arthritis of both shoulders. She reported no pain but a feeling of instability in her shoulders.

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She had to use a wheelchair and had to trigger defecation with her right hand, which required sufficient internal rotation of the right shoulder. Physical examination revealed a marked decrease in motion in both shoulders (Table I). The activity of both deltoid muscles was adequate. Radiographs and computed tomography scans demonstrated severe destructive changes of the shoulders, including formation of Charcot's joint with partial resorption of the humeral head, cysts, bony erosion, and osteophytes (Figure 1, A and B). Magnetic resonance imaging showed severe degeneration of the rotator cuff but a good deltoid muscle (Figure 1, C). In the setting of a rounded glenoid and a collapsed humeral head on imaging studies, we considered the implantation of a reverse prosthesis.

In July 2002, a reverse Delta III shoulder prosthesis (DePuy, Leeds, England) was implanted in the right shoulder, via a transdeltoid anterior approach. Postoperative treatment consisted of active assisted physiotherapy limited to 45° abduction and flexion with 0° external rotation and free internal rotation for the first 7 days, increasing gradually in the following weeks. At 6 months' follow-up after surgery, the range of motion of the shoulder had clearly improved (Table I). Three months after the operation, the patient could reach comfortably to her mouth, hair, and neck with the right arm (Figure 2, A). At that point, the range of motion of the left shoulder was still limited (Figure 2, B).

In November 2002, reverse shoulder arthroplasty of the left shoulder was done. Postoperative radiographs demonstrated good positioning of both prostheses (Figure 3). In January 2003, a fixation screw of the right glenoid component had to be changed because of loosening. Thereafter, both prostheses were stable, and no further revision was necessary.

Table I Data from American Shoulder and Elbow Surgeons evaluation form obtained preoperatively and 6 months and 24 months after surgery

	Pain	Daily function	Anterior flexion (°)	External rotation (°)	Internal rotation (°)
Right shoulder					
Preoperatively	Moderate	4/15	30	0	10
Postoperatively					
6 mo	None	9/15	80	40	60
24 mo	None	11/15	110	45	80
Left shoulder					
Preoperatively	Slight	3/15	20	0	15
Postoperatively					
6 mo	None	6/15	60	70	80
24 mo	None	9/15	110	70	80



Figure 1 **A** and **B**, Radiographs of right and left shoulders demonstrating partial resorption of humeral head, para-articular calcification, and dislocation. **C**, A magnetic resonance image of the right shoulder revealed severe degeneration of the rotator cuff but a good deltoid muscle.

We found a gradual improvement in range of motion and daily activity until the most recent clinical attendance at 24 and 30 months after surgery (Table I). Radiographs revealed no loosening of the prostheses and screws. The patient reported no pain but had functional strength and a stable feeling in both shoulders. Daily activity was improved, and triggering defecation with her right hand

was possible, but rising from the wheelchair was still problematic.

DISCUSSION

Neuropathic shoulder arthropathy is classic for syringomyelia, although exact numbers cannot be found. Because



Figure 2 Postoperative clinical control of right shoulder (**A**) and preoperative range of motion of left shoulder (**B**).

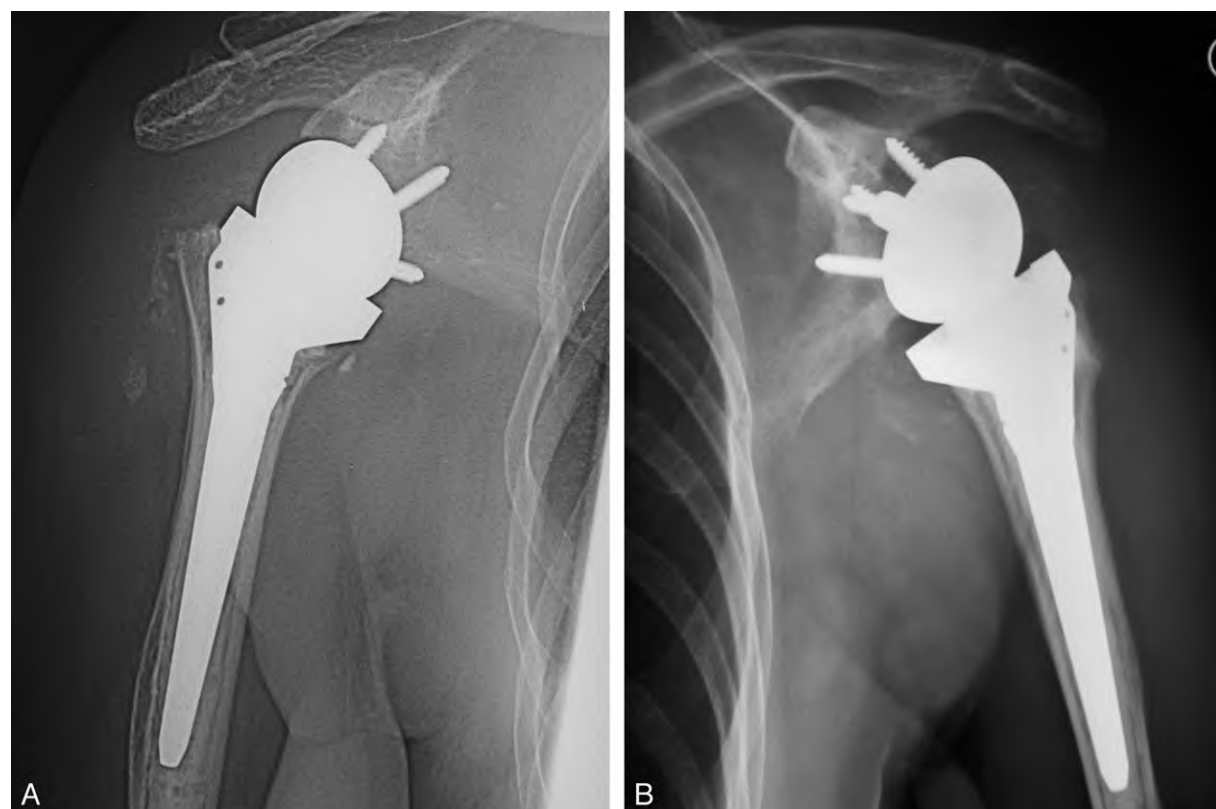


Figure 3 Postoperative radiographs demonstrating intact prostheses of right (**A**) and left (**B**) shoulders.

syringomyelia is rare, there are only periodic case reports in the literature about shoulder arthropathy in patients with this disease.^{3,5,7,9,10,16,18,19,21,23} To our knowledge, there are only two reports in the literature about operative treatment of shoulder arthropathy in syringomyelia patients, managed by either arthrodesis¹² or total shoulder arthroplasty.¹⁵ Mau and Nebinger¹³ reported failure when arthrodesis was performed to treat neuropathic arthropathy of the shoulder and concluded that this condition is a contraindication for arthrodesis.

In our patient, shoulder function was extremely limited. Conservative treatment did not improve her condition. A traditional shoulder prosthesis would have failed because of the massive tear and degeneration of the rotator cuff in these neuropathic joints. The condition of a rounded glenoid and a collapsed humeral head with severe degeneration of the rotator cuff, but a good deltoid muscle, led us to implant a reverse prosthesis, even though joint replacement is generally considered risky in syringomyelia patients because of the lacking sensorimotor feedback.

In 1993, Grammont and Baulot⁶ first reported their development of the reverse shoulder prosthesis (Delta) for rotator cuff rupture. The principle is to reverse the articular surfaces of the humeral head and glenoid to improve the function of the deltoid muscle.² There are few reports about this prosthesis.^{1,3,8,11,14,17,20,22,24-26} However, we could not find any report about the implantation of this prosthesis in a neuropathic shoulder.

Given the disparate condition of our patient, we decided to implant a reverse prosthesis first on the right shoulder and then on the left. We observed our patient for 26 and 30 months and could demonstrate survival of both prostheses over that period. We found a gradual functional improvement with a marked improvement in range of motion. Most of the demands of this paraplegic patient, who depends on her arms to function and transfer to and from her wheelchair, were fulfilled after this treatment. However, it has to be mentioned that we are only reporting on the short- to medium-term outcome in this patient.⁴

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