



# Shoulder arthropathy secondary to syringomyelia: case series of 10 patients

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Received: 2 May 2021 / Accepted: 16 August 2021 / Published online: 24 August 2021

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## Abstract

**Purpose** Neuroarthropathy is a progressive joint degeneration secondary to neurological diseases. In the upper extremity, the shoulder is the most exposed, and it is mainly caused by syringomyelia. This condition is rare; therefore, the literature has documented only a few case reports or case series of small groups of patients.

**Methods** We collected data about patients with shoulder arthropathy due to syringomyelia who were treated in our two institutes and collected among members of the Polish Shoulder and Elbow Society. Our analysis was based on epidemiological data, symptoms, and clinical examinations. We also examined the results of diagnostic tests, including—spinal cord MRI and shoulder X-ray, and treatment methods and their effectiveness.

**Results** The examined group included 10 women with an average age of 63 years. Of these, nine patients reported pain, seven reported—swelling, and nine reported—weakness. In every patient, diagnosis was confirmed by X-ray of the shoulder with joint degeneration and MRI of the spinal cord with syrinx. Two patients were operated with reverse shoulder arthroplasty; the first one had excellent result—significant active range of motion improvement and reduction of symptoms, and the second one had a good result—pain relief and moderate range of motion improvement. Other patients were conservatively treated, resulting in total or partial symptoms relief but without significant range of motion improvement.

**Conclusion** Charcot shoulder secondary to syringomyelia was mainly manifested by range of motion limitation, swelling, and pain. Both conservative and surgical treatments could be a good solution. However, if reverse arthroplasty is technically possible, it seems to be the most promising treatment for recovering function.

**Keywords** Charcot joint · Syringomyelia · Shoulder neuroarthropathy

## Introduction

The most common etiology of shoulder neuropathic arthropathy is syringomyelia. It is a rare and slowly progressive spinal cord disease that destroys the spinal cord's substance

and leads to neurological symptoms and abnormalities, such as loss of pain and temperature sensation, weakness, and arthropathy [1, 2]. Among a group of patients with syringomyelia, approximately 20–30% eventually develop a typical Charcot shoulder [3]. Joint-related symptoms are sometimes the first manifestation of syringomyelia. Typically, patient presents to an orthopedic surgeon with shoulder pain, stiffness, and swelling [1, 4–6]. When the shoulder is affected, it will usually lead to progressive degeneration of the humeral head and glenoid. Bone fragmentation and absorption or separation of large, displaced bone fragments have been diagnosed on radiographic evaluation [7, 8]. The neuroarthropathic shoulder differs from idiopathic osteoarthritis of the shoulder in the presence of additional neurological symptoms, massive bone loss and high inflammation.

So far, there have only been a few case reports of shoulder degeneration due to syringomyelia, and our understanding of pathophysiology, clinical picture, and management is still

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quite limited. Therefore, the aim of our study is to present our case series of neuroarthropathic shoulders secondary to syringomyelia, with a focus on epidemiological data, clinical picture presentation, treatment methods, and their effectiveness. To the best of our knowledge, this is the first such extensive case series described in the literature. Some surgeons avoid surgery on patients with a Charcot joint because of reports of early loosening due to the progressive bone loss seen with this disease. In our series, we present two patients successfully treated with shoulder arthroplasty.

## Materials and methods

This was a retrospective study based on a series of 10 patients with shoulder neuroarthropathy related to syringomyelia. The patients were qualified based on the following inclusion criteria:

- radiographic features of shoulder arthropathy based on shoulder X-ray (AP and axial views),
- characteristic neurological symptoms of syringomyelia,
- syrinx in the cervical spine based on magnetic resonance imaging (MRI),
- and the minimal time of onset 2 years. Most journals require at least 2 years of observation, so we have applied this period of observation. However, the shortest observation time in our group was 3 years. Time of follow-up was considered as the time since onset of the shoulder symptoms. This was not always the time of an established diagnosis, since shoulder symptoms are initially not specific to syringomyelia.

Patients' data were collected from both institutions (seven patients) and also from the survey conveyed among members of the Polish Shoulder and Elbow Society (three patients). For the initial data collection, we used a unified questionnaire sent to orthopedists, containing questions about epidemiological data, clinical symptoms, shoulder degeneration on X-ray, cervical spine MRI, and type and effect of treatment.

All patients were specifically examined for the presence of particular symptoms: shoulder pain, swelling, weakness, sensory dysfunction, and balance problems. Clinical examination relied on active range of motion (ROM), including flexion, and internal and external rotations. In every case, pain and temperature sensation was assessed. Two patients treated with reverse shoulder arthroplasty (RSA) and two patients treated conservatively were followed prospectively for a minimum of 3 years.

The degree of joint destruction of both the humeral head and glenoid was rated in every case based on the most recent

shoulder X-ray (two views). For both components, three stages were distinguished: no X-ray changes, partial degeneration (< 50%), and a massive degeneration (> 50%). This evaluation was a part of a larger project.

The evaluation of cervical spine MRI included the length and localization of the syrinx and identification of Arnold Chiari malformation.

This study was approved by Ethical Committee of Poznan University of Medical Sciences (number 162/17 from 02.02.2017).

No advanced statistical analysis or correlations were performed due to the low number of patients; however, complete patients' data are presented in Table 1.

## Results

### Demographic data

All of our patients were women, with an average age of 63 years (min. 24, max. 76). One patient had bilateral shoulder arthropathy. Among the rest, the left shoulder was affected in five patients, and the right shoulder was affected in four patients.

### Clinical picture

The most common shoulder complaints were: significant active and passive range of motion limitations, which occurred in all 11 arthropathic shoulders. Other complaints included—pain in nine shoulders, swelling—in seven, and weakness—in nine shoulders. Several additional symptoms, characteristic of syringomyelia were reported: sensory dysfunction (lack of pain and temperature sensation)—eight patients, balance problems—four patients, and lower limbs weakness—one patient. Both shoulder pain and limitation of active motion were the most disabling complaints in nearly all patients. Average ranges of active motion were limited to 65° (± 29, 20–140) for flexion, 36° (± 18, 20–80) for internal rotation, and 26° (± 22, 0–60) for external rotation.

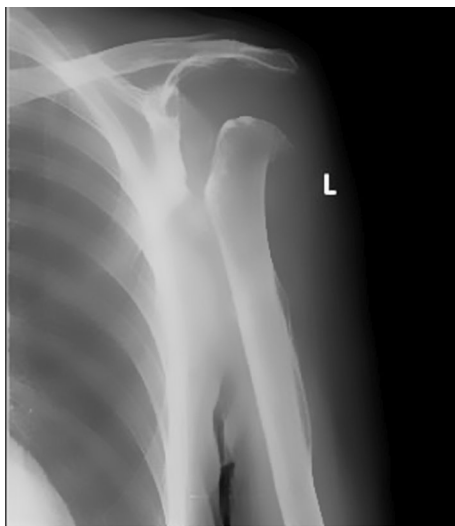
### Imaging

In all cases, the humeral head showed significant signs of degeneration. In eight shoulders, the humeral head was completely destroyed, so its shape could not be identified (Fig. 1). Based on an X-ray view, in three cases, partial head degeneration occurred, affecting over 50% of the humeral head cortex. The glenoid was intact on the X-ray in three cases. Partial loss of glenoid shape occurred in five shoulders. In three cases, the radiological shape of the glenoid was lost entirely.

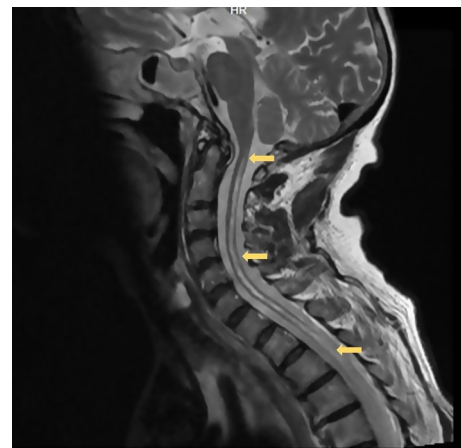
**Table 1** Patients' data

Patient	Age	Sex	Side	Symptoms before treatment	ROM before treatment (Flexion/Internal rotation/External rotation)	Treatment	Symptoms after treatment	ROM after treatment (Flexion/Internal rotation/External rotation)
1	76	Female	Left	Swelling, weakness	20/20/0	Reverse arthroplasty	Relief	140/45/40 <i>improved</i>
2	58	Female	Right	Pain, swelling	60/50/0	Reverse arthroplasty	Relief	90/50/40 <i>improved</i>
3	70	Female	Right	Pain, swelling, weakness	60/30/50	Conservative (physiotherapy, drugs)	Relief	120/60/60 <i>improved</i>
4	69	Female	Right, Left	R—weakness L—pain, swelling, weakness	R—50/60/45 L—45/20/0	Conservative (physiotherapy)	Partial relief	R—50/60/45 L—45/20/0 <i>no change</i>
5	68	Female	Right	Pain, swelling, weakness	50/30/40	Conservative (physiotherapy)	Relief	50/30/40 <i>no change</i>
6	72	Female	Left	Pain, weakness	70/30/50	Conservative (physiotherapy)	Partial relief	70/30/50 <i>no change</i>
7	68	Female	Left	Pain	80/30/10	Conservative (physiotherapy, drugs)	Partial relief	80/30/10 <i>no change</i>
8	64	Female	Left	Pain, swelling, weakness	80/20/10	Conservative (physiotherapy, drugs)	No relief	80/20/10 <i>no change</i>
9	24	Female	Left	Pain, weakness	140/80/60	Conservative (physiotherapy)	No relief	140/80/60 <i>no change</i>
10	61	Female	Right	Pain, swelling, weakness	60/20/20	Conservative (physiotherapy, drugs)	Partial relief (recurrent inflammation)	90/30/30 <i>improved</i>

R—right, L—left, ROM—range of motion

**Fig. 1** X-ray of a completely destroyed humeral head

The MRI of every patient showed syrinx in the cervical spinal cord expanding through the average length of 7, 5 segments ( $\pm 4,5$ , 4–20). (Fig. 2) We identified one patient with Arnold Chiari malformation on a spinal cord MRI.

**Fig. 2** MRI scan of a spinal cord with a syrinx

## Treatment

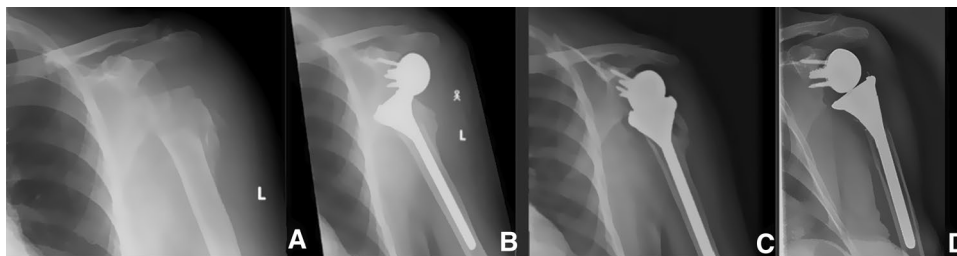
Eight patients were treated non-operatively with physiotherapy and/or anti-inflammatory medications. In two cases, it resulted in an improved active range of flexion: in one from 60 to 120° and the second from 60 to 90°. Five patients (out of eight) remained with significantly limited ROM despite an attempt of therapy. None of them were able to lift an arm to the shoulder level. One patient had a fairly functional

elevation that was not further affected by exercises. Six patients reported significant partial or complete pain relief over the course of the treatment and time.

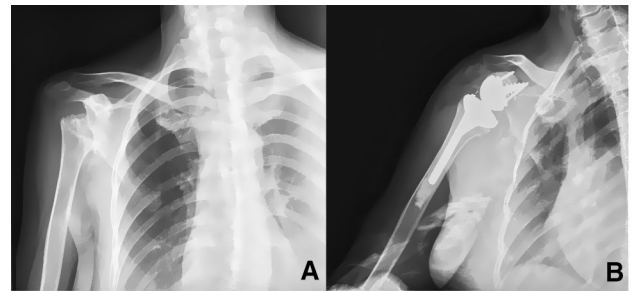
The two most recent patients were treated surgically by reverse shoulder arthroplasty (RSA). One of them experienced immediate dislocation that had been treated with closed reduction and 4 weeks immobilization in cast, eventually followed by a successful rehabilitation program. At their 4 years follow-up, this patient improved active flexion from 20 to 140°, internal rotation from 20 to 45°, and external rotation from 0 to 40° (Fig. 3). Her UCLA score improved from 6 to 27 [9], and her shoulder remained stable and functioned well at 4 years mark. (Fig. 4) Another patient treated with RSA also achieved good treatment results with improvement of active flexion from 60 to 90° and external rotation from 0 to 40°. Internal rotation remained on the level of 50° 3 years after the surgery. Her UCLA score improved from 8 to 22. (Fig. 5).



**Fig. 3** Patient 1's—clinical picture before and after RSA at the 4-year follow-up



**Fig. 4** Patient 1's **a**—preoperative X-ray, complete destruction of the humeral head, partial destruction of the glenoid; **b**—immediate postoperative X-ray, dislocated RSA; **c**—X-ray at follow-up after 4 weeks



**Fig. 5** Patient 2's **a**—preoperative X-ray, complete destruction of the humeral head, partial destruction of the glenoid; **b**—postoperative shoulder X-ray

## Discussion

Shoulder neuroarthropathy is a slowly progressive joint degeneration, often diagnosed at an advanced stage, occurring in patients with neurological disease (mainly syringomyelia localized in the cervical segments) [10, 11]. This condition develops over the years, causing significant limitations in shoulder function [1, 7, 11]. Syringomyelia is a rare disease (8, 4/10000) and only 20–30% of patients will develop a secondary shoulder arthropathy [3, 7]. Within the upper limb, the shoulder is the most common localization, although it is still an uncommon problem [2, 8, 10, 11]. No guidelines have been published so far about the diagnosis and treatment of such a group of patients. There have been only a few case reports, with very few cases described and one systematic review that summarized cases available in the literature [1]. The largest published groups included five patients (six shoulders) reported by Atalar et al. [3], three patients by Matsushashi et al. [12], and three patients by Ruetten et al. [8]. To the best of our knowledge, this is the largest series of patients with shoulder arthropathy secondary to syringomyelia reported so far in the literature. We have attempted to summarize the most common complaints and presented a standardized approach to diagnostics and the possibilities of treatment of the disorders.

of case immobilization and rehabilitation program; and **d**—X-ray at follow-up 4 years after the surgery

Our patients complained about the two most clinically relevant problems: pain and major reduction of shoulder function, especially by limitation of the active range of motion. We confirmed that patients still experienced severe pain despite typical for syringomyelia pain and temperature sensory loss. This was also confirmed by Atalar et al. [3] and Wang et al. [2]. We found that eight out of ten patients had sensory disorders, and only one patient did not report shoulder pain. Pain in syringomyelia has been previously reported as a common complaint and considered as central [13, 14]. Therefore, the pain level does not have to be related to the grade of joint destruction. The possible reasons for feeling pain in syringomyelia might be the loss of sensory balance due to disjunction between the anterolateral and dorsal columns of the spinal cord or abnormal levels of some neurotransmitters, such as substance P [15–17]. Previous reports indicate that neurosurgical treatment should be considered in syringomyelia. According to current knowledge, early neurosurgical treatment often results in stabilization or a slight improvement of symptoms for most patients, provided that there is no irreversible damage to the spine [16]. The most important goal of neurosurgical treatment is to prevent further enlargement of the cavity and damage to the remaining parts of the spinal cord. However, in our case studies, both conservative and surgical shoulder treatment, conducted without neurosurgical intervention, eliminated shoulder pain in four patients and reduced it in another four patients within a period longer than 3 years. Based on our analyses of the cases available in the literature, we believe that pain reduction in Charcot shoulder is a natural stage in the course of the disease and often does not depend strongly on the type of treatment used. Most of the patients experienced the following course of the disease: slight pain and limitation of shoulder function, sudden deterioration of function with severe pain and swelling, and then obtaining a stable reduction of pain with a significant limitation of the range of motion and weakness.

Shoulder X-ray analysis showed that humeral head destruction is more common and severe than glenoid destruction. Every patient with significant glenoid destruction also showed advanced head degeneration. Similar finding was reported by Atalar et al. [3] and Matsushashi et al. [12]. It is highly probably that the destruction process of the shoulder in this condition begins within the head and secondarily involves the glenoid. This phenomenon is hard to explain. If the microvascular disturbance was part of the process, then features similar to avascular head necrosis, which typically involves the humeral head and not glenoid, should be presented. Another similar condition in which the head is first affected is primary osteoarthritis of the glenohumeral joint, which also starts mostly at the head and affects the head more severely.

A significant number of cases presented in the literature showed advanced shoulder degeneration, so it can be expected that the natural course of shoulder arthropathy evolves to its total degeneration [1, 6, 8, 18–20]. These observations suggest that this condition should be treated early. Decision about potential reverse arthroplasty should be considered and not postponed before glenoid becomes completely destroyed.

Most orthopedic surgeons seem to treat this arthropathy with physiotherapy and anti-inflammatory medications [1, 4]. Conservative treatment can be a good solution for patients suffering mostly from pain. Six out of eight patients in our series who were treated conservatively had partial or total pain relief, but only two of them experienced improvement in active shoulder mobility. This approach may not meet the more functional expectations of patients. From our experience, there is no optimal and particularly effective conservative treatment that can be recommended. However, we believe that at a very early stage, some form of physiotherapy and anti-inflammatory medication should be the first line of treatment. Such treatment may also be effective to some extent at late stages, at which no surgical treatment is possible or is contraindicated.

For select patients, shoulder arthroplasty may be a better option. Some authors presented satisfactory results after humeral head replacement with preserved or repaired rotator cuff [1, 12]. With the advances and growing experience with reverse shoulder arthroplasty, the latter may be a better option in the view of severe bone destruction, instability, and soft tissue damage that occurs in neuropathic shoulder. Ueblacker et al. also described a good result of bilateral RSA in a 62-year-old woman with advanced shoulders neuroarthropathy [11]. Both of our patients regained function after RSA, and in the short-term follow-up, they remained satisfied.

The option of RSA should be considered early, before complete destruction occurs. There is one major prerequisite for installing the baseplate: the glenoid must be in a well-preserved condition. Humeral state is much more forgiving and easier to properly address with modular stem designs. In most cases, patient evaluations should include thorough imaging (MRI, CT) of both bone and soft tissue [12, 21, 22]. Published data on RSA, or arthroplasty in general, in neuropathic shoulder is very limited. Schoch et al. published a study based on a group of ten patients with Charcot shoulder treated with arthroplasty. The study included three patients with syringomyelia, with results encouraging consideration of arthroplasty in this condition [21]. No strict recommendations on the application of arthroplasty can be offered yet; however, for such a disabling shoulder disorder, arthroplasty might be a good solution, provided that the glenoid is well preserved. Moreover, Walecka et al. proved that RSA could

restore shoulder proprioception, which is very valuable in this condition [23]. However, we believe that a longer follow-up after RSA is important, particularly for evaluation of possible glenoid resorption. The balance of the benefits and risks of the procedure have to be considered with caution. Other contradictions to RSA may also appear. One of our patients was disqualified from surgery based on a general health condition and serious neurogenic balance problems.

In this study, one of the deciding factors in choosing a treatment method after conservative treatment failed was glenoid condition. Among conservatively treated patients, radiographic images varied: three shoulder had preserved intact glenoid, three had partial glenoid degeneration, and three had total or almost total glenoid degeneration. Patients without at least a partially preserved glenoid were disqualified from RSA. Other reasons for withdrawing from surgery included: sufficient improvement after conservative treatment, general health conditions, and lack of patient consent. At the beginning of our study, we also believed that conservative treatment was promising. However, an analysis of the clinical picture and the increasingly frequent reports of arthroplasty in the neuroarthropathic shoulder prompted us to change our perception of treatment for this condition.

This study has several limitations. The series was not large enough to draw conclusions based on advanced statistical analysis. The group was also quite heterogeneous in terms of the stage of arthropathy and treatment. Only two patients underwent surgery, and the rest of the patients were conservatively treated. Another limitation is the retrospective nature of this research. Despite these limitations, to the best of our knowledge, this study is the largest case series reported so far. All patients had fairly uniform evaluations with numerous variables analyzed, including patients symptoms, shoulder image, and morphology of the spinal cord. We believe these analyses also provided a general view of treatment options and their effectiveness in shoulder arthropathy secondary to syringomyelia.

## Conclusions

Charcot shoulder secondary to syringomyelia is a devastating disease with severe pain, ROM limitations, swelling, and weakness. A diverse radiological picture is expected in clinic, with the head usually more affected than the glenoid. The main clinical problems are: pain and major reduction of shoulder function, especially active range of motion limitation. Both conservative and surgical treatments may be a good solution. However, if reverse arthroplasty is technically possible, it seems to be the most promising treatment for recovering function

**Supplementary Information** The online version contains supplementary material available at <https://doi.org/10.1007/s00590-021-03102-0>.

**Acknowledgements** Alicja Viljoen

**Author contributions statements** All authors contributed to the study's conception and design. Material preparation, data collection and analysis were performed by AW and PL. The first draft of the manuscript was written by AW, and all the authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

**Funding** No funds, grants, or other support was received for this study.

**Availability of data and material** Associated data have been deposited in a data repository.

## Declarations

**Conflict of interest** All authors certify that they have no affiliations with or involvement in any organization or entity with any financial or non-financial interest in the subject matter or materials discussed in this manuscript.

**Ethics approval** Ethical Committee of Poznan University of Medical Sciences: 162/17 on 02 February 2017.

**Consent** The consent of the patient to publish her image is attached.

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