

## Posterior shoulder dislocation revealing syringomyelia

### Une syringomyélie révélée par une luxation postérieure de l'épaule

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#### ABSTRACT

**Introduction:** Syringomyelia is a potential cause of neuropathic osteoarthropathy of the shoulder, or "CHARCOT Shoulder." After swelling and pain, stiffness and decreased range of motion were the most frequent presenting symptoms, but the posterior dislocation of the shoulder had not been described. We report a case of posterior shoulder dislocation that revealed a syringomyelia and discuss the method of diagnosis and treatment.

**Case report:** A 50-year-old woman presented to the emergency room with a posterior right shoulder dislocation occurring after a minor trauma. Closed reduction was easy, but the shoulder was very unstable. After failed posterior bone block, shoulder arthrodesis as a salvage operation was done. X-rays taken 6 weeks after operation showed bone non-union with resumption of the proximal shaft of the humerus and a fracture of the arthrodesis plate. Diagnosis of "CHARCOT Shoulder" was evoked and MRI had showed a CHIARI I malformation with cervical and upper thoracic syrinx.

**Discussion:** Once a neuropathic joint has been diagnosed, its etiology should be pursued with aseptic joint aspiration to look for infection or tumor, and a spinal MRI to search for syringomyelia if the etiology remains in doubt. Arthroplasty is generally not advisable for destroyed dislocated syringomyelic joints, and the major complications occurring after attempted shoulder arthrodesis are non-union.

**Conclusion:** Diagnosis and treatment of the neuropathic arthropathy caused by syringomyelia are difficult.

#### RÉSUMÉ

**Introduction:** La syringomyélie est une cause potentielle d'arthropathie neurogène de l'épaule. Le gonflement articulaire, la douleur, la rigidité et la diminution de la mobilité sont les symptômes de présentation les plus fréquents, mais la luxation postérieure de l'épaule n'a jamais été décrite. Nous rapportons un cas de luxation postérieure de l'épaule qui a révélé une syringomyélie et nous discuterons le diagnostic et le traitement.

**Observation :** Il s'agit d'une femme de 50 ans, qui s'est présentée aux urgences avec une luxation postérieure de l'épaule droite suite à un traumatisme minime. La réduction était facile, mais l'épaule était très instable. Après l'échec d'une butée iliaque postérieure, une arthrodèse a été pratiquée comme une opération de sauvetage. Les radiographies de contrôle à 6 semaines, ont montré une résorption de l'extrémité supérieure de l'humérus et une fracture de la plaque d'arthrodèse. Le diagnostic d'arthropathie neurogène a été évoqué et une IRM médullaire a été faite et a montré une malformation de CHIARI de type I et des cavités de syringomyélie cervicale et thoracique.

**Discussion :** Devant une destruction articulaire peu douloureuse touchant l'épaule et après avoir éliminé une origine infectieuse et tumorale, il est nécessaire de penser à l'étiologie neurogène. Si l'étiologie reste dans le doute, une IRM vertèbro-médullaire doit être alors pratiquée à la recherche de cavité syringomyélique. Sur ces articulations syringomyéliques détruites et disloquées, l'arthrodèse ne consolide pas en général et le recours à des arthroplasties est globalement déconseillé.

**Conclusion :** Le diagnostic et le traitement de l'arthropathie neurogène causée par une syringomyélie sont difficiles.

## I. INTRODUCTION

Syringomyelia is a degenerative disorder of the spinal cord characterized by an abnormal longitudinal cavitation (syrinx) filled by cerebrospinal fluid in the central canal [1]. It has been described as a cause of neuropathic arthropathy in upper limbs and predominantly affecting shoulders or elbows [1, 2]. After swelling and pain, stiffness and decreased range of movement are the most frequent presenting symptoms, but a posterior dislocation of the shoulder had not described. We report a case of syringomyelia revealed by a posterior shoulder dislocation and discuss the method of diagnosis and treatment.

## II. CASE REPORT

A 50-year-old woman presented to the emergency with a posterior right shoulder dislocation happening after a minor trauma (Figure 1).



**Fig. 1:** anteroposterior radiograph of the right shoulder shows posterior dislocation

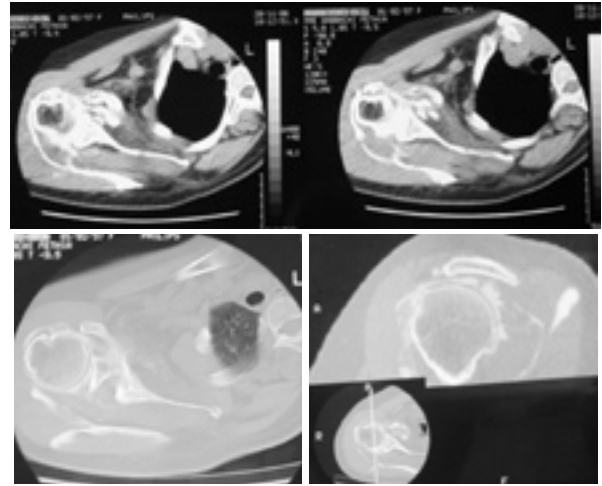
Closed reduction was easy (Figure 2), but the shoulder was very unstable. In her past medical history, the patient had chronic shoulder's moderate pain and there was no history of spinal surgery. She did not take medications.



**Fig. 2:** anteroposterior radiograph of the shoulder after reduction

On physical examination, she had full range of motion of her cervical spine. The left upper extremity was normal with full range of motion, 5/5 strength, and intact sensation. Examination of the patient's right upper extremity revealed normal skin and no abnormal masses. She had 4/5 biceps strength, 5/5 triceps strength, and 5/5 motor strength distally in the right upper extremity. Her median, ulnar, and radial nerves are all intact at the level of the hand.

An arthroscan of the right shoulder showed rotator cuff damage with a GOUTALLIER fatty degeneration grade (Figure 3).



**Figure 3 :** A, B, C: arthroscanner of the right shoulder shows rotator cuff damage with state IV degenerate grease

A posterior approach was used and a bone graft was then harvested from the iliac crest and inserted into glenoid posterior face. Fixation of the graft to the posterior border of the glenoid was obtained with two parallel postero-anterior screws after adequate rasping of the upper surface and the posterior border of the glenoid (Figure 4A). Bone and synovial biopsies were taken for histology and bacteriology exams, which showed evidence of aseptic inflammation. An internal rotation sling was maintained for 6 weeks after surgery. Passive forward flexion and external rotation to neutral position were obtained after one month and the patient experienced then after excellent initial relief from pain. The postoperative radiography was satisfactory (Figure 4B).



**Figs 4-A and 4-B:** anteroposterior and lateral radiographs of the shoulder shows fixation of the iliac crest bone graft to the posterior border of the glenoid with two screws

Three months later, examination revealed a swollen right shoulder with discomfort on passive and active motion.

The shoulder was warm to the touch and the patient was afebrile. Palpation revealed a generally tender shoulder with no obvious localized bone or joint tenderness. Clinically, there was 60 degrees of flexion and abduction, with normal external rotation. The internal rotation was painful. Gross neurovascular examination was normal as a more detailed examination was precluded by the patient's discomfort.

Laboratory data showed negative inflammatory signs, no leukocytosis, and culture of aspirated fluid showed negative findings. In addition, there were no crystals, no pus cells and no organisms. However, the X-rays (Figure 5) showed signs resorption of the graft and the humeral head and destructive joint changes as compared with the previous plain radiographs taken only 2 weeks previously.

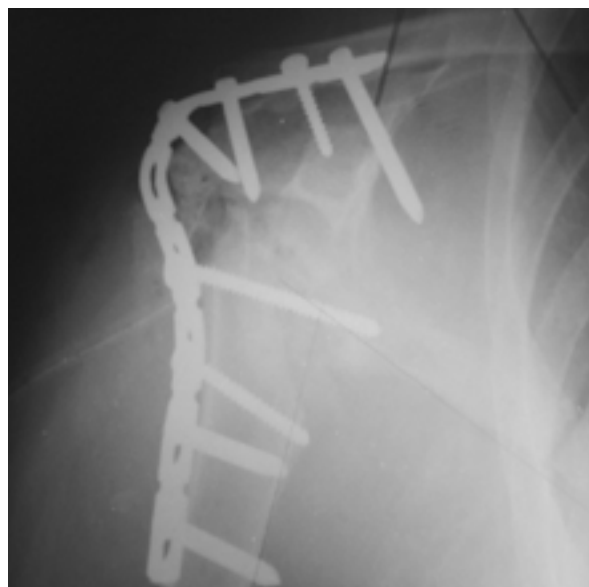


**Fig. 5:** anteroposterior radiograph of the shoulder shows signs resorption of the graft and the humeral head and destructive joint changes

The patient was reoperated with the same posterior approach; loosening of the iliac graft, joint destruction, disorganization, and effusion with osseous debris were found. Shoulder arthrodesis was decided and articular surfaces of the glenoid and humeral head were then denuded from cartilage, and the undersurface of the acromion was decorticated in a similar fashion. A 12-hole large-fragment dynamic compression plate was needed for osteosynthesis (Figure 6).

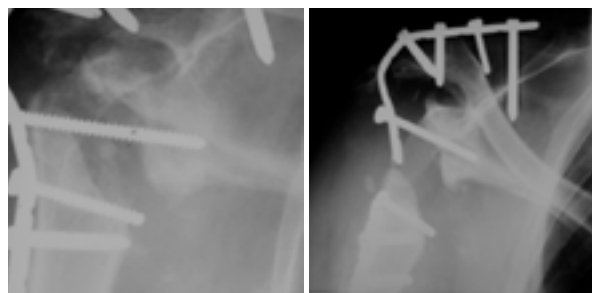
With the denuded humeral head pressed into the glenoid and acromion, two K-wires were passed to help maintain the position. Then, with the plate in place, a 6.5 mm partially threaded cancellous screws were placed across the gleno-humeral joint.

One or two screws were placed across the plate and through the acromion into the humeral head. Finally, the remaining screw holes along the spine of the scapula and shaft of the humerus were filled with cortical screws. At least 3 cortical screws were placed in the spine of the scapula. Bone autograft from the humeral head and iliac crest was placed in the space about the head, glenoid, and acromion. Abduction angle between the lateral border of the scapula and the humerus was 45°.



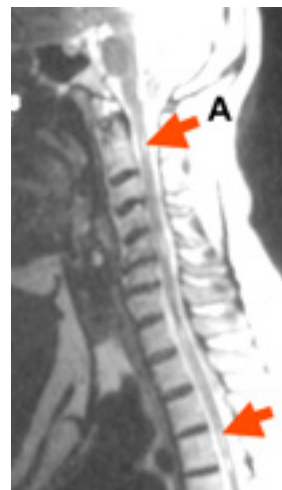
**Fig. 6:** anteroposterior radiograph of the shoulder shows 12-hole dynamic compression plate was contoured to run along the spine of the scapula, over the top of the acromion, and down the anterior shaft of the humerus

The X-rays taken 6 weeks after operation showed non-union, resorption of the graft and the proximal shaft of the humerus with a plate fracture (Fig. 7).



**Figs 7-A and 7-B:** anteroposterior radiograph of the shoulder made 6 weeks after operation (fig. 7-A) and 8 weeks (fig. 7-B) showed non-union, signs resorption of the graft, the proximal shaft of the humerus and a fracture of the plate (7-B).

Neuropathic arthropathy was evocated and sagittal T1-weighted MRI with contrast had demonstrate a CHIARI I malformation with syringomyelia. The position of the syrinx was found at the lower cervical and upper thoracic level (Fig. 8).



**Figure 8:** sagittal T1-weighted MRI with contrast demonstrating a Chiari I malformation with syringomyelia. The position of the syrinx was found at the lower cervical (A) and upper thoracic level (B).

### III. DISCUSSION

In this case, posterior dislocation of the shoulder had revealed syringomyelia and Arnold CHIARI I malformation. In 1998, HATZIS et al. [3] had reviewed the literature and found only 31 documented cases of neuropathic arthropathy of the shoulder. Swelling of the shoulder and pain were the most frequent presenting symptoms with respectively 29 and 25 of the 31 cases. For ALNOT [4] a neurological cause might be involved when joint destruction of the shoulder or elbow produces little pain. In the review of HATZIS et al. [3], stiffness and decreased range of movement were the most frequent presenting symptoms after swelling and pain. However, posterior dislocation of the shoulder had not been described.

CHIARI I malformation is a congenital anomaly, resulting in the downward displacement of the lower most portion of the cerebellum, with elongation of the fourth ventricle and lower brain stem, in the absence of intracranial space-occupying lesions [5, 6]. Patients with CHIARI I malformation develop syringomyelia in about 75-85% of cases [7]. Syringomyelia is a cavitation (syrinx) within the spinal cord and most commonly presents in the third to fourth decade [8]. These syrinxes interrupt the decussating fibres of the lateral spinothalamic tracts that mediate pain and temperature sense [6, 9]. Neuropathic joints develop in 25% of patients with syringomyelia, and 80% involve the upper limb mainly the shoulder [3]. Neuropathic arthropathy, also known as CHARCOT's joint, is a progressive degenerative arthritis associated with loss of nociceptive stimulus at the affected region [9]. The pathogenesis of this condition is attributed to an initial phase of neural mediated vascular changes. After sensitive loss, fractures and joint damage occur leading to bone fragmentation, instability, joint dislocation, and secondary degenerative changes [10, 11].

The differential diagnosis will often include tumour, tuberculosis, infection and GORHAM disease [6]. Once a neuropathic joint has been diagnosed, its etiology should be pursued with aseptic joint aspiration to look for infection or tumor, and a spinal MRI to evaluate for syringomyelia if the etiology remains in doubt. Tabes dorsalis, diabetes mellitus, leprosy, multiple sclerosis, and rare congenital sensory neuropathies are recognized as other causes [12].

Treatment success had been reported with arthrodesis [13] and humeral head prosthesis [8, 14, 15] but in our case, the result of surgery was poor. We believe that surgical treatment of neuropathic arthropathy caused by syringomyelia is difficult and, indeed, we think that it has to be often contraindicated.

In conclusion, when shoulder dislocation after a minor trauma produces little pain, a neurological cause might be involved.

### IV. REFERENCES

- 1) Neves F.deS., Gonçalves D.P., Gonçalves C.R. Syringomyelia, neuropathic arthropathy and rheumatoid arthritis as diagnostic dilemmas in two different cases: confounding factor and true coexistence. *Clin Rheumatol* 2007; 26:98-100.
- 2) Ekim A., Armagan O. Neuropathic arthropathy caused by syringomyelia in different joints and lesion of brachial plexus at right upper extremity: A case report. *Agri* 2007; 19:54-9.
- 3) Hatzis N., Kaar T.K., Wirth M.A., Toro F., Rockwood C.A.J. Neuropathic arthropathy of the shoulder. *J Bone Joint Surg* 1998; 80A:1314-9.
- 4) Alnot J.Y., Rossarie R., Welby F. Syringomyelia and associated bone and joint diseases. *Rev Chir Orthop* 2007; 93:269-76.
- 5) Cheng K.C.K., Douglas C., Barnes S.J. Delayed presentation of neuropathic arthropathy of shoulder secondary to syringomyelia. *Injury Extra* 2005; 36:42-4.
- 6) Bowen J.D., Malanga G.A. Spondylolysis associated with Arnold Chiari malformation and syringomyelia: a report of 2 cases. *Spine* 1997; 22:2458-63.
- 7) Klekamp J., Iaconetta G., Samii M. Spontaneous resolution of Chiari I malformation and syringomyelia: case report and review of the literature. *Neurosurgery* 2001; 48:664-7.
- 8) Crowther M.A., Bell S.N. Neuropathic shoulder in syringomyelia treated with resurfacing arthroplasty of humeral head and soft-tissue lining of glenoid: A case report. *J Shoulder Elbow Surg* 2007; 16:e38-40.
- 9) Jones J., Wolf S. Neuropathic shoulder arthropathy (Charcot joint) associated with syringomyelia. *Neurology* 1998; 50:825-7.
- 10) Brower A.C., Allman R.M. Pathogenesis of the neuropathic joint: neurotraumatic vs neurovascular. *Radiology* 1981; 139:349-54.
- 11) Jones E.A., Manaster B.J., May D.A., Disler D.G. Neuropathic osteoarthropathy: Diagnostic dilemmas and differential diagnosis. *Radiographics* 2000; 20:S279-93.
- 12) Guille J.T., Forlin E., Bowen J.R. Charcot joint disease of the shoulders in a patient who had familial sensory neuropathy with anhidrosis. *J Bone Joint Surg* 1992; 74A:1415-7.
- 13) Kuur E. Two cases of Charcot's shoulder arthropathy. *Acta Orthop Scand* 1987; 58:581-3.
- 14) Levy O., Copeland S.A. Cementless surface replacement arthroplasty of the shoulder. *J Bone Joint Surg* 2001; 83B:213-21.
- 15) Burkhead W.Z., Hutton K.S. Biologic resurfacing of the glenoid with hemiarthroplasty of the shoulder. *J Shoulder Elbow Surg* 1995; 4:263-70.

