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Case Report

Bilateral shoulder pain after a generalized tonic-clonic seizure: case report of an acute undiagnosed bilateral fracture of proximal humerus

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Bilateral shoulder fracture is a rare event and convulsive epileptic seizure is one of the most common causes. Detailed physical and neurovascular examination are important but shoulder's anteroposterior and axillary radiographs are essential to reveal a possible fracture and/or dislocation. Diagnosis could be difficult and often missed because of the importance given to patient's neurological issue. We present a case of a young man with an acute undiagnosed bilateral fracture of proximal humerus as complication of a generalized tonic-clonic seizure. After seven days we performed a surgical procedure of bilateral reduction and osteosynthesis with angular stability plate and screws.

Keywords: Shoulder dislocation; Proximal humeral fracture; Seizure.

INTRODUCTION

Simultaneous bilateral fracture with a possible dislocation of shoulder is a rare event and it has been described in literature since 1950 (Gehr et al., 1999; Shaw, 1971; Mackmull and Weeder, 1950). According to the Literature, convulsive epileptic seizure is one of the common causes of these kind of fractures and could be associated with posterior humeral head dislocations. Diagnosis is difficult and often missed (Kokkalis et al., 2012), because after seizure, the medical attention is aimed at investigating neurological sequelae and so consequences of fall take second place.

Epylepsy is one of risk factor for proximal humerus

fractures (Chu et al., 2004) and it is considered to increase a person's risk of fracture because of falls caused by seizures (Finelli and Cardi, 1989).

Patient's history, detailed physical and neurovascular examination are important but an antero-posterior and axillary radiographs of each shoulder are essential to reveal a possible fracture and/or dislocation of shoulders.

In case of anterior or posterior dislocation, a procedural sedation, reduction and immobilization are possible, but with a fracture, it is mandatory an orthopaedic evaluation.

CASE PRESENTATION

A 31-years-old man was admitted to our Emergency Department (ED) after a generalized tonic-clonic seizure.

On patient arrival at ED, he was post-ictal and unable to offer either a complaint or a clinical history. He

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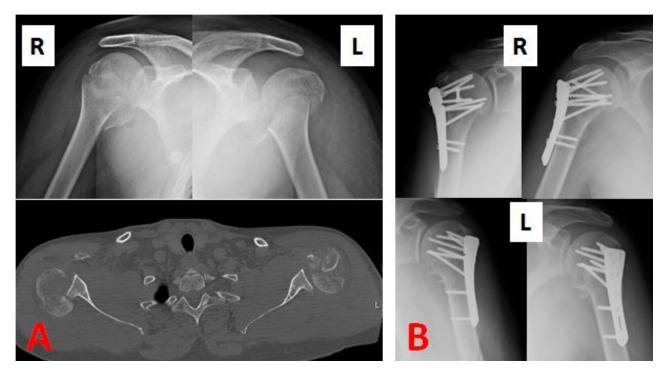


Figure 1. A) preoperative X-Rays and CT scan images. B) postoperative X-Rays showing adeguate reduction and internal fixation with angular stability plate

presented diffuse rigidity, froathing with tremor and, on physical examination, his vital signs were: pulse, 80 beats/min; blood pressure, 150/90 mm/Hg; breathing, 25/min; temperature, 35.9°C.

Initial examination of heart and abdomen were unremarkable but lungs presented reduced bases breath sound. A fingerstick glucose was not obtained on arrival in ED.

Patient's mental status improved as his post-ictal state resolved and he reported a history of epilepsy. He also reported bilateral shoulder pain with an alteration of the normal anatomical profile and reduced motility of both upper limb. His neurological examination was normal.

Multiple EEGs, cheast X-Rays and brain Magnetic Resonance Imaging (MRI) did not reveal any abnormalities.

He was given a benzodiazepine and admitted to a department of internal medicine without a shoulders X-Rays and an orthopaedic evaluation.

After three days, patient continued to report shoulders pain and at this time it was performed an orthopaedic examination that detected an important inability to mobilization of both shoulders without neurological deficits. So bilateral shoulder X-Rays were performed that revealed a right posterior shoulder dislocation associated with fracture of humeral head and a left fracture of proximal humerus.

It was performed a Computed Tomography (CT) to achieve better depiction of shoulder fractures and to plan

surgery.

According to AO fracture classification, he had a 11-C3.3 fracture on right side and a 11-C2.3 fracture on left side (Figure 1A).

Seven days after arrival in ED it was performed a treatment of both fractures with open reduction and internal fixation (O.R.I.F.) with angular stability plate (O'Nil Proximal Humerus Fixation, made by Intrauma).

There haven't been any intraoperatively complications and postoperative X-rays showed adequate fractures reduction and correct implant position (Figure 1B).

Patient was discharged 3 days post-operatively, shoulders were immobilized in a sling and it was allowed only a passive mobilization and Codman's exercises with a limitation for abduction to 90° and extrarotation for 2 weeks. After that it was permitted a careful active mobilization.

There were no more signs of epilepsy in the follow-up. The patient was followed with clinical examination and AP and axillary X-Rays of both shoulders at 1, 3, 6 and 12 months after surgery. We did not reveal signs of necrosis or osteomalacia of both humeral head, clinically and radiologically.

Fractures healed after 90 days (Figure 2) and we performed removal of internal devices at 15 months. At time of removal, patient was free of pain with a bilateral range of motion of 180° abduction, 160° right and 180° left elevation and bilateral 45° external rotation.



Figure 2. Clinical results after 90 days from surgery

DISCUSSION

According to the Literature, simultaneous bilateral fracture with a possible dislocation of shoulder is a rare event (Gehr et al., 1999; Shaw, 1971) and it has been described in literature since 1950 (Mackmull and Weeder, 1950).

The diagnosis is difficult and often missed, as stressed by Kokkalis et al. (2012).

This injury needs high forces and epileptic seizure is one of common cause (Finelli and Cardi, 1989; Lindholm and Elmstedt, 1980). This has been described by Brackstone et al. as the "triple E syndrome": epilepsy, electrocution and extreme trauma (Brackstone et al., 2001).

As written by Noachtar, during generalized tonic-clonic seizures the humerus is adducted, rotated inward and bent, while the muscle of scapular region pull on head. The infraspinous, teres minor, deltoid, latissimus dorsi and teres major muscles collaborate in a posterior direction and humeral neck can then fracture (Noachtar, 1998).

As described by the major Authors, displaced 3- and 4part proximal humerus fractures are aggravated by important complication rate and avascular necrosis of the head (AVN) is a common one (Meffert and Eden, 2009; Burkhart et al., 2013).

Other complications are related to choice of surgical treatment with open reduction and internal fixation with plate and are reported in a review of 2011 by Sproul et al. (varus maluonion, AVN, screws cut-out or perforation, infection, subacromial impingment, non-union, device failure, adhesive capsulitis, nerve palsy, pulmonary embolus and stiffness) (Sproul et al., 2011).

In our experience, displaced 3- and 4-part proximal humerus fractures are challenging even for experienced surgeon and there are many surgical strategies ranging from percutaneous fixation, intramedullary nail, O.R.I.F. and prosthetic replacement, but uniform results have not been obtained with any fixation device.

Chances of success are related to pattern of fracture, like involvement of medial part of humeral neck or tuberosities displacement or humeral head angle and anteversion, to time from fracture, to bone mineral density, to patient age and to personal surgeon experience.

In our case, fractures were not been revealed at the arrival in ED and this delayed diagnosis was an additional

risk factor for AVN.

We decided for surgical plating treatment for both fractures, despite high incidence for humeral head necrosis after O.R.I.F. This option was chosen based on pattern of fracture and patient young age.

According to Duralde and Leddy, in displaced unstable proximal humeral fractures the goals of O.R.I.F. are an acceptable alignment and fracture union in order to obtain early rehabilitation (Duralde and Leddy, 2010).

We support treatment with open anatomical/functional reduction and internal fixation with plate or intramedullary nail when allowed by fracture pattern and we refer shoulder prosthetic replacement only in some elderly patients and in few selective young patient cases.

CONCLUSIONS

As stressed by O'Connor-Read et al, full musculoskeletal examinations are not routinely performed following a seizure and the reported rate of late diagnosis is greater than ten percent in patients with direct trauma (O'Connot-Read et al., 2007).

So we suggest all patients with shoulder pain after epileptic seizures should be evaluated with X-rays and/or orthopaedic evaluation because delayed or not diagnosed fracture/dislocation is a medical negligence.

We do not agree with Jansen et al. that sustain prosthetic replacement as primary choice in treatment of shoulder dislocation with a complex fracture after an epileptic seizure (Jansen et al., 2012).

Our surgical proposal is to attempt open reduction and internal fixation where possible and to reserve choice of prosthesis in elderly patients or failures of other fixation devices.

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