

# Bilateral posterior fracture - dislocation of the shoulders after epileptic seizure in patients with cerebral meningiomas. Triple E syndrome – Review of the literature

M. Petrov<sup>a,\*</sup>, M. Laleva<sup>a</sup>, M. Fidanov<sup>b</sup>, V. Gerganov<sup>a,c</sup>, N. Velinov<sup>a</sup>, A. Baltov<sup>b</sup>, N. Gabrovsky<sup>a</sup>

<sup>a</sup> Department of Neurosurgery, University Multiprofile Hospital for Active Treatment with Emergency Medicine N. I. Pirogov, Sofia, Bulgaria

<sup>b</sup> Department of Orthopedics and Traumatology, University Multiprofile Hospital for Active Treatment with Emergency Medicine N. I. Pirogov, Sofia, Bulgaria

<sup>c</sup> International Neuroscience Institute, Hannover, Germany

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## ABSTRACT

Bilateral posterior fracture-dislocation of the shoulders is a very rare entity with an average rate of 0.6/100000 per year. It was first described in 1902 by Mynter. Only a few cases have been published so far. The “triple E syndrome” is used to describe the causative factors involved in this injury – epilepsy, electrocution, extreme trauma. We present our experience since 2019 with 2 cases of bilateral posterior fracture-dislocation of the shoulders after an epileptic seizure in patients with cranial meningiomas. Total removal of the meningiomas was performed in both cases and afterwards the patients were operated on by the traumatology team.

The shoulder joint is the most commonly dislocated joint in the body with less than 4 % being posteriorly dislocated. Bilateral fracture-dislocation of the shoulders is associated with “triple E syndrome” and 90 % of the cases are related to seizures. The diagnosis is usually delayed due to the lack of signs of trauma. Early diagnosis and proper surgical treatment can maximize the final functional results and patient recovery.

## Introduction

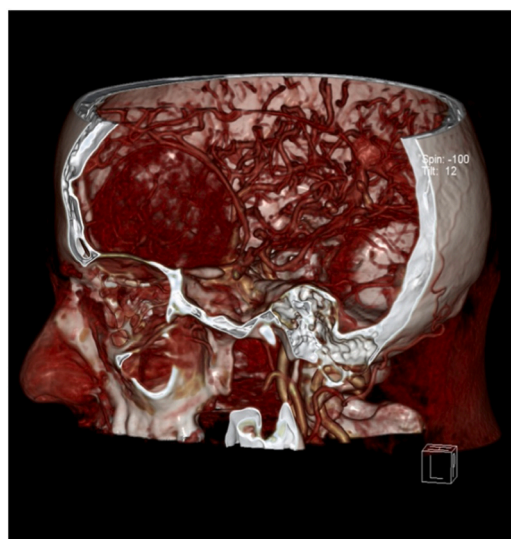
Posterior fracture-dislocation of the shoulder is a very rare entity with an average rate of 0.6/100000 per year and represents 2–4 % of all types of dislocation of the shoulder [1]. Bilateral posterior fracture-dislocation of the shoulders is encountered in less than 1 % of the above-mentioned cases and is usually the result of epileptic seizures. It was reported for the first time in the beginning of the 20th century by Mynter [2]. Only around 35 cases have been reported so far [3]. The term “Triple E” syndrome was introduced in 2001 by Brackstone [3]. The name is based on the three main etiologies causing bilateral fracture-dislocation of the shoulders – epilepsy [3–5], electrocution [1], and extreme trauma. We present 2 cases of patients with cranial meningiomas that presented after epileptic seizures. After the seizures both patients had posterior fracture-dislocation of the shoulders without any previous trauma or fall during the event.

\* Corresponding author.

E-mail address: [mihail5rov@gmail.com](mailto:mihail5rov@gmail.com) (M. Petrov).



**Fig. 1.1.** Axial reconstruction of the preoperative CT scan of the patients from clinical case 1.



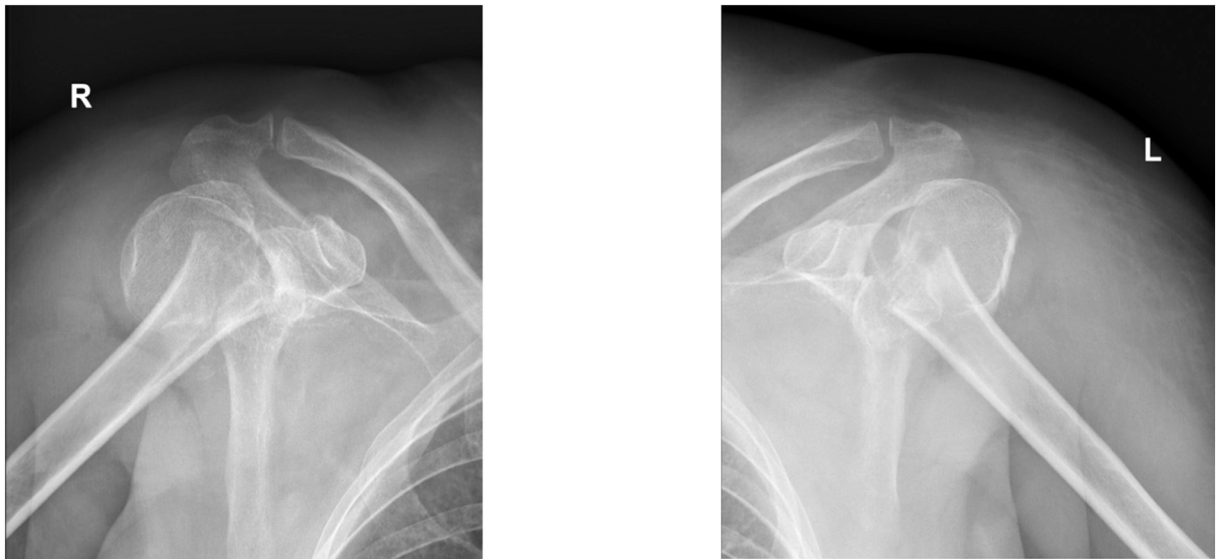
**Fig. 1.2.** 3D-reconstruction of the contrast-enhanced preoperative CT scan of the patient from clinical case 1.

### Clinical case 1

A 49-year-old female patient was admitted after 2 tonic-clonic epileptic seizures. The patient has had intermittent headache for a period of 2 years without seeking medical attention. Some behavioral changes have also been noted.

At admission the patient was GCS 13 points, disoriented, with no motor deficit. An emergency contrast-enhanced CT (computed tomography) scan was performed (Figs. 1.1 and 1.2). A frontobasal contrast-enhanced lesion was diagnosed with considerable perifocal oedema. After conducting dehydration therapy (including Mannitol and Dexamethason) the patient was GCS 15, neurologically intact. On the second day the patient had another generalized epileptic seizure. Postictally the patient complained about pain in both shoulders. Fig. 2.1 and Fig. 2.2 show the anteroposterior (AP) view radiographs of the patient's shoulders. A bilateral 4-part posterior fracture-dislocation of the proximal humerus according to the Neer classification system [6] was diagnosed and confirmed by the CT scan of the shoulders (Fig. 3.1 & Fig. 3.2).

After an interdisciplinary discussion it was decided that due to the frequent epileptic seizures, the mass effect of the lesion and the present focal neurological signs, the tumor removal should be performed first. A bicoronal skin incision was performed in order to



**Fig. 2.1 & 2.2.** Preoperative AP view radiographs of the right and left shoulder of the patient from Clinical case 1.



**Fig. 3.1.** 3D-reconstruction of the CT scan of the right proximal humerus. (Clinical case 1).



Fig. 3.2. 3D-reconstruction of the CT scan of the left proximal humerus. (Clinical case 1).

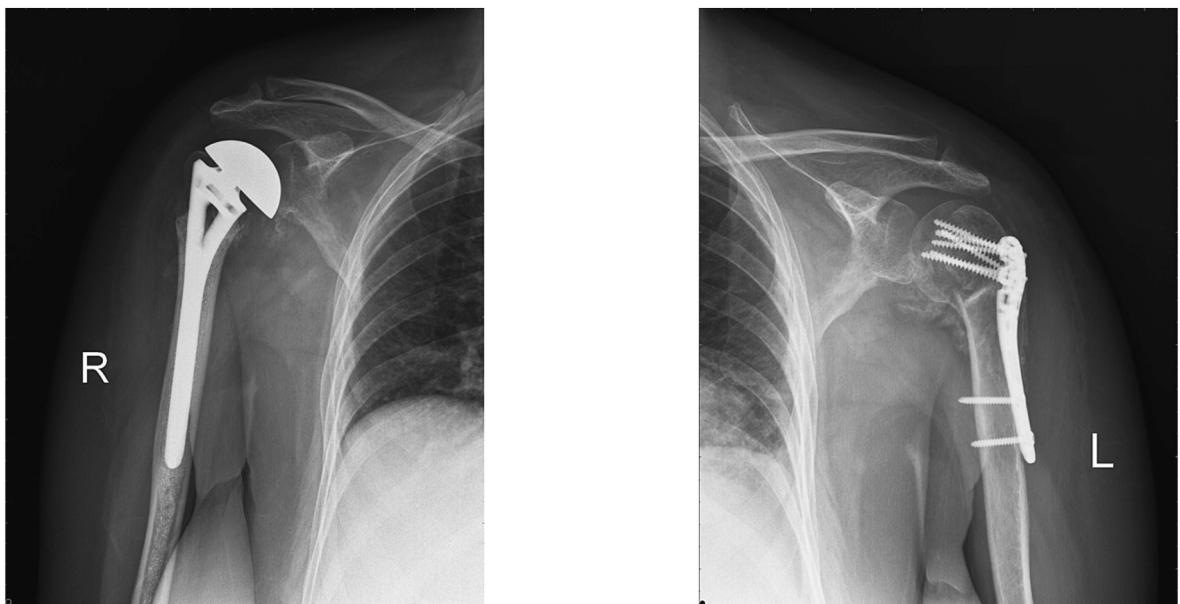
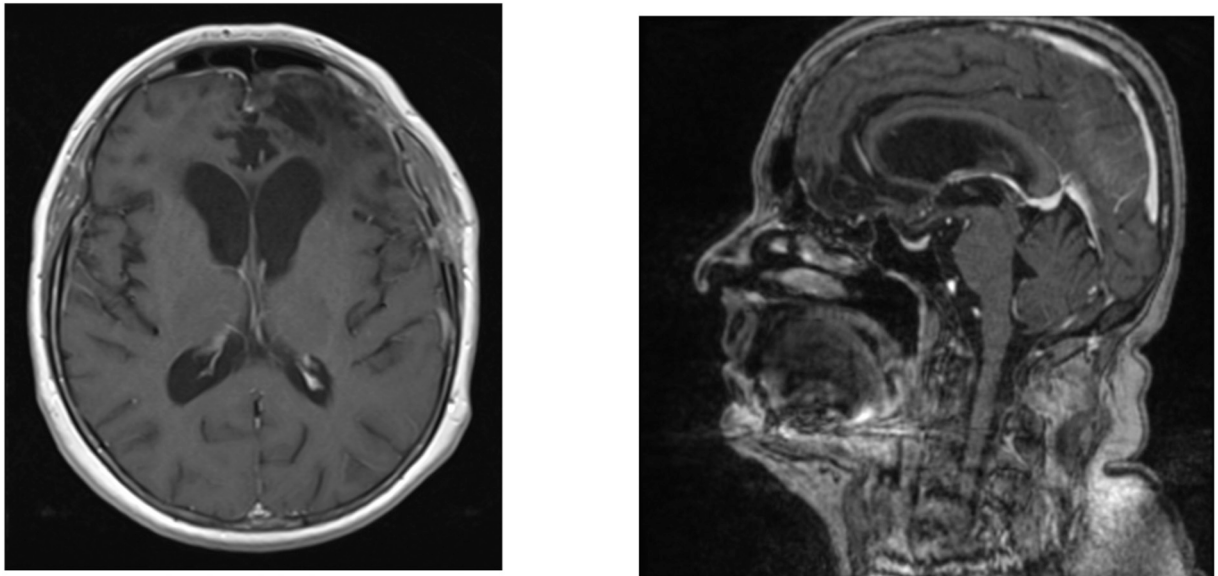
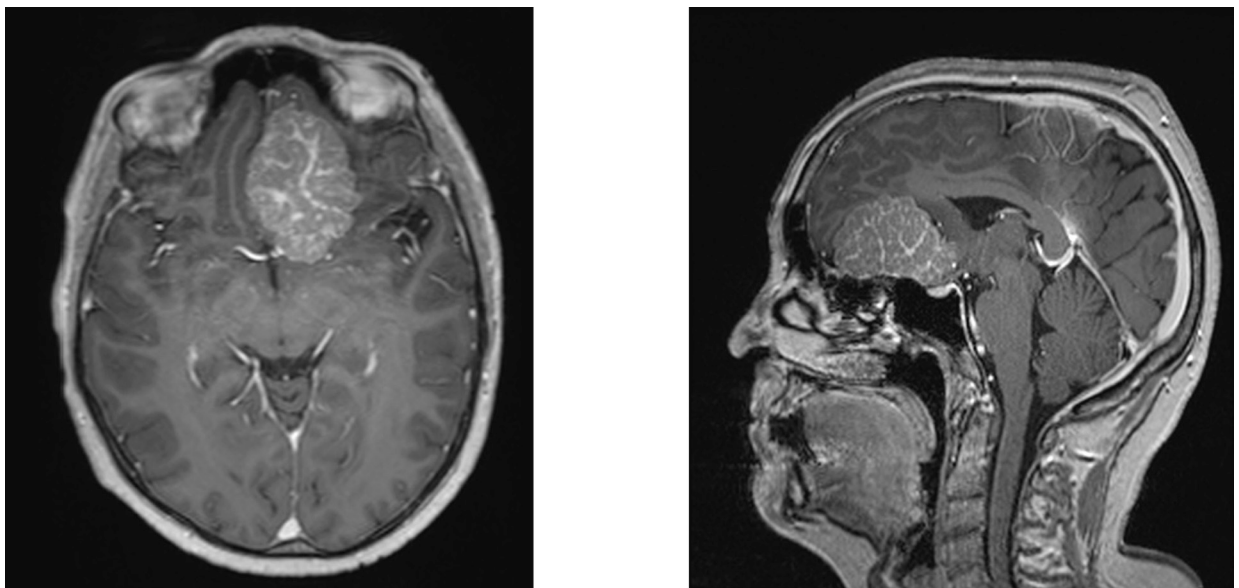


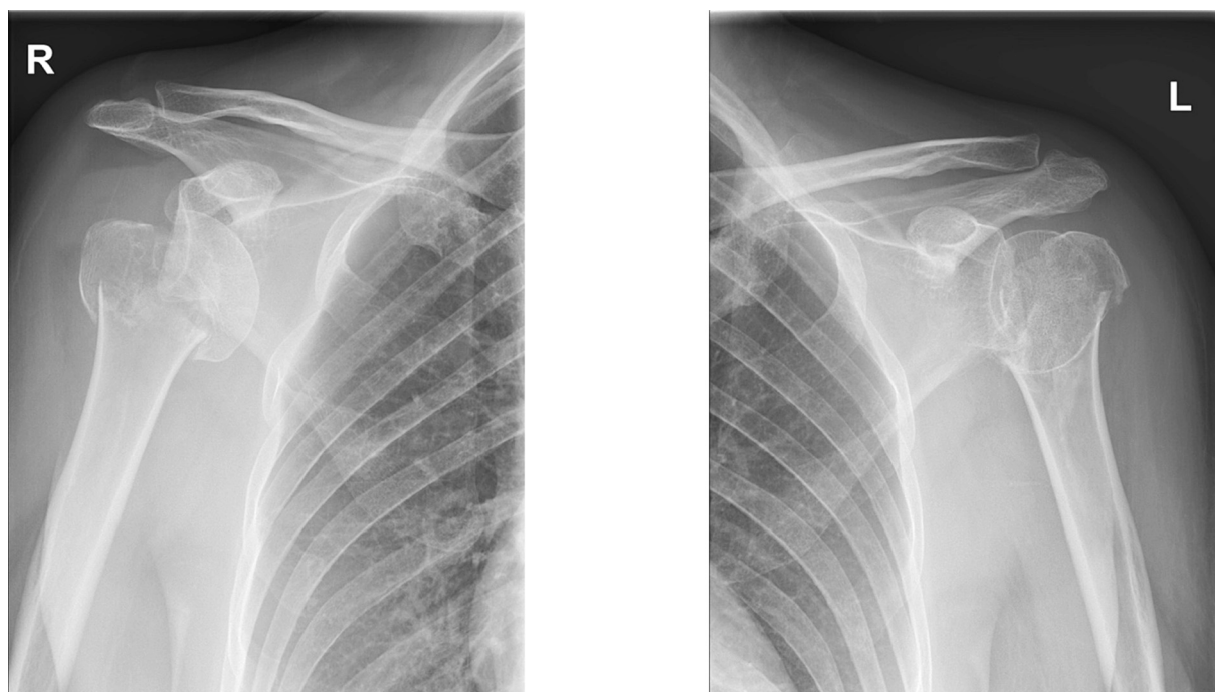
Fig. 4. Postoperative AP view radiographs of right and left shoulder of the patient from Clinical case 1.



**Fig. 5.** Axial and sagittal reconstructions of an MRI scan of the patient from Clinical case 1, 3 months postoperatively.



**Fig. 6.** Axial and sagittal reconstructions of the preoperative MRI scan of the patient from Clinical case 2. An extra-axial frontobasal lesion is noted with heterogenous contrast-enhancement.



**Fig. 7.1 & 7.2.** Preoperative AP view radiographs of the right and left shoulders of the patient from Clinical case 2.

harvest as much galeal flap as possible for the eventual reconstruction of the skull base. A left pterional craniotomy was conducted. A total tumor removal (Simpson II) was achieved. The histological result was WHO Grade I transitional cell meningioma. The postoperative period of the patient was uneventful. A control MRI scan of the patient was conducted 3 months postoperatively (Fig. 5).

In a second stage both shoulders of the patient were operated. The CT scan of the right shoulder confirmed a split fracture of the right proximal humerus. Considering the higher risk for development of avascular necrosis of the humeral head associated with this fracture pattern, a hemiarthroplasty was performed through a deltopectoral approach [13,14]. Open reduction and internal fixation (ORIF) was used as treatment method for the left proximal humerus fracture again through a deltopectoral approach (Fig. 4). At the 3-rd month follow-up Constant-Murley Score of the right shoulder was 62 points, while of the left - the score was 84 points [7].

### Clinical case 2

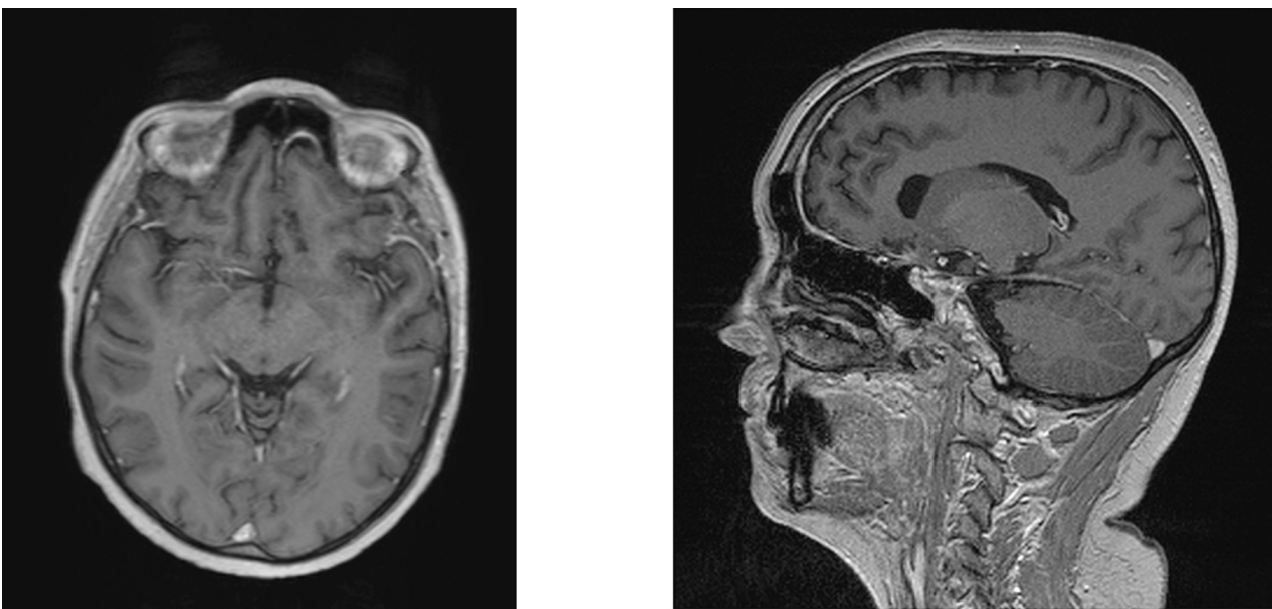
A 47-year-old female patient was admitted after a generalized epileptic seizure. Postictally the patient had restricted range of motion of both shoulders. AP view radiographs of the shoulders confirmed the presence of bilateral 4-part posterior fracture-dislocations according to the Neer classification system [6] (Fig. 7.1 & 7.2). The preoperative cerebral MRI scan of the patient is shown on Fig. 6. Total tumor removal (Simpson I) was achieved through a left pterional craniotomy. The histological result was transitional cell meningioma with a low proliferative activity, Ki-67 less than 1 %.

Shortly afterwards, the patient was referred to the traumatology department. Due to the split fracture with concomitant impaction of the right humeral head Reverse Total Shoulder Arthroplasty was performed as a treatment method (Fig. 9) [13,14]. A few months later, in another hospital, the same procedure was performed for the fracture of the left proximal humerus (Fig. 10). A control MRI scan was conducted 1 month postoperatively (Fig. 8). On her 3-rd month follow-up visit the patient was neurologically intact with preserved mobility of the right shoulder, Constant – Murley Score of 85 points. The patient is currently conducting physiotherapy for the left shoulder as there is still some limitations in the range of motion, Constant – Murley Score of 58 points.

### Discussion

Only 1.1 % of the patients admitted after an epileptic seizure have some kind of bone fracture – 0.5 % due to direct trauma, 0.3 % as a complication of the seizure and in 0.3 % the cause is not defined [8]. In the group lacking evidence of trauma, 6 from the 9 fractures included the humerus [9,11,12].

The shoulder is the most mobile joint in the body which is the reason for its frequent dislocation. In less than 4 % of the cases a posterior dislocation is diagnosed. A bilateral fracture-dislocation of the shoulder is observed in less than 1 % of the cases [3]. Only 35 cases have been documented so far and in 90 % of the cases the cause for this type of fracture was an epileptic seizure [12]. Frequently there is a concomitant vertebral, acetabular, scapular, femoral or pelvic fracture. During an epileptic seizure the usual position of the



**Fig. 8.** Axial and sagittal reconstructions of a contrast-enhanced MRI scan of the patient from Clinical case 2, 1 month postoperatively.

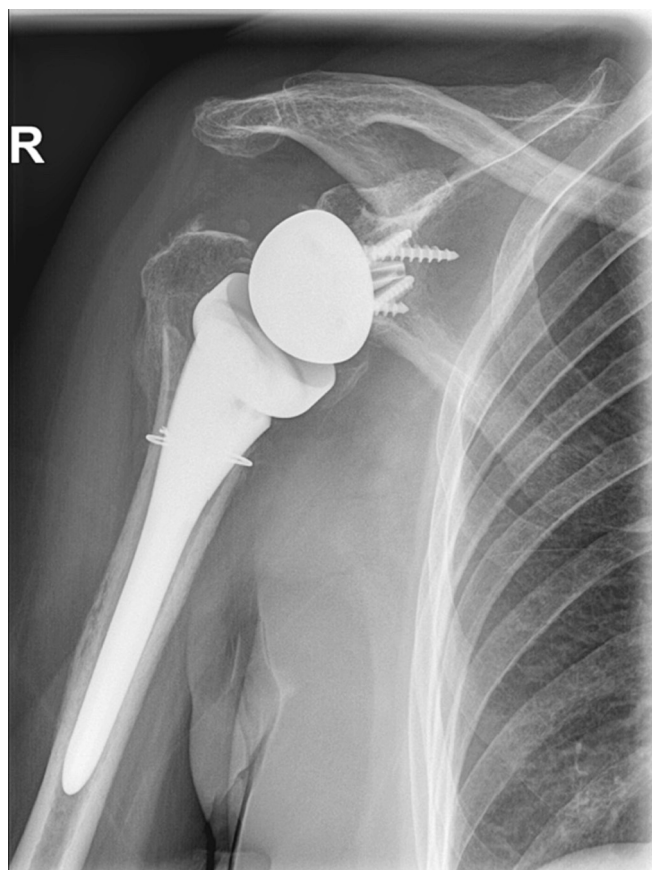


Fig. 9. Postoperative AP view radiographs of the right shoulder.

shoulder is adduction, internal rotation and flexion. The muscle spasm compresses the humerus upwards and posterior to the cavitas glenoidalis [10]. The diagnosis is usually confirmed late due to the lack of evidence for trauma. A characteristic feature for this type of shoulder fracture is the impossible external rotation of the shoulder. Most of the cases reported in the literature (<75 %) were diagnosed late, some even 1 year after the epileptic seizure. Most of these cases were incorrectly diagnosed as a syndrome of the “frozen shoulder”. For this reason, the posterior fracture-dislocation should be included in the differential diagnosis of patients with restricted shoulder movement after epileptic seizure.

We have presented two almost identical cases of patients with meningiomas that presented with epileptic seizures. Fortunately, the bilateral fracture-dislocations of the shoulders were diagnosed early in both cases and the patients were immobilized properly and referred to the traumatology department on time. This was the prerequisite for the full functional preservation in both patients. In both clinical cases there was no fall of the patient during the epileptic seizure and there were no external signs of trauma. Therefore, a high level of suspicion should be kept for any patient with shoulder restriction postictally.

## Conclusion

The shoulder joint is the most commonly dislocated joint in the body with less than 4 % being posteriorly dislocated. Bilateral fracture-dislocation of the shoulders is associated with “triple E syndrome” and 90 % of the cases are related to seizures. The diagnosis is usually delayed due to the lack of signs of trauma. Early diagnosis and proper surgical treatment can maximize the final functional results and patient recovery.

## Declaration of competing interest

We have no conflicts of interest to disclose.



Fig. 10. Postoperative AP view radiographs of the left shoulder.

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