Seizure: European Journal of Epilepsy 70 (2019) 38–42

Contents lists available at ScienceDirect

Seizure: European Journal of Epilepsy

journal homepage: www.elsevier.com/locate/seizure

Short communication

Seizure-induced shoulder dislocations – Case series and review of the literature

Lisa Langenbruch[a,1], Carolin Rickert[b,1], Georg Gosheger[b], Dominik Schorn[b], Benedikt Schliemann[c], Tobias Brix[d], Christian E. Elger[a,e], Sven G. Meuth[a], Stjepana Kovac[a,*]

[a] Department of Neurology, University of Münster, Münster, Germany
[b] Department of General Orthopedics and Tumor Orthopedics, University of Münster, Münster, Germany
[c] Department of Trauma, Hand and Reconstructive Surgery, University of Münster, Münster, Germany
[d] Institute of Medical Informatics, University of Münster, Münster, Germany
[e] Kompetenzzentrum für Epileptologie, Beta Neurologie, Bonn, Germany

ARTICLE INFO

Keywords:
Fracture dislocation
Injury
Epilepsy
Comorbidity

ABSTRACT

Purpose: We aimed to identify clinical characteristics of patients with shoulder dislocations caused by an epileptic seizure.
Methods: In our retrospective analysis, we identified 15 patients, recorded over an 8-year period, who were diagnosed with shoulder dislocations in the setting of a bilateral tonic-clonic seizure.
Results: Patients were almost exclusively male (13/15) and drug-naïve patients suffering their first or second seizure (14/15). Epilepsy was diagnosed in five of these 14 patients after further diagnostic tests, four patients were diagnosed with a provoked or acute symptomatic seizure and five patients with an unprovoked seizure. Treatment with anticonvulsant drugs (AED) was initiated in 10/15 patients after the first seizure, without recommendation for tapering, although long-term treatment was retrospectively judged to be appropriate for only four of those cases. Posterior dislocations – usually rare – were seen in 12/15 patients and often required complex orthopedic interventions.
Conclusions: We conclude that in particular posterior shoulder dislocations are often caused by a first seizure and should always raise the suspicion of an epileptic seizure even in the absence of a clear history. AED treatment likely has a protective effect against this type of injury, even if seizure-freedom is not achieved.

1. Introduction

Seizures and epilepsy are a burden for patients not only due to the debilitating nature of seizures and their unpredictable course, but also because of seizure-related injuries. Injuries occur more often in patients suffering from seizures and epilepsy compared to the general population and have been linked to bilateral tonic-clonic seizures and atonic seizures [1–3], both of which are associated with falls.

The tonic, unnatural and forceful contractions during seizures exert a strain on joints and bones and result in luxations and fractures. A forceful contraction of one muscle group, such as in the tonic phase of a seizure, is able to dislocate the shoulder joint. Anecdotal evidence underlines that the usually rare posterior shoulder dislocations are seen more often in epilepsy patients [4]. A bilateral posterior shoulder dislocation has been claimed to be almost proof of a seizure as the underlying cause [5].

Surprisingly, despite the impact of these injuries on patients' lives, a more detailed investigation into the circumstances of seizure-related shoulder dislocations is lacking. The existing larger studies have mostly focused on the orthopedic patterns of shoulder injuries [6]. We thus set out to examine the clinical characteristics of patients presenting with shoulder dislocations in the setting of an epileptic seizure.

* Corresponding author at: Department of Neurology with Institute of Translational Neurology, University Hospital Münster, Albert-Schweitzer-Campus 1, Gebäude A1, 48149 Münster, Germany.
E-mail addresses: lisamarie.langenbruch@ukmuenster.de (L. Langenbruch), carolin.rickert@ukmuenster.de (C. Rickert), georg.gosheger@ukmuenster.de (G. Gosheger), dominik.schorn@ukmuenster.de (D. Schorn), benedikt.schliemann@ukmuenster.de (B. Schliemann), tobias.brix@ukmuenster.de (T. Brix), christian.elger@betaklinik.de (C.E. Elger), sven.meuth@ukmuenster.de (S.G. Meuth), Stjepana.Kovac@ukmuenster.de (S. Kovac).
[1] These authors contributed equally.

https://doi.org/10.1016/j.seizure.2019.06.025
Received 25 April 2019; Received in revised form 18 June 2019; Accepted 20 June 2019

Table 1
Demographics and clinical characteristics of the study cohort. M: male; F: female; AED: antiepileptic drug; NA: not applicable; ICH: intracerebral hemorrhage; TBI: traumatic brain injury; ORIF: open reduction and internal fixation; DVT: deep vein thrombosis; NSAID: non-steroidal anti-inflammatory drugs; CRF: chronic renal failure; CAD: coronary artery disease; ASA: acetylsalicylic acid.

Pt. No.	Sex	Age	Side	Direction	Dislocation at first seizure	AED intake prior to first dislocation	Epilepsy diagnosed at time of dislocation	Trigger factor	Seizure/epilepsy classification	Orthopedic management	Comorbidities	Concomitant medication
1	M	68	Bilateral	Posterior	No	No	Yes	None	Temporal lobe epilepsy	Left: ORIF + locking plate + Allograft Right: reversed shoulder arthroplasty	DVT, pulmonary embolism	Phenprocoumon
2	M	28	Left	Posterior	Yes	No	No	Meningo-encephalitis	Acute-symptomatic seizure	ORIF + Allograft	None	None
3	F	34	Right	Anterior	Yes	No	Yes	Post-partum	Genetic generalized epilepsy	Closed reposition	Glaucoma	None
4	M	54	Left	Posterior	Yes	No	Yes	None	Temporal lobe epilepsy	Closed reposition	Arterial hypertension	Anti-hypertensive drugs
5	M	43	Right	Posterior	Yes	No	No	Not known	Unprovoked seizure	Open reposition + Allograft	NSAID overuse	Diclofenac
6	M	33	Left	Posterior	Yes	No	No	Sleep deprivation	Provoked seizure	Closed reposition	None	None
7	M	38	Left	Posterior	No	Yes	NA (Known epilepsy)	None	Focal epilepsy due to astrocytoma	Closed reposition	None	None
8	M	38	Right	Posterior	Yes	No	No	Cerebral biopsy	Acute symptomatic seizure	ORIF + locking plate	None	None
9	M	41	Left	Anterior	Yes	No	Yes	None	Focal Epilepsy due to ICH or TBI	Closed reposition	None	None
10	M	23	Left	Anterior	Yes	No	No	None	Unprovoked seizure	Closed reposition	None	None
11	M	33	Bilateral	Posterior	Yes	No	No	Systemic infection	Provoked seizure	Left: ORIF + Allograft Right: ORIF + Allograft	CRF	Anti-hypertensive drugs, Calcitriol, Sodium hydrogen carbonate
12	M	67	Left	Posterior	No	No	Yes	Not known	Epilepsy of unknown classification	Closed reposition	Arterial hypertension, CAD	Anti-hypertensive drugs
13	M	45	Right	Posterior	Yes	No	No	Not known	Unprovoked seizure	ORIF + locking plate	CAD	Esomeprazole
14	M	61	Bilateral	Posterior	Yes	No	No	None	Unprovoked seizure	Left: ORIF + locking plate, right: closed reposition	Arterial hypertension	Anti-hypertensive drugs
15	F	81	Bilateral	Posterior	Yes	No	No	None	Unprovoked seizure	reversed shoulder arthroplasty bilateral	Arterial hypertension, CRF	Anti-hypertensive drugs, Pantoprazole, Simvastatin, Mirtazapine, ASA, Ropinirole, Salmeterol/Fluticasone

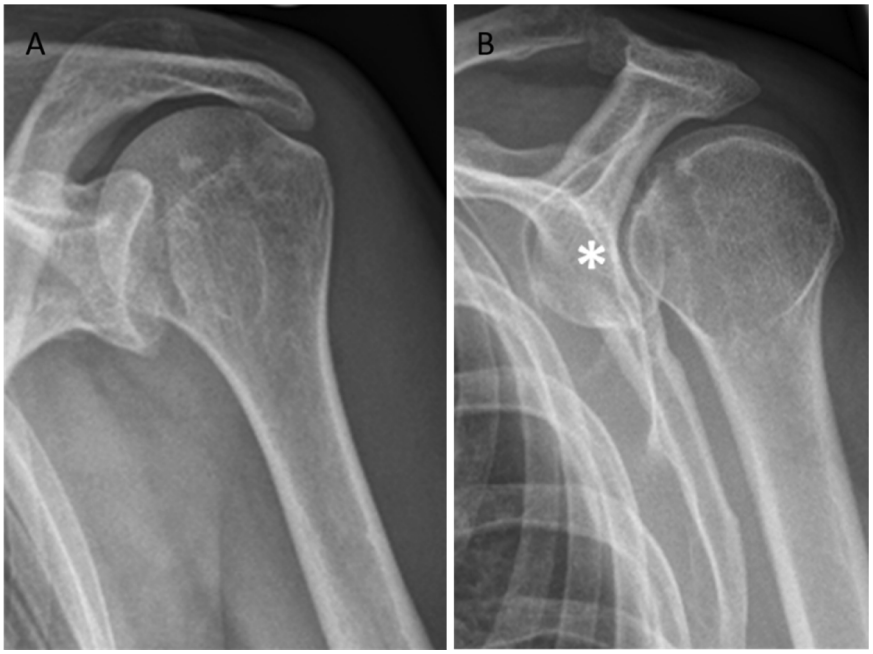

Fig. 1. Posterior shoulder dislocation (patient 12), in antero-posterior (A) and lateral view (B). The white asterisk in A indicates the empty glenoid cavity.

2. Material and methods

We screened all cases of the Department of Neurology at the University Hospital Münster, Germany, between 01.01.2010 and 15.11.2018, for the disease specific code (International Classification of Diseases; ICD-10) for shoulder dislocation (S43). We then manually identified those patients who were admitted due to an acute shoulder dislocation in the course of an epileptic seizure. Additionally, we included patients who presented to the Surgery Department for an acute shoulder dislocation and received a neurological consultation because of a related epileptic seizure. These patients were identified by screening for the ICD-10 disease codes for epilepsy (G40) and seizure (R56) and by manual data review. Between 01.01.2010 and 15.11.2018, we identified 4416 patients with an epilepsy or seizure code in the Neurology Department and 1263 patients with a shoulder dislocation code in the Surgery and Orthopedics department. To assess the proportion of a seizure-induced dislocation among all acute shoulder dislocations, we manually identified all acute dislocation cases presenting from 01.01.2017 to 31.12.2017 and calculated an estimate for the total period. Likewise, we manually assessed the number of patients presenting with an acute bilateral tonic-clonic seizure between 01.01.2017 and 31.03.2017 for extrapolation.

The resulting 15 patients with a seizure-induced shoulder dislocation were evaluated independently by two epileptologists (LL and SK), who were blinded to each other's evaluation. The independent raters defined whether the diagnostic criteria for epilepsy were fulfilled at the time the patient presented with the seizure-induced shoulder dislocation. If no epilepsy could be diagnosed, the two raters had to classify whether the seizure was considered an acute symptomatic seizure, a provoked seizure, or an unprovoked seizure. In addition, they independently assessed whether long-term anticonvulsive medication was justified in these cases.

3. Results

In the selected study period, we identified 15 patients with a first acute shoulder dislocation. As described above, the total number of patients presenting with an acute bilateral tonic-clonic seizure was estimated as 1634 and the total number of patients with an acute shoulder dislocation regardless of etiology as 290. Thereby, we reached a proportion of 0.92% of shoulder dislocations among patients presenting with a seizure. Seizures accounted for 5.2% of acute shoulder dislocations. The patients' clinical data is summarized in Table 1. Briefly, the majority of patients were male (87%), the median age was 41 years (range 23–81 years). 14/15 (93%) patients experienced their index shoulder dislocation due to their first or second seizure and prior to any diagnosis of epilepsy. In one patient only, the epilepsy diagnosis had been established prior to the shoulder dislocation. This patient had received treatment with Levetiracetam. No other patient was treated with anticonvulsants at the time of the shoulder dislocation.

The two independent raters (LL and SK) yielded a 100% agreement on the diagnoses of epilepsy, acute symptomatic seizures, provoked seizures, or unprovoked seizures. Five patients without prior diagnosis fulfilled the diagnostic criteria for epilepsy at the time of seizure-related shoulder dislocation [7]. In four patients, a provoked or acute symptomatic seizure was diagnosed. Five patients suffered an unprovoked first seizure and the diagnostic work-up did not indicate an increased risk of seizure recurrence. In 10 out of 15 patients (67%) anticonvulsive medication was initiated after seizure-related shoulder dislocation. Discontinuation of anticonvulsant medication was not planned or attempted in any of the patients who had been started on the medication – even in the setting of acute symptomatic seizures. According to the retrospective assessment of the two independent raters, long-term treatment was not indicated for six out of 10 patients who had been started on anticonvulsant medication after the shoulder dislocation.

The majority of patients (12/15) suffered from a posterior shoulder dislocation (as shown in Fig. 1). Four out of 15 dislocations were bilateral. Eight (53%) patients required operative treatment of the shoulder dislocation. (see Table 1).

4. Discussion

Injuries, especially dislocations and fractures, substantially add to the burden of disease in patients with epilepsy. The literature on shoulder dislocations and epileptic seizures is heterogenous (see Table 2) [4,6,8–13]. Posterior dislocations are reported to be caused by seizures in 24–47%. Other common causes include falls, motor vehicle accidents, sports injuries and other injuries. It has been speculated that the axial force of the adducted and internally rotated arm during the tonic phase of a seizure is the main culprit for shoulder dislocations in

Table 2
Review of the literature on seizures and shoulder dislocation.

Author and year of publication	Type of Study	Number of patients	Number of shoulder dislocations	Direction of dislocation	Proportion of seizure-related dislocations
Dimon 1967	Case series	25	25	Posterior	24%
Bühler 2002	Case series	26	34	17 anterior, 17 posterior	NA (epilepsy cohort)
Kwon 2010	Population-based case control study	1831 epilepsy patients, 4273 controls	NA	Not reported	NA (0.6% dislocations of upper limb in epilepsy patients vs. 0.3% in controls)
Robinson 2011	Case series	112	120	Posterior	31%
Rouleau 2012	Systematic Review of the literature	477	547	Posterior	39%
Thangarajah 2015	Case series	33	49	36 anterior (73%), 8 posterior (16%), 5 multidirectional (10%)	NA (epilepsy cohort)
Basal 2018	Systematic Review of the literature	353	353	Posterior (locked)	47%
Kelly 2019	Systematic Review of the literature	182	216	Posterior	38%

this setting [14]. In our study, 5.2% of acute shoulder dislocations – both anterior and posterior - were caused by a seizure. As expected, the proportion of patients with a shoulder dislocation among all patients presenting with a bilateral tonic-clonic seizure is small (0.92%) and in line with the existing literature (see Table 2) [12].

We were able to show that shoulder dislocations in the setting of bilateral tonic-clonic seizures are mostly seen in the context of the first occurring seizure – and not in patients with an established diagnosis of epilepsy. In fact, we only saw one patient who suffered a seizure-related first shoulder dislocation despite anticonvulsive medication. This strongly points to a protective effect of anticonvulsive treatment against this type of injury. We speculate that one reason is the less violent nature of seizures in patients who are under anticonvulsive treatment. From a clinical perspective, this would strongly encourage the use of anticonvulsant drugs in the setting of bilateral tonic-clonic seizures, even if seizure freedom may not be achieved. We believe that the potential prevention of serious shoulder injury is an additional reason for the initiation of anticonvulsive treatment in patients who are at risk for recurring seizures and who are reluctant to start medication.

A diagnosis of epilepsy according to current ILAE criteria [7] was established for one third of the patients after their seizure-related shoulder dislocation. Together with the patient for whom a diagnosis of epilepsy was established prior to the seizure leading to a shoulder dislocation, 40% of the patients with shoulder dislocation were thus diagnosed with epilepsy. The remaining 60% suffered an acute symptomatic, provoked or unprovoked first seizure. Their seizure recurrence risk would have been formally calculated as below 60%, and therefore initiation of anticonvulsant treatment would not have been necessary. In our cohort, 11/15 (73%) patients received anticonvulsants after the shoulder dislocation, including six patients who did not meet the diagnostic criteria for epilepsy. These patients would usually have qualified for a watchful strategy if the seizure had not resulted in an injury. Previous work indicates a higher risk of recurrent shoulder instability in patients with uncontrolled seizures [11,13]. In order to reduce the seizure recurrence risk in the post-operative period and to improve orthopedic outcome, one may support this clinical management, i.e. starting anticonvulsive treatment even in the absence of an established diagnosis of epilepsy. However, this should prompt the treating physician to reevaluate the treatment and consider tapering anticonvulsant drugs after a certain period.

Previous studies have shown that fractures in general occur more often in patients taking enzyme-inducing medication [15]. This does not seem to hold true for shoulder dislocations or fracture dislocations, since none of our patients received enzyme-inducing medication. In addition, relevant comorbidities which may contribute to bone instability were scarce. In particular, osteoporosis was not mentioned as a comorbidity in the clinic letters of any of our patients at the time of the shoulder dislocation. In this context, we are surprised that the patients suffering a seizure-related shoulder injury do not fit the category 'older epilepsy patient with multiple comorbidities or polypharmacy' as may have been expected. In contrast, we found a clear predominance of relatively young men (87%). Taking these two aspects together, muscle strength must be considered a more relevant factor in the pathophysiology of shoulder dislocation than osteopenia.

One limitation of our study may be the reliance on diagnosis codes for patient identification. Since a shoulder dislocation and a seizure constitute relevant comorbidities that will lead to consultation of a neurologist and surgeon/orthopedic respectively, we believe that the risk of underreporting is low.

5. Conclusion

We here show that patients with a seizure-related shoulder dislocation are – in the vast majority of cases – male and drug naïve patients suffering their first bilateral tonic-clonic seizure without a prior history of epilepsy. Clinicians should be aware that some of these

patients do not fulfill the diagnostic criteria for epilepsy and long-term anticonvulsive treatment may therefore not be required. Posterior dislocations are overrepresented in this patient group, hence this injury should always raise the suspicion of an epileptic seizure.

Author contributions

SK, LL and CR conceived the study and defined the concept. TB and LL performed the database query, LL and CR screened the clinic letters and analyzed the data. LL wrote the initial draft of the manuscript and prepared the tables for the manuscript, CR prepared the figure. SK extensively revised the manuscript for intellectual content. SGM, CEE, DS, BS and GG advised on the study concept and interpreted some of the data. All authors contributed to the concept of the work and writing the manuscript, critically discussed the data and approved the version to be published.

Acknowledgements

This work was supported by the medical Faculty of the University of Münster (fellowship to SK) and the Ursula von Euch Stiftung (fellowship to SK).

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

References

[1] Wirrell EC. Epilepsy-related injuries. Epilepsia 2006;47(Suppl 1):79–86.
[2] Baraldi S, Farrell F, Benson J, et al. Drop attacks, falls and atonic seizures in the Video-EEG monitoring unit. Seizure 2015;32:4–8.
[3] Lawn ND, Bamlet WR, Radhakrishnan K, et al. Injuries due to seizures in persons with epilepsy: a population-based study. Neurology 2004;63:1565–70.
[4] Kelly MJ, Holton AE, Cassar-Gheiti AJ, et al. The aetiology of posterior gleno-humeral dislocations and occurrence of associated injuries. Bone Jt J 2019;101-B:15–21.
[5] Finelli PF, Cardi JK. Seizure as a cause of fracture. Neurology 1989;39:858–60.
[6] Dimon JH. Posterior dislocation and posterior fracture dislocation of the shoulder: a report of 25 cases. South Med J 1967;60:661–6.
[7] Fisher RS, Acevedo C, Arzimanoglou A, et al. ILAE official report: a practical clinical definition of epilepsy. Epilepsia 2014;55:475–82.
[8] Basal O, Dincer R, Turk B. Locked posterior dislocation of the shoulder: a systematic review. EFORT Open Rev 2018;3:15–23.
[9] Rouleau DM, Hebert-Davies J. Incidence of associated injury in posterior shoulder dislocation: systematic review of the literature. J Orthop Trauma 2012;26:246–51.
[10] Robinson CM, Seah M, Akhtar MA. The epidemiology, risk of recurrence, and functional outcome after an acute traumatic posterior dislocation of the shoulder. J Bone Jt Surg Am 2011;93:1605–13.
[11] Bühler M, Gerber C. Shoulder instability related to epileptic seizures. J Shoulder Elbow Surg 2002;11:339–44.
[12] Kwon C-S, Liu M, Quan H, et al. The incidence of injuries in persons with and without epilepsy–a population-based study. Epilepsia 2010;51:2247–53.
[13] Thangarajah T, Lambert S. The management of recurrent shoulder instability in patients with epilepsy: a 15-year experience. J Shoulder Elbow Surg 2015;24:1723–7.
[14] Rockwood CA. The shoulder. 3rd ed Philadelphia, [London]: Saunders; 2004.
[15] Nicholas JM, Ridsdale L, Richardson MP, et al. Fracture risk with use of liver enzyme inducing antiepileptic drugs in people with active epilepsy: cohort study using the general practice research database. Seizure 2013;22:37–42.