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ORIGINAL ARTICLE / KLİNİK ÇALIŞMA

Epilepsy Related Shoulder Dislocation: Demographic and Clinical Analysis of 21 Patients

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Epilepsi ile İlişkili Omuz Dislokasyonları: 21 Olgunun Demografik ve Klinik Analizi

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Summary

Objectives: There are multiple consequences of epilepsy that impact quality of life. While psychiatric comorbidities are some of the most commonly studied aspects, orthopedic complications are less frequently documented. The focus of the present study was shoulder dislocation related to epilepsy, and, unlike that of previous studies, the aim was to identify epilepsy-dependent risk factors in the development of joint disability.

Methods: Twenty-one patients of the Cerrahpaşa Faculty of Medicine epilepsy outpatient clinic were retrospectively identified as having had epilepsy-related shoulder dislocation between 1990 and 2013. Probable risk factors were evaluated in terms of demographic and clinical features. A report published by the International League Against Epilepsy (ILAE) in 2010 was used to classify seizure and epilepsy syndromes.

Results: Median age at onset of epilepsy was 20 years, while median age at shoulder dislocation was 24. The seizure type that most commonly led to dislocation was bilateral convulsive (81%). Initial dislocations (IDs), those following any of the first 3 seizures, were identified in 45% of the population. Patients with ID tended to be older than those with late dislocation (LD) ($p<0.001$). Rate of recurrent dislocation was found to be higher in patients with early onset epilepsy, compared to those older than 20 years at diagnosis ($p<0.05$).

Conclusion: While, as expected, shoulder dislocation was most commonly caused by bilateral convulsive seizures, factors thought to impact severity of dislocation including seizure frequency, occurrence of status epilepticus (SE), response to treatment, and use of antiepileptic drugs (AEDs) that affect bone metabolism were found to have no influence on temporal development, affected side (bilateral or unilateral), or recurrence rate.

Keywords: Epilepsy; epilepsy-related complications; joint instability; seizure; shoulder dislocation.

Özet

Amaç: Epilepsi, bireyin hayatını çok yönlü etkileyen bir hastalıktır. Çalışmalarda sıklıkla epilepsinin yol açtığı psikiyatrik sorunlar ön plana çıkmıştır, ancak ortopedik komplikasyonlara çok fazla değinilmemiştir. Çalışmamızda epilepsi ile ilişkili omuz dislokasyonları ele alınmış olup; daha önce ortopedi alanında yayınlanan çalışmalardan farklı olarak, bu eklem hasarına yol açabilecek epilepsi ile ilişkili risk faktörlerinin anlaşılması hedeflenmiştir.

Gereç ve Yöntem: Çalışmamızda, 1990–2013 yılları arasında İstanbul Üniversitesi Cerrahpaşa Tıp Fakültesi, Nöroloji Anabilim Dalı'nın Epilepsi Polikliniği'ne başvuran hastalar geriye dönük olarak değerlendirilmiş ve 21 epilepsi ile ilişkili omuz dislokasyonu olgusu saptanmıştır. Hastalara ilişkin demografik ve klinik özellikler olası risk faktörleri kapsamında ele alınmıştır. Epilepsi nöbetleri ve sendromları ILAE-2010'a göre sınıflandırılmıştır.

Bulgular: Çalışmamızda; epilepsinin başlangıcı median 20 yaş, omuz dislokasyonunun gelişimi ise median 24 yaş olarak bulunmuştur. Omuz dislokasyonuna en sık neden olan nöbet tipi iki taraflı konvülzif nöbetlerdir (%81). Hastaların %45'inde ilk üç nöbette (erken dönem) dislokasyon gelişmiş olup, bu gruptaki hastaların geç dislokasyon gelişen gruba göre daha yaşlı olduğu gözlenmiştir ($p<0.001$). Omuz dislokasyonlarındaki tekrar oranı ise; erken yaşta dislokasyon görülen grupta, 20 yaş sonrasında tanı alan gruba göre anlamlı düzeyde artmıştır ($p<0.05$).

Sonuç: İki taraflı konvülzif nöbetler en sık omuz dislokasyonuna neden olan epilepsi tipidir. Ancak sık nöbet tekrarı, status varlığı, bağ dokusu ve kemik metabolizmasını etkileyebilecek antiepileptik ilaçların kullanımı ve tedavi yanıtının azlığı gibi faktörler incelendiğinde; dislokasyonların daha erken dönemde, iki yanlı veya sık tekrarlarla seyretmesi beklenirken, çalışmamızda bu değişkenlere ait anlamlı bir farklılık saptanmamıştır.

Anahtar sözcükler: Epilepsi; epilepsi ile ilişkili komplikasyonlar; eklem instabilitesi; nöbet; omuz dislokasyonları.

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Introduction

Several complications may occur with epileptic seizures. Shoulder dislocation related to epileptic seizures is primarily described from an orthopedic approach. The glenohumeral joint is the most mobile component of the shoulder joint complex and is therefore the most commonly dislocated joint in the body. Static (ligaments, labrum, capsule, negative pressure), dynamic (rotator cuff muscles), and proprioceptive factors collectively support joint stability. [1-3] While the most common etiology is traumatic injury, in the absence of a history of trauma, joint instability is usually caused by violent muscle contractions in patients with epileptic seizure or in those who have been exposed to electric shock.[4,5]

Direction and radiographic measurement of instability are studied in order to determine rate of recurrence and inform choice of therapy.[6-8] Neurologic aspects are otherwise frequently overlooked, and likely risk factors, including type of seizure and epileptic syndrome, presence and frequency of status epilepticus (SE), duration of disease, choice of antiepileptic drug (AED), treatment response, presence of additional systemic disorders, and family history are merely documented. The demographic and clinical analysis of 21 patients diagnosed with epilepsy-related shoulder dislocation is described in the present report.

Materials and Methods

Included in the present retrospective study were 21 patients identified as having experienced epilepsy-related shoulder dislocation between 1990 and 2013. Patient records maintained by a senior neurologist (S.N.Y.) experienced in treating epilepsy and related disorders were analyzed, and demographic and clinical findings were documented. Neither obtaining approval from the ethics committee nor informed consent were deemed necessary.

No pediatric patients were included, as follow-up in the epilepsy outpatient clinic is only conducted with patients older than 15 years. However, retrospective design provided the documentation of reported shoulder instability that had occurred before the age of 15. Diagnosis of shoulder dislocation was based on data obtained from patient registries. While each patient had orthopedic consultation, diagnostic measurements and details of radiologic investigations were not included in the documentation. Data regarding traumatic shoulder dislocation not caused by seizure were excluded from the present study.

Age at onset of initial seizure, age at first shoulder dislocation (if remembered precisely, otherwise age at dislocation during any of the first 3 seizures), seizure type and phenomenology, history of SE, types of medication, treatment response, neuroimaging features, accompanying systemic diseases, and family history were used to identify the relationship between dislocation and patient characteristics. An International League Against Epilepsy (ILAE) report published in 2010 was used to classify seizure and epilepsy syndrome.[9]

Median age at epilepsy onset (20 years) was utilized in order evaluate the effect of aging and impact of disease duration. Patients were also grouped according to approximate seizure frequencies: daily, frequent (at least 1 per month), rarely occurring (at least 1 per year), occasional (1 or 2 in a period of 5 years), and remission (no seizure in over 5 years).

To determine the relationship between the effects of high seizure frequency, long-term use of AEDs, and time to development of shoulder dislocation, patients were analyzed in 2 groups: patients with initial dislocation (ID), occurring during the any of the first 3 seizures, and late dislocation (LD), occurring after multiple seizures.

Poor response to treatment was defined as the occurrence of seizure without any provoking factor. Good response was defined as freedom from seizures for more than 6 months.

Continuous seizure activity lasting longer than 5 minutes or recurrent seizures exceeding 5 minutes with no intervening interictal state were considered SE. Non-convulsive SE was not included in documentation.

Statistical analysis

Descriptive statistical analyses of data, including percentages, median, and frequency distribution, were calculated with SPSS software (version 15.0; SPSS Inc., Chicago, IL, USA). Significance of numerical data was calculated using Student's t-test when distributed homogeneously and with Mann–Whitney U test when distributed heterogeneously. Chi-square test was used for non-parametric data. A p value of <0.05 was considered statistically significant. Pearson linear correlation was used to analyze correlation between demographic/clinical features and epilepsy-related dislocation.

Results

Fifteen male and 6 female patients were diagnosed with epilepsy-related shoulder dislocation (male/female ratio, 2.5:1). Actual mean age was 33 years (range 16–71 years). Median age at onset of first seizure was 20 years (range 10–68 years), and median age at shoulder subluxation was 24 years (range 14–71 years; Table 1).

Type of seizure and epileptic syndrome

Thirteen patients (62%) had focal epilepsy, 6 (29%) had generalized epilepsy, and 2 had unclassified bilateral convulsive seizures. Of the generalized epilepsy patients, 2 had juvenile absence epilepsy, 1 had juvenile myoclonic epilepsy, and 3 had epilepsy with generalized tonic-clonic seizure alone. Structural lesion was revealed on cranial magnetic resonance imaging in 38% of patients with focal epilepsy, 2 of whom had hippocampal sclerosis, 2 of whom had focal cortical atrophy, 2 of whom had cortical dysplasia, and 2 of whom had ischemic vascular lesion.

The type of seizure that most commonly led to shoulder dislocation (81%) was bilateral convulsive seizure. Three patients experienced initial shoulder dislocation during focal asymmetric tonic postural seizure.

Seizure frequency

Two patients (9.6%) with cortical dysplasia experienced bilateral convulsive seizure up to several times per day, comprising the daily group. Five patients (24%) experienced seizure 1—3 times per month (frequent group), 7 patients (33%) experienced seizure 1—3 times per year (rare group). Five patients (57%) experienced seizure as a result of lack of compliance (occasional group), and 2 patients (9.6%) were seizure-free (remission group) (Table 2).

Antiepileptic drug use

Fifteen patients (71%) received monotherapy, and 5 received polytherapy of 2 or 3 drugs. Two monotherapy patients were in remission. One patient refused treatment due to rare seizures. The most frequently prescribed drugs were levetiracetam (LEV, n=7); valproic acid (VPA, n=5); oxcarbazepine (OXC, n=5); carbamazepine (CBZ, n=4); lamotrigine (LTG, n=3); and diphenylhydantoin (DPH, n=2).

Presence of status epilepticus

SE was verified in 8 patients (38%), and had never been experienced by 9 (43%). A definitive decision regarding the SE status of 4 patients could not be made due to insufficient data.

Characteristics of shoulder dislocations

Five patents experienced ID during the 1st seizure, 3 patients during the 2nd, and 1 during the 3rd, with 45% of the population experiencing ID during their first 3 seizures. The rest experienced shoulder instability after multiple seizures, with 7 patients experiencing instability between the 1st and 4th year after onset of seizure, 3 patients experiencing instability after 5 years, and 2 patients experiencing instability after 10 years.

Median age at first dislocation was 24 years (range 14–68 years), and 6 patients were older than 30 years. Age at epilepsy onset was compared between patients whose ID occurred during any of their first 3 seizures (ID group) and patients whose ID occurred only after multiple seizures (LD group) (Table 3). The ID group was found to have been older at epilepsy onset ($p < 0.001$). Differences between the ID and LD groups regarding gender, type of epilepsy, type of seizure at dislocation, occurrence of SE, seizure frequency, and treatment responses were not significant ($p > 0.05$).

Dislocation severity and recurrence

Recurrence rate of dislocations was found to be higher in patients with early onset epilepsy (83%), compared to those aged older than 20 years at diagnosis (22%; $p<0.05$). Seizure frequency did not differ between patients aged younger or older than the median 20 years at epilepsy onset ($p = 0.174$).

Supposedly high-risk factors for recurrent dislocations including time to development of dislocation, presence of SE, seizure frequency, and bilateral convulsive seizures were not different between the groups ($p > 0.05$).

Eight patients had left-sided, and 4 patients had bilateral dislocations. Twelve patients (57%) experienced recurrent dislocations during multiple seizures, as well as spontaneously. Unilateral (n=17) and bilateral (n=4) dislocations were compared, taking gender, duration of epilepsy, age at onset of epilepsy, type of epilepsy, seizure type at dislocation (focal, generalized, or SE), seizure frequency, history of SE, and treatment responses into consideration. No significant differences were determined ($p > 0.05$).

Twelve patients experienced recurrent shoulder disloca-

Table 1. Demographic and clinical characteristics

Patient age in years/sex	Epilepsy onset-age (years)	Age at dislocation (years)	Type of epilepsy syndrome	Type of seizures at dislocation	Aura	Seizure frequency	History of SE	Cranial MRI	Site of dislocations	Recurrence	AED
32/Female	15	23	JAE	BC	None	Rare	–	N	Bilateral	+	VPA
49/Male	33	33	IFE	FS	+	Rare	–	N	Unilateral	–	CBZ, LEV
37/Male	34	34	IFE	BC	+	Frequent	+	N	Unilateral	–	LTG
27/Male	18	21	JME	BC	None	Occasional	Unknown	MTS	Unilateral	+	VPA
47/Female	44	44	Structural etiology	BC	None	Remission	+	N	Unilateral	–	LEV
30/Male	20	27	Structural etiology	BC	+	Frequent	+	Cortical dysplasia	Unilateral	+	OXC, DPH
24/Male	23	24	JME	BC	None	Remission	–	Unknown	Unilateral	–	VPA
20/Male	15	15	IFE	FS	+	Occasional	–	N	Unilateral	+	CBZ
46/Male	21	22	Structural etiology	BC	+	Frequent	–	MTS	Bilateral	+	CBZ, LEV
20/Male	13	15	IFE	BC	+	Frequent	–	N	Bilateral	+	LEV
38/Male	32	32	IFE	BC	None	Occasional	+	N	Unilateral	+	OXC
71/Male	68	68	IGE	BC	None	Rare	Unknown	CVD sequel	Unilateral	Unknown	LEV
30/Male	14	18	IGE	PS	+	Daily	+	Cortical dysplasia	Unilateral	Unknown	LEV, OXC, DPH,
33/Male	17	29	Unknown	BC	None	Frequent	+	Focal atrophy	Unilateral	Unknown	LTG
21/Male	16	19	IFE	FS	None	Frequent	–	N	Unilateral	+	CBZ
29/Male	25	25	IFE	BC	None	Occasional	–	Focal atrophy	Unilateral	-	OXC
24/Female	12	18	JAE	BC	None	Occasional	+	N	Unilateral	+	VPA, LTG
33/Female	10	27	Structural etiology	BC	+	Occasional	Unknown	CVD sequel	Unilateral	+	OXC
35/Female	20	22	IGE	BC	None	Rare	Unknown	N	Unilateral	+	None
16/Female	11	14	IGE	BC	None	Occasional	–	N	Unilateral	+	VPA
37/Male	37	37	Unknown	SE	None	Rare	+	N	Bilateral	–	LEV

SE= status epilepticus; AED= antiepileptic drugs; M= male; F= female; JAE= juvenile absence epilepsy; JME= juvenile myoclonic epilepsy; IGE= idiopathic/genetic generalized epilepsy; IFE: idiopathic/genetic focal epilepsy; BC= bilateral convulsive seizure; PS= focal seizure; SE= status epilepticus; N= normal imaging; MTS= mesial temporal sclerosis; CVD= cerebrovascular disease; VPA= valproic acid; CBZ= carbamazepine; OXC= oxcarbazepine; LEV= levetiracetam; LTG= lamotrigine; DPH= diphenylhydantoin.

Table 2. Seizure frequencies of the patient group

	Daily	Frequent: 1–3 seizures/month	Rare : 1–3 seizures/year	Occasional seizures*	Remission
Number of patients	1	6	7	5	2

*This group of patients has experienced seizures usually as a result of lack of compliance with treatment.

Table 3. Age-dependent comparison of early and late dislocations

		Onset age of initial seizure	Age at the first subluxation	p
Early dislocation (n=9)	Median (IQR)	33 (24–40.5)	33 (24.5–40.5)	<0.001
Late dislocation (n=12)	Median (IQR)	15 (12.25–17.5)	20 (15.75–26)	

tion, 4 (19%) of whom had daily seizures. However, the remaining 8 (38%) had only 1 or 2 seizures per year (rare group) and no history of SE. The recurrence rate of shoulder dislocation in patients with convulsive SE was 50%. Only 4 of the 12 patients with recurrent dislocation had undergone an operation, as the others had refused surgical treatment.

Systemic disorders and family history

Among the 21 patients, 2 had hypertension and diabetes mellitus, and 1 had ankylosing spondylitis. One patient had epilepsy, and 1 had spontaneous shoulder dislocation in first-degree relatives.

Discussion

The humeral head has a large surface area and loose contact with the glenoid surface. This disproportion and capacity for multiaxial movement can result in injuries when exposed to trauma or the powerful muscle contractions seen in epilepsy. The humeral head can be dislocated in any direction, most commonly anteriorly and posteriorly, less commonly inferiorly and superiorly, of the glenoid.[1,2] While anterior dislocations comprise 85% of shoulder dislocations, posterior dislocations (comprising less than 5%) are considered to be pathognomonic for epileptic seizures.[3,4] Data regarding surgical relevancies and orthopedic features of dislocations were beyond the scope of the present study.

In accordance with the findings of others,[5,6] epilepsy-related shoulder dislocation was more prevalent in males, with a proportion of 2.5:1. Interestingly, in 45% of the present population, dislocations developed during the first 3 seizures experienced (initial seizures). A strong correlation was found regarding age at epilepsy onset among the ID and LD groups; those who experienced shoulder instability following initial seizure tended to be older. This finding could be associated with decreased elasticity and compliance of the shoulder joint complex against traumas with aging.

Increased seizure activity and prolonged convulsion time, as seen in SE or in restrained patients during the dynamic ictal period, are frequently emphasized risk factors for shoulder instability.[7,8] However, type of seizure and epilepsy syndrome, disease duration, occurrence of SE, and AED response had no influence on temporal development, bilateral or unilateral involvement, or recurrence of dislocation in the present population.

In contrast, good disease outcome did not prevent patients from developing shoulder instability or recurrence. Risk of dislocation was significantly higher in the year following the first epileptic seizure. In addition, rate of recurrence was higher in patients with early onset epilepsy than in those older than 20 years at diagnosis. However, a correlation with seizure frequency, type of seizure, or epilepsy syndrome was not observed. While therapy-resistant epilepsy is supposedly a high-risk factor for shoulder instability, no significant difference was found between time to dislocation and rate of recurrence, severity of seizure frequency, and/or poor response to AED therapy.

Collagen tissue disorders such as Dupuytren's contracture, frozen shoulder, Peyronie's disease, and fibromas have been linked to long-term use of AEDs, particularly older generics such as phenobarbital, DPH, and CBZ.[10] In the present population, 9 patients (43%) were on OXC or CBZ, 2 of whom were on combined treatment with DPH, with no barbiturate. No differences in median age at shoulder dislocation

were found among patients taking the older generics and those taking LTG, LEV, and VPA. LEV was prescribed for 2 patients with ankylosing spondylitis who experienced dislocation at first seizure before the drug was administered, and a correlation could not be made.

In conclusion, the most common type of seizure leading to shoulder dislocation was bilateral convulsive seizure, as expected. However, factors thought to have an impact on severity of dislocation did not affect time of development, side (bilateral or unilateral), or recurrence rate of shoulder dislocation in the present population. Patients with older age at onset of epilepsy were found to have earlier dislocations, occurring during the first 3 seizures, in association with age-related increased vulnerability of the shoulder joint complex. Recurrence rate of dislocations was found to be higher in patients younger than 20 years of age at onset of epilepsy, independent of seizure frequency or accompanying SE.

In future studies with larger patient populations, it will be important to investigate factors such as individual and glenohumeral joint characteristics, allowing for a clearer understanding of factors that contribute to the development and recurrence of epilepsy-related shoulder dislocation.

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