

Prone sleeping and SUDEP risk: the dynamics of body positions in non-fatal convulsive seizures

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1 **ABSTRACT**

2 *Background*

3 Most victims of sudden unexpected death in epilepsy (SUDEP) are found prone with signs
4 suggestive of an unwitnessed convulsive seizure (CS). Prone sleeping has been proposed as a
5 risk factor for SUDEP. Little is known, however, about the change of body position during
6 the course of CSs.

7 *Methods*

8 We retrospectively reviewed video EEG data and assessed body positions during the course of
9 CSs, until there was a physical interaction by nursing staff with the subject.

10 *Results*

11 We identified 180 CSs in 90 individuals. In 16 of the 180 CSs (9%) the subject started in or
12 turned to the prone position. Of the seven CSs that started in prone position, three turned to a
13 lateral position during the CS. In 13 CSs the subject was in prone position at time of nursing
14 intervention; nine (69%) of these started in a non-prone position.

15 *Discussion*

16 Our data suggest that the prone position occurs infrequently in closely supervised non-fatal
17 CSs, a notable contrast to the number of SUDEP victims found prone. Whether prone
18 sleeping prior to CSs increases SUDEP risk, however, remains speculative, as body position
19 during the course of a CS appeared to be dynamic.

20

21 **Key words:** sudden unexpected death in epilepsy (SUDEP); epilepsy; semiology; prone
22 position

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1. INTRODUCTION

Sudden unexpected death in epilepsy (SUDEP) is a poorly understood complication of epilepsy. The majority (73%) of all reported SUDEP cases are found in the prone position.[1] In view of a possible association between this and SUDEP, it has been debated whether prone sleeping increases SUDEP risk and the prone position has even been included in safety checklists. [2-4] Due to similarities with sudden infant death syndrome (SIDS) a “back-to-sleep” campaign to prevent SUDEP has been promoted by some[1] while others argue that prone body turning rather than prone sleeping is a SUDEP risk factor.[5]

In non-fatal convulsive seizures (CSs) body version seems common: prone body turning (up to 90 degrees) was found in 23 of 51 focal-onset CSs[6] and whole body version (of at least 180 degrees) in 12 of 277 epilepsy surgery candidates.[7] Body position changes in non-fatal CSs have not been reported. We aimed to explore the dynamics of body position in CSs. We hypothesized that body positions vary during the course of CSs.

2. METHODS

2.1 Selection

We reviewed the video-EEG database from two tertiary epilepsy referral centres, in Bonn, Germany and Heemstede, Netherlands. The databases were previously described.[8, 9] Pre-surgical video-EEG reports from between 2003 and 2011 of all people aged 15 years and older were reviewed and reports mentioning one or more recorded CSs were selected.[8, 9] Those with whom the nursing staff had a physical interaction (such as touching) prior to seizure onset, and those with video recordings that did not allow assessment of body positions, were excluded.

2.2 Collection of variables

We collected data on: sex, age, epilepsy classification (symptomatic or cryptogenic/idiopathic), age at onset, duration of epilepsy, CS frequency, state of wakefulness before seizure onset (awake/asleep), learning disability (yes/no), lesion on MRI (yes/no), localization of EEG seizure onset (temporal/extra-temporal), and the occurrence of postictal generalized EEG suppression (PGES) lasting more than 20 seconds (yes/no).

Body position categories were defined as: (1) prone: lying on the front, upper body lifted less than 45° from horizontal plane and angle between shoulders and horizontal axis $<45^\circ$; (2) supine: lying on the back, upper body lifted less than 45° from horizontal plane and angle between shoulders and horizontal axis $<45^\circ$; (3) lateral: right or left, angle between shoulders and horizontal axis $>45^\circ$ and $<135^\circ$ and upper body lifted less than 45° from horizontal plane; (4) sitting: angle between upper body and horizontal axis $>45^\circ$ and $<135^\circ$; (5) standing: standing or walking.

Body positions were scored by one author (SS) and the timing was related to the phase of the CS. In case of uncertainty either RDT or RS was consulted to reach consensus. Body position data were included until there was a physical nursing intervention.

2.3 EEG evaluation

Conventional scalp EEG recordings (International 10–20 System) (Stellate Harmonie, Stellate Systems, Montreal, QC, Canada) were performed at a sampling rate of 200 Hz. The end of the CS was defined as the time of the last muscular jerk.

2.4 Statistical analysis

We assessed the association between prone position during the CS and other seizure characteristics with the Mann-Whitney U test for continuous and χ^2 test for categorical variables. Only those variables with $p < 0.05$ were considered significant. Correction for multiple testing was made using the Bonferroni method. Where significant associations

occurred we corrected for the correlation between seizures in the same individual using generalized estimating equations (GEE). Statistical analysis was performed with IBM SPSS Statistics 23.

3. RESULTS

We identified 189 CSs in 92 individuals. Six CSs were excluded as there was a physical interaction prior to seizure onset and three as the video did not allow assessment of body position. After exclusions, 180 CSs in 90 individuals remained. Most had a focal onset (n=171, 95%). Clinical and seizure characteristics are shown in Supplementary Table 1.

3.1 Nursing intervention

Nearly all CSs prompted the nursing staff to intervene (n=174, 97%): at the start of the clonic phase 43% remained untouched and at offset only 16%. As a result of the nursing interventions most of the body position data are based on the first part of the CS (Figure 1).

figure1

3.2 Body position change

In 45% of all 180 CSs (n=81) the body position changed spontaneously during the course of the seizure. The most common transition was from lateral to supine (n=37). The majority of position changes were observed during the tonic (n=35) and focal phase (n=32).

3.3 CS and prone position

Seven of the 180 CSs started in prone position (4%) and in nine CSs the subject turned prone during the CS (5%). The median time spent in the prone position was 32 sec (IQR 14-29 sec).

Of the 7 CSs in which the subjects started in the prone position, three turned to a non-prone position. At the time of the nursing intervention prone position was noted in 13 CSs (Table 1).

Of these 13 CSs, the subject started in a non-prone position in nine (69%).

In CSs arising from sleep (116 of 180 CSs, 64%), the individuals were more likely to be in prone position at some point during the course of the CSs (14 of 16, 88%) than non-prone (102 of 116, 62%; χ^2 , $p=0.044$). This difference, however, was not significant after correction for multiple testing (significance level $p=0.007$).

During seizures in which the subject was prone at some point, a higher rate of spontaneous body position changes was seen compared to the non-prone seizures (χ^2 , $p=0.002$), while nursing intervention did not occur sooner (MW, $p=0.7$). When accounting for correlation between seizures in the same individual using GEE the difference in spontaneous body position changes remained significant (adjusted $p=0.001$).

Six of 90 subjects (7%) started prone and 7 subjects (8%) turned prone during at least one of their recorded CSs. A total of 31 CSs were recorded in these 13 subjects; eleven had multiple CSs. The consistency of starting or ending prone in these subjects is shown in Table 1.

4. DISCUSSION

Our data suggest that prone position occurs infrequently closely supervised non-fatal CSs, a notable contrast to the number of SUDEP victims found prone. We found that body position often changes during the course of CSs. In most seizures in which the subject was prone at time of nursing intervention, the subject started in non-prone. A high intra-individual variability of body positions was seen in the subjects with multiple recorded CSs. Whether prone sleeping prior to CSs increases SUDEP risk, thus, remains speculative.

Ictal body turning may explain why body position appeared to be dynamic. Body turning is notably seen in (mesial) frontal lobe seizures.[10, 11] The dynamic nature of the body position during the course of CSs is not only explained by the seizure onset zone as some subjects turned prone in the tonic, clonic or postictal phase.

1 A major, but inevitable, limitation of our study is that we have little information on the
2 positions at seizure end as all seizures are witnessed and nurses intervene before seizure end
3 in most. If we had been able to assess CSs without intervention, the proportion of CSs in
4 which the subject turns to another position, including from and to the prone position, would
5 have likely been higher. It could be argued that the hospitalisation may have biased our results
6 due to sleep disturbances or altered sleep behaviour. This effect seems to be minor as the
7 proportion of cases sleeping prone prior to CS (4%) was quite similar to the figures obtained
8 in a population and in a home-based study (9%).[12]

9 Another inevitable limitation is the absence of body position data of SUDEP cases. This will
10 remain challenging since SUDEP is mostly unwitnessed, recordings are rare, and often lack
11 details on body positions.

12 In only 12 of 253 published SUDEP cases body position prior to death was reported.[1] Ten
13 of these 12 cases were found prone and most of these cases (6 of 10) started in a non-prone
14 position.[5, 13, 14]

15 We found a low percentage of subjects who were prone at some point in 180 CSs (9%); thus
16 contrasting with SUDEP where the majority of cases (73%) is found prone. This contrast is
17 probably largely explained by the nursing interventions preventing the prone position. This
18 intervention may be one of the reasons why SUDEP tends to be unwitnessed, as the prone
19 position, along with the patient's immobility, potentially compromises respiration.

20 In SIDS, non-prone sleeping reduces mortality.[15] An important difference between SUDEP
21 and SIDS is the cause of the immobility: new-born infants are not able to change body
22 position during normal sleep. In people with epilepsy body movement is only beyond
23 voluntary control during the seizure itself or postictally when unconsciousness prevents the

subject arousing if hypoxia occurs.[16] The effects of CSs on body position and postictal recovery seem of greater importance than position prior to the CS.

The validity of a back-to-sleep campaign, therefore, remains unproven. We believe that sleeping in a non-prone position will not prevent SUDEP, but preventing a prone position may reduce SUDEP risk in those who are immobile in the postictal phase.

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6. CONFLICT OF INTEREST

SS reports no conflict of interest. RS has received speaker fees from Cyberonics, Eisai, Novartis and UCB Pharma, served on the advisory board of Eisai and is member of the editorial board of Epilepsy & Behavior and Epilepsia. JWS reports personal fees from Lundbeck and Teva, grants and personal fees from UCB, Eisai, grants from GSK, WHO and Dutch National Epilepsy Fund, outside the submitted work; JWS's current position is endowed by the Epilepsy Society, he is a member of the Editorial Board of the Lancet Neurology, and receives research support from the Marvin Weil Epilepsy Research Fund. RDT receives research support from the Dutch National Epilepsy Fund, NUTS Ohra Fund, Medtronic, and AC Thomson Foundation, and has received fees for lectures from Medtronic, UCB and GSK.

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Table 1. Body positions during the course of all CSs of the 13 subjects who were prone at some point during at least one of the recorded CSs. Seven CSs were prone prior to seizure onset (1, 4, 6, 9, 11, 13, and 14) and 9 CS turned prone during the course of the CS (16, 17, 18, 20, 22, 24, 25, 29, and 30). LT left temporal; RT right temporal; LFT left frontotemporal; RFT right frontotemporal; LP left parietal; LF left frontal; Non-lat non-lateralizing. Body positions: P prone; Su supine; LR right lateral; LL left lateral; Si sitting; * touched.

Subject #	Seizure #	EEG seizure onset	Time periods				
			Pre-ictal phase	Focal phase	Tonic phase	Clonic phase	Postictal phase
1	1	LP	P	P	P	P, LR	LR, *
1	2	LP	Su	Su	Su	Su, *	
1	3	LP	LR	LR	LR	LR, *	
2	4	RFT	P	P	P, RL, P	P, *	
2	5	RFT	Su	Su	Su	Su, *	
3	6	LF	P	P	P	P, *	
3	7	LF	Su	Su	Su, *		
3	8	LF	Su	Su	Su	Su, *	
4	9	LT	P	LR	LR, *		
4	10	LT	Su	Su, *			
5	11	RFT	P	P	P	P, *	
5	12	RFT	Si	Si, *			
6	13	LFT	P	P	P, *		
6	14	LFT	P	P	P, LL, *		
6	15	LT	Su	Su	Su	Su, *	
7	16	Non-lat	LL	P, *			
7	17	Non-lat	Su	P, *			
7	18	Non-lat	LL	P, *			
7	19	Non-lat	Su	Si, *			
8	20	LFT	LR	P	P, *		
8	21	LFT	Su, LL	LL	LL	LL	LL, *
9	22	LFT	Su	P, *			
9	23	LFT	Su	Si, *			
10	24	LT	LR	LR	P	P	P, *
11	25	LF	LR	LR	P	P, *	
11	26	LF	Su	Su	Su	Su	Su
11	27	LF	Si	Si	Si, *		
11	28	LF	LL	LL	LL	LL, Su, *	
12	29	RT	LL	LL	P, *		
13	30	RT	Su	P, *			
13	31	RT	Su	Si, *			

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3 **FIGURE LEGEND**

4 **Figure 1.** Number of CSs in which no physical nursing intervention has occurred at seven
5 time points during the course of the CS. For the seizures without focal onset (n=9) the time of
6 focal onset was taken as the same time the tonic phase starts. CS=convulsive seizure.

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- 1 **Supplementary table 1.** Clinical and seizure characteristics of the prone and non-prone group. χ^2 , Chi-square
 2 test; MW, Mann-Whitney U Test; CS, convulsive seizure; TC, tonic-clonic; N/A, not applicable; PGES, postictal
 3 generalized EEG suppression.

Variables seizure characteristics	Prone (n=16)^b	Non-prone (n=164)	All (n=180)	Test
State of wakefulness (% asleep)	14 (87.5)	102 (62.2)	116 (64.4)	χ^2 , p = 0.044
Ictal EEG onset, n (%)				χ^2 , p = 0.019
Temporal	13 (81.3)	83 (50.6)	96 (53.3)	
Non-temporal	3 (18.8)	81 (49.4)	84 (46.7)	
Duration of seizure, s (median; range)	96.5 (68 – 149)	93 (25 – 720)	93 (25 – 720)	MW, p = 0.631
Duration of TC phase, s (median; range)	69.5 (34 – 112)	58,5 (11 – 138)	61 (11 – 138)	MW, p = 0.229
PGES > 20 s, n (%)	7 (43.8)	72 (43.9)	79 (43.9)	χ^2 , p = 0.991
Touched by nurse, s after onset (median; range)	48 (5 – 127)	48 (1 – 640)	48 (1 – 640)	MW, p = 0.737
Change of body position prior to touch, n (%)	13 (81.3)	68 (41.5)	81 (45)	χ^2 , p = 0.002*
Variables clinical characteristics	Prone (n=13)^a	Non-prone (n=77)	All (n=90)	Test
Sex male, n (%)	9 (69.2)	40 (51.9)	49 (54.4)	N/A
Age at onset of epilepsy, years (median; range)	15 (3 – 25)	13 (0 – 55)	13 (0 – 55)	N/A
Age at time of EEG, years (median; range)	25 (16 – 32)	34 (15 – 63)	32 (15 – 63)	N/A
Duration of epilepsy, years (median; range)	11 (0 – 27)	19 (1 – 53)	18 (0 – 53)	N/A
Epilepsy classification (%)				N/A
Symptomatic	9 (69.2)	47 (61)	56 (62.2)	
Cryptogenic/idiopathic	4 (30.8)	30 (39)	34 (37.8)	
Lesion on MRI, n (%)	9 (69.2)	44 (57.9)	53 (58.9) (53/89)	N/A
Frequency of CS, n (%)				N/A
1-2 CS/year	9 (69.2)	33 (42.9)	42 (46.7)	
≥ 3 CS/year	4 (30.8)	44 (57.1)	48 (53.3)	

Learning disability, n (%)	2 (18.2) (2/11)	3 (6.4) (3/47)	5 (8.6) (5/58)	N/A
Epilepsy center, n (%)				N/A
Bonn	6 (46.2)	33 (42.9)	39 (43.3)	
Heemstede	7 (53.8)	44 (57.1)	51 (56.7)	

1 ^a subjects who were prone at some point during at least one of the recorded CS.

2 ^b CSs in which the subjects was prone at some point.

3 *significance level after correction for multiple testing using the Bonferroni method $p=0.007$. P-values based on
 4 univariate analysis.

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