DISTINGUISHING SLEEP-RELATED HYPERMOTOR EPILEPSY FROM DISORDERS OF AROUSAL AND REM SLEEP BEHAVIOR DISORDER: THE ROLE OF CLINICAL HISTORY

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Aims

Identifying sleep-related hypermotor epilepsy (SHE) presents a particular challenge for the clinician, mainly with respect to parasomnias. Proposed diagnostic criteria for SHE give new importance to clinical history, which represent the first step in the diagnostic pathway. In this scenario, the present study tests the accuracy of anamnestic features for the diagnosis of SHE, disorders of arousal (DA), and idiopathic REM sleep behavior disorder (iRBD).

Materials and Method

We retrospectively reviewed all patients evaluated with video-polysomnography for abnormal paroxysmal motor events in sleep, from January 2011 since December 2015, in the Clinic of Neurology of the University Hospital of Udine. Patients without a confirmed diagnosis (26 subjects), with extrapyramidal signs or cognitive decline (19 subjects) were excluded. If data weren't already available, patients and witnesses were reinterviewed for the specific purposes of the study. We used frontal lobe epilepsy and parasomnias (FLEP) scale and structured interview for nocturnal frontal lobe epilepsy (SINFLE) to collect anamnestic information. Moreover we asked for dream enactment and reported in detail age at onset.

	SHE	DA	iRBD
Patients	7	31	24
Gender male	3 (42.86%)	21 (67.74%)	19 (79.17%)
Age (mean ± s.d.)	35.43 ± 13.26	32.06 ± 13.17	68.46 ± 8.64
Age (median, range)	28, 23-56	31, 13-75	70.5, 48-82
Diagnosis	3 paroxysmal arousals (PA) 2 PA + hypermotor seizures + ENW 2 hypermotor seizures	11 sleepwalking + confusional arousals 2 sleep terror 11 confusional arousals 3 sleep terror + confusional arousals 4 sleepwalking + sleep terror + confusional arousals	

Results

Out of 107 subjects evaluated, 62 patients (7 SHE, 31 DA, and 24 iRBD) were recruited. The sensitivity of the FLEP scale and SINFLE, as diagnostic tests for SHE, was respectively 57.1% vs 28.6%, the specificity 92.7% vs 98.2%, the positive predictive value 50% vs 66.7%, the negative predictive value 94.4% vs 91.5%. **"Stereotypy" (*p* 0.024)** and **"aura" (*p* 0.001)** were best predictors for SHE compared with DA, instead **"wandering" (*p* 0.008)** favored the diagnosis of DA. **"Age at onset" (*p* <0.001)**, **"stereotypy" (*p* <0.001)**, **"event duration" (*p* 0.003)**, **"aura" (*p* <0.001)**, **"dream enactment" (*p* <0.001)** distinguished SHE from iRBD.

Diagnostic validity of anamnestic characteristics

SHE vs DA

	Sens	Spec	LR +	LR -	DOR
Duration	28.6% (3.7-71)	87.1% (70.2-96.4)	2.21 (0.50-9.79)	0.82 (0.50-1.34)	2.70 (0.39-18.93)
Clustering	0% (0-41)	80.6% (62.5-92.5)	0.23 (0.03-1.67)	1.24 (1.04-1.47)	0.20 (0.02-1.57)
Timing	0% (0-41)	96.8% (83.3-99.9)	1.41 (0.15-12.91)	1.03 (0.97-1.10)	1.43 (0.14-14.43)
Aura (FLEP)	**57.1%** (18.4-90.1)	**96.8%** (83.3-99.9)	**17.71** (2.32-135.16)	**0.44** (0.19-1.04)	**40.00** (3.31-483.47)
Aura (SINFLE)	**71.4%** (29.0-96.3)	**93.5%** (78.6-99.2)	**11.07** (2.68-45.80)	**0.31** (0.09-0.99)	**36.25** (4.11-319.94)
Wandering	**71.4%** (29.0-96.3)	**83.9%** (66.3-94.5)	**4.43** (1.75-11.22)	**0.34** (0.10-1.11)	**13.00** (1.95-86.81)
Complex directed behavior	71.4% (29.0-96.3)	64.5% (45.4-80.8)	2.01 (1.03-3.92)	0.44 (0.13-1.47)	4.55 (0.75-27.43)
Dystonic or hyperkinetic pattern	28.6% (3.7-71.0)	96.8% (83.3-99.9)	8.86 (0.93-84.54)	0.74 (0.46-1.18)	12.00 (0.91-158.44)
Stereotypy	**42.9%** (9.9-81.6)	**93.5%** (78.6-99.2)	**6.64** (1.35-32.58)	**0.61** (0.32-1.17)	**10.87** (1.37-86.38)
Recall	0% (0-41)	96.8% (83.3-99.9)	1.41 (0.15-12.91)	1.03 (0.97-1.10)	1.43 (0.14-14.43)
Unstructured vocalisation	14.3% (0.4-57.9)	61.3% (42.2-78.1)	0.37 (0.06-2.39)	1.40 (0.93-2.11)	0.26 (0.03-2.47)

SHE vs iRBD

	Sens	Spec	LR +	LR -	DOR
Age at onset	**100%** (59.0-100)	**87.5%** (67.6-97.3)	**8.00** (2.78-23.06)	**0.05** (0.01-0.35)	**168.00** (20.19-1398.01)
Duration	**28.6%** (3.7-71.0)	**100%** (85.7-100)	**20.86** (2.66-163.75)	**0.71** (0.45-1.14)	**28.80** (3.23-257.05)
Clustering	0% (0-41.0)	95.8% (78.9-99.9)	1.09 (0.12-9.97)	1.04 (0.96-1.13)	1.10 (0.11-11.09)
Aura (FLEP)	**57.1%** (18.4-90.1)	**100%** (85.7-100)	**41.71** (5.75-302.57)	**0.43** (0.18-1.01)	**96.00** (11.13-827.88)
Aura (SINFLE)	**71.4%** (29.0-96.3)	**100%** (85.7-100)	**52.14** (7.31-372.12)	**0.29** (0.09-0.92)	**180.00** (20.17-1606.59)
Complex directed behavior	71.4% (29.0-96.3)	4.2% (0.1-21.1)	0.75 (0.46-1.20)	6.86 (0.72-64.93)	0.11 (0.01-1.45)
Dystonic or hyperkinetic pattern	28.6% (3.7-71.0)	87.5% (67.6-97.3)	2.29 (0.47-11.08)	0.82 (0.50-1.34)	2.80 (0.36-21.49)
Stereotypy	**42.9%** (9.9-81.6)	**100%** (85.7-100)	**31.29** (4.20-233.08)	**0.57** (0.30-1.09)	**54.00** (6.26-465.68)
Recall	0% (0-41.0)	95.8% (78.9-99.9)	1.09 (0.12-9.97)	1.04 (0.96-1.13)	1.10 (0.11-11.09)
Unstructured vocalisation	14.3% (0.4-57.9)	50% (29.1-70.9)	0.29 (0.04-1.83)	1.71 (1.04-2.83)	0.17 (0.02-1.60)
Dream enactment	**100%** (59.0-100)	**75%** (53.3-90.2)	**4.00** (2.00-8-00)	**0.06** (0.01-0.41)	**63.00** (7.90-502.19)

Discussion and Conclusions

Semeiological similarities make it difficult to differentiate SHE not only from DA but also from iRBD. However DA and iRBD core features differ each other with respect to SHE. A rigorous report of age at onset and dream enactment can improve diagnostic accuracy. We hypothesize that dystonic or hyperkinetic pattern didn't reach statistical significance because, as for other features, observers may not be reliable. Our study confirms the usefulness of anamnestic information for the differential diagnosis of SHE, DA, and iRBD. If we imagine a structured algorithm, clinical core features are crucial for the correct selection of further diagnostic investigations.

References

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