J Neurol (2000) 415 : 878–879

Bernd M. Sanner
Nikolaus Büchner
Sylvia Kotterba
Walter Zidek

Polysomnography in acute African trypanosomiasis

Received: 16 December 1999
Received in revised form: 10 May 2000
Accepted: 4 June 2000

Sirs: Sleeping sickness is one of the sleep disorders listed in the International Classification of Sleep Disorders (3.C.1). To date there have been only few reports of sleep studies in chronic sleeping sickness [1, 2, 3] and, to our knowledge, no reports of sleep studies in acute sleeping sickness. We describe a case of acute African trypanosomiasis with disturbed sleep following travel to the tropics and the results of the sleep studies in this patient.

A 47-year-old woman was admitted to hospital because of high remittent fever, insomnia during night, hypersomnia by day, and jaundice. Symptoms had started 5 days prior to admission, 7 days after returning from a 20-day trip to Zambia, Zimbabwe, and Tanzania. The patient had a fever of 39.2°C, an ulcerated and indurated lesion of 3 cm in diameter at the right first carpometacarpal joint, discrete nontender axillary lymphadenopathy on the right side, and jaundice. Abdominal ultrasound, chest radiography, and echocardiography on the day of admission were completely normal. Three blood cultures and microscopy of blood smears isolated no organisms.

During the next 2 days her condition deteriorated, fever rose to 40.9°C, she became more sleepy during the day and was restless at night, and developed multiorgan failure with hepatitis, myocarditis, nephritis, pancreatitis, polyserositis, and disseminated intravascular coagulation. Again, microscopy of peripheral blood smears was performed, and this time trypanosomes were found. The indirect fluorescent antibody test for African trypanosomiasis was initially negative but turned positive (1:20) during the following 3 days.

Due to the history, clinical findings, and microscopy results acute infection with *Trypanosoma brucei rhodesiense* with septic and multiorgan involvement was diagnosed. Treatment with suramin was initiated immediately, and she recovered completely over the following days. Lumbar puncture was performed on day 9 of suramin treatment, when there were no more trypanosomes in the peripheral blood smear, and the platelet count was normal; this yielded no direct evidence of central nervous system involvement. Total protein level of the spinal fluid was slightly elevated (0.65 g/l), but there was an increase neither in cells nor in the IgM level. Free immunoglobulin light chains were not present.

Polysomnography was performed according to widely accepted methods on days 7, 9, and 15 and 6 months after the onset of trypanosomiasis [4]. Sleep was staged manually using the methods of Rechtschaffen and Kales [5], and arousals and leg movements were classified according to ASDA recommendations [6, 7].

Sleep examination revealed a poor sleep efficiency and decreased slow wave sleep, while the amount of arousals and awakenings was increased (Table 1). Furthermore, the patient had periodic limb movements (PLM) during sleep with a PLM index of 27.2/h and a PLM arousal and wake index of 11.7/h. Repeat polysomnographic measurements during follow-up showed an increase in sleep efficiency and amount of slow wave sleep and a decrease in arousals and awakenings, but PLM remained. MRI of the brain conducted on day 10 of disease onset showed a small angioma at the right frontal pole and was otherwise unremarkable. Mobile long-term EEG recording (24 h duration) was performed on day 22. The eight-channel monitoring revealed an alpha rhythm (10/s) with recurrent short naps (non-rapid eye movement sleep 2) during the day. Night recording confirmed polysomnographic data showing a poor sleep efficiency with frequent awakenings and arousals. At sleep onset generalized high theta activity and steep potentials were observed. There were no signs of encephalitis. Laboratory examination on day 25 after onset of the disease showed normal values for vitamin B_1, vitamin B_6, and vitamin E.

The nerve conduction study using surface electrodes to record compound muscle action potentials revealed prolonged conduction velocity of the right peroneal nerve (38 m/s), left tibial nerve (34 m/s), and left

Table 1 Results of the polysomnographic examinations (*REM* rapid eye movement, *PLM* periodic limb movement)

	Day 7	Day 9	Day 15	6 Months
Total sleep time (min)	307	278	393	397
Sleep efficiency (%)	71	73	97	91
Wake (%)	26	22	3	5
Sleep stages 1+2 (%)	54	52	69	55
Sleep stages 3+4 (%)	2	8	4	24
Sleep stage REM (%)	18	18	24	16
Arousal index	28.3	27.5	20.1	11.3
PLM index	27.2	26.2	34.7	19.5
PLM arousal + wake index	11.7	9.7	10.4	8.9

sural nerve (41 m/s) with normal amplitudes. There were no pathological changes in the median nerves. Thus electroneurographic results indicated multifocal demyelinating neuropathy. A follow-up nerve conduction study after 12 weeks demonstrated similar results with prolonged conduction velocity of both peroneal nerves (37 m/s) and of the left sural nerve (32 m/s). Electromyography of the tibialis anterior muscle was normal.

Sleeping sickness in its chronic form is commonly seen in Africa, but an acute course of African trypanosomiasis and especially of imported sleeping sickness in temperate countries are few [8]. Sleeping sickness due to *Trypanosoma brucei rhodesiense* usually causes a rapid progressive disease often resulting in cardiac failure and acute neurological manifestations, while patients with a chronic course are classically described as sleepy by day and restless by night. Untreated, African sleeping sickness is almost invariably fatal. It is worth noting that our patient also had the classical symptoms of the disease in addition to the signs of multiorgan failure.

Waking electroencephalograms in the chronic stages of African trypanosomiasis are characterized by (I) sustained low-voltage background similar to that seen during light sleep (II) paroxymal waves, or (III) various types of delta wave (similar to those seen in demyelinating encephalitis) and rapid, intermittent high voltage delta bursts between periods of lower-voltage delta activity, probably dependent on the degree of cerebral involvement of the disease [9].

To our knowledge, this is the first report of sleep studies in a patient with an acute course of African trypanosomiasis. The initial sleep study was characterized by a decrease in sleep efficiency and slow-wave sleep, while the amount of brief arousals and awakenings was increased. Rapid eye movement sleep was normal. Furthermore, the patient had periodic limb movements. After 1 week, the sleep fragmentation improved, but delta-wave sleep remained greatly reduced. The latter returned to normal after 6 months. Given these results, the question arises whether the changes in the sleep structure and the periodic limb movements are an epiphenomenon of the severe disease with multiorgan failure and fever or are directly related to or induced by trypanosomiasis.

There are only few reports on periodic limb movements in trypanosomiasis, but that may be because most of the polysomnographic studies performed in trypanosomiasis have not recorded electromyograms of the legs. Interestingly, a French group recording leg activity during the night observed myoclonic jerks associated with arousals in the two studied patients with chronic sleeping sickness [2]. Our patient had periodic limb movements not only in the first polysomnographic recording performed on day 2 after the disease was diagnosed, and these limb movements were sustained during follow-up, even after 6 months. Therefore it may be speculated that periodic limb movements – and perhaps also neuropathy – are part of the clinical manifestations of trypanosomiasis. On the other hand, since there is a high prevalence of PLMs during sleep, their pre- or coexistence in our patient cannot be ruled out.

With travel to and from Africa, trypanosomiasis has begun to present more frequently in temperate countries. Polysomnographic recordings may be characterized by a decrease in slow-wave sleep and sleep efficiency along with frequent arousals and awakenings. In addition, periodic limb movements may be part of the disease and partially responsible for the clinical symptoms.

References

1. Buguet A, Bert J, Tapie P, Tabaraud F, Doua F, Lonsdorfer J, Bogui P, Dumas M (1993) Sleep-wake cycle in human African trypanosomiasis. J Clin Neurophysiol 10:190–196
2. Montmayeur A, Brosset C, Imbert P, Buguet A (1994) The sleep-wake cycle during Trypanosoma brucei rhodesiense human African trypanosomiasis in 2 French parachutists. Bull Soc Pathol Exot 87:368–371
3. Tapie P, Buguet A, Tabaraud F, Bogui P, Doua F, Bert J (1996) Electroencephalographic and polygraphic features of 24-hour recordings in sleeping sickness and healthy African subjects. J Clin Neurophysiol 13:339–344
4. American Thoracic Society – Medical Section of the American Lung Association (1989) Indications and standards for cardiopulmonary sleep studies. Am Rev Respir Dis 139:559–568
5. Rechtschaffen A, Kales A (1968) A manual of standardized terminology, techniques and scoring system for sleep stages of human subjects. NIH publication no. 204. United States Government Printing Office, Washington
6. ASDA Task Force (1992) EEG arousals: scoring rules and examples. Sleep 15:174–184
7. ASDA Atlas Task Force (1993) Recording and scoring leg movements. Sleep 16:749–759
8. Spencer HC, Gibson JJ, Brodsky RE, Schultz MG (1975) Imported African trypanosomiasis in the United States. Ann Intern Med 82:633–638
9. Hamon JF, Camara P, Gauthier P, Arnaud C, Gottesmann C (1993) Waking electroencephalograms in the blood-lymph and encephalitic stages of Gambian trypanosomiasis. Ann Trop Med Parasitol 87:149–155

B. M. Sanner (✉) · N. Büchner · W. Zidek
Department of Medicine I,
Ruhr University Bochum,
Marienhospital Herne,
Hölkeskampring 40,
44625 Herne, Germany
e-mail: Bernd.Sanner@ruhr-uni-bochum.de
Tel.: 49–23–234990
Fax: 49–23–23499302

S. Kotterba
Department of Neurology,
Ruhr University Bochum,
Bergmannsheil Bochum, Germany