

Case report

Sleep disorder in alien hand syndrome

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Received 19 June 2002; received in revised form 19 September 2002; accepted 25 September 2002

Abstract

A 63-year-old right-handed woman developed an alien hand syndrome (AHS) after an acute infarction in the territory of the left anterior cerebral artery. The uncontrolled hand movements were present during the daytime and eventually disturbed sleep. Polysomnography revealed that these motor actions only appeared when the patient was awake. These awakenings emerged mostly from NREM sleep stage 2 during the first half of the night. There was no evidence of any epileptiform activity, dyssomnia or parasomnia. These movements were controlled making her wear an oven mitt during sleep. The temporal distribution of this motor activity seems to follow the progressive hyperpolarization of anterior horn neurons that occurs when sleep deepens. The accommodation of the grasp reflex in AHS probably helps control this unwanted motor activity.

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Keywords: Abnormal movements; Alien hand; Electroencephalogram; Polysomnography; Sleep; Stroke

1. Introduction

The alien hand syndrome (AHS) [1] is a motor phenomenon in which one hand performs seemingly purposeful actions that are beyond the patient's voluntary control. This rare condition has been related to a variety of lesions of the corpus callosum, sometimes involving the supplementary motor areas as well, or affecting posterior brain territories [2–4]. Stroke is the most frequent cause. In AHS the contralateral hand to the brain lesion performs annoying free movements like grasping or groping; the patient feels forced to grab it with the other hand in order to control them. These motor actions can be very disturbing, particularly at night, when the alien hand can continuously disrupt sleep [3,5].

2. Clinical case

A 63-year-old right-handed woman with a previous history of diabetes mellitus, hypertension and obesity presented with a mild right hemiparesis and a self-limited speech disorder. Magnetic resonance imaging (including

diffusion-weighted and vascular sequences) revealed a 6 × 2 cm acute infarction in the territory of the left anterior cerebral artery. Single photon emission computed tomography showed significant low blood flow in the left frontal and anterior parietal cortex, as well as frontal and parietal parasagittal cortex. Computed tomography added no further information. A couple of days after admission she complained that her right hand had started moving around in an uncontrolled, spontaneous fashion, crawling around her head and grasping her bed sheets and clothes, so that she had to grab it with her left hand in order to hold it. Electroencephalogram (EEG) showed high voltage delta waves on the left frontal and temporal-parietal regions, suggestive of lesion, with a normal background activity, but no epileptiform discharges. Her husband later reported that he had witnessed an increase of these involuntary movements during the night. A nocturnal polysomnogram was subsequently performed. EEG was recorded from surface electrodes placed at Fp1, Fp2, C3, C4, A2, O1 and O2, according to the international 10–20 system. Electrocogram and submental electromyogram (EMG) were recorded for sleep stage scoring, which followed common standards [6]. Surface EMG was recorded from bilateral flexor carpi radialis and extensor digitorum communis, and right deltoid muscles. Electrocardiogram was also recorded. Bed time was 7 h 17 min, with a total sleep time of 3 h 7 min. Sleep

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Fig. 1. Recording shows how the alien hand starts moving (right arrow) *after* the patient has awakened (left arrow). EEG prior to awakening corresponds to NREM sleep stage 2. EEG electrodes nomenclature according to 10/20 international system. EOG, electrooculogram; EMG, electromyogram; EKG, electrocardiogram; FCR, flexor carpi radialis muscle; EDC, extensor digitorum communis muscle; R, right; and L, left.

latency was almost 17 min. NREM stage 1 sleep time was 32.5 min (17.4%), stage 2 was 63.5 min (34%), and stages 3–4 were 47.5 min (25.5%). REM sleep time was 43 min (23.1%), with two complete REM/NREM cycles. Eighteen awakenings were recorded, each of them followed by a long period of wakefulness. Total awakening time was 4 h 10 min, resulting in a low sleep efficiency of 43%. Awakenings arose from NREM stages 1 (22.2%) and 2 (77.8%). Sleep was reasonably continuous when present, with few stage changes. Arousal index was under 5 per hour of sleep. Video-EEG recording showed a nearly continuous right hand movement, but only when the patient was awake (Fig. 1). This activity was more frequent in the first awakenings than later on. The patient said she was afraid of falling asleep in case her right hand clutched her neck and suffocated her during sleep. Three weeks later, with no subsidence of her hand's movements, a second polysomnography was performed. Oral/nasal airflow, thoracic and abdominal ventilatory effort, arterial oxyhemoglobin saturation (SaO_2) and pulse rate were recorded in addition to previous standards. Surface EMG was limited to bilateral extensor digitorum communis and right tibialis anterior muscles. On this occasion the patient wore an oven mitt on her right hand. No spontaneous hand movements were recorded; the patient slept all night long, and sleep architecture was completely normal. An obstructive sleep apnea syndrome was ruled out since no apneic episodes were recorded. SaO_2 was not under 97%. No periodic limb

movements were noted. Previous EEG lesional findings had already disappeared.

So far, this patient is able to sleep quietly wearing her oven mitt, and only suffers from AHS when her hand receives tactile stimuli during the daytime.

3. Conclusion

Three forms of AHS have previously been described [4, 7]. Our patient presented the frontal form, which results from damage to the supplementary motor area, anterior cingulate gyrus, and medial prefrontal cortex of the dominant cerebral hemisphere and anterior corpus callosum and occurs in the dominant hand. Callosal AHS results from an isolated anterior callosal lesion and is manifested by intermanual conflict. The third form involves lesion of posterior cortical or subcortical areas. It has been proposed that the grasping in AHS is a cutaneous reflex mediated at a subcortical or spinal cord level [8]. An alien hand-like syndrome has also been described in the context of a partial status epilepticus, with spike activity on the EEGs [9].

Alien hand motor activity may disturb the continuity of sleep at night. We have only found two published cases of AHS disturbing sleep. Both were frontal and clinically similar to ours, but did not include any reference to polysomnographic studies [3,5]. Ours showed that the alien hand movements only occurred during wakefulness

emerging from superficial sleep, particularly stage 2, and that these movements were more frequent early at night than later on. These findings may express the normal gradual hyperpolarization of anterior horn cells that occurs when sleep deepens from superficial stages to slow and REM sleep, causing a progressive decline in motor activity. Habituation of cutaneous reflex to scant afferent stimuli after some time in bed may also contribute to this temporal distribution. An oven mitt probably controls alien hand movements because it provides enough sensory stimulation to the spinal grasp reflex to reach a level of accommodation that stops the unwanted motor activity. Our study indicated that the sleep disturbance in AHS might also arise from the patient's fear of being attacked by her hand while asleep, rather than solely from its motor activity. Accordingly, AHS can benefit from some psychological assurance.

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