Two Faces of Nocturnal Tongue Biting

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Abstract

Tongue biting associated with tonic / clonic movements of the limbs is common in epileptic patients, however nocturnal tongue biting as the only manifestation of epilepsy is rare. It can be found in frontal lobe epilepsy. Two cases with the same manifestation of nocturnal tongue biting were presented. One was the result of parasomnias-rhythmic movement disorders (RMD) and the other was a result of nocturnal frontal lobe epilepsy. The definite diagnosis of these abnormal nocturnal events was documented by prolonged EEG monitoring and polysomnography with simultaneous video studies. To our knowledge, RMD presenting with nocturnal tongue biting has never been reported in Thailand. Even in the foreign journals it has rarely been reported. It is crucial to make a definite diagnosis of RMD and nocturnal epilepsy to avoid overtreatment in the former and undertreatment in the latter. Symptomatology, diagnostic approach with therapy of these disorders were reviewed.

A wide variety of paroxysmal movements in children for example : epilepsy, parasomnias including rhythmic motor movement, sleep bruxism, periodic limb movements and gastro-esophageal reflux may occur during sleep. Among these, parasomnias share the most common features with nocturnal epilepsy. Common symptoms of both conditions are recurrent, stereotypical, paroxysmal, transient disturbance of behaviour with return to baseline function between attacks[1-3]. In most cases of both epilepsy and parasomnias, causative underlying systemic or progressive neurologic diseases are absent and the patients are normal between the attacks. Often, neurological examinations and standard clinical neuroimaging studies in both conditions are unremarkable[3-5].

We describe 2 children presenting with the same manifestation, repeated nocturnal tongue biting, but of different etiologies in whom the specific diagnosis were clearly established by pro-

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longed electroencephalography (EEG) monitoring and polysomnography (PSG)-video studies.

CASE REPORTS

Patient 1, a 1 $^{1}/_{2}$-year-old girl, presented with tongue biting during night sleep for 2 months. The symptoms occurred unpredictably 5-6 times per night or during day nap on some occasions. Her mother was awakened by loud crying and gurgling sounds and found the child very distressed with a deep bite on the anterior aspect of her tongue and fresh blood stains on the pillow case. These events recurred nearly every night. Her mother reported the child's head banging and head rolling sometimes but no tonic-clonic phenomenon.

The past medical history was unremarkable except for slight psychomotor delay and underweight. She had just been able to walk steadily for 1 week and spoke only 2 meaningful words. Her 3-year-old older sister had 1 episode of unprovoked seizure at 2 years of age but no recurrence since then. There was no family history of sleep bruxism or other sleep disorders.

A 16-channel EEG according to the international 10/20 system showed no abnormal activity. No tongue biting occurred during daytime study. However, due to clinical suspicion, sodium valproate as well as phenobarbital and diazepam were introduced but were of no benefit. The child was admitted for diagnostic evaluation for nocturnal epilepsy or rhythmic movement disorders (RMD). Physical examination revealed a malnourished girl : weight of 9.2 kg, height of 80 cm, macrocephaly: head circumference of 49 cm with anterior fontanelle of 1x1 cm and chronic ulcer at tip of the tongue diameter of 2 x 0.8 cm (Fig. 1). Neurological examination was normal. Complete blood count revealed nutritional anemia. Normal serum chemistries included electrolytes, calcium, phosphate, alkali phosphatase and thyroid function. Chromosomal study was normal. Urinalysis was normal. Computerized brain tomography as well as cranial magnetic resonance imaging revealed no abnormality of parenchymal and ventricular systems.

The child underwent all night PSG study and simultaneous video monitoring for 2 consecutive nights. The montage (12-channel study) for recording sleep-related seizure activity, according to Keenan[6] was used. No epileptiform activity was seen but increased muscle activities of chin

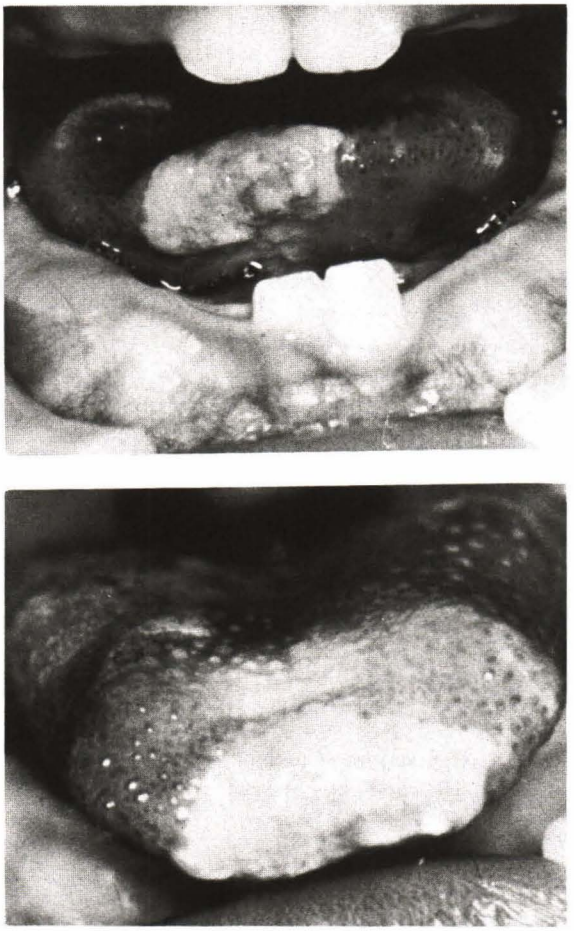

Fig. 1. The tongue of patient 1 shows a chronic ulcer with diameter of 2x0.8 cm.

was detected during the tongue biting attacks. The events occurred just after sleep onset and through the night 5-6 times during stage 1 and 2 of non-rapid eye movement (NREM) sleep and between changing the stage of sleep from stage 1 NREM to wakening (Fig. 2) or after turning over into the prone position. After the attacks, the child awakened and cried then returned to sleep again. There were also increased rhythmic movements of her extremities during light sleep. Rapid eye movement (REM) sleep was not observed may be because of frequent awakening. During stage 3,4 of NREM sleep some snoring was detected. The diagnosis of RMD was considered because of the sleep stage in which the attacks occurred and the absence of simutaneous abnormal EEG activity during the attacks. Due to frequent and severe attacks the child was initially

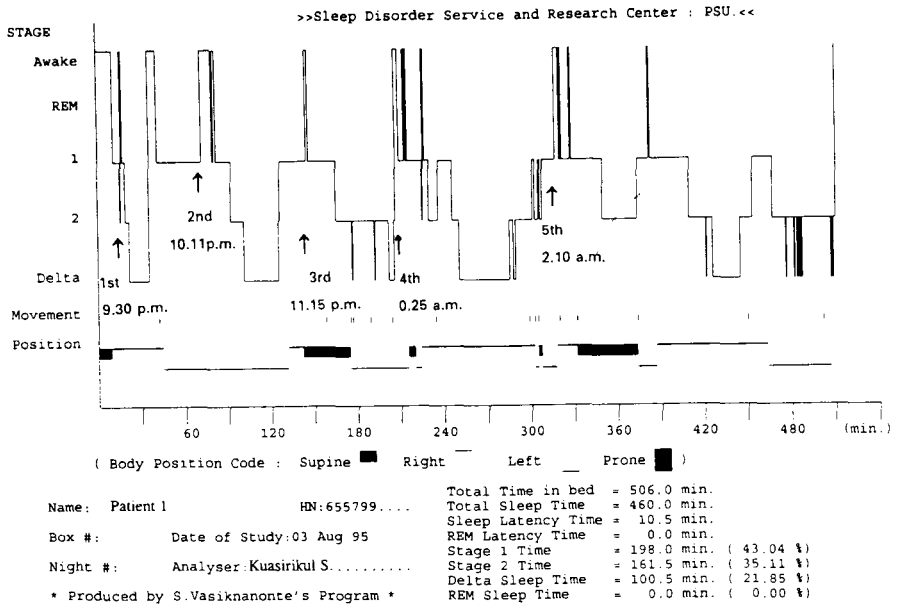

Fig. 2. Hypnogram of patient 1 shows episodic tonic tongue biting occurring 5 times / night in light NREM sleep and during brief arousals or transition to lighter stages of sleep.

treated with oral clonazepam 0.075 mg/kg/day. After one week of treatment, she responded with decrement of the attack to 1/night. Dental guard was also used as initial adjunctive management. When increasing the dose of clonazepam to 0.1 mg/kg/day, the attacks disappeared and the dental guard was not necessary. One month later, the mother herself stopped clonazepam, the child's symptoms recurred again, however, it was resolved after the drug was re-introduced. We have so far followed this girl for 6 months. Her clinical symptom was still uneventful and her psychomotor development was much improved.

Patient 2, a 2-year-3-month-old girl, had a 4 $^{1}/_{2}$ -month history of nocturnal tongue biting nearly everynight. The symptom usually attacked half an hour after falling asleep, but occurred during day nap on some ocassions. The mother described the child's gurgling sounds and jittering before the deep tongue biting then followed by loud crying and awakening. No tonic-clonic movements of extremities nor somnambulism were observed. There was no family history of epilepsy nor sleep disorders. Her past medical history was unremarkable. She had been treated with sodium valproate 20 mg/kg/day for 1 $^{1}/_{2}$ month with unsatisfied clinical response and was referred to our center with suspected diagnosis of RMD during sleep. Physical together with neurological examination were normal except for a shallow ulcer at left anterior of her tongue.

A prolonged 16-channel EEG, according to the international system showed some paroxysmal short bursts of spikes from the left frontal area without clinical seizure (Fig. 3). PSG for recording sleep-related seizure activity, the method according to Keenan[6] and simultaneous video monitoring showed left frontal spikes with clinical tongue biting. No rhythmic movement of extremities was observed. The paroxysmal spikes were in NREM sleep. These confirmed the diagnosis of nocturnal frontal lobe epilepsy. After increasing the dosage of sodium valproate to 30 mg/kg/day, no symptoms recurred.

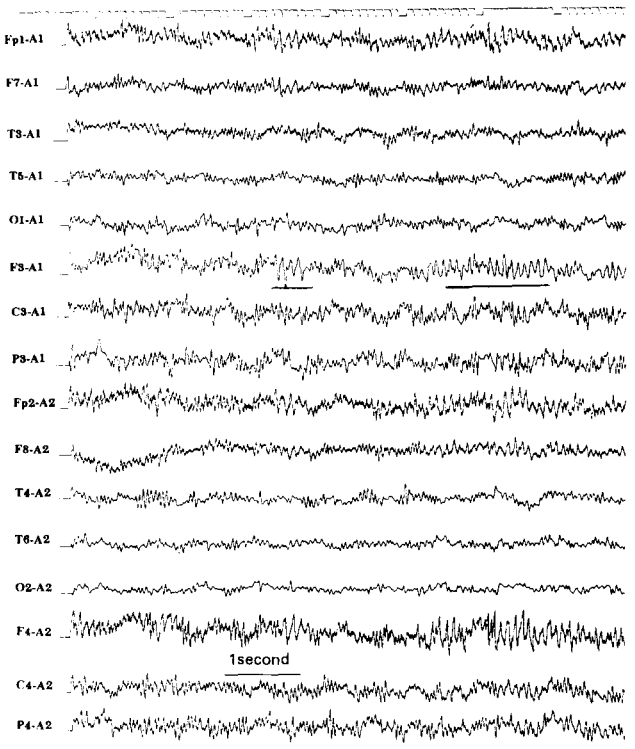

Fig. 3. EEG recording during sleep of the patient 2 illustrates some paroxysmal spikes from left frontal area.

DISCUSSION

In terms of episodic and paroxysmal symptoms occuring during sleep, several non-epileptic disorders resembling childhood epilepsy should be considered in the differential diagnosis. These include sleep parasomnias such as rhythmic movement disorders, nightmares, sleep terror, sleep bruxism and benign neonatal sleep myoclonus. Parasomnias in childhood are frequent and usually harmless. They are the result of immaturation of the central nervous system[3] and are usually self limited when the child grows older[3,4]. Only some cases need treatment. In contrast, some childhood epilepsy may persist through adultlife and increase considerably in severity and neurosequlle if untreated. In both conditions, the physical including neurological examination are usually normal but in some cases delayed psychomotor development might be found.

The fundamental clues to differentiate parasomnia from epilepsy based on detailed history of the episodic phenomena. These include : 1) level of consciousness and type of behaviour exhibited during the attack 2) dreaming and dream content associated with the attack 3) associated typical epileptic phenomenon (witnessed generalized tonic clonic seizure) 4) post ictal stage 5) number of attacks in a single night 6) overall recurrent pattern (every night or sporadically) 7) time during the sleep cycle that the attacks occur 8) age of the patient 9) family history of sleep disorders or epilepsy 10) associated psychiatric or neurologic condition. The detailed history together with the physical and neurological examination are helpful for making the initial diagnosis, however, many cases have episodes that are difficult to diagnosis by the history and physical examination alone, even by standard EEG[1,2,5]. With the aid of

modern PSG and video-EEG monitoring during sleep, a precise diagnosis can be made[1,5,6]. The two cases we described here presented with the same manifestation, recurrent nocturnal tongue biting: one as a result of RMD, the other as a result of frontal lobe epilepsy.

RMD are classified as sleep-wake transition disorders, a group of disorders that is a subcategory of the parasomnias[4]. The onset of RMD is usually in infancy between 8-18 months[4,7]. Two forms of the disorder have been categorized according to the association with the time of occurence[7]. The first occurs predominantly during the daytime which does not necessarily occur in relation to sleep and may, infact be inhibited by sleep. This form is more typically seen in individuals who are either mentally retarded, or have major psychiatric disorders and are commonly associated with self injurious behaviour. The second occurs predominantly during the night sleep. Sallusto[8] suggested that the etiology of RMD is different in the noctunal form, however, more information is needed to confirm that suggestion. From the literature, it is clear that even in those individuals who have the disorders predominantly during sleep also manifest the disorders at times of drowsiness and in wakefulness[7-9]. The vast majority of individuals with RMD are otherwise normal infants and up to 67 per cent of normal children experience the disorders[9]. Although the persistence of the disorder from infancy to older children or adulthood may be associated with mental retardation, autism or other major psychopathology, many adults with the disorder do not have such associating factors[7,9,11]. Nocturnal RMD in children includes head banging (jactatio capitis), head rolling, body rocking, body rolling, leg banging, leg rolling. Tongue biting as a manifestation of nocturnal RMD is rarely reported [12]. The repetitive and violent nature of the rhythmic movements may lead to injury of the involved body part[12-14]. With rhythmic movements of the muscles of mastication leading to forceful chewing and jaw closure which results in tongue injuries [12]. This may be misdiagnosed as sleep bruxism or nocturnal epilepsy. Usually, the onset of sleep bruxism begins in older children and persists to adulthood[3,4,15]. Presentation of the loud grinding sound may not be associated in some cases of sleep bruxism[15]. Differential diagnosis of rhythmic nocturnal tongue biting from sleep bruxism is based on evidence of tooth wearing on dental examination[15]. Frequently, the RMD, starts with rhythmic movement of the muscles of the head and neck in supine position[3,4,7]. Episodes of repetitive movements recorded by PSG with video monitoring typically occur in presleep drowsiness, in light NREM sleep, or during brief arousals or transition to lighter stages of sleep, but the activity rarely occurs in deep slow wave sleep or solely during REM sleep[3,4,7,16]. A cluster of movements may last only a few minutes or longer than 30 minutes[3,4,6].

The cause of RMD is unknown. Golbin[9] hypotheorized it as a biological state, that is an attempt of transitioning or maintaining various states of vigilance. Some evidence suggested that vestibular stimulation may have a positive effect upon the development of the vestibulococchlear reflex[17]. Sallusto[8] postulated, based on the high prevalence of RMD in early infancy, that an innate tendency of the behaviour and the persistence of the behaviour into older childhood or adolescence is a result of learning behaviour. RMD occuring after head injury has been reported by Drake[18], however, it is possibly a result of a prior mild tendency for RMD from infancy exacerbated by the head injury. No neurological lesion has been mentioned in RMD although an abnormality of the basal ganglia has been described[19].

RMD is usually diagnosed by its characteristic clinical features[1,3,4,7,9]. Nevertheless, in some circumstances PSG evaluation may be necessary to differentiate this disorder from an epileptic disorder[1,2,6,7]. The treatment of RMD depends on the age of onset and the severity. Young infants or children require no treatment as the condition will usually resolve spontaneously in the second or third year of life[3,4,7,9]. RMD can be prevented in some individuals by avoidance of emotional stress and environmental stimulation. Short acting benzodiazepines such as clonazepam can be helpful in severe cases[7].

Nocturnal seizure, is commonly seen in frontal lobe epilepsy. Frontal lobe epilepsy is associated with a wide spectrum of clinical features that reflects the aura of the frontal lobe involved[20]. Frontal lobe complex and frontal lobe supplementary motor epilepsy are the forms of epilepsy particularly misdiagnosed as parasomnias or pseudoseizure[20-23]. The typical features of frontal lobe supplementary motor epilepsy include an abrupt

onset of tonic posturing of one or more extremities[5,20,21], manifestations of crying out or being unable to speak but usually preserving consciousness[5,20,21], and a brief attack (10-40 seconds); usually less than 2 minutes[20,21]. Nocturnal seizures typically occur on awakening from NREM sleep stage 2 in most instances[5]. The tonic component predominates but only rhythmic movement is observed in most children[24]. When the rhythmic movement involves muscles of the chin, tongue biting occurs. In some patients, seizures occurs in clusters[20-24]. The high incidence of normal or nonspecific interictal EEG findings[21-23] made standard interictal EEG study less valuable in making the diagnosis or localizing the epileptical focus. Even studying during an ictal attack, the EEG abnormality may be only subtle and interfered by muscle activities[22,23]. However, the prolonged EEG with video-PSG monitoring may demonstrate ictal activity and the relationship with the clinical manifestations[5,21]. To demonstrate precise epileptic foci for surgical treatment in some cases, subdural or depth electrodes EEG are needed [22,25]. Repeated observations of video-EEG may be necessary in some patients. Drug treatment of frontal lobe epilepsy include carbamazepine, phenytoin, valproate, phenobarbital, primidone, or some of the newer antiepileptic drugs such as lamotrigine, vigabatrin, felbamate[22,26]. In selected uncontrolled patients, surgery may be needed[27].

Finally, the clinician should keep in mind that : 1) There may be rhythmic movement disorders coinciding with epilepsy in some cases [22,28]. 2) An EEG overnight sleep study is not equivalent to a clinical standard EEG evaluation. The limited number of electrodes (usually only 2 to 8 channels in sleep studies) and slow paper speed (to permit all night recording) do not permit the specific EEG information obtained in a full 10/20 international system EEG for a neurologic evaluation. 3) Definite diagnosis of unusual events during sleep essentially needs the co-operation between a neurologist and sleep clinician.

SUMMARY

Many infants and children exhibit unusual mannerism behaviours and spells.These events cause concern to their parents and they are frequently brought to a physician. If the spells are infrequent and do not interfere with their daily activities, no intervention is necessary. On the other hand, a definite diagnosis and treatment are mandatory if the symptoms are severe. Many spells that are misdiagnosed as epilepsy by a pediatrician, but are RMD or another parasomnia. Definite diagnosis is based on the detailed history, complete physical examinations and appropriate investigations. The diagnostic methods currently available, such as prolonged EEG and PSG with video-EEG monitoring, frequently provide specific diagnosis and lead to appropriate treatment. In some cases, even after the sophisticated diagnostic tests have been performed, no diagnosis is forthcoming. In these cases, reassurance and follow-up are helpful to the young patient and their families.

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อาการกัดลิ้นขณะนอนหลับ

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อาการกัดลิ้น เป็นอาการแสดงออกที่พบร่วมได้บ่อยในภาวะชักซึ่งมีการเกร็งหรือกระตุกของแขนขาด้วย แต่ภาวะชักแสดงออกโดยการกัดลิ้นขณะนอนหลับเพียงอย่างเดียวพบได้ไม่บ่อยนัก พบได้ในการชักแบบ frontal lobe epilepsy รายงานนี้ได้นำเสนอผู้ป่วยเด็กสองรายซึ่งมีอาการแสดงออกโดยการกัดลิ้นในขณะนอนหลับเหมือนกัน แต่มีสาเหตุต่างกัน รายหนึ่งเป็น frontal lobe epilepsy อีกรายหนึ่งเป็น parasomnias (rhythmic movement disorders) พิสูจน์ยืนยันจากการตรวจ prolonged EEG และ polysomnography video–monitoring สำหรับ rhythmic movement disorders พบได้บ่อยในเด็ก แต่ rhythmic movement disorders แสดงออกโดยการกัดลิ้นขณะนอนหลับพบน้อยมากแม้ในรายงานต่างประเทศ ในประเทศไทยยังไม่มีผู้รายงาน แม้ว่าอาการแสดงกัดลิ้นขณะหลับจะเป็นอาการนำของผู้ป่วยที่มาพบแพทย์น้อยแต่มีความจำเป็นที่แพทย์ต้องให้การวินิจฉัยแยกสองภาวะนี้ให้ได้เนื่องจากการรักษาแตกต่างกัน รายงานนี้ได้ทบทวนและรวบรวมอาการแสดงออก การวินิจฉัย และการรักษาของ frontal lobe epilepsy และ rhytmic movement disorders ไว้

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