SHORT REPORT

Activity patterns of leg muscles in periodic limb movement disorder

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The movements of leg muscles in reference to periodic limb movement disorder (PLMD) have only been described in global terms. The sequences of contracting muscles that cause the PLMs are said to be stereotypical. There is, however, doubt about this fixed sequencing in PLMD. Our goal was to define the sequence of muscle movements in PLMs and then analyse their patterns. We recorded with surface EMG all movements of the muscles said to be involved in PLMs (extensor digitorum brevis, EDB; tibialis anterior, TA; biceps femoris, BF; tensor fasciae latae; TFL) as well as the quadriceps (Q) and soleus (S) muscles in 12 patients with restless legs syndrome combined with PLMD. Accompanying polysomnography provided the sleep parameters. In total, 469 movements were analysed. In only 12% was there the appearance of the classic movement (EDB-TA-BF-TFL) or its direct variants. The most frequent sequences were characterised by contraction of only the TA, TA-EDB only, or TA-EDB followed by all other combinations (32%). The pattern EDB only, EDB-TA, or EDB-TA followed by contraction of one or more other muscles, was seen in 18%. All other combinations appeared in much smaller numbers or only once. Eight patients had specific patterns. Three consistently started with the same muscle. One patient always contracted all six muscles. Six patients never contracted more than three muscles. The number of muscles contracted correlated positively with the appearance of arousal from sleep. The interval between onset of contractions within the PLMs varied randomly in a range of 0–1 s. Within PLMs many variations of muscle movements were documented. Patterns were recognisable, individually determined, and related to arousal from sleep.

Periodic limb movement disorder (PLMD) is characterised by periodic episodes of repetitive and highly stereotyped limb movements that occur during sleep.[1] Periodic limb movements (PLMs) are thought to be connected to nocturnal awakenings and unrefreshing sleep. Individuals with restless legs syndrome (RLS) usually have PLMs. As RLS has been identified in 5–15% of the population and in up to 80% of particular groups, such as those receiving dialysis for end stage renal disease,[2–4] PLMD is probably clinically relevant in many persons.

PLMs appear in all non-REM sleep stages and are well defined in terms of duration of the movements, interval between movements, and association with arousal from sleep.[5] Most PLMs are seen in the legs and are described as an extension of the great toe followed by partial flexion of the ankle, knee, and sometimes the hip. In its complete form, the movement is said to resemble the so-called triple response. From our clinical observation, we doubted whether this fixed combination or sequence of muscle contractions actually occurs in every PLM. Our impression was further endorsed by the recent publication by Provini *et al*,[6] who found large variations in the recruitment of leg muscles in PLMD.

The purpose of the study was to describe the sequences of muscle activation during PLMs and to correlate these muscle activation patterns with sleep parameters. We assumed that the findings could be of interest from a nosological point of view and might also give insight in the pathophysiology of PLMs.

PATIENTS AND METHODS

Twelve patients (median age 55 years, range 45–68 years; 6M, 6F) with idiopathic RLS combined with PLMD underwent a 24 hour polygraphy at home. RLS and PLMD were defined according to the International Classification of Sleep Disorders published by the American Sleep Disorders Association (ASDA).[1] All patients had a score of 15 or higher on the 40 points RLS Severity Score[7] and had a mean of at least 25 PLMs per hour of sleep during the polysomnography (electroencephalograph (EEG), electro-oculograph (EOG), chin electromyograph (EMG), respiration, and EMG of the leg muscles). The Rechtschaffen and Kales analysis was applied, as were the ASDA PLM scoring rules[5] including those for scoring arousal from sleep.[8] Surface EMG electrodes were used to evaluate six muscles of the right leg (extensor digitorum brevis (EDB), tibialis anterior (TA), biceps femoris (BF), tensor fasciae latae (TFL), soleus (S), and quadriceps (Q), and also the TA of the left leg. The number of PLMs per hour did not differ significantly for both TAs, thus, owing to technical limitations, we chose to analyse only the right leg. The onset of the muscle activation was taken as the moment at which the amplitude of EMG activity was double that of the preceding baseline, and was measured with an accuracy of 1 millisecond. Associated EEG arousals were reported. PLMs occurring in non-REM stages 2, 3, and 4 were analysed. Within PLMs, the muscle activation patterns were described in terms of the order of muscles contracting and time differences in onset of EMG activity in the muscles involved.

RESULTS

We evaluated 469 PLMs in total. The TA was most frequently identified as the muscle that contracted first (n = 151; 32% of all PLMs). The contracting patterns starting with the EDB occurred in 18% (n = 85 PLMs) of the recordings. Only 55 of the PLMs (12%) could be described as the classic triple response or its variants. The full triple response (EDB-TA-BF-TFL, in that order) was found only once. The other

Abbreviations: ASDA, American Sleep Disorders Association; BF, biceps femoris; EDB, extensor digitorum brevis; EEG, electroencephalograph, EMG, electromyograph; EOG, electro-oculograph; PLMD, periodic limb movement disorder; PLMs, periodic limb movements; Q, quadriceps; RLS, restless legs syndrome; S, soleus; TA, tibialis anterior; TFL, tensor fasciae latae

Table 1 Onset muscle and sequences of muscle contractions in PLMs

Onset muscle	Following muscles	Number of PLMs
EDB	None	20
EDB	TA, BF*	9
EDB	TA, BF, TFL*	1
EDB	TA	29
EDB	(TA and) all other sequences	26
Total EDB		85
TA	None	60
TA	BF*	22
TA	EDB, BF*	18
TA	EDB, BF, TFL*	3
TA	BF, TFL*	2
TA	(EDB and) all other sequences	46
Total TA		151
BF, TFL, Q or S (no EDB or TA)	All other sequences	233
total PLMs		469

*Sequences belonging to the classic triple response or its variants.
EDB, extensor digitorum brevis; TA, tibialis anterior; BF, biceps femoris; TFL, tensor fasciae latae; Q, quadriceps; S, soleus.

combinations in this category were seen somewhat more frequently (table 1). Thus, 50% of the PLMs did start with toe (EDB) or foot (TA) lifting muscles; the other half did not. In the latter category, all other combinations were found, often only once or in low numbers. The different forms of PLMs occurred in all stages of non-REM sleep.

Quite unexpectedly, we often found successive contracting of agonists and antagonists at onset of the PLMs, giving to and fro movement in the ankle or knee. These sequences (successive contractions of EDB and S, TA and S, Q and BF) occurred in 77 PLMs (16%).

The differences in onset time of each pair of consecutively contracting muscles were calculated. These time differences could have positive or negative values depending on which muscle contracted first; for example, in the TA-BF combination the time difference was negative when the TA moved before the BF. As we analysed contractions of six different muscles, intervals were calculated for 15 pairs of muscles. Some pairs were found only a few times, while others occurred frequently. For the latter, the differences in onset time invariably had a Gaussian distribution, as did the grand mean of all combinations (fig 1).

We discovered personal patterns. Three patients always started with the same muscle: EDB, TA, or Q. One patient contracted all six muscles in every PLM, but in various sequences. For all patients, the number of muscles contracted was associated with EEG arousals from sleep. When four or more muscles were part of the PLM ($n = 220$), arousals occurred in 139 instances. Movements of three muscles or less led to an arousal in 68 of the 249 movement episodes in this category (difference between both groups: $p<0.01$; χ^2 test). One hundred and seventy-eight arousals occurred in NREM 2 sleep. Sequences with four or more muscles were seen more frequently in superficial than in deep sleep (NREM 2: 54% , NREM 3+4: 33% of all PLMs in that sleep stage), but, independent of the sleepstage, PLMs with at least four muscles involved were associated more frequently with arousals than the less extended PLMs (NREM 2: $p<0.01$, NREM 3+4: $p<0.05$).

DISCUSSION

Approximately 50% of the PLMs started in the extensors of the toes, dorsiflexors of the ankle, or both. Even when these muscles initiated the sequence of contractions and the PLM started in the classical way, various patterns still occurred. In the other half of the PLMs studied, we found that nearly all

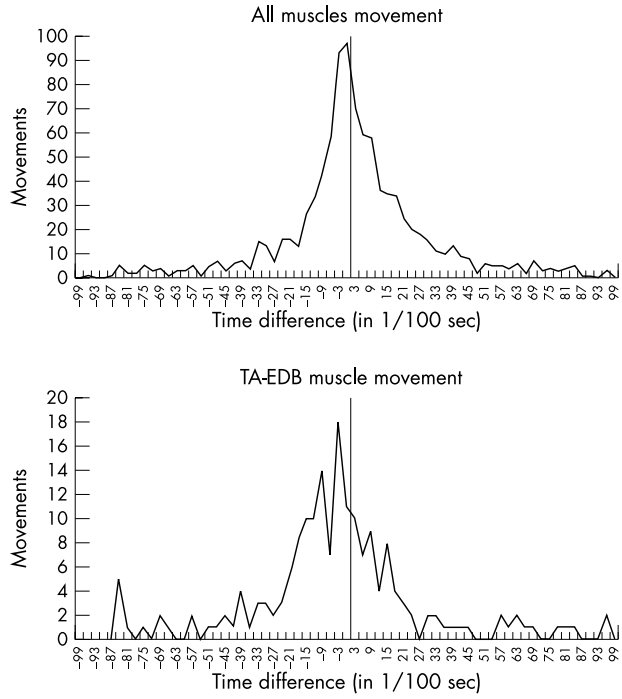

Figure 1 Intervals between onset of EMG activity in consecutively contracting muscles. (A) Distribution of intervals of all possible pairs (grand mean). (B) Distribution of intervals for the frequently occurring pair of extensor digitorum brevis (EDB) and tibialis anterior (TA) muscles.

muscle movements were random. Our findings and those by Provini *et al*,[6] which were based on surface EMG, contrast with the classical description, [1] which is based on clinical observations of muscles moving during sleep. EMG may be a more sensitive way of detecting muscle action, but it is difficult to understand how this explains the large discrepancies in findings.

Although we could not confirm the classical description of PLMs with its suggested order of movements, we did find some patterns in the muscle contractions. Awaiting corroboration of our results, we propose to change the description of PLMs as follows: ''The movements usually occur in the legs and consist of varying combinations of contracting muscles. In nearly half of the PLMs, extension of the great toe, and partial dorsiflexion of the ankle occur. Flexion of the knee and hip may be part of the movement. There is no fixed order in these movements during the PLM''. In the definition of PLMD, the part of ''repetitive and highly stereotyped limb movements'' should be simplified to ''repetitive limb movements''. For comparison, see the original definitions.[1]

The large variation in the intervals between muscle contractions and their Gaussian distribution make ''prewired'' patterns in the sequence of movements less likely. Up to now this idea was part of the theories on the origin of PLMs. The concept of more or less fixed patterns in the muscle activation in PLMs was based on functional connections between motor neurones in the spinal segments L4–S1 or on cooperating nuclei in the medulla oblongata that influence these neurones, the latter theory being more probable.[3] Obviously the anatomical aspects are correct, but our findings cast doubt on the physiological part of these notions. According to the results of previous studies, we still think that a pathological pacemaker in the medulla oblongata is the origin of the PLM. The spinal segment is activated through descending tracts. This simple wiring is under the influence of inhibiting and excitatory input at least at three levels: the basal ganglia, the medulla oblongata, and

the spinal segment L4–S1 itself.[3 9] This complex input makes the recruitment of motor neurones variable, and consequently the movements are inconsistent in terms of muscles involved, sequences of muscles, and intervals between onset of activation.

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HISTORICAL NOTE

Barré-Liéou ''syndrome''

Sometimes, non-entities are instructive. Barré's name is commemorated in the Guillain-Barré eponym,[1] but unfortunately, also in the contentious Barré-Liéou syndrome.[2 3] Barré described a syndrome of the posterior cervical sympathetic nerves and its frequent cause—chronic cervical arthritis. Barré-Liéou syndrome has the synonyms brachialgia paraesthetica nocturna, cervical migraine, chronic cervical arthritis, migraine cervicale, neurovertebral dystonia, syndrome sympathique cervicale postérieur,[4] and vertigo of cervical arthrosis. The number of synonyms appears to be inversely related to the soundness of the diagnosis.

It has not been agreed as a neurological entity, but is used as a repository for undiagnosed symptoms including those succeeding acute neck injuries, despite the term *chronic* originally described. Symptoms alleged[5 6] are occipital headaches,[7] nystagmus on head movement, tinnitus, spasms, blurred vision, corneal hyperaesthesia, and corneal ulcers. Other symptoms (Neri) are anxiety, depression, and memory and cognitive disorders.[8] This non-specific constellation has been ascribed to trauma or arthritis of the third and fourth cervical vertebrae or disks, which in turn cause a disturbance of the cervical sympathetic nerves and disordered circulation,[2 3] in the cranial nuclei V and V111.

Much of the literature predates 1990 and appears in journals of chiropraxis, acupuncture, and manipulation (references available from the author). Although the validity of Barré-Liéou syndrome is not generally accepted,[9] the term still appears in the literature. Yang Choen Liéou[3] described a sympathetic disorder associated with cervical ''arthritis'' in his thesis of 1928. The Bärtschi-Rochaix syndrome is very similar and of equally dubious substance.

Jean Alexandre Barré (1880–1967) was born in Nantes and died in Strasbourg.[10] He is well known for his work with Guillain on inflammatory polyneuritis (1916). Barré was an intern with Babinski and in 1912 published his thesis on *Les ostéoarthropathies du tabes*. He met Guillain in the first world war at the centre neurologique of the 6th army. There he served with an ambulance unit caring for patients with major injuries, with outstanding bravery and skill, acknowledged by the *Légion d'Honneur*, France's most important military distinction. Because France reoccupied Alsace-Lorraine at the end of the war, a chair was needed in Strasbourg. On his return from the front he was appointed Professor of Neurology in Strasbourg, at that time unique because only in Paris was such a chair extant.[1 10]

His interests were also in neuro-otology and vestibular syndromes, and he founded *Revue d'Oto-Neuro-Ophthalmologie*. He was also preoccupied by semiotics, an interest probably stimulated by Babinski.

In world war two, Barré rejoined the army but was sent to Clermont-Ferrand in the free zone, ''Vichy-France'', seat of the new government. Barré retired in 1950 and sadly suffered a stroke, which left him dysphasic.[1 10]

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