

POLYSOMNOGRAPHY IN IDIOPATHIC MUSCLE PAIN SYNDROME (FIBROSITIS)

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SUMMARY — Muscle pain occurs in various neuromuscular disorders with characteristic physiological or biochemical abnormalities. There is, however, a group of patients in whom there is no clear physiological or structural basis for their pains. This syndrome has been called fibrositis or fibromyalgia. Sleep abnormalities have been reported in some of these patients, but have not been confirmed by others. We studied 8 patients with this disorder and found sleep abnormalities that were characterized by nocturnal myoclonus, alpha-delta sleep, and abnormalities compatible with depression. Polysomnography was, therefore, instrumental in helping direct the treatment of these patients. Therapeutic approaches aimed to correct the specific disorders were effective in improving the pain symptoms

Polissonografia na síndrome de dor muscular idiopática (fibrosite)

RESUMO — Dor muscular ocorre em diversas doenças neuromusculares com anormalidades fisiológicas e bioquímicas características. Contudo, há um grupo de pacientes nos quais não são detectadas claramente bases fisiológicas ou estruturais que expliquem suas dores. A síndrome tem sido também chamada fibrosite ou fibromialgia. Anormalidades do sono têm sido relatadas em alguns desses pacientes, embora nem sempre confirmadas. Estudamos 8 pacientes com esse quadro e encontramos anormalidades do sono caracterizadas por mioclono noturno, sono alfa-delta e anormalidades compatíveis a depressão. Assim, a polissonografia é método auxiliar útil no estabelecimento do modo de tratar desses pacientes. As medidas terapêuticas adotadas visavam a corrigir as desordens específicas encontradas, tendo sido eficazes para aliviar os sintomas de dor.

Muscle pain is a symptom in a number of neuromuscular disorders that are usually diagnosed clinically or in combination with electromyography or muscle histochemistry and biochemistry 23. There are, however, some patients for which there is not a clear, organic explanation for their muscle pain. This syndrome, which is frequently called fibrositis 3,4 or fibromyalgia 3,25, has a fairly characteristic clinical presentation in which the presence of «trigger points» seem to be an important diagnostic feature 4,8. Recently, Moldofsky et al.^{1,2} found sleep abnormalities in fibrositis, characterized by alpha activity on the EEG during non-REM sleep, an electroencephalographic pattern that was initially described by Hauri and Hawkins and called alpha-delta sleep 10.

Moldofsky and his colleagues postulated that patients with these abnormalities had a lack of restorative sleep that caused secondary fatigue and muscle pains^{1,2}. Recently, however, Golden and colleagues reported that they were unable to confirm these earlier findings 6. In order to clarify further the sleep abnormalities in this condition, we performed polysomnographic (PSG) studies in a group of patients with idiopathic muscle pains who fulfilled the diagnostic criteria of fibrositis as outlined by others^{1,2}.

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PATIENTS AND METHODS

We studied 8 patients with chronic muscle pains and fatigue who had a history of trigger points that fulfill the criteria of Moldofsky and Lue 17 for «fibrositis». The sex, age, and pain duration for these eight patients are summarized in Table 1. All patients were studied clinically by ischemic exercise tests, by muscle biopsy for biochemistry and biochemical tests, and by electromyography. All tests were negative, and the etiology for muscle pains in these patients could not be demonstrated.

All of these subjects were submitted to polysomnography for 1 or 2 nights at their regular bedtimes and for the duration of their average sleep in the circadian cycle. Polysomnography comprised EEG activity, recorded by C3-A2 and A4-A2 scalp electrodes, eye movement recorded by right eye to frontal pole and frontal pole to left eye electrodes. Muscle activity was recorded by chin and leg electrodes. Respirations were monitored by nasal and buccal thermistors and by chest and abdominal strain gauges. EKG activity was recorded by two chest electrodes. Stage Rapid Eye Movement (REM) latency in this study was measured from stage 2. All patients were monitored by video camera with infrared illumination and microphones. The room temperature setting was 73°F.

The records were scored according to standard criteria 22. Data from these individuals were compared with those from normal patients previously studied in our laboratory. With one exception, all 8 patients under study also had psychiatric surveys and responded to Beck's Inventory for depression. Results of polysomnography and psychiatric evaluation on the patient population studied are shown in Table 2.

	Male	Female	Total
Number of patients	7	1	8
Mean age (years)	45.6	40	44.9
Range (years)	31-55	—	31-55
Mean pain duration (years)	8.4	10	8.6
Range (years)	1-27	—	1-27

Table 1 — Sex, age, and pain duration in eight fibrositis patients.

Patient number	Polysomnography						Psychiatric	
	Sleep latency (min)	REM latency to Stage 2 (min)	Arousals (#)	Awakenings (#)	Alpha-Delta sleep	Myoclonus	Psychiatric interview * diagnosis	Beck Inventory Score
1	6	66	65	3	No	Yes	A	8
2	35	112	108	7	No	Yes	A+D	14
3	1	58	86	1	No	Yes	D	6
4	10	214	214	17	No	Yes	A	13
5	7	53	42	0	No	No	—	2
6	10	168	47	4	Yes	No	D	16
7	10	214	14	14	Yes	No	A+D	6
8	5	65	18	3	No	No	D	2

Table 2 — Polysomnographic and psychiatric findings in eight patients with fibrositis.

* A, anxiety; D, depression.

For purposes of this research, REM latency less than 70 minutes and Beck Inventory Score more than were considered to indicate depression. More than 30 arousals and more than 3 awakenings were considered abnormal. Normal sleep latency, <10 min; normal REM latency to Stage 2, >70 min.

Patient number	Medication *	Fibrositis symptoms
1	CZ + AMT	Disappearance of symptoms
2	CZ + AMT	Mild improvement
3	CZ + AMT	Mild improvement
4	CZ + AMT	Mild improvement
5	AMT	Moderate improvement
6	AMT	Lost follow-up
7	ALP	Significant improvement
8	AMT	Disappearance of symptoms

Table 3 — Response to therapy in eight fibrositis patients.

* CZ, clonazepam; AMT, amitriptyline; ALP, alprazolam.

The decision to treat patients with clonazepam, amitriptyline, and alprazolam, was based upon the clinical diagnosis and psychiatric condition, aided by PSG. The degree of improvement in fibrositis symptoms and response to therapy were established as follows: no improvement; minimal improvement; mild improvement; moderate improvement; significant improvement; and disappearance of symptoms.

RESULTS

As shown in Table 2, psychiatric interviews revealed signs of both anxiety and depression in two patients (patients 2 and 7). Depression was documented during psychiatric interview in three patients (patients 3, 6, and 8). Beck's Inventory revealed high scores for depression in patients 2, 4, and 6. Four individuals showed REM sleep latency less than 70 minutes, which is consistent with depression 5,8,15. It is to be noted that depression in two of these subjects was not detected by psychiatric interview or by Beck's depression inventory. Other causes for short REM latency, such as narcolepsy, schedule disorders, and effect of drugs or alcohol, were excluded in all cases. Alpha-delta activity was present in only two patients; four subjects exhibited nocturnal myoclonus. Neither parameter appeared to correlate with psychiatric results.

Responses to therapy in these patients are summarized in Table 3. Except for one patient whose follow-up was lost, some degree of improvement in symptoms of fibrositis was noted in all patients. Significant improvement or disappearance of symptoms was noted in three subjects and mild to moderate improvement in the remainder.

COMMENTS

Patients with muscle pains are often a challenge to neuromuscular specialists and neurologists, and when these are not explained by standard diagnostic tests for metabolic physiological or structural muscle diseases, the management could be rather frustrating. The patients studied belong to such group, with unexplained muscle pain and the characteristic of «fibrositis» syndrome 3,4. Although our population was somewhat unusual because of its male predominance 8, it fulfilled the diagnostic criteria outlined by others 17. Reported studies of muscle biopsy mainly show nonspecific findings and type II fiber atrophy 13,14 with somewhat abnormal oxidative enzyme staining, muscle necrosis, and subsarcolemmal accumulation of mitochondria U-13. The abnormalities are by no means specific.

Because the diagnosis of «fibrositis» is based on clinical findings, groups of patients in previous studies have not always been similar. In some studies, all patients with unexplained muscle pains were included. In others, strict clinical criteria were used to define the study population 17. Even when strict criteria were applied, however, the groups have not been homogenous. Some investigators have reported a high incidence of depression 1,7,12,2^ while others have separated «fibrositis» patients from those with depression-induced muscle pain 9,25. The present study demonstrates

that such separation is artificial, based on the high incidence of depression in patients with classical fibrositis.

In agreement with Moldofsky and colleagues^{1^} we have found that sleep abnormalities are common in the «fibrositis» syndrome. These authors considered that alpha-delta sleep patterns characterize this syndrome. However, in our patient population, the alpha-delta sleep pattern was only present in two patients. We also found that nocturnal myoclonus was present in half of our patients. Moldofsky et al.²⁰ found similar abnormalities in their older population, suggesting that there is a group of «fibrositis» patients in which these, rather than the alpha-delta sleep is more characteristic. It is conceivable that nocturnal myoclonus precedes the alpha-delta sleep pattern and that our patients would acquire abnormalities later. However, nocturnal myoclonus could, by itself, trigger muscle pains and fatigue, as occurred in our patients; these individuals did improve with therapy aimed to suppress myoclonus.

We cannot determine if depression was a consequence or the cause of muscle pains, but the finding by others of alpha-delta sleep pattern in patients with rheumatoid arthritis¹⁸ and post-traumatic pains²⁴ suggest that painful conditions can produce this sleep abnormality, which leads to more muscle pains, thereby creating a vicious cycle.

Our studies indicate that polysomnography appears to have importance in the evaluation of patients with unexplained muscle pains which often present to neuromuscular clinics and can aid in the determination of therapeutic intervention by demonstrating leg myoclonus in some patients, as in 50% of our cases, or aiding and reaffirming the diagnosis of depression in others by showing short REM latency, also true of 50% of our patients.

While the number of patients in this study is relatively small, it represents a unique group of individuals fitting the diagnosis of fibrositis and who were thoroughly evaluated by neurological means; by muscular pathology, including muscle biopsy and psychiatric evaluation; and by polysomnography. Except for chronic pain, as a group these patients did not show another diagnostic finding clearly defining the syndrome (Table 2). Specifically, while 50% of the patients displayed leg myoclonus in their sleep, only two had documented alpha-delta sleep, pointing to the nonspecificity of this finding in the disorder. On the other hand, clinical psychiatric evaluation and polysomnography profile pointed to a diagnosis of anxiety, depression, or both. Also, 50% of the patients demonstrated leg myoclonus during the PSG, which helped the physicians to direct the treatment, with some success. We recognize the need for studies of additional cases in order to firmly establish the diagnostic criteria for this condition. The findings in this study, however, permitted more defined therapeutic management, with overall improvement in 7 patients; one additional patient was lost to follow-up.

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