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Case Report

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Painful Hand and Moving Fingers: Case Report and Literature Review

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Abstract

We describe a 41 year-old man with painful hand and moving fingers (PHMF) syndrome. He presented with pain in the ulnar distribution of the left forearm and hand, and continuous abduction-adduction confined to the left IV finger. The symptoms started several months after trauma of the left cubital fossa with ulnar nerve injury. Response to therapy has remained suboptimal. We summarized the reported cases of the PHMF.

Keywords

Painful hands and moving fingers, Painful legs and moving toes

Introduction

PHMF syndrome, first reported by Verhagen et al. in 1985 [1], is relatively uncommon with features of aching pain in the upper extremity and spontaneous involuntary movements in fingers, with a paucity of published reports to date. The syndrome is akin to a similar syndrome involving lower extremities, which is known as painful legs moving toes (PLMT) syndrome. It was initially coined by Spillane et al. in 1971 [2], and is more common. We report a case of the PHMF, and provide summary of the published cases of this disorder.

Case Report

A 41 year-old man presented with a 12-year history of pain in the ulnar region of the left forearm and hand, numbness in the left IV and V digits, and continuous abduction-adduction movements of the left IV finger. He was not able to voluntarily suppress the movements. The symptoms started months after a traumatic crash injury of the left cubital fossa, resulting in the lesion of the ulnar nerve. Pain was partially relieved by duloxetine, but failed to respond to therapeutic doses of gabapentin, pregabalin, and venlafaxine. Physical examination revealed a normal mental status, cranial nerves, reflexes, coordination, and motor power. There were continuous rhythmic or semi-rhythmic abduction-adduction movements of the left IV finger. Sensation to touch was decreased in the left IV and V fingers. Electroencephalogram, and MRI of the neck, left brachial plexus, and head were unremarkable. Motor nerve conduction studies of the ulnar nerve using inching technique showed a delay in the conduction velocities above the medial epicondyle of the elbow (Table 1).

Discussion

PHMF was reported in the middle-aged female with left breast

Table 1: Ulnar motor inching technique across the elbow.

	Latency (ms)	Distance (mm)	Velocity (ms)
6 cm to 4 cm distal	0.4	20 mm	50 m/s
4 cm to 2 cm distal	0.3	20 mm	67 m/s
2 cm to elbow	0.3	20 mm	67 m/s
Elbow to 2 cm prox.	0.4	20 mm	50 m/s
2 cm to 4 cm prox.	0.9	20 mm	22 m/s
4 cm to 6 cm prox.	0.3	20 mm	67 m/s

Note: Elbow: medial epicondyle, distal: below medial epicondyle, proximal: above medial epicondyle, ms: milliseconds, mm: millimeters

adenocarcinoma treated with surgery and radiation therapy [1]. Seven months later, she developed pain in the distal left forearm and involuntary continuous movements of her left fingers at the metacarpophalangeal joints, and was found to have brachial plexopathy. She remained refractory to therapy. Although it is widely accepted that this is the first published case report of the PHMF, in fact, the syndrome was reported previously [3, 4], in both cases associated with the PLMT. Since the original report, there were only a few scattered published cases, all sharing some commonalities. Table 2 summarized all the reported cases of the PHMF in chronological manner, with the most relevant features. The

Table 2: Summary of the published reports of the PHMF syndrome.

Study	Age Sex	Related event	Pain	Abnormal movements	Associated features
Montagna et al. [3]	74 F	Polyneuropathy, B12 deficiency	Feet	L fingers IV and V adduction/abduction	Paresthesia evolving from the L hand to feet to R hand PLMT
Schoenen et al. [4]	68 F			L thumb flexion/ extension	PLMT
Verhagen et al. [1]	54 F	Post-radiation brachial plexopathy	Left forearm	Fingers at MCP joints	Left forearm paresthesias
Funakawa et al. [15]	52 M	Left hand crash injury and amputation	Left hand	Fingers at MCP joints	
Ebersbach et.al. [5]	64 M	No trauma or neuropathy	Painless	Left I, II, III digits	Right PLMT
Jabarri et.al. [16]	35 F	Right shoulder trauma after a fall	RUE pain, followed by LUE pain	Right III-V digits, followed 10 months later by the left III-V digits	Both upper limbs involved
Supiot et al. [7]	68 M	CABG surgery	Distal medial hand and forearm	IV and V digits	
	25 F	Neck trauma, C8 radiculo-pathy	Medial right arm	IV and V digits	
	49 M	Left forearm injury	IV and V digits and palm.	IV and V digits abduction/ adduction	Hand and forearm paresthesia
	66 F	PCA CVA	Left hand	III-V digits.	Left superior quadrantanopsia
Sudo et al. [13]	56 F	Radiculopathy/ myelopathy	Right forearm	Right III finger adduction/abduction,	Temporary suppression by voluntary effort Symptoms resolved after cervical laminoplasty
Wider et al. [8]	55 F	Radial bone fracture and carpal tunnel release	Left hand and middle finger	III digit	CRPS-I Wearing glove or tactile stimulation of medial nerve region relieved pain and movement
Singer and Papapetro-poulos [11]	20 F	Muscle strain	Painless	MCP joints of the R III, IV, V fingers > L IV finger	Improvement after Botox injections
Alvarez et al. [6]		Polyneuropathy Sjogrens disease			PLMT Improved with gabapentin
		Polyneuropathy Sjogrens disease			PLMT Refractory
		Polyneuropathy B12 deficiency	Painless		PLMT Refractory
		Lumbar stensis/ radiculopathy	Painless		PLMT Improved with Gabapentin
Miyakawa et al. [9]	36 M	Cervical spondylosis, C6 radiculopathy	R arm, fingers I, II, neck and scapula	R thumb	Not suppressible Disappeared in sleep Resolution after Laminoplasty and Foraminotomy
Hayat et al. [10]	40 F	None	Painless	II digit	Present during sleep; Voluntary partial suppression of movement for seconds PLMT

Note: F: female: M: male, CRPS-I: complex regional pain syndrome type-1 CVA: cerebrovascular accident; MCP – metacarpophalangeal; PCA: posterior cerebral artery, PLMT – painful legs moving toes

etiology of the PHMF is heterogenous, and often involves trauma, compression neuropathies, peripheral nerve, plexus, and/or root disease. In the majority of cases pain precedes involuntary finger movements by several months or years, and is usually the primary reason for seeking medical advice. Pain is intense, constant, pulling, throbbing, burning, or crashing in nature, and located at the distal forearm and hand (ulnar or radial region). However, pain is not a mandatory component, and painless variant of PHMF syndrome was reported in 4 patients, interestingly, always in association with the PLMT [5, 6]. Movements are involuntary, complex (writhing, flexion/extension, abduction/adduction), difficult to imitate, only partially, if at all, suppressed by the voluntary effort, and are often present during sleep. Both pain and involuntary movements are aggravated by cold, and relieved by the heat and tactile stimulation [1, 5-10]. The symptoms are often refractory to therapy. The following oral medications were tried for the management of the PHMF, often unsuccessfully: anticonvulsants, antidepressants, muscle relaxants, neuroleptics, benzodiazepines, and opioids [1, 5-10]. Limited success was reported with nerve blocks, epidural blocks, sympathetic blockade, and spinal cord stimulator. Botox therapy [11] and decompressive surgery (foraminotomy and laminoplasty) [9, 12] resulted in a significant improvement or even complete resolution of the symptoms.



Video: Video displays involuntary intermittent rhythmic adduction-abduction of the left IV digit.

The pathophysiology of the PHMF is complex and diverse, with involvement of both peripheral and central mechanisms - spinal and supraspinal. The fact that PHMF often occurs in the setting of painful neuropathies, suggests a peripheral source, probably involving C and delta fibers [13]. On the other hand, the latency period between the injury and the onset of clinical symptoms, as well as complex pattern of movements, suggests a central source [14]. While PNS trauma or injury is often a predisposing factor for the PHMF, idiopathic cases were also reported. The most accepted pathophysiology is the alteration in afferent sensory information with subsequent reorganization of segmental and suprasegmental efferent motor activity [14]. It was suggested that in this process of sensory-motor pathological synaptic reorganization, spinal cord may work as a pacemaker. This may result from segmental sensory inputs from lesion(s) in afferent sensory fibers of the posterior root that generates spontaneous motor responses in anterior horns cell with repetitive muscle movements [15]. Abnormalities of the somatosensory pathway affecting the brain stem, thalamus, and sensory cortex may also be involved. The spreading of a rhythmic PHMF syndrome from one hand's muscles to the other suggests the same central generator [16]. However, the asynchrony movements in hand and leg muscles in individuals affected by the periodic leg movement syndrome (PLMS) and PHMF suggests different central generators [5]. Whether resulting dysfunction occurs at the spinal or supraspinal level, is not clear, but coexistence of central and peripheral mechanisms points to the abnormal sensory motor integration.

Conclusion

PHMF syndrome is a rare disorder that often starts after PNS injury and presents with forearm and/or hand pain and involuntary finger movements. The pathophysiology is unknown and therapy is often ineffective.

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