

Pain syndrome and focal myokymia due to anterior interosseous neurovascular relationships: report of a case and neurophysiological considerations

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✓ The anterior interosseous nerve can become entrapped within the antecubital fossa at its origin from the median nerve, which results in the so-called Kiloh-Nevin syndrome. In this report, the authors describe an atypical anterior interosseous nerve syndrome due to neurovascular relationships with the anterior interosseous artery. The patient complained of unbearable analgesic-resistant pain within the forearm and focal myokymia in muscles innervated by branches of the anterior interosseous and distal median nerves. Pain and myokymia were alleviated by inflated blood pressure cuff compression in the bicipital region when the arterial pulse was abolished distally. Microsurgical correction of the pulsatile arterial compression resulted in relief from pain and myokymia. Neurophysiological considerations of the mechanism underlying "irritative" neuropathy and myokymia are discussed.

KEY WORDS • anterior interosseous nerve • myokymia • pulsatile arterial compression

KILOH and Nevin⁹ in 1952 reported that lesions of the anterior interosseous branch of the median nerve "have been recorded so infrequently that [their] description of two cases of isolated neuritis of the nerve seem[ed] justifiable." Subsequently, approximately 100 cases of anterior interosseous nerve syndrome have been reported in the literature. The spontaneous appearance of the condition is now considered to be an entrapment neuropathy caused by fibrous bands at the so-called "arcuate ligament" in the antecubital fossa.^{5,13,16,17} The major features of the syndrome are motor deficits of the flexor pollicis longus, pronator quadratus, and flexor digitorum profundus muscles. There is a resultant loss of hand dexterity and pinch attitude of the thumb and index finger. This syndrome is also characterized by a lack of sensory deficits although the anterior interosseous nerve is considered to have mainly motor function.⁹ Pain may occur but it is transitory and confined within the anterior forearm and elbow, typically heralding paresis.^{16,17}

In this report we describe a patient who complained of persistent and unbearable pain in the lateral forearm and wrist, associated with segmental myokymia without sensory or motor deficits. Clinical and neurophysiological assessment led to surgical exploration within the antecubital fossa, where atypical relationships between the anterior interosseous nerve and the anterior interosseous artery were found. No similar cases of anterior interosseous nerve involvement with focal myokymia and pain were

found in the literature. This case suggests that there are neurophysiological considerations regarding the so-called "neurovascular conflict" as a cause of atypical peripheral nerve entrapment, originally reported by Segal, *et al.*,¹⁴ in 1992.

Clinical Material and Methods

Illustrative Case Report

This 60-year-old white woman was referred to our institution complaining of a 2-month history of severe aching and recurrent cramping pain within the anterior left forearm and wrist. She was a right-handed housewife without significant anamnestic data for trauma or heavy utilization of her left arm.

Examination. General physical examination revealed only easily provoked sweating in limb extremities. Neurological examination revealed a dystonic-like attitude of the left wrist and first fingers. At rest the patient had difficulty extending the left wrist and fingers due to hyper-tonia and continuous vermiciform muscle movements involving the flexor and thenar muscles; no involuntary movements were observed in other muscles of the hand and forearm. No signs of peripheral neuropathy were detected. The dexterity of the left hand was compromised because of pain and postural analgesic supination of the forearm. The pain was resistant to antiinflammatory drugs

Anterior interosseous neurovasculopathy in painful myokymia

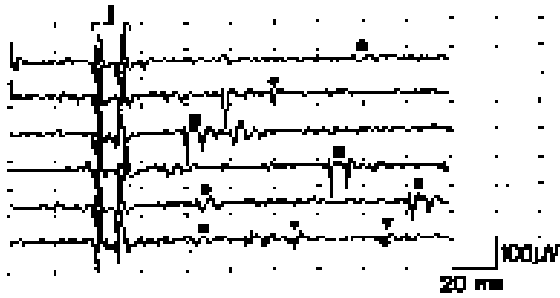


FIG. 1. Electromyographic recording showing neuromuscular hyperactivity in the opponens pollicis muscle. The traces were triggered by the rhythmic electrical discharge of a doublet near the needle recording electrode (*arrow*). Other rhythmic, asynchronous electrical discharges were also recorded both near and distant from the needle electrode (*boxes, circles, arrowheads*). These findings are consistent with myokymia.

including corticosteroids; carbamazepine and phenytoin, administered for the recurrent cramping and neuralgic pattern of pain, were also unsuccessful. Magnetic resonance imaging and ultrasonography ruled out schwannoma or gross compression of the nerve by soft tissue or bone masses.⁴

Neurophysiological assessment showed normal distal motor latency and normal sensory action potential of the median nerve. Segmental sensory and motor conduction of the median nerve at the wrist were also normal. No sub-clinical peripheral nerve involvement was found. Using a concentric needle electromyographic evaluation, very intense, continuous, spontaneous discharges were recorded. These were characterized by rhythmic doublets (Fig. 1) and triplets, which were noted in hemithenar and flexor muscles but not in hypothenar, interossei, or other muscles. This focal myokymia^{1,2} involved muscles innervated by both the median nerve and the anterior interosseous nerve.

It was peculiar that the ischemia test, conducted by inflated blood pressure cuff compression at the bicipital region, resulted in evident attenuation of both myokymic activity and pain when the inflated pressure was higher than arterial pressure. A pharmacological block of the median nerve using a local anesthetic (2 cc lidocaine) within the antecubital fossa led to transitory total abolition of spontaneous muscle activity within 1 to 2 minutes; this result led to the exploration of the median nerve at the origin of the anterior interosseous nerve.

Operation. While the patient was under general anesthesia, surgery was performed using microsurgical techniques. The left median nerve was exposed immediately below the lacertus fibrosus between the retracted brachioradialis and pronator teres muscles. The flexor digitorum superficialis muscle was sectioned; a true arcuate ligament, as described by Fearn and Goodfellow,⁵ was not found. The origin of the anterior interosseous nerve was easily identified lying on the flexor digitorum profundus muscle. The radial artery and anterior interosseous artery appeared larger than normal. The latter indented the nerve and one or two fibrous bundles were observed crossing the artery at its inferior surface (Fig. 2). The close adherence

between nerve and artery was relieved through sectioning the small fibrous bundles under the vessel; the nerve trunk was mobilized and a pedunculated fat graft was interposed to avoid recurrence of the pulsatile arterial compression.

Results

The patient's myokymia resolved after surgery. The pain became less severe without cramping features and disappeared completely after approximately 1 month. The patient recovered full dexterity of the hand within 3 weeks. She is still doing well at 1-year follow up without analgesic drugs.

Discussion

The hallmark of this patient's condition was the presence of neurovascular relationships as pathogenic events of the very unusual syndrome characterized by pain unresponsive to analgesic agents and neuromuscular hyperactivity consistent with segmental myokymia involving muscles innervated by median nerve and anterior interosseous nerve branches. The provoked ischemia test that temporarily abolished pain and myokymia was the only factor suggesting this peculiar neurovascular peripheral nerve entrapment syndrome. In fact, the same test is known to increase focal pain secondary to conventional entrapment or compression by nonvascular structures.

Focal myokymia is usually reported as occurring after radiotherapy of the brachial plexus and in chronic as well as acute peripheral nerve injury.¹⁰ In our patient the condition was not associated with nerve damage: electrophysiological examination revealed normal nerve function.

There are three indicators in this case that suggest that myokymia and pain may be due to neurovascular compression: 1) the surgical findings of anomalous arterial-nerve contact; 2) the transient attenuation of bioelectrical activity on ischemic testing; and 3) the abolition of both myokymia and pain following resolution of the compression. It must be pointed out that although the site of compression involved only the anterior interosseous nerve, myokymia was also present in muscles innervated by the median nerve. The diffusion of pathological neuromuscular activity may be explained by the well known mechanism of ephaptic and antidromic conduction, as suggested in hemifacial spasm and spasmodic torticollis.^{6,11,12,15} In other words, an ephaptic irritative focus at the site of the peripheral neurovascular entrapment may antidromically kindle wider pools of motor neurons, resulting in spatial extension of abnormal muscle activity. Pain origin may also be explained by ephaptic interaction between major motor fibers and the nociceptive fibers of the anterior interosseous nerve that provide sensory innervation of periosteum, as suggested by Granit, *et al.*,⁷ and more recently by Calvin and colleagues.³

In conclusion, our data stress the hypothesis that pulsatile arterial compression may act on peripheral nerves in a fashion similar to its action on cranial nerves.^{11,12,14} In fact, it has been reported recently that vascular compression in paroxysmal cranial nerve syndromes may also arise beyond the root entry zone.⁸ Although peripheral nerve neurovascular compression is a rare pathological

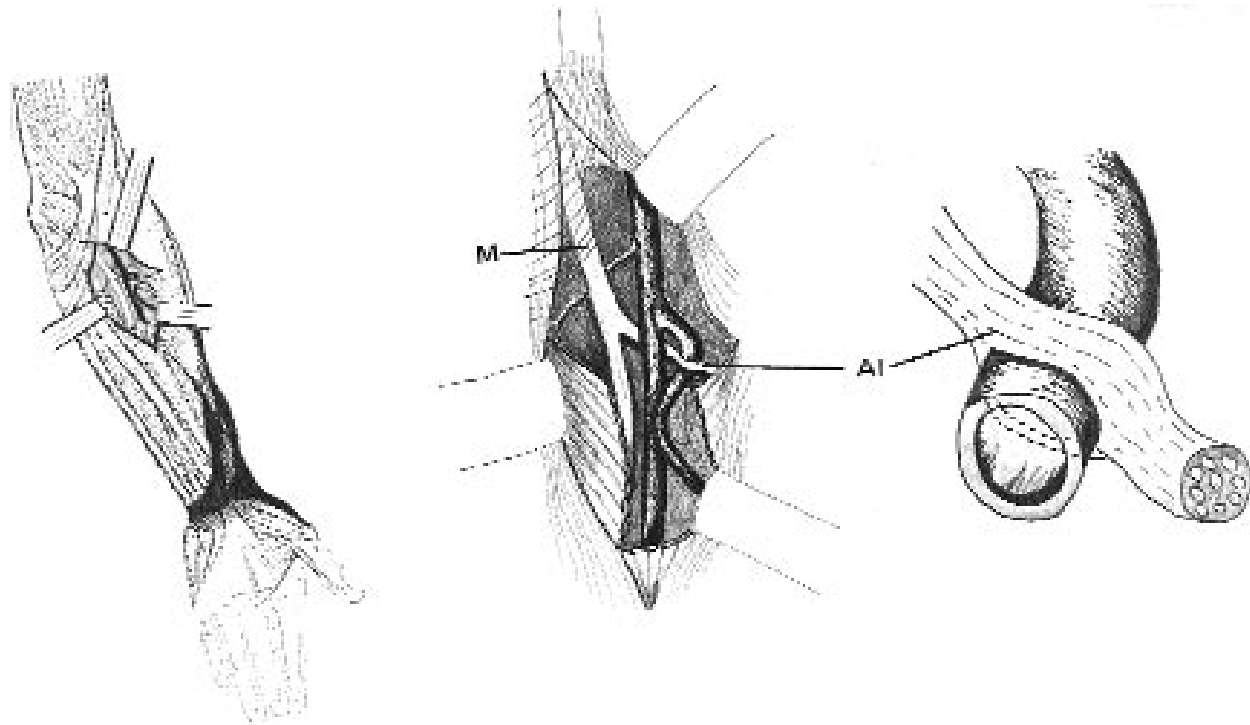


FIG. 2. Surgeon's drawing illustrating the operative field. *Left:* The surgical approach below the lacertus fibrosus in the antecubital fossa; the painful area is marked in black on the lateral forearm and wrist. *Center:* The median nerve (M) and the anterior interosseous nerve (AI) as exposed in the antecubital fossa. The radial artery and the anterior interosseous artery are represented by deep gray shading. *Right:* The neurovascular relationship between the anterior interosseous nerve and the anterior interosseous artery. The diameter of these vessels was larger than normal and a fibrous bundle from the perineurium was present, linking the nerve to the artery.

condition, we agree with the authors¹⁴ who included it among the causes of "irritative" entrapment neuropathies.

Acknowledgment

The authors thank Miss Phil Glynn for reviewing the manuscript.

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Manuscript received March 14, 1994.

Accepted in final form July 6, 1994.

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