Idiopathic Isolated Flexor Pollicis Longus Denervation (Incomplete Kiloh-Nevin Syndrome)—A Rare Presentation Managed by Tendon Transfer

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Abstract

This case report describes a rare presentation of isolated denervation of the Flexor Pollicis Longus (FPL), known as incomplete Kiloh-Nevin syndrome. The FPL, responsible for thumb flexion, is innervated by a motor branch of the anterior interosseous nerve (AIN), a branch of the median nerve. A 19-year-old female presented with sudden difficulty in holding a pen, and electromyographic findings revealed complete denervation of the FPL.

Tendon transfer from the flexor digitorum superficialis to the FPL was performed, followed by physiotherapy. Postoperatively, full active flexion was achieved at the thumb's distal interphalangeal joint. The discussion explores AIN dysfunction causes, diagnostic considerations, and treatment options, emphasizing the importance of surgical intervention when paralysis persists. This case highlights the significance of recognizing and addressing uncommon neuromuscular conditions for effective management.

Keywords

- Kiloh-Nevin syndrome
- flexor pollicis longus (FPL)
- anterior interosseous nerve
- distal interphalangeal flexion
- median nerve

Introduction

Flexor pollicis longus (FPL) is a muscle in the volar compartment of the forearm which is responsible for the flexion of the distal phalanx of the thumb. The innervation of FPL is provided by an isolated motor branch of the anterior interosseous nerve (AIN), a branch of the median nerve. AIN carries median nerve supply to the FPL, the flexor digitorum profundus, and pronator quadratus. Solitary paralysis of the FPL is very uncommon in clinical practice. A circular OK sign with the thumb and the second finger is used to assess the integrity of AIN and its motor branches. If any dysfunction is present, the person will not be able to flex the distal phalanx of the involved fingers (Kiloh-Nevin syndrome).[1,2]

Case Report

A 19-year-old right-handed female student presented with sudden onset of difficulty in holding the pen on the right hand while writing. There was no history of trauma. No history of loss of higher mental status. No associated neck pain, numbness, or any sensory symptoms. There was no history of other weakness. On examining the patient, there was loss of active flexion at the distal interphalangeal joint of the thumb (►**Fig. 1A, 1B**). Forearm pronation and distal interphalangeal flexion of other digits were normal. There were no distal sensory deficits.

Electromyographic findings revealed complete isolated denervation of the FPL. This represents a lesion involving the

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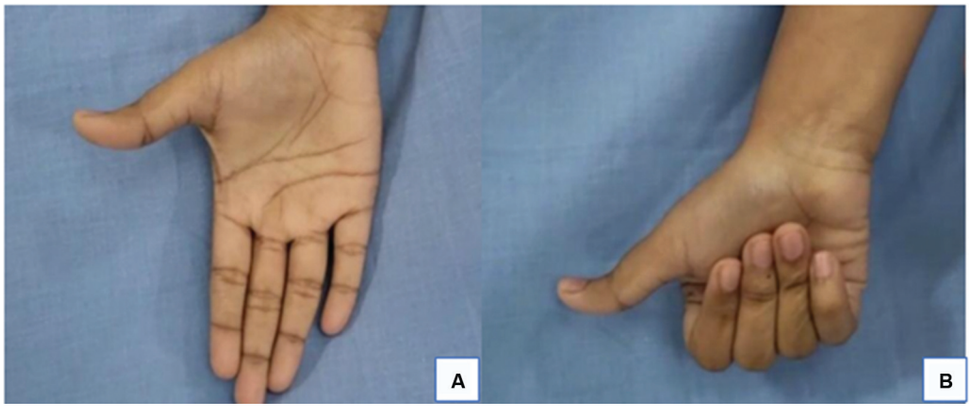

Fig. 1 (**A**, **B**) Preoperative images showing absence of flexion at the distal inter phalangeal joint of thumb.

branch or branches of the AIN to the FPL. Magnetic resonance imaging showed hyperintensity in the FPL and pronator quadratus muscle belly. No features of muscle wasting or fatty infiltration. All these features were suggestive of incomplete AIN syndrome.

The patient underwent tendon transfer. Flexor digitorum superficialis (FDS) of ring finger was transferred to the FPL of the thumb. Postoperatively, physiotherapy was done, and the patient was followed up for 3 months. On follow-up, there was full active flexion at the distal interphalangeal joint of the thumb (►**Figs. 2A, 2B** and **2C**).

Discussion

We reported an uncommon case of isolated denervation of FPL or incomplete Kiloh-Nevin syndrome. AIN of the median nerve originates a short distance below the elbow and runs distally, accompanied by the anterior interosseous artery. It lies between flexor digitorum profundus and FPL and passes beneath pronator quadratus, its terminal twigs supplying the wrist joint. No sensory supply for AIN. AIN compression can be caused due to several causes—tendon anomalies, as in the case of the deep head of the pronator teres or the origin of the head for the superficial flexor of the third finger; the presence of accessory muscles as in the case of the Gantzer muscle, an accessory head of the FPL that may originate from either the FDS muscle, coronoid process of the ulna, or medial epicondyle of the humerus; vascular problems, such as anomalous radial artery or vascular alterations of the ulnar collaterals' branches; and acute direct trauma with consequent neurotmesis.[3]

AIN dysfunction causes symptoms of exclusive motor involvement, concerning the FPL muscle and the FPD in its component for the second and third finger; the involvement of the pronator quadrate muscle is expected but it remains mostly asymptomatic. Only a few cases are reported in which unilateral FPL palsy occurs for acute lesion involving the branch or branches of the AIN to the FPL,[4] after complex forearm fractures[5] or during surgical procedures at the elbow or at the wrist.[6] There was no history of trauma or surgical procedures in our case.

In view of the sudden onset of the symptoms, it would appear to be a case of an interstitial neuritis. It is probable that examples of neuritis of the nerve occur more commonly than is generally recognized, for the resulting disability is not severe, accommodation occurring quickly in most patients, while recovery follows in many.[7]

Diagnosis of incomplete Kiloh-Nevin syndrome has been based upon history, physical findings, and sometimes operative findings. Treatment options are exploration of FPL tendon and muscle, exploration of AIN, resection of fibrous band, or tendon transfer. When paralysis of the muscles

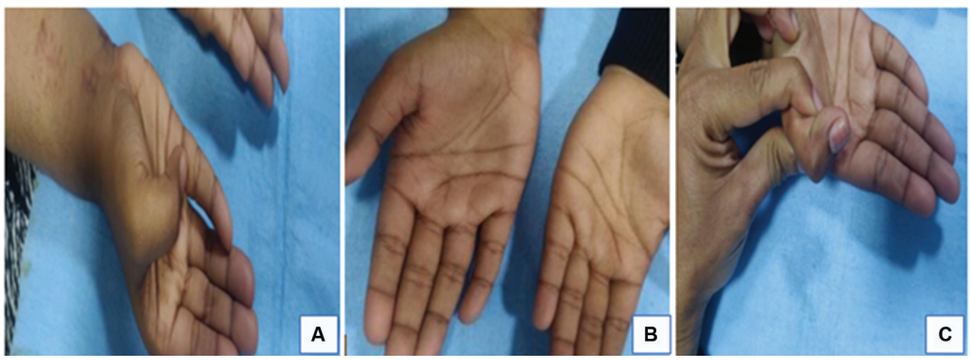

Fig. 2 (**A**, **B**, **C**) Post operatives image showing active flexion at the distal inter phalangeal joint of thumb.

supplied by the AIN is suspected, observation for a period of 6 to 8 weeks for possible improvement of the condition cannot be contraindicated.[8] When opting for surgical intervention, compression of AIN should be sought, at or distal to the branching off from the median nerve.

Declaration of Informed Consent

Written informed consent was obtained from all subjects before the study. The author(s) hereby declare that there is no information (names, initials, hospital identification numbers or photographs) in the submitted manuscript that can be used to identify patients.

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Conflict of Interest

None declared.

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