SAGE Open Medical Case Reports
Volume 6: 1–3

DOI: 10.1177/2050313X18777416
journals.sagepub.com/home/sco
SAGE

Bilateral anterior interosseous nerve syndrome with 6-year interval

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Abstract

Flexor pollicis longus paralysis related to idiopathic anterior interosseous nerve syndrome is well known, but few reports exist on bilateral disease. A 24-year-old man with no personal or family history of neurological disease developed isolated total loss of active flexion of the right thumb's interphalangeal joint after undergoing a wrist arthroscopy. Surgical exploration 5 weeks after onset showed flexor pollicis longus tendon to be intact; anterior interosseous nerve decompression was done with no abnormalities found. Because of persistent paralysis, electromyography was performed showing findings consistent with anterior interosseous nerve syndrome. After 7 months without recovery, the patient underwent tendon transfer. After 6 years, the patient presented with left-sided isolated flexor pollicis longus paralysis and electromyography indicated anterior interosseous nerve syndrome. Examination 9 months after onset showed persistent complete flexor pollicis longus paralysis but by 15 months spontaneous complete recovery had occurred. Anterior interosseous nerve syndrome can occur bilaterally and is likely to resolve completely without intervention but recovery may take longer than a year.

Keywords

Flexor pollicis longus, median nerve, anterior interosseous nerve, FPL, paralysis

Date received: 22 October 2017; accepted: 23 April 2018

Introduction

The anterior interosseous nerve (AIN) is a mainly motor nerve that arises from the median nerve in the proximal forearm.[1–2] The AIN syndrome is an uncommon condition that is characterized by weakness or paralysis of one or more of the AIN-innervated muscles, most commonly the flexor pollicis longus (FPL). It may also involve the flexor digitorum profundus (FDP) of the index finger, the pronator quadratus (PQ), and occasionally the FDP of the middle finger. Although the etiology of the AIN syndrome is still unclear, it is now believed that the underlying pathology is likely to be neuritis rather than compression.[3]

The clinical features of the AIN syndrome include forearm pain and weakness or paralysis involving flexion of the thumb's interphalangeal (IP) joint, and in some cases, flexion of the index and middle finger distal IP joints. Electromyography (EMG) and nerve conduction studies (NCS) are important in establishing the diagnosis.[1] According to current literature, nonoperative treatment is generally recommended as initial treatment for the AIN syndrome, with surgical exploration of the AIN advocated when recovery does not occur.[4] Some authors have proposed surgical exploration if no recovery had occurred 3 months after onset,[4] whereas others have advocated waiting up to 12 months.[5] There is no evidence as to what extent surgery influences the recovery and the course of the disease.

Although the literature contains several case reports and small series of AIN syndrome, only few cases of bilateral disease have been reported.[6–8] We report the case of a young adult with bilateral non-simultaneous idiopathic AIN syndrome.

Case report

A 24-year-old man was referred by a primary-care physician to our orthopedic department because of a 4-year history of non-traumatic ulnar-sided pain in the right wrist. The patient

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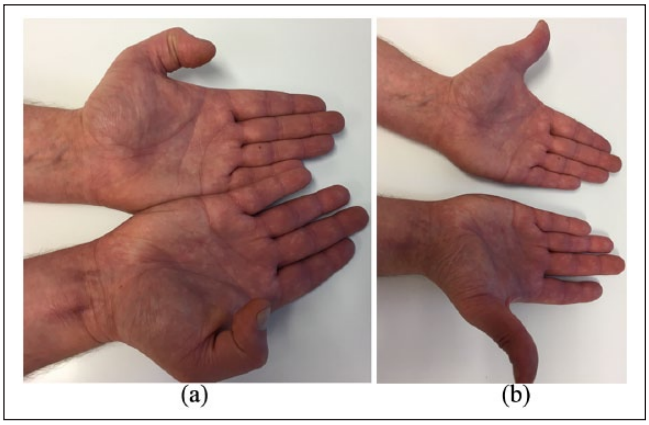

Figure 1. Patient with bilateral non-simultaneous anterior interosseous nerve syndrome that manifested as complete flexor pollicis longus paralysis; active flexion (a) and extension (b) of the interphalangeal joint after tendon transfer (right thumb) and spontaneous recovery without intervention (left thumb).

was a smoker and worked as a carpenter. He had no other medical conditions and did not use medications. He had no family history of any neurological disease. Physical examination revealed normal findings with no signs of carpal instability or distal radioulnar joint disorders. Tests of sensation and muscle strength were normal. A plain X-ray (antero-posterior and lateral views) of the wrist was normal. Magnetic resonance imaging (MRI) of the right wrist showed edema in the triangular fibrocartilage complex (TFCC). A wrist arthroscopy was done during which a TFCC tear was repaired. An above-elbow cast was applied. One week after the surgery the patient felt pain, described as a burning sensation, in the proximal forearm and noticed that he could not actively flex his right thumb's IP joint. The pain disappeared after a day. Physical examination of the hand showed total loss of the thumb's active IP flexion but no other abnormalities.

MRI did not show any abnormalities involving the FPL. EMG done 2 weeks after the onset of paralysis showed delayed insertion activity in the FPL suggesting possible AIN lesion. A repeat EMG, 2 weeks later, could not confirm AIN lesion, but the examiners reported difficulties in locating the FPL due to the arm cast. At 5 weeks after onset, no improvement had occurred and the treating surgeon decided to proceed to surgery on suspicion of tendon rupture. During surgical exploration, the FPL tendon was found to be intact; exploration and decompression of the AIN was then performed with no abnormalities found.

A repeat EMG, 6 months after onset, showed denervation activity in the FPL indicating severe axonal loss to the FPL with no reinnervation, grade-2 fibrillation potentials (graded 0–4), and absent motor unit potentials. At 6 months after AIN decompression, no improvement in the thumb's IP flexion had occurred. A tendon transfer using the flexor digitorum superficialis (FDS) of the ring finger was performed. Physical examination 14 months after onset showed strong active flexion in the right thumb's IP joint and no other AIN-related symptoms. However, the patient complained of recurrent swelling and pain in the palm and ring finger and subsequently underwent surgery with excision of the remaining part of the FDS, synovectomy and FDP tenolysis.

Contralateral AIN syndrome

Six years after the right-sided episode, the patient presented with symptoms in his left arm. He reported that he had felt pain in the left arm followed by loss of active flexion in the left thumb's IP joint. Physical examination of the patient's left hand showed total paralysis of the FPL but normal strength of the index finger and middle finger FDP and thenar muscles. EMG showed denervation activity in the FPL and PQ with no voluntary activity, findings that were interpreted as AIN lesion; all other muscles were normal and NCS of the median and ulnar nerves were normal. The pain gradually subsided. At 9 months after onset, the patient was examined by a hand surgeon at another hospital. The surgeon recorded presence of total FPL paralysis and scheduled the patient for AIN decompression. However, 15 months after onset of paralysis, the patient regained active IP flexion. On examination immediately before the planned surgery, the surgeon found good FPL strength and canceled the surgery. The patient has full strong active flexion and extension in the IP joints of both thumbs (Figure 1). He was able to return to work as a carpenter but works currently as a truck driver for employment-related non-medical reasons.

Discussion

We report the case of a young adult with non-traumatic bilateral AIN syndrome presenting with isolated total FPL paralysis with an interval of 6 years between the two episodes. In the side that was not subjected to surgery, total paralysis was still present 9 months after onset, but complete recovery occurred by 15 months. This is longer than the proposed waiting time before proceeding to surgery, commonly advocated in the literature.

Very few cases of non-simultaneous bilateral idiopathic AIN syndrome with no other neurological features have been reported. Recently, Chung et al.[6] reported a similar case of a 42-year-old man with bilateral AIN syndrome with about 4 years interval between the episodes, but the patient was treated surgically within 6 months of onset on both sides. Sood and Burke[7] reviewed 16 cases of AIN syndrome, of which one case was bilateral with an interval of 3 years between the two episodes; the authors reported that both hands were affected in the "postnatal period," but provided no case-specific information about treatment or outcome. Barbour et al.[8] reported a case of a 59 year old man with bilateral AIN syndrome with an interval of 13 years between the episodes, but they presented the case as possibly related to Parsonage–Turner syndrome.

Another condition that might have similar presentation is hereditary neuropathy with liability to pressure palsies (HNPP), but the patient in our case report did not have any history or symptoms and signs suggestive of HNPP. Although there was neither history nor clinical or neurophysiological findings suggestive of any other neurological disorder, it might not be possible to entirely exclude that the isolated AIN neuropathy was a manifestation of a rare neurological disease. Although EMG is the main diagnostic test in AIN syndrome, there is also a potential role of ultrasound and magnetic resonance (MR) neurography in the diagnosis and differential diagnosis. Ultrasound could have been helpful in our case as tendon exploration may have been avoided.

Recent research on AIN syndrome has suggested that abnormalities can be seen in the median nerve trunk more proximal to the AIN.[3,9] Based on current literature, we do not believe there is evidence to support that AIN decompression has any role in the treatment of AIN syndrome. Recovery after surgical decompression reported in previous studies[4,10] could have been spontaneous and unrelated to the surgery. It can be argued that tendon transfer surgery provides more rapid restoration of thumb function. However, surgery is not always successful and may be associated with complications, as was seen following the tendon transfer done on the first-affected arm in this case. Patients can regain full FPL function without surgery.

Conclusion

AIN syndrome can occur bilaterally and is likely to resolve completely without intervention, but recovery may take longer than a year. Thus, a conservative approach in the management of AIN syndrome might be considered.

Acknowledgements

The authors thank Associate Professor Gert Andersson, Department of Neurophysiology at Lund University for his help with the manuscript.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

The Regional Ethical Review Board waived the need for ethics approval.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Informed consent

Written informed consent was obtained from the patient for publication of the case report and the accompanying image.

References

1. Rodner CM, Tinsley BA and O'Malley MP. Pronator syndrome and anterior interosseous nerve syndrome. *J Am Acad Orthop Surg* 2013; 21: 268–275.
2. Dang AC and Rodner CM. Unusual compression neuropathies of the forearm, part II: median nerve. *J Hand Surg Am* 2009; 34: 1915–1920.
3. Pham M, Baumer P, Meinck HM, et al. Anterior interosseous nerve syndrome: fascicular motor lesions of median nerve trunk. *Neurology* 2014; 82: 598–606.
4. Park IJ, Roh YT, Jeong C, et al. Spontaneous anterior interosseous nerve syndrome: clinical analysis of eleven surgical cases. *J Plast Surg Hand Surg* 2013; 47: 519–523.
5. Seki M, Nakamura H and Kono H. Neurolysis is not required for young patients with a spontaneous palsy of the anterior interosseous nerve: retrospective analysis of cases managed non-operatively. *J Bone Joint Surg Br* 2006; 88: 1606–1609.
6. Chung DW, Han CS, Lee JH, et al. Bilateral anterior interosseous nerve syndrome: a case report. *J Korean Soc Surg Hand* 2015; 20: 180–185.
7. Sood MK and Burke FD. Anterior interosseous nerve palsy: a review of 16 cases. *J Hand Surg Br* 1997; 22: 64–68.
8. Barbour JR, Gontre G and Mackinnon SE. Delayed bilateral anterior interosseous neuritis: an atypical presentation. *J Hand Surg Eur* 2013; 38: 206–207.
9. Ochi K and Horiuchi Y. Fascicular constrictions in spontaneous anterior interosseous nerve palsy and spontaneous posterior interosseous nerve palsy. *Neurosurgery* 2014; 75: E737–E738.
10. Ulrich D, Piatkowski A and Pallua N. Anterior interosseous nerve syndrome: retrospective analysis of 14 patients. *Arch Orthop Trauma Surg* 2011; 131: 1561–1565.