Clinical Case Report

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# Bilateral idiopathic neuralgic amyotrophy involving selective branches of peripheral nerves with a stepwise progression

## A case report

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**Abstract**

**Rationale:** This is a report about a rare case of idiopathic neuralgic amyotrophy (INA) involving selective peripheral nerve branches of bilateral upper extremities, which exhibited a stepwise progression.

**Patient concern:** A 66-year-old woman presented with paresis of selective branches of bilateral median nerves, followed by paresis of bilateral posterior interosseous nerve (PIN) 8 weeks later.

**Diagnoses:** We diagnosed it as INA involving the selective motor branches of bilateral median nerves and bilateral PINs. Forearm magnetic resonance imaging combined with electrodiagnostic testing helped accurately identify the affected regions, and ultrasonography demonstrated a severe constriction of the left PIN.

**Interventions:** Intravenous methylprednisolone partially relieved the pain and paralysis. Surgical neurolysis of the constricted left PIN was done for persistent paralysis.

**Outcomes:** The muscle power of the bilateral median nerve territories was recovered to nearly normal, but the muscle power of the left PIN territories remained at grade 1.

**Lessons:** This case indicates that INA can manifest as a multiple mononeuropathy involving individual fascicular levels of peripheral nerve branches with focal constriction, and electrodiagnostic study combined with forearm MRI and ultrasonography can help in identifying affected lesion and predicting the prognosis.

**Abbreviations:** AIN = anterior interosseous nerve, APL = abductor pollicis longus, ECR = extensor carpi radialis, EIP = extensor indicis proprius, EMG = electromyography, FCR = flexor carpi radialis, FDP = flexor digitorum profundus, FDS = flexor digitorum sublimis, FPL = flexor pollicis longus, INA = idiopathic neuralgic amyotrophy, MRC = Medical Research Council, MRI = magnetic resonance imaging, NCS = nerve conduction study, PIN = posterior interosseous nerve, PQ = pronator quadratus.

**Keywords:** anterior interosseous nerve, case report, magnetic resonance imaging, neuralgic amyotrophy, posterior interosseous nerve, ultrasonography

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## 1. Introduction

Idiopathic neuralgic amyotrophy (INA) is a clinically defined syndrome with the acute onset of a painful neuropathy, predominantly involving the upper extremities.[1] INA is thought to be related to immune-mediated disorders of some peripheral nervous system, and the extent and distribution of affected nerves is quite variable.[2] Either diagnosis or treatment of INA has not yet been proved "gold standard."[2] The purpose of this case report is to illustrate a rare finding of bilateral distal INA exhibiting a stepwise progression, and to discuss diagnostic and therapeutic approach to the disease. The patient has provided written informed consent for the publication of the case with anonymity, and the ethical approval of this study was exempt by the ethics committee of Incheon St. Mary's Hospital because this study was a single case report (Ethical approval number: OC17ZSI0119).

## 2. Case report

A 66-year-old woman presented with weakness of flexion of both thumbs and the second and third fingers at the interphalangeal joints for 7 weeks. She had experienced severe burning pain in both shoulders 1 week before the paresis developed. Pain and

motor weakness had started on the left but had shortly progressed to the right. Her medical history was unremarkable except for diabetes mellitus.

On examination, muscle strength was assessed as Medical Research Council (MRC) grade 3 in the right flexor pollicis longus (FPL) and flexor digitorum profundus (FDP) muscles of the second and third fingers, and MRC grade 2 in the left FPL, FDP, and flexor digitorum sublimis (FDS) muscles of the second and third fingers. Both shoulder presented normal ranges of motion and normal scapular movements. She complained of dull ache on both forearms from the antecubital fossa to 1st to 3rd fingers but there was no discrepancy on sensory tests. Magnetic resonance imaging (MRI) of the cervical spine which was taken in the other hospital showed no significant abnormality.

Sensory nerve conduction study (NCS) was normal in the bilateral median, ulnar, and radial nerves. Motor NCS showed decreased amplitudes of compound motor action potentials (CMAPs) in both median nerves recorded on the pronator quadratus (PQ) muscles (Table 1). Needle electromyography (EMG) revealed abnormal spontaneous activity in the left pronator teres, flexor carpi radialis (FCR), second FDS and FDP, as well as in the right FPL and pronator quadratus (PQ) muscles (Tables 2 and 3). These findings indicated neuropathies of the left median nerve and the right anterior interosseous nerve (AIN). On ultrasonography, the bilateral median nerves were traced from the wrist to the axillar region. There was no definite swelling or constriction of the bilateral median nerves, but the bilateral FPL, FDP, and PQ muscles showed hyperechogenicities. The patient was diagnosed with INA on the basis of the characteristic clinical features and was treated with oral prednisolone (20 mg/d).

A week later, the patient reported a slight improvement of right thumb IP flexion, but noted weakness of the left thumb extension, with a new finding of grade 1 muscle power on examination. T2-weighted contrast-enhanced magnetic resonance imaging (MRI) of both forearms was done at 8 weeks from the first symptom, and demonstrated high signal intensities correlating with denervation injury and edema within the left FDS, FDP, FCR, FPL, PQ, extensor indicis proprius (EIP), and APL as well as within the right FDP, FPL, PQ, supinator, and extensor carpi radialis (ECR) muscles (Fig. 1A–F, Table 3). The second NCS showed decreased amplitudes of CMAP in the left radial nerve recorded on the EIP muscle (Table 1). Follow-up needle EMG revealed denervation potentials in the left EIP and APL muscles, suggesting neuropathy of the left posterior interosseous nerve (PIN) (Tables 2 and 3). Although there was no definite motor weakness, right supinator and ECR brevis muscles were sampled considering MRI findings and showed denervation potentials, suggesting neuropathy of the partial branches of the right PIN. Also, additional positive sharp waves were found in left FPL muscle at the more proximal site than where the muscle was sampled in the former study (Table 3). The serologic tests including autoantibodies and viral markers showed no significant findings.

Intravenous methylprednisolone (1 g/d) was administered for 3 days, which was switched to oral prednisolone (60 mg/d), tapered over 9 days. The patient did not report significant adverse effect including gastrointestinal problems during the steroid pulse therapy. Physical therapy including electrical stimulation on the left PIN was continued. During the treatment, muscle power in the right and left FPL and second FDP muscles improved from MRC grade 3/2 to 4/3, and the left second FDS muscles from MRC grade 2 to 3. However, muscle power in the left APL did not improve.

Six months from symptom onset, the muscle power in the second FDS and FDP and FPL recovered to nearly normal, but the muscle power of the left APL remained at MRC grade 1. On ultrasonography, an incomplete fascicular constriction of the left PIN within the supinator muscle was detected (Fig. 1G, Supplementary video, http://links.lww.com/MD/C965). She underwent surgical neurolysis of the left PIN. Intraoperatively, the nerve was seen to be constricted and edematous at the entering

**Table 1**

**Results of nerve conduction studies in the upper extremities.**

| Nerve/site | Initial study (June 21, 2017) | | | | Follow-up study (July 6, 2017) | | | |
|---|---|---|---|---|---|---|---|---|
| | Latency (ms) | | Amplitude† | | Latency (ms) | | Amplitude† | |
| | Right | Left | Right | Left | Right | Left | Right | Left |
| Sensory nerve | | | | | | | | |
| Median (wrist) | 2.81 | 2.81 | 37.4 | 47.9 | 2.85 | 2.85 | 40.0 | 48.7 |
| Ulnar (wrist) | 3.13 | 3.13 | 16.6 | 33.3 | 3.10 | 3.15 | 38.2 | 39.2 |
| Radial (forearm) | 2.03 | 1.82 | 63.4 | 55.3 | 2.25 | 2.10 | 55.4 | 47.2 |
| Motor nerve | | | | | | | | |
| Median-APB (wrist) | 2.71 | 2.66 | 14.2 | 13.4 | 2.7 | 2.85 | 9.0 | 11.6 |
| Median-APB (elbow) | 5.83 | 5.83 | 14.8 | 14.6 | 5.75 | 5.85 | 9.0 | 11.5 |
| Median-PQ (wrist) | 3.54 | 3.39 | **1.0*** | **1.4*** | 3.4 | 3.6 | **1.3*** | **1.0*** |
| Ulnar-ADM (wrist) | 2.5 | 2.86 | 12.0 | 9.3 | 2.4 | 2.75 | 10.4 | 8.5 |
| Ulnar-ADM (below elbow) | 5.73 | 5.42 | 11.5 | 8.5 | 5.85 | 5.7 | 9.8 | 7.1 |
| Radial-EIP (forearm) | | | | | 1.9 | 2.3 | 5.0 | **0.7*** |
| Radial-EIP (elbow) | | Not tested | | | 3.5 | **7.0*** | 4.7 | **0.6*** |
| Radial-EIP (upper arm) | | | | | 5.35 | **9.1*** | 4.3 | **0.6*** |
| F-wave | | | | | | | | |
| Median-APB | 24.01 | 23.49 | | | | | | |
| Ulnar-ADM | 24.95 | 24.84 | | | | | | |

ADM = abductor digitorum minimi, Amp = amplitude, APB = abductor pollicis brevis, CV = conduction velocity, EIP = extensor indicis proprius, Lat = latency (motor, onset latency; sensory, peak latency), PQ = pronator quadratus.

* Indicates abnormal data based on our reference values.

† Amplitudes are measured in millivolt (mV, motor) and in microvolt (μV, sensory).

The bold values mean significant findings in electrodiagnostic studies and MRI studies, which prove neuropathies.

**Table 2**

**Results of needle electromyography in the upper extremities.**

| | Initial study (June 21, 2017) | | | | | | | Follow-up study (July 6, 2017) | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Spontaneous activity | | Motor unit action potential | | | | | Spontaneous activity | | Motor unit action potential | | | | |
| | Fib | PSW | Amp | Dur | Poly | Recruitment pattern | Interferential pattern | Fib | PSW | Amp | Dur | Poly | Recruitment pattern | Interferential pattern |
| B. Cervical paraspinalis | N | N | | | | | | N | N | | | | | |
| B. Deltoid | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| B. Biceps brachii | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| R. Pronator teres | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| L. Pronator teres | **1+** | **1+** | N | N | N | N | N | N | N | N | N | N | N | N |
| R. Flexor carpi radialis | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| L. Flexor carpi radialis | **4+** | **4+** | N | N | N | **Reduced** | **Reduced** | N | N | N | N | N | **Reduced** | **Reduced** |
| R. Flexor digit profundus (I-II) | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| L. Flexor digit profundus (I-II) | **4+** | **4+** | | | | **No Activity** | | | | | | Not tested | | |
| R. Flexor pollicis longus | **4+** | **4+** | N | N | N | **Reduced** | **Discrete** | **1+** | **1+** | N | N | N | **Reduced** | **Discrete** |
| L. Flexor pollicis longus | N | N | N | N | N | N | **Reduced** | **3+** | **3+** | N | N | N | **Reduced** | **Reduced** |
| R. Pronator quadratus | **4+** | **4+** | | | | **No Activity** | | **2+** | **2+** | | | | **No Activity** | |
| L. Pronator quadratus | **4+** | **4+** | | | | **No Activity** | | **4+** | **4+** | N | N | N | **Reduced** | **Discrete** |
| B. Abductor pollicis brevis | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| B. Triceps brachii | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| B. Brachioradialis | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| B. Extensor digitorum communis | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| R. Extensor indicis proprius | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| L. Extensor indicis proprius | N | N | N | N | N | N | N | **1+** | **1+** | N | N | N | **Reduced** | **Discrete** |
| L. Abductor pollicis longus | | | | | Not tested | | | **3+** | **3+** | | | | **No Activity** | |
| B. Flexor carpi ulnaris | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| R. Flexor digit profundus (III-V) | N | N | N | N | N | N | N | N | N | N | N | N | N | N |
| B. First dorsal interosseous | N | N | N | N | N | N | N | N | N | N | N | N | N | N |

Amp = amplitude, B = both, Dur = duration, Fib = fibrillation, Lt = left, N = normal, N = normal, Poly = polyphasic pattern, PSW = positive sharp wave, Rt = right.
The bold characters mean positive findings of denervation potentials in sampled muscles, which confirms neuropathies involving the distribution of each peripheral nerves.

**Table 3**

**Results of electromyography and magnetic resonance imaging of the upper extremities in a case of idiopathic neuralgic amyotrophy.**

| | Right | | | | | | Left | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Median nerve territory | | | Radial nerve territory | | | Median nerve territory | | | Radial nerve territory | | |
| | + | − | NT | + | − | NT | + | − | NT | + | − | NT |
| First EMG<br>June 21, 2017 | **FPL, PQ** | APB, FCR, FDP, PT | PL | | EDC | ANC, BR, ECR, EDM, EIP, EPL, SP, TB | **FCR, FDP, FDS, PQ, PT** | APB, FPL | PL | | EDC | ANC, BR, ECR, EDM, EIP, EPL, SP, TB |
| Contrast-enhanced MRI (T2)<br>July 5, 2017 | **FDP, FPL, PQ** | APB, FCR, FDS, PL, PT | | **ECR, SP** | ANC, APL, BR, EDC, EDM, EIP, EPL, TB | None | **FCR, FDP, FDS, FPL, PL, PQ, PT** | APB | | **APL, EIP** | ANC, BR, ECR, EDC, EDM, EPL, SP, TB | |
| Second EMG<br>July 6, 2017 | **FPL, PQ** | | APB, FCR, FDP, FDS, FPL, PL, PT | **ECR, SP** | EDC, EIP | ANC, APL, BR, EDM, EIP, EPL, TB | **FCR, FPL, PQ, PT** | APB | PL | **APL, EIP** | BR, TB | ANC, BR, ECR, EDM, EPL, SP |

+ = positive for denervation, − = negative for denervation, ANC = anconeus, APB = abductor pollcis brevis, APL = abductor pollicis longus, BR = brachioradialis, ECR = extensor carpi radialis, EDC = extensor digitorum communis, EDM = extensor digiti minimi, EIP = extensor indicis proprius, EMG = electromyography, EPL = extensor pollicis longus, FCR = flexor carpi radialis, FDP = flexor digitorum profundus, FDS = flexor digitorum sublimis, FPL = flexor pollicis longus, MRI = magnetic resonance imaging, NT = not tested, PL = palmaris longus, PQ = pronator quadratus, PT = pronator teres, SP = supinator, TB = Triceps brachii.
The bold characters mean positive findings of needle electromyography and MRI imaging, in combination.

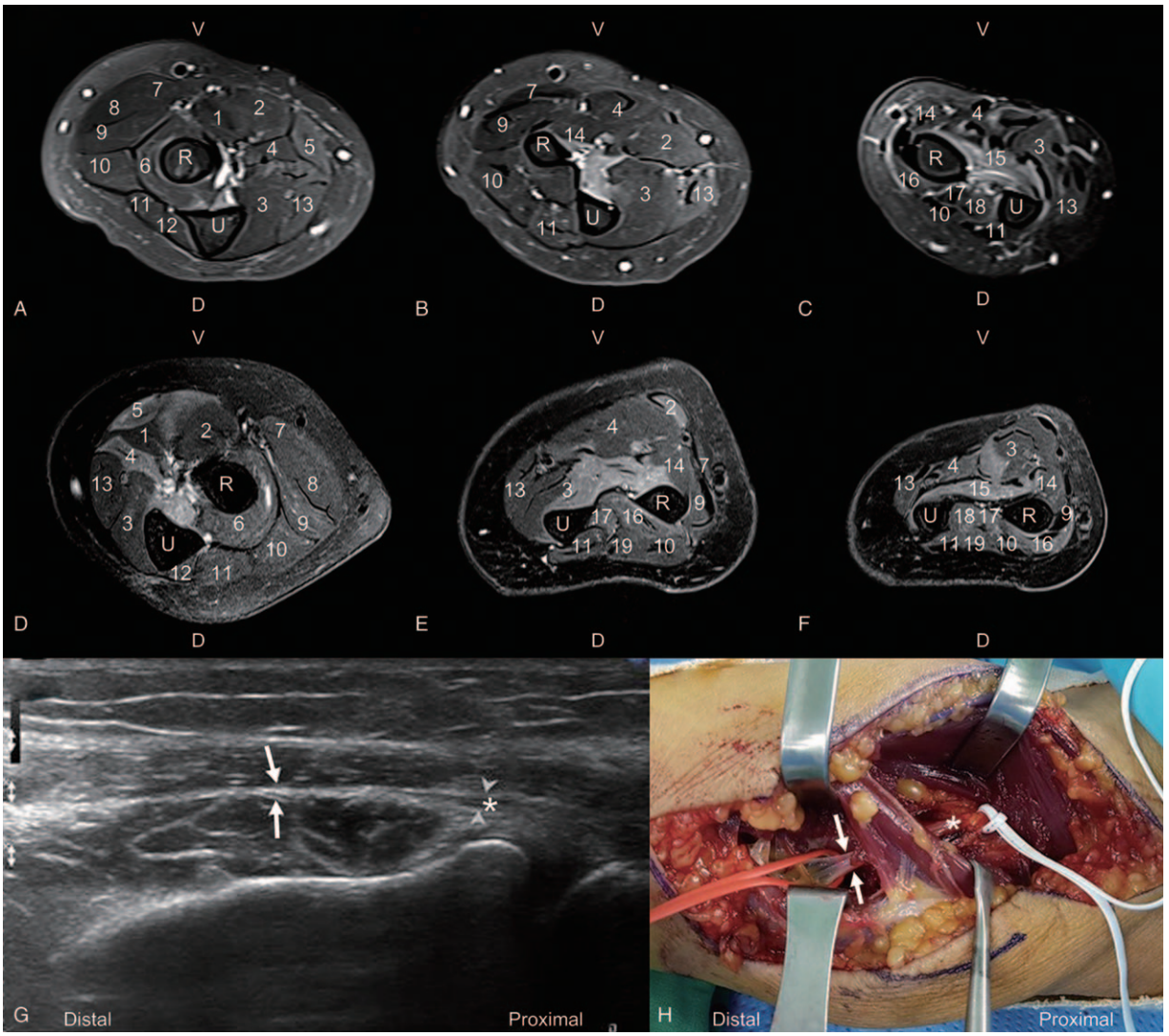


**Figure 1.** T2-weighted contrast-enhanced MRI of both forearms (A–F), preoperative ultrasonographic findings of the left posterior interosseous nerve (G), and intraoperative findings of the left posterior interosseous nerve (H). 1; Pronator teres (PT), 2; Flexor carpi radialis (FCR), 3; Flexor digitorum profundus (FDP), 4; Flexor digitorum sublimis (FDS), 5; Palmaris longus (PL), 6; Supinator (SP), 7; Brachioradialis (BR), 8; Extensor carpi radialis longus (ECRl), 9; Extensor carpi radialis brevis (ECRb), 10; Extensor digitorum communis (EDC), 11; Extensor carpi ulnaris (ECU), 12; Anconeus (ANC), 13; Flexor carpi ulnaris (FCU), 14; Flexor pollicis longus (FPL), 15; Pronator quadratus (PQ), 16; Abductor pollicis longus (APL), 17; Extensor indicis proprius (EIP), 18; Extensor pollicis longus (EPL), 19; Extensor digiti minimi (EDM), V; Volar, D; Dorsal, R; Radius, U; Ulna. A–C, Right forearm; (D–F), left forearm. G, Preoperative ultrasonography of the left posterior interosseous nerve (PIN; arrowheads), Showing the constriction of the left PIN within the supinator muscle (arrows) and swelling of the nerve (*) just proximal to the muscle. H, Intraoperative findings of the left PIN, with constriction (arrows) within the muscle and swelling (*) proximal to the muscle.

point to the supinator muscle, consistent with the ultrasonography findings (Fig. 1H). At 3 months after the surgery, muscle power in the left thumb extension did not improve (Table 4).

## 3. Discussion

The presented case described progressive bilateral INA involving the right AIN, left median, and both PIN, which was confirmed by electrodiagnostic studies and imaging studies. Administration of intravenous methylprednisolone and oral prednisolone had a favorable effect on AIN and median nerve palsy, but left PIN palsy persisted despite a surgical treatment.

The distribution of abnormalities in INA can vary from an isolated nerve to the widespread involvement of the brachial plexus.[3] Isolated cases of median or radial involvement in INA have been reported, but the combination of these 2 is relatively rare.[1,4–7] Bilateral INA occurs in approximately 30% of patients,[7] and some case reports of bilateral AIN involvement exist.[8] To the best of our knowledge, this is the first report of bilateral median and radial involvement in INA.

The patient experienced the right finger flexion weakness a week after the initial event of severe pain. And then, after 8 weeks the left finger extension weakness developed. Note that the motor weakness after initial pain can develop from a day to 2 weeks or later,[7] delayed or stepwise progression of the PIN palsy could be possible. Nevertheless, one should keep in mind that recurrence of idiopathic NA is not so uncommon than expected.[7] The 8-week interval between the left median and PIN palsies suggests that peripheral nerve involvement in INA can progress in a delayed or stepwise manner.

**Table 4**

**Timeline table.**

| Dates | Relevant past medical history and interventions | | |
|---|---|---|---|
| | Diabetes mellitus<br>No specific family history | | |
| Date | Summaries from initial and follow-up visits | Diagnostic testing (including dates) | Interventions |
| June 21, 2017 | Weakness of flexion of both thumbs and the second and third fingers at the interphalangeal joints, following severe burning pain in both shoulders. | Magnetic resonance imaging (MRI) of the cervical spine which was taken in the other hospital showed no significant abnormality.<br>Electrodiagnostic study revealed bilateral idiopathic neuralgic amyotrophy. | Oral prednisolone (20 mg/d) |
| July 6, 2017 | A slight improvement of right thumb IP flexion, but noted weakness of the left thumb extension, with a new finding of grade 1 muscle power on examination. | T2-weighted contrast-enhanced magnetic resonance imaging (MRI) of both forearms demonstrated high signal intensities correlating with denervation injury and edema within the left FDS, FDP, FCR, FPL, PQ, extensor indicis proprius (EIP), and APL as well as within the right FDP, FPL, PQ, supinator, and extensor carpi radialis (ECR) muscles.<br>Follow-up needle EMG revealed denervation potentials in the left EIP and APL muscles, suggesting neuropathy of the left posterior interosseous nerve (PIN). | Intravenous methylprednisolone (1 g/d) was administered for 3 d, which was switched to oral prednisolone (60 mg/d), tapered over 9 d |
| October 6, 2017 | The muscle power in the second FDS and FDP and FPL recovered to nearly normal, but the muscle power of the left APL remained at MRC grade 1. | Ultrasonography revealed an incomplete fascicular constriction of the left PIN within the supinator muscle | Surgical neurolysis of the left PIN |
| January 2018 | Three months after the surgery, muscle power in the left thumb extension did not improve. | | |

Although a thorough and extensive electrodiagnostic examination is required for determining the lesion distribution and the time course of INA, there are limitations in choosing target muscles with respect to technique, time, and patients' compliance. Furthermore, sampling error in needle EMG may be common.[3] Recent studies have shown that imaging studies of the brachial plexus and peripheral nerves with either MRI or ultrasonography may show abnormal findings of the affected muscles and nerves in INA.[9–15] In the present case, the T2-weighted forearm MRI revealed high signal intensities of the clinically affected muscles as well as the subclinical involvement of the right supinator and ECR. Correlation with the forearm MRI findings improved the accuracy of the second EMG in that denervation potentials were found in the left FPL more proximally than on the first examination. Although brachial plexus MRI is frequently used for investigating NA, previous studies reported a low percentage of significant findings in plexus MRI.[7,16] As the involvement of the extraplexal nerves rather than the brachial plexus proper has been described in INA,[5,11,16] high-resolution MRI of the upper extremities may help identify muscles that are affected by the selective fascicular level of the peripheral nerves distal to the plexus. Ultrasonography, although highly dependent on the skills of the examiner, is a good technique for investigating individual peripheral nerves involved in NA, especially when looking for nerve swellings or constrictions preoperatively.[11,17,18] In this case, a constriction and swelling of the left PIN was identified, which is a condition thought to be caused by inflammation and to be indicative of poor prognosis.[11,17,18]

INA treatment is empirical according to published case series. Some articles have reported favorable outcomes of oral or intravenous steroid treatment for INA.[19–21] Considering the rarity of the disorder, no randomized controlled trials assessing the efficacy of steroid therapy have been conducted; thus, further studies are warranted to establish a guideline for steroid use in INA. Surgery may be considered in cases of prolonged paresis despite steroid treatment. However, no definite consensus exists regarding either the indications for or the optimal timing of surgery. Some authors recommend surgery of the AIN or PIN in cases without any recovery at 3 months after onset.[4,18,22] Ochi et al[23] have recommended the neurolysis of the PIN in patients under 50 years of age if performed within 7 months of onset. Wu et al[18] have suggested that older patients are more likely to present with a worse operative outcome. They have argued that neurorrhapy or autografting is more effective than neurolysis in cases of severe hourglass-like fascicular constriction.[18] The patient demonstrated no recovery of the left PIN at least 3 months after neurolysis despite relatively early intervention. This poor outcome may be because of her age or simply because the nerve did not adequately recover. There has been emerging views that AIN and PIN palsy is not an entrapment neuropathy at the usual compression site, but a consequence of the selective motor fascicular neuropathy of more proximal nerve trunk.[12,14] The left PIN palsy presented in this case may have involved both pathologies, although more proximal lesions in the upper arm level could not be identified on ultrasonography. High-resolution ultrasonography can detect fascicular lesions of the distal nerve,[24] but advanced neuroimaging such as magnetic resonance neurography may be considered to provide additional diagnostic information about far proximal nerve lesions.

## 4. Conclusion

The clinical features and diagnostic studies presented in this case support the contention that INA can manifest as a multiple mononeuropathy associated with inflammation and constriction

affecting multiple peripheral nerves. The presented case indicates that INA can progress in a stepwise manner. Forearm MRI and ultrasonography may help identify the specific lesions, improving the accuracy of EMG and perhaps indicating whether surgery should be undertaken. Our patient seemed to benefit from intravenous methylprednisolone, but the constriction of the left PIN contributed to a poor outcome despite surgical neurolysis.

## Author contributions

**Conceptualization:** Min-Wook Kim, Dae-Hyun Jang.
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**Writing – original draft:** Mee-Gang Kim.
**Writing – review & editing:** Mee-Gang Kim, Dae-Hyun Jang.

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