INTRODUCTION

The Kiloh-Nevin Syndrome or Anterior interosseous nerve (AIN) syndrome is a proximal mononeuropathy affecting the AIN at its emergence from the median nerve at the proximal forearm [1]. This syndrome is due to the compression and damage of the AIN. In 1952, Kiloh (Australian physician) and Nevin (English neurologist) published two cases with similar clinical characteristics and attributed them to a mononeuritis of idiopathic etiology [2]. Four years before, Parsonage and Turner had published 6 cases of paralysis of the thumb finger and the index finger that they had attributed to an anterior spinal cord injury in the context of an abscess of the spinal cord. The AIN is a pure motor nerve that arises from the median nerve 6–8 cm below the level of the elbow’s lateral epicondyle [3], in the proximal forearm between the two heads of the pronator teres muscle to run deep along the interosseous membrane where it innervates mostly the deep muscles of the forearm anterior compartment as known, the flexor pollicis longus (FPL), the flexor digitorum profundus (FDP) and the pronator quadratus (PQ), as shown in the figure 1 below. AIN syndrome is an isolated palsy of these three muscles showed at the (Figure 1) above. The etiology of AIN syndrome is still poorly understood. It is one of the less frequent compressive pathologies of the upper extremity. Its clinical diagnosis is difficult since, as AIN is a pure motor nerve, it lacks of sensitive signs in an area where could be easily identified. It can occur secondary to a primary AIN entrapment, direct trauma, or in more ambiguous or vague clinical presentations. Controversy and different explanations have been proposed as the etiology; nevertheless, the condition is considered a transient neuritis in most cases. Among the traumatic causes are supracondylar fractures, penetrating injuries, stab wounds, cast fixation, venipuncture, and surgical reduction of fractures. Other causative factors are brachial plexus neuritis, compartment syndrome, and compression neuropathy.
ies (ENG/EMG testing) must be included, as they make an important role in the workup for AIN syndrome and support the diagnosis. In this context it should be known that imaging studies are not typically helpful for an accurate assessment and subsequent diagnose. Radiographic findings are usually normal, MRI may show edema in the AIN innervated muscles, and rarely compression or inflammation signs along the nerve pathway.

The medical management is conservative in the majority of cases and thus, surgical management should be considered with an evident diagnosed compressive (space-occupying) lesion or traumatic lesion or in those patients who fail to recover approximately 3 months of nonsurgical treatment. The general consensus includes a period of rest and splinting of the elbow. The majority of patients may experience improvement between 6 and 12 weeks [5,6], although the prognosis will depend on the degree of the damage. In cases with partial affectionation of presumably neuritic etiology, compression and subsequent axonotmesis, functional recovery occurs in an average of 6 to 12 months. In traumatic perforating cases due to injections of catheterisms, needles or stabbing weapons, the prognosis, according to the literature, is worse. There are no adequate studies that have compared the clinical benefit between conservative and surgical management so there is no consensus on finding the right timing to perform surgical treatment. Several reports [7,8] have described spontaneous recovery in most patients with AIN syndrome. Therefore, in general, due to the high probability of spontaneous recovery at least one year after the onset of symptoms, a prolonged observation period (together with conservative treatment of the symptoms) should be considered in the management of this condition.

2 CASE REPORT

A 33-year-old male patient, with no previous relevant medical history of diseases was admitted to the Emergency ward of the University Hospital of Fuenlabrada, Madrid (Spain), showing

Figure 1: Image of Anterior Interosseous Nerve in the forearm [4].

Figure 2: A correct “Ok” / “O” sign (left side) versus “Ok” / “O” sign with Anterior Interosseous Nerve injury (right side).
a clinical picture of sudden characteristically neuropathic pain in the right scapular region reflecting to the right arm and cubital region of forearm until the wrist and 5th finger. He also described a significant loss of distal strength when performing manual activities such as housekeeping tasks. The previous month, the patient had been admitted at the Intensive Care Unit at the Severo-Ochoa Medical hospital in Madrid, due to right-side hemotorax. Further anamnesis referred to previous spontaneous pneumothorax in 2002 and 2003, smoker and patient operated from disc hernia surgery some time ago. This previous medical information gave rise to initially rule out a pneumothorax relapse as the first medical approach. A chest Rx showed no evidence of pneumothorax. The physical examination showed no further information into the patient’s history reasonably consistent with the previous pneumothorax. Furthermore, a cranial CT scan was performed showing no apparent evidence of intracranial damage. At that time the patient was hospitalized at the neurology ward and received medical treatment with analgesia and antiplatelet drugs.

The physical examination was described as “vague forearm neuropathic pain which increases with the flexion-extension movement of the elbow, weakness and distal motor deficit when making the clamp gesture with the 1st and 2nd fingers of the right hand, and 1st and 3rd fingers so the patient was unable to make an “OK” or “O” sign with his thumb and index finger of his right hand. Myotactic reflexes and global sensitivity were preserved”.

It was decided to rule out a possible discopathy/acute radiculopathy, as well as other possible pathologies affecting the central nervous system (CNS) thus; a list of laboratory tests was requested (full battery of: CBC, biochemistry, serologies, metabolism tests, PCR tests, autoantibody/autoimmunity and ELISA tests). A CT scan of cervical cord was performed. After the result, it was concluded that there was an annular posterior cranial protrusion of C4-C5 discs, imprinting the anterior spinal surgery, a cranial CT scan was performed showing no evidence of traumatic phenomena as the patient was under metamizole as painkiller and still unable to make the “OK” sign with his right hand. The full blood test showed no alterations of any parameter. The next day, the pain in the right wrist and elbow continued while taking painkillers at the hospital stay. A deep physical examination showed that apparently, no traumatic etiology was related as there was no swelling, erythema or external inflammatory signs. There was pain on deep palpation when performing active flexion-extension of the elbow in the mediolateral bregmatic side, and limitation in active flexion of the tip of the 1st and 2nd right fingers with difficulty in performing the clamp. Patient’s arm showed no musculare atrophies. After ruling out all possible pathologies at the central CNS level (i.e. including as well as discopathy/radiculopathy), a mononeuropathy was under consideration, therefore an ENG/EMG and a brachial plexus & right superior limb MRI were requested with the aim of completing the diagnostic study as far as possible. The right superior limb ENG/EMG study showed active denervation of Pronator quadratus (PC), flexor digitorum profundus (FDP) and flexor pollicis longus (FPL); concluding in an anterior interosseous nerve (AIN) lesion. The brachial plexus MRI showed no evidence of measurable injuries. The right superior limb MRI showed a hyperintensity signal in muscles FPL and FDP. There were no alterations in the path of the anterior interosseous nerve. These findings were also suggestive of acute denervation of the muscles innervated by the AIN, without muscle atrophy that would indicate chronicity. After having ruled out other severe and chronic conditions, the AIN syndrome was diagnosed and revealed as the cause of this patient’s consultation.

Subsequently as a conservative therapeutic measure, a rehabilitation therapy which consisted of 24 occupational therapy sessions was carried out. A slow gradual complete functional improvement of the affected limb took place over the period of one year and a half. The subsequent control EMG/ENG tests conducted over 2 to 4 months after the first diagnostic EMG/ENG study showed a progressive reinnervation pattern.

3 | DISCUSSION

The Kiloh-Nevin Syndrome or Anterior interosseous nerve (AIN) syndrome is a rare entity of a pure distal dynamic motor mononeuropathy [9]. This case report points out the importance of a careful and exhaustive systematical differential diagnosis and diagnostic algorithm for these patients. Within this case report, the main task of performing a comprehensive diagnostic algorithm as followed is to ensure that there is no other underlying medical condition which could lead to another under-diagnosed pathology. On the other hand, as for the clinical signs in this case report, the patient was asked to make a circle with the thumb and index finger of the hand and he was unable to make it. The complete paralysis of the long thumb flexor and the deep flexor of the 2nd finger prevents this simple maneuver because the patient cannot actively flex neither the thumb’s interphalangeal nor the distal index’s interphalangeal, describing a kind of flattened “OK”/”O” sign, as shown in the right side of the (Figure 2). Since the very characteristic clinical sign which leads to the inability to make a complete “OK”/”O” sign is almost a pathognomonic sign of an AIN injury (as classically referred by the literature) the absence of local injuries, such as traumatic, neurological, tumor nodules or other lesions, makes it decisive to initiate the present diagnostic medical algorithm as followed for this clinical case. Furthermore, a concrete etiology of the AIN injury was still unknown for this patient. Nonetheless, considering the recent hospital admission to the patient in the ICU ward for a right hemоторax, it was suggested that a damage at some point on the right-hand AIN nerve pathway could have been produced within the context of peripheral arterial/venous catheters and/or drainage placements used during the hospital stay. In this case the therapeutic management was at all-time conservative with analgesia and rehabilitation therapy consisted of 24 sessions of occupational therapy (OT). Overall, a slow, gradual but complete functional improvement of the affected limb was observed over a wide period of one year and a half. The subsequent control EMG/ENG tests, performed in the following months, showed a progressive reinnervation pattern of the AIN, demonstrating that the distal motor paresia was transient over time.

4 | REFERENCES