

Kiloh-Nevin syndrome : a compression neuropathy or brachial plexus neuritis ?

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Kiloh-Nevin syndrome was first described in 1948 by Parsonage and Turner and further defined in 1952 by Kiloh and Nevin. The aetiology is highly debated. Two common causes of Kiloh-Nevin syndrome are compression neuropathy and brachial plexus neuritis.

In this study the results in six patients who were treated operatively and were evaluated after a mean follow-up of 42 months (range: 24 to 60), are presented.

Retrospectively, two subgroups could be identified based on initial clinical presentation and EMG : one subgroup of patients presented with a compression neuropathy and the other subgroup presented with brachial plexus neuritis. An important difference in outcome was seen between the two subgroups. There were excellent results after surgical decompression in the patients with a compression neuropathy, whereas the results after surgical decompression in the subgroup presenting with brachial plexus neuritis were less predictable.

The data presented here and the data found in the literature, suggest that if the clinical image and the EMG suggest a compression neuropathy, surgical decompression, after two months of conservative treatment, will give excellent results. If the clinical image and EMG suggest brachial plexus neuritis, a conservative treatment may be more appropriate.

Keywords : Kiloh-Nevin syndrome ; compression neuropathy ; brachial plexus neuritis ; surgical decompression.

INTRODUCTION

In 1948 Parsonage and Turner (12) described 'neuritis' of the anterior interosseous branch of the median nerve. In 1952 Kiloh and Nevin (9) further defined the anterior interosseous nerve syndrome. The clinical presentation includes inability to flex the thumb at the interphalangeal joint and inability to flex the index at the distal interphalangeal joint, caused by a complete palsy of the flexor pollicis longus and the flexor digitorum profundus of the index. Patients are unable to form an O with the thumb and index finger (O sign or Spinner's sign). There is also a palsy of the pronator quadratus which can be tested with the elbow in a flexed position to neutralise the humeral head of the pronator teres muscle.

Since these descriptions of the clinical entity, there has been much debate about the aetiology and treatment.

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Posttraumatic haematoma (3), traction and posterior contusion of the median nerve combined with transient ischemia after supracondylar humeral fractures (7), arm bandages (1) and direct trauma during elbow arthroscopy (14) have all been described as possible causes of a palsy of the anterior interosseous branch of the median nerve.

The aetiology of the so-called 'idiopathic' anterior interosseous nerve palsy is more debated. Different aetiologies and different subsequent recommendations for treatment are highly specialtyoriented.

The results achieved in six patients treated operatively, and a literature review regarding aetiology and treatment are presented.

MATERIAL AND METHODS

Patients

Six patients, five male and one female presenting with Kiloh-Nevin syndrome were treated operatively. In all cases a preoperative EMG was performed. Four patients filled out a preoperative DASH score. The mean age of the patients at the time of operation was 41.3 years (range : 25 to 57). The time interval between the onset of symptoms and operation was 2 to 6 months. All patients were invited for evaluation after a mean follow-up of 42 months (range : 24 to 60). Five patients agreed to participate. One patient replied he was satisfied with the treatment and did not wish to participate further.

Surgery

The operation was performed under tourniquet with the limb exsanguinated. A zigzag volar incision on the proximal forearm was used. The laceratus fibrosus was identified and divided. The median nerve and the anterior interosseous branch were explored and decompressed from proximal to distal. The arcuate ligament, a fibrous insertion of the pronator teres and fibrous bands in the flexor digitorum sublimis were released. In two cases a Gantzer's muscle was found and released.

RESULTS

Five patients were re-evaluated, one patient replied he was satisfied but denied further evaluation. The mean preoperative DASH score was 57.2; the mean postoperative DASH score was 37.8.

Acta Orthopædica Belgica, Vol. 73 - 3 - 2007

Two patients had excellent results. One of them filled in a DASH score preoperatively, which was 40.4. This patient had complete recovery ; his post-operative DASH score was 25. Key pinch strength was 13 kg on the affected, dominant side compared to 12 kg on the unaffected side. The other patient also had full functional recovery, his postoperative DASH score was 24.3.

One patient had a good result. Postoperatively, there was a normal function with minimal loss in strength. Postoperatively, the key pinch of the affected, dominant side was 11 kg, compared to 12 kg on the non-dominant, unaffected side. The DASH score was 83.3 preoperatively and 31.6 postoperatively.

One patient had a moderate result. He regained the ability to flex the tip of the thumb and the index finger (O sign), but he did not regain full strength. Key pinch of the affected, dominant side was 5 kg, compared to 13 kg on the non-dominant, unaffected side. He also had some persistent paraesthesia in the forearm. His DASH score was 74.2 preoperatively and became 52.5 postoperatively.

One patient had only minimal improvement and was unable to flex the tip of the index finger after 50 months. His postoperative DASH score was 55.8.

DISCUSSION

One of the common causes of the anterior interosseous nerve syndrome is a compression neuropathy. Structures that can compress the anterior interosseous nerve are a tendinous origin of the deep head of the pronator teres or of the flexor digitorum sublimis, Gantzer's muscle, thrombosis of the ulnar collateral vessels, an aberrant radial artery, accessory muscles and tendons from the flexor digitorum sublimis, a tendinous origin of the palmaris longus or of the flexor carpi radialis and an enlarged bicipital bursa (18). Initial treatment should be conservative for at least 8 weeks. If no improvement occurs, surgical decompression should be performed. The results of this treatment as reported in the literature are very good (2, 4, 6, 13, 15-17, 19-21).

Hourglass-like fascicular constriction of the fascicle of the median nerve destined to become the

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anterior interosseous nerve more distally, without signs of external compression, is also described as a cause of anterior interosseous nerve syndrome. In these cases interfascicular neurolysis leads to good results (11).

Another common cause of the anterior interosseous syndrome is brachial plexus neuritis or neuralgic amyotrophy. Conservative treatment with spontaneous recovery occurring up to 2-3 years following onset of the symptoms has been described. Overall results after conservative treatment are good (5, 8-10, 12, 15, 16).

All the patients in this study were treated with surgical nerve decompression after an initial conservative treatment of at least two months, treating the anterior interosseous syndrome as a compression neuropathy. Retrospectively, two subgroups could be identified based on the initial clinical presentation and the EMG. In the first group of three patients, EMG showed complete denervation of the anterior interosseous branch of the median nerve without proximal abnormalities. These patients also had no other clinical signs than forearm pain combined with the inability to flex the interphalangeal joint of the thumb and the distal interphalangeal joint of the index. These findings confirm an isolated compression neuropathy.

A second group of three patients had a history of shoulder and upper arm pain, followed by the inability to flex the thumb interphalangeal joint and the index distal interphalangeal joint (O sign). Besides complete denervation of the anterior interosseous nerve, diffuse proximal abnormalities were found on EMG, suggesting brachial plexus neuritis. In two of the three patients the clinical image was even more complex. One of these patients had an anterior interosseous syndrome combined with a palsy of the long thoracic nerve, causing scapular winging. The other patient also showed altered sensibility in the thumb. This altered sensibility in the thumb does not exclude a compression site, because an arcuate ligament can compress both the median nerve and the anterior interosseous branch (6), nor does it exclude brachial plexus neuritis.

Interestingly, the preoperative DASH score divides the patients, who filled in a preoperative

DASH score, into the same subgroups. In both groups two out of three patients filled in a DASH score preoperatively. In the first group with compression neuropathy, the preoperative DASH scores were 31.6 and 40.4. In the second group with brachial plexus neuritis, DASH scores were 83.3 and 74.2. This difference in preoperative DASH scores also confirms the difference in clinical presentation and in dysfunction caused by two different aetiologies.

An important difference in outcome between the two groups was seen. In the group with compression neuropathy, two patients had excellent results after surgical nerve decompression with recovery of mobility and strength. One patient replied he was satisfied but denied further evaluation. Although the preoperative DASH score of these patients was better than in the other group, the postoperative DASH scores still improved to 24.3 and 25.

The results in the group with brachial plexus neuritis were mixed. One patient had a good result with recovery of strength and mobility. His DASH score improved from 83.3 to 31.6. Another patient had a moderate result. He regained mobility, but recovery of strength was only partial. He also had some persistent paresthesia in the forearm. His DASH score improved from 74.2 to 52.5. The third patient had only minimal improvement. Even after a follow-up period of 50 months, he was still unable to flex the tip of the index finger and he had a postoperative DASH score of 55.8.

This study is retrospective, the number of patients is very limited and there was no control group. All patients were treated operatively after an initial conservative treatment of two to six months. It is impossible to tell if the patients with a compression neuropathy would have had the same outcome after conservative treatment. It is also impossible to tell if the patients with brachial plexus neuritis would have done better or worse without operative treatment.

Combining these data with the literature, two types of anterior interosseous syndrome could be identified. The first is a compression neuropathy which can be treated operatively with very good results, as found here and confirmed in the literature review (2, 4, 6, 13, 15-17, 19-21).

The second type of anterior interosseous nerve syndrome is a presentation of brachial plexus neuritis, giving a more complex clinical image. According to the data presented here and the data found in the literature (*5*, *8-10*, *12*, *15*, *16*), the results of surgical nerve decompression in these patients are less predictable.

In conclusion, we would suggest surgical nerve decompression, after an initial conservative treatment of two months, in patients with the anterior interosseous nerve syndrome, if the clinical image and EMG suggest a compression neuropathy. If the clinical image and the EMG suggest brachial plexus neuritis, conservative therapy may be more appropriate.

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