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Case report

Neuralgic amyotrophy and COVID-19 infection: 2 cases of spinal accessory nerve palsy

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A R T I C L E   I N F O

Article history:
Accepted 12 April 2021
Available online 24 April 2021

Keywords:
COVID-19
Neuralgic amyotrophy
Parsonage–Turner syndrome
Spinal accessory nerve
Trapezius muscle palsy
Peripheral neuropathy

A B S T R A C T

Objective: Neuralgic amyotrophy (NA), also known as Parsonage–Turner syndrome is often triggered by mechanical stress or viral infections. We reported 2 cases of shoulder weakness and amyotrophy related to spinal accessory nerve (SAN) palsy due to neuralgic amyotrophy occurring after COVID-19 infection.

Methods: For both patients, clinical history, clinical examination, electrodiagnostic (EDX), and imaging examinations invalidated other diagnoses but confirmed NA diagnosis.

Results: The NA involved only the SAN in both cases. EDX revealed a characteristic axonal lesion found in SAN. SAN conduction study revealed normal latencies and low compound motor action potential amplitude for trapezius muscle when needle examination demonstrated a neurogenic pattern and denervation signs in the trapezius muscle. Both patient’s MRI revealed denervation T2 hyper signal in impaired muscles, without any mass, cyst, injury, fibrous band, or tearing signs along SAN course.

Conclusions: The COVID-19 infection could be the trigger for NA as many other viruses, and as it is a possible trigger for Guillain–Barré syndrome.

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1. Introduction

The most frequent and serious symptoms due to COVID-19 infection are related to severe acute respiratory syndrome (SARS) [1]. Neurological disorders have also been described, which may involve the central and the peripheral nervous systems, from the most frequent and benign such as anosmia to the rare and severe Guillain–Barré syndrome (GBS) [1,2]. Neuralgic amyotrophy (NA) is defined as an acute and painful monophasic peripheral axonal neuropathy, with single or multiple nerve lesions that cause weakness, amyotrophy, and sensory loss in an asymmetric and patchy distribution, involving especially the upper limbs [3]. As GBS, it is presumed to have autoimmune and inflammatory pathophysiology. It is usually triggered by mechanical stress or viral infections [3,4]. Three cases of neuralgic amyotrophy (NA) related to COVID-19 respiratory infections have been reported [5–7]. One was purely sensitive [5], the second one involved supraspinatus, infraspinatus, teres minor, teres major, and trapezius muscles [6]; and the third one involved the median nerve [7]. Hereafter, we have reported two cases of NA involving spinal accessory nerve (SAN) following documented SARS related to COVID-19 infection.

2. Case reports

2.1. Case 1

A 63-year-old man presented with a SARS related to COVID-19 infection, documented by positive nasopharyngeal swab PCR and suggestive chest CT-scan. Coronavirus infection was treated with hydroxychloroquine, azithromycin, baricitinib, ceftriaxone, and dexamethasone. Due to worsening of respiratory distress, he required mechanical ventilation resuscitation for 6 weeks; first by orotracheal intubation, then by percutaneous tracheotomy. When discharged from the intensive care unit (ICU), the patient experienced a period of mental confusion and agitation [8,9]. Lumbar punction, electroencephalogram, and neuro-imagings were normal. So, it is only one month after ICU discharge that the patient...
first complained of pain and disability in his right shoulder. The first clinical evaluation found an active and passive limitation of shoulder range of motion, and muscular atrophy of upper trapezius muscle and supra-infra-spinous fossae [10,11]. Other muscles’ strength was normal especially the sternocleidomastoid (SCM) muscle. Adhesive capsulitis was diagnosed but this did not explain the importance of shoulder myopathy. Tendon reflexes were preserved. The patient also complained of paresthesia in the ulnar area of both forearms. Dynamic examination of the shoulder showed altered scapulothoracic rhythm: the scapula was mildly winged in the lateral position, and abruptly slid during the lateral elevation of the upper limb (Fig. 1) [11,12]. An electrodagnostic examination (EDX) was performed 5 months after the beginning of the COVID-19 infection and revealed an isolated important right spinal accessory nerve (SAN) axonal involvement: nerve conduction study found normal latencies with marked reduction of compound action potentials (CMAP) amplitude of right upper and lower trapezius muscles compared with the left side (Fig. 2). Needle examination found much-reduced interference patterns and denervation signs in upper and lower trapezius muscles. Other muscles (especially infraspinatus, deltoïd, and serratus anterior), and nerves (long thoracic, axillary, supraspinacular, median, and ulnar nerves) were normal. MRI of the cervical spine and brachial plexus performed seven months after infection onset showed no inflammatory modification of cranial nerve XI (normal signal on MR neurography and no gadolinium intake on T1 with DIXON fat subtraction). This was compatible with delay superior to 3 months. Nevertheless, myopathy and fatty infiltration on T1 sequence and high signal intensity on STIR sequence of right trapezius were noted, in favor of semi-recent denervation in the distal territory of cranial nerve XI. Other viral serologies (hepatitis B, C, and E, HIV, CMV, B19 virus) were negative. X-rays invalidated diaphragmatic paralysis and MRI of the cervical spine and right shoulder eliminated other neurological, bone, and joint pathologies.

2.2. Case 2

A 74-year-old man presented with SARS related to COVID-19 infection documented by positive nasopharyngeal swab PCR and suggestive chest CT-scan. He required mechanical ventilation resuscitation for 5 weeks, by oral-tracheal intubation, and was treated with cefotaxime, dexamethasone, and tocilizumab. He had an ICU-acquired transient tetraparesis that involved especially the lower limbs. One week after ICU discharge, the patient complained of pain and disability of his left shoulder, and clinical examination revealed muscular atrophy of upper trapezius muscle and supra-infra-spinous fossae [10,11]. Active and passive shoulder ranges of motion were limited in the three spatial planes. Muscle testing of the shoulder showed an important weakness and a mild winged scapula in a lateral position. One could see an abrupt sliding of the scapula laterally and downward during upper limb lateral elevation [11,12]. Tendon reflexes were preserved. EDX performed 4 months after the COVID-19 infection onset, revealed an isolated important left SAN axonal lesion: nerve conduction study found normal latencies with marked reduced of compound action potentials amplitude of left upper and lower trapezius muscles compared with the right side. Needle examination found much-reduced interference patterns and denervation signs in upper and lower trapezius muscles. Other muscles (especially infraspinatus, serratus anterior, and sternocleidomastoid muscles), and nerves (long thoracic, axillary, supraspinacular, median and ulnar nerves) were normal. MRI of brachial plexuses performed one month later showed muscular atrophy of left trapezius in T1 and high signal intensity on STIR sequence, in favor of semi-recent denervation in the distal territory of cranial nerve XI. MRI of the left shoulder found a distal deep partial tear of the infraspinatus tendon and enthesisopathy of the supraspinatus. This radiological assessment eliminated other bone, spinal and endo-canal differential diagnoses. It also eliminated injury of the spinal ganglia, post-ganglion roots, and plexus trunks. Diaphragmatic paralysis was ruled out thanks to X-rays. The serological assessment found no other recent viral infection (hepatitis B, C, and E, HIV, CMV, B19 virus).
3. Discussion

The SAN is the external terminal division of the eleventh cranial nerve; it crosses the cervical area to innervate the sternocleidomastoïd muscle (SCM) and the three bundles of the trapezius muscle [11,12]. When it is damaged, consequences are atrophy and weakness of the trapezius muscle. It causes limitation of the elevation of shoulder with lateral position of the scapula at rest, mild lateral winging, and abrupt sliding of the scapula during lateral elevation. This clinical pattern is typical of what was found in both reported cases. As usual, the muscular strength of SCM was preserved [10–12].

Three cases of neuralgic amyotrophy (NA) related to COVID-19 infections have already been reported [5–7]. One case was purely sensitive [5] and involves only the antebellum cutaneous nerve. Another case involved supraspinatus, infraspinatus, teres minor, teres major, and trapezius muscles. MRI abnormalities were typical of NA but there was no EDX data available [6]. The third one [7] was described as an astonishing mild lesion (for NA diagnosis) of the median nerve with a partial conduction block in the forearm, and no denervation signs with needle examination.

NA is frequently an elimination diagnosis [3]. Before evoking NA in our two patients, we have ruled out damage of the SAN, with clinical examination, EDX, and MRIs. We first eliminated cervical spine or brachial plexus trauma or stretching, due to patient mobilizations in prone position, while they were under curare. History found neither surgery of the cervical region, nor venous jugular access, which could have been the cause of a SAN injury. The patients carried homolateral subclavian catheters, distant from the path of the SAN. One of them also had a controlateral catheter. Both patients had other associated upper limb conditions that could not explain the pain and that have been confounding for a time. In case 1, the patient had adhesive capsulitis that explained the limitation of passive shoulder mobility. This association is described in 17% of NA cases [3]. The second patient had a rotator cuff impairment, which is a very common pathology according to his age group, and certainly participating in the shoulder painful symptomatology. But it did not explain the muscular atrophy of the supra and infra-spinal fossae as there was no tendon rupture. He also had an ICU neuro-myopathy that could explain initially a part of shoulder disability. Finally, the NA diagnoses of our two case reports were quite typical and well documented.

SAN palsy related to NA is underdiagnosed, especially when it is isolated [11]. SAN palsy was found in 20% of NA cases by van Allen and van Engelen [3]. Muscular atrophy of infra-supra-spinal fossae and winging scapula can clinically evoke suprascapular or long thoracic nerve palsy. This could explain why NA is frequently misdiagnosed. Serious damages of peripheral nervous system related to COVID-19 infection appears to be rare and consists mainly of Guillain–Barré syndrome or its variants [2]. Peripheral nervous system involvement varies from 8.9% to 0.05% [2,13]. NA is probably underdiagnosed because its disability remains usually mild and limited [5]. The fact that COVID-19 may be responsible for Guillain–Barré syndrome is an argument in favor of its possible responsibility in NA, as their pathophysiology seems to be very close, and as the same viruses can be the trigger for both pathologies [3,4,14].

However, there were two limitations to these case reports. The first one was the impossibility to determine the exact delay between COVID-19 infection onset and NA occurrence: the patients in ICU were unable to communicate with practitioners. Furthermore, upon discharge of ICU, patients had numerous disabilities [9], and they needed to have recovered sufficient autonomy, and a clear mind, to become aware of their shoulder disability. The second limitation is that NA is sometimes triggered by a surgical event or traumatism. So, intensive care could appear to be a sufficient trauma to trigger NA. But astonishingly a Medline query found no case reported in the literature.

Disclosure of interest

The authors declare that they have no competing interest.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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