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A CASE OF NEURALGIC AMYOTROPHY AFTER VACCINATION AGAINST COVID-19

UN CASO DE NEURALGIA AMIOTRÓFICA TRAS VACUNACIÓN FRENTE A COVID-19

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ABSTRACT

Introduction: neuralgic amyotrophy is an inflammatory disorder of the brachial plexus with a poorly understood pathophysiology, possibly related to immunological phenomena. It is characterized by acute and intense pain, accompanied by muscle weakness and altered results in electromyographic and nerve conduction studies. Having no specific treatment, it is approached through physiotherapy and symptomatic control.

Case history: we present the case of a 37-year-old woman, with no relevant history, who developed the condition after being vaccinated against SARS-CoV-2. The patient responded discreetly to physical therapy, conventional analgesia and corticosteroids. After 10 months of treatment, the decision was made to use pulsed radiofrequency

and ultrasound-guided plexus block of the plexus, which achieved better results. A year after the onset of pain, the patient's symptoms had improved, although she had not been able to return to her professional activity as a policewoman.

Discussion: very few cases of neuralgic amyotrophy have been reported after SARS-CoV-2 vaccination. The differential diagnosis includes many common muskuloskeletal and neurologic disorders, which can mislead professionals and delay identification of the disease, especially in the context of massive vaccination campaigns. Conventional analgesia is often insufficient to address these patients' complaints; thus, scheduled invasive techniques need to be considered. Given the rarity of the condition and its impact on the personal and professional life of the patient, we highlight the importance of an early diagnosis and smooth communication with the patient. Finally, we stress the value of declaring adverse reactions as a sign of professionalism and an asset in establishing a constructive doctor-patient relationship.

Conclusion: neuralgic amyotrophy is a disorder of the brachial plexus, possibly related to immunological phenomena such as SARS-CoV-2 vaccination. In patients with insufficient response to physical therapy and pharmacological treatment, interventions such as pulse radiofrequency or ultrasound-guided plexus blocks can be valuable. A swift diagnosis and a transparent doctor-patient relationship are highlighted as key assets in managing these cases.

Keywords: Neuralgic amyotrophy, Parsonage-Turner syndrome, adverse reaction, vaccination, SARS-CoV-2.

RESUMEN

Introducción: La neuralgia amiotrófica es un trastorno inflamatorio del plexo braquial con una fisiopatología poco conocida, posiblemente relacionada con fenómenos inmunológicos. Se caracteriza por dolor agudo e intenso, acompañado de debilidad muscular y alteración de los resultados en los estudios electromiográficos y de conducción nerviosa. Al no tener un tratamiento específico, se aborda a través de fisioterapia y el control sintomático.

Caso clínico: Presentamos el caso de una mujer de 37 años, sin antecedentes relevantes, que desarrolló el cuadro tras ser vacunada contra el SARS-CoV-2. La paciente respondió de forma discreta a la fisioterapia, analgesia convencional y corticosteroides. Después de 10 meses de tratamiento, se tomó la decisión de utilizar radiofrecuencia pulsada y bloqueo eco-guiado del plexo, lo que logró mejores resultados. Un año después de la aparición del dolor, los síntomas de la paciente habían mejorado, aunque no había podido reincorporarse a su actividad profesional previa como policía.

Discusión: Se han publicado muy pocos casos de neuralgia amiotrófica después de la vacunación contra el SARS-CoV-2. El diagnóstico diferencial incluye diversos trastornos musculoesqueléticos y neurológicos comunes, que pueden despistar al profesional y retrasar la identificación de esta entidad, especialmente en el contexto de campañas de vacunación masivas con gran volumen de reacciones adversas. La analgesia convencional a menudo es insuficiente para abordar los problemas de estos pacientes, por lo que se debe prever la necesidad de programar técnicas invasivas. Dada la rareza de la afección y su impacto en la vida personal y profesional del paciente, se destaca la importancia de un diagnóstico precoz y una comunicación fluida. Finalmente, se subraya el valor de la declaración de reacciones adversas como un signo de profesionalidad y un activo para establecer una relación médico-paciente constructiva.

Conclusión: La neuralgia amiotrófica es un trastorno del plexo braquial, posiblemente relacionada con fenómenos inmunológicos, como la vacunación contra el SARS-CoV-2. En casos como el presentado, en que la fisioterapia prolongada y el tratamiento farmacológico son insuficientes, pueden resultar valiosas intervenciones como la radiofrecuencia pulsada o el bloqueo eco-guiado del plexo braquial. Se señalan la rapidez del diagnóstico y la transparencia en la relación médico-paciente como elementos clave en el abordaje de estos casos.

Palabras clave: Neuralgia amiotrófica, síndrome de Parsonage-Turner, reacción adversa, vacunación, SARS-CoV-2.

INTRODUCTION

Neuralgic amyotrophy is an inflammatory disorder of the brachial plexus also known as paralytic brachial neuritis, idiopathic brachial plexopathy, and acute brachial radiculitis. Although Dreschfeld published the first case in 1886, it is often called Parsonage-Turner Syndrome, due to these authors describing it in 1948. It is a rare entity, with an incidence of around 1.64 cases per 100,000 inhabitants (1).

The pathophysiology of neuralgic amyotrophy is poorly understood, and more histopathological evidence is needed to identify its causes. The available literature suggests that it is a multifocal process that primarily, but not exclusively, affects secondary motor neurons (2). Among its etiopathogenic factors, events with immunological repercussion stand out, particularly a history of recent vaccination, infection, strenuous physical exercise, surgical interventions, gestation and puerperium (3,4).

Neuralgic amyotrophy is characterized by a sudden clinical onset, consisting of acute and intense pain followed by muscle weakness. It is usually distributed in a patched form throughout the nerve territories dependent on the superior and middle roots of the brachial plexus. The classical presentation usually includes a *scapula alata*, due to involvement of the long thoracic nerve and the consequent atrophy of the anterior serratus muscle. The pain, which is preponderant in the lateral area of the arm and rotator cuff, is severe and wakes the patient up at night, reaching its peak intensity in the first month. In most cases, sensory symptoms are associated, such as allodynia or paresthesia, which indicates that not only motor neurons are affected. Bilateral manifestations exist, but are rare (4).

Muscle weakness occurs shortly after the onset pain, usually during the first two weeks. It is especially apparent in the territories innervated by the suprascapular, axillary, long thoracic, musculocutaneous, radial and anterior interosseous nerves. In a minority of patients, there are manifestations occurring outside the brachial plexus. These typically include the lumbosacral plexus, the phrenic nerve, and the recurrent laryngeal (5). Diaphragmatic dysfunction due to phrenic neuropathy often goes unnoticed, but is present in up to 7 % of cases (6).

The diagnosis is established on clinical and electrophysiological grounds. The suspicion is based on the previously described pattern of sudden, severe pain, followed by

atrophic muscle weakness and a slow recovery. Conduction studies can help exclude other more common neuropathies; needle electromyography (EMG) is important for documenting denervation.

Analytical and imaging tests, on the other hand, allow for the exclusion of alternative causes of acute plexopathy, including neoplasms and some infections such as those caused by HIV or *Borrelia burgdorferi* (7,8). In most idiopathic cases, the image allows to rule out lesions with a mass effect, especially when the anamnesis is atypical or suggests malignancy. If neurophysiology leads to the suspicion of intraplexual injury, magnetic resonance imaging (MRI) is the most useful test; the studied region should include the cervical spine to rule out concomitant radiculopathy (9).

There is no specific treatment for neuralgic amyotrophy, so management is based on physical therapy and the control of symptoms. Some authors advocate the use of corticosteroids, although there are no studies to support their efficacy (10). Recovery occurs slowly and spontaneously, usually requiring 1 to 3 years until fully achieved, something which does not always happen (11).

CASE HISTORY

We present the case of a 37-year-old woman, working as a police officer at an Air Base, of athletic complexion, and with a history of smoking (20 packs-years) and polycystic ovary syndrome in treatment with oral contraceptives. There were no other elements of interest in her history and no known drug allergies. Her immunization status was appropriate for her age and sex, and included all the standard vaccines required for personnel belonging to the Spanish Armed Forces (12).

In the context of a Covid-19 vaccination, she received a first dose of AstraZeneca (VaxZevria), with mild initial pain at the point of punction. At approximately 7 days, the patient began to perceive cervical discomfort, for which she consulted in the emergency room. Pain was located around the left trapezius area and was accompanied by mild paresthesia in the shoulder and upper arm. Initially, a mechanical origin was attributed to cervicalgia, and treatment with dexketoprofen and methocarbamol was prescribed.

Given the persistence of the pain and the appearance of a subjective sensation of loss of strength in the hand, the patient consulted again two days later. At that time, cervical radiographies were obtained, which did not demonstrate pathological findings; an MRI and a neurophysiological study were also scheduled, orienting the diagnosis towards radiculopathy.

The MRI, performed 3 weeks after the onset of symptoms, revealed mild nonspecific protrusions in C4 and C5, without root involvement. Pending the neurophysiological study, medication was changed to etoricoxib and a combination of paracetamol and tramadol, as the previous analgesic regimen was judged insufficient.

The nerve conduction study carried out 33 days post-clinical debut showed, after transdermal stimulation of the axillary nerve at Erb's point, a prolonged distal latency measured at the middle deltoid. No alterations in amplitude were observed in comparison to the contralateral shoulder. A decrease in amplitude did appear in the lateral antebrachial cutaneous nerve. A needle EMG study was performed as well on the deltoid muscle, showing an undetected decrease in amplitude, and an unspecified reduction in motor unit recruitment. Motor unit potentials (MUP) measured at this level showed widespread and significant increase in polyphasia and duration of the potential. In the upper fascicle of the trapezius muscle (spinal nerve territory, C3-C4), the same alterations as those in the deltoid were observed, although of lower intensity. In summary, the neurophysiological study showed partial, patchy involvement of the upper trunk of the brachial plexus and C3-C4, with evolutionary neurogenic involvement and signs of reinnervation in progress, of subacute evolution, and without denervation. These findings, contextualized, led to the diagnosis of neuralgic amyotrophy.

Initially, analgesia was prescribed with pregabaline and paracetamol, with metamizole as a rescue drug. In subsequent visits, given the little improvement observed, prednisone was added to this regimen. A physical therapy plan consisting of active and passive mobility exercises, rotator cuff strengthening, and transcutaneous nerve stimulation was established. Throughout the first three months the patient developed concomitantly an anxiety-depression disorder derived from the sudden functional impairment, requiring psychotherapy and treatment with duloxetine.

A new neurophysiological study conducted 6 months after the onset of symptoms showed improvement in the conduction values of the lateral antebrachial cutaneous nerve. There was, however, newly found electromyographic evidence of neurogenic involvement of the biceps brachii; clinical involvement of the median nerve with paresthesia at the palmar-lateral aspect of the hand had also become apparent. Moreover, the findings from the previous study persisted, with an increase in the amplitude, duration and polyphasia of MUPs in the territories innervated by C3-C4 (trapezius muscle) and C5-C6 (deltoid, brachial and infraspinous muscles).

Over the following 10 months, 135 physical therapy sessions were carried out, achieving a slow and scarce improvement, with persistence of pain and the appearance of multiple myofascial trigger points with neuropathic irradiation. It was finally decided to refer the patient to a Pain Clinic with the aim of exploring other options.

At the Pain Clinic, pulse radiofrequency was chosen as the next step, and a first session was performed on the upper trunk of the brachial plexus, with an associated ultra-sound guided nerve block. A second radiofrequency sessions was performed, with remarkable amelioration. In addition to said techniques, cannabidiol oil was prescribed as an analgesic adjuvant. One year after the onset of the syndrome, and after all the interventions mentioned above, the patient presented remarkable improvement, but had not achieved complete remission and was still unfit for her previous professional activities.

DISCUSSION

To date, 9 cases of neuralgic amyotrophy secondary to a Covid-19 vaccine have been reported (1). The vaccine given to our patient was based on a chimpanzee adenovirus coding for the spike glycoprotein of SARS-CoV-2, manufactured in lines from genetically-modified human kidney embryonic cells (HEK 293). Its use has been linked to very rare adverse neurological reactions; in particular, several cases of Guillain-Barré Syndrome (GBS) and Transverse Myelitis (TM) have been reported (13).

The differential diagnosis of neuralgic amyotrophy includes both musculoskeletal pathologies (subacromial bursitis, fascioscapulohumeral dystrophy, adhesive capsulitis) and neurological (radiculopathies and compressive, hereditary or inflammatory neuropathies). The personal and professional history of the patient, with a long military career in tactical roles, initially led to a presumptively mechanical cervicalgia of traumatic or degenerative origin, a fact which underlines the importance of the neurophysiological study.

It is important to highlight the importance of adequate analgesia and good planning of the diagnostic process, given that the main complementary tests (MRI and EMG) are rarely available in emergency departments, requiring the scheduling of appointments and specialized assessments.

The case presented is illustrative because of its representativity. Knowing the disease and its functional prognosis allows the doctor to accompany the patient and inform him properly to make decisions regarding his personal and professional life. Individuals with physically demanding professions will usually require long absences from work, sometimes having to completely reorient their professional career. Adequate communication contributes to the patient's well-being and reduces the impact of problems typically associated with chronic pain, such as anxiety and depression.

As a final note, the importance of reporting adverse drug reactions to the relevant health authorities is highlighted. This is a key element of pharmacological vigilance and constitutes a sign of professionalism that contributes to strengthening the doctor-patient relationship, especially in the case of adverse events caused by medical actions.

CONCLUSION

Neuralgic amyotrophy is a disorder of the brachial plexus with a pathophysiology possibly related to immunological phenomena, including vaccination. Our patient was a 37-year-old woman who developed the entity after receiving the AstraZeneca vaccine for SARS-CoV-2, which has been related to other adverse reactions with neurological manifestations. The patient required prolonged physical therapy and

pharmacological treatment with conventional analgesia and corticosteroids, which could not fully control the symptoms. Invasive interventions such as pulse radio frequency and ultrasound-guided plexus blocks achieved relative success in our patient, and could be a valuable tool. A swift diagnosis and familiarity with the disorder's natural course can help with the management of these patients. A transparent doctor-patient relationship is emphasized as an important asset, especially in the context of an adverse reaction to a medical intervention (such as a vaccination campaign).

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