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Parsonage-Turner Syndrome, an Unusual Infant Impairment - A Case Report and Review of Literature

F. A. B. Garcia & G. R. L. Barroso

[1]Federal University of Mato Grosso, Health Sciences Institute

Author for correspondence: feraubigar@yahoo.com.br

Abstract. The Parsonage-Turner Syndrome, also known as Neuralgic Amyotrophy, is classified as a plexopathy. A multifactorial arrangement of trigger factors are proposed, including traumatic injury and an autoimmune hypothesis. Treatment still lays on the empirical practice and the preventive measure is unclear and not available. The objectives of this article is describing an unusual case report of a patient, female, 12-years-old, who was diagnosed with Parsonage-Turner Syndrome, and more important, to display the fact that not only young adults and elderly individuals are predisposed to develop this injury.
Keywords: plexopathy, electroneuromyography, infant

Introduction

The Parsonage-Turner Syndrome (PTS), also known as Neuralgic Amyotrophy, is classified as a plexopathy and classically runs with a sudden onset of pain, which lasts for hours or days, multifocal paresis, sensorial loss on the distribution of brachial plexus, would also runs over another portions of the Peripheral Nervous System, and cause muscular atrophy of the upper limbs, specially those associated to the shoulder, normally these symptoms occur unilaterally, which can last weeks to years (SEROR, 2016). These symptoms are related to the nerve inflammation and due to the axonal loss (SEROR, 2016; van EIJK, 2016). PTS can be divided in two main disorders, the idiopathic and the hereditary forms, being the first one 10 times more common. It can predominate in a single episode or recurrent episodes during life (SEROR, 2016; EIJK, 2016). Its etiology had not been elucidated since 1948 when Maurice Parsonage and John Turners first established its major clinical aspects (PARSONAGE & TURNER, 1948), but a multifactorial arrangement of trigger factors are proposed: biomechanical injury, autoimmune response, upper respiratory tract virus infections, genetic susceptibility (van ALFEN, 2011; van EIJK, 2016). Treatment still lays on the empirical practice, with few evidences and absence of guidelines, and the preventive measure is unclear and not available. Its incidence varies around 1 to 3 cases in 100,000 individuals per year, but it is thought that the incidence might be 20 to 30 cases in 100,000 individuals, principally due to the under-recognition and misdiagnosis (van ALFEN, 2011; van EIJK, 2016). There is also a major incidence in men than women, with ratios of 2:1 until 4:1 (TILBERY, 2000).

The case described bellow demonstrate a rare condition of a young girl presenting her first PTS attack, who had already been examined by an orthopedist and then referred to a neurology centre, where a hypothesis of PTS was raised and confirmed by the electroneuromyography. Treatment was initiated, there was a pain symptom reduction, but increase in muscle atrophy. Meanwhile the patient suffered a relapse, in a short period of time.

Methods

There was made an analysis of the case described below, focusing the importance of being aware that young patients can present and develop this injury, and also the rarity involving a young girl and her recidivism in a short period of time.

A bibliographic survey was made in PubMed and MEDLINE platforms, looking for case relates and reviews associated to PTS, looking for its progression, management, etiology, treatment and also child injury and recidivism.

Case Report

JLC, a 12-year-old Caucasian female student, presented to the Integrated Neurology Center - Sinop MT, complaining of sudden onset of

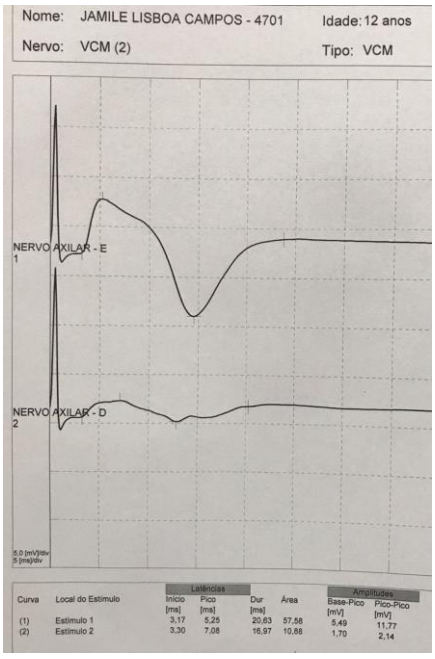
severe pain in the shoulder, progressing to the cervical region in one hour and then radiating to distal Region of the upper right limb, worsening during the night time, being awakened by pain, associated with progressive loss of force on the muscles of the right upper limb.

The patient reported a history of non-specific trauma due to repetitive movements associated with the use of school bag and an infectious disease, an upper respiratory tract infection, ethnologically not described 10 days before the onset of PTS. It is reported that forty days before the beginning of the clinical condition she had Herpes Zoster on her…

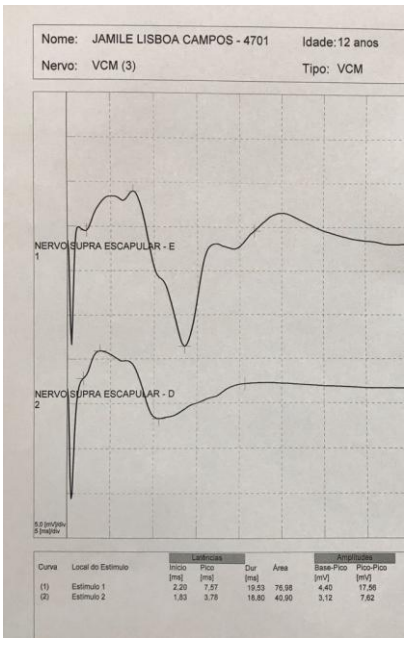
Another important point is the fact that the patient recently experienced the loss of a relative, which could have initiated a condition of discomfort or depression that may be related to the onset and intensification of pain. It has been reported that Ibuprofen and Diclofenac sodium were administered to control pain in the initial phase of the symptoms. On physical examination: Deltoid's shoulder asphyxia and right muscular atrophy were observed; Passive mobilization presented limitations due to pain; Measurement of muscle strength III on the right shoulder instead of the forearm and muscle strength of the preserved hand, other muscles still have muscle strength V; Sensitivity was preserved in the upper and lower limbs; The biceps, triceps and brachioradial reflexes were normal; Nerve compression test Tinel and Phalen were negative; Anxiety was noted during the evaluation of the muscular maneuver of the right Supraspinatus (Figure 2A).

It was suspected on tumor compression of the Brachial Plexus, Shoulder Belt Syndrome, Impact Injury Syndrome and PTS. A Brachial Plex Magnetic Resonance Imaging (MRI) pointed to an asymmetry of Short Tau Inversion Recovery (STIR) and T2 signal from the plexus components.

Probably due to the involvement of the inflammatory process, with a lower signal on the right side, however, no compressible evidence was identified. An X-ray of the shoulder and cervical / thoracic spine usually presented and a cervical spine MRI discarded spinal cord or root compression. A therapeutic regimen was established with Pregabalin 75mg daily and Duloxetine 30mg every 12 hours during 10 days. Side effects were presented during this period of time, making the use of the medication unfeasible. Predinisolone 30mg per day was also introduced for 15 days and then reduced gradually. A second pharmacological treatment regimen was started, Carbamazepine 200mg every 12 hours, Amitriptyline 25mg day, and Tramadol 50 mg every 8 hours, physiotherapy was also prescribed when the pain began to alleviate. The results of the electromyography showed a reduction in the amplitude of the motor action potential of the right axillary (Figure 1A) and suprascapular nerves (Figure 1B), which, together with the clinical presentation of the patient, contributed to the diagnosis of PTS.



A



B

Figure 1: Electroneurography performed on the right and left axillar nerve observing reduction of amplitude of motor potential right side in relation to the left in A. Electroneurography performed on the right and left suprascapular nerve observing a reduction in the amplitude of the motor potential on the right side in relation to the left one in B.

The patient improves pain, but the Deltoid, Infraspinatus, Supraspinatus and Serratus right worsened. Around 30 days after the start of the second treatment, the patient returned presenting

intense pain after doing stretching exercises in the physiotherapy, progressed with worsening pain and accentuated the muscular atrophy (Figure 2B). An adjustment was made on the dose of Tramadol to 50 mg every 6 hours.

It is also known that PTS can present cases of recurrence during life (SEROR, 2016), but during the literature review was found any case of short-term PTS relapse attack, combined with the young patient's presentation, We consider this an unusual condition And rare. New insights into the development and recurrence of PTS, especially in young patients, should be evaluated, focusing on the presentations and songs that may aid in diagnosis. It is important to remember that children's presentations of PTS may occur, but most of the time it is misdiagnosed or even not cogitate in the differential diagnosis (SALEEM & MOZAFFAR, 1999; NORD, 2017) Treatment and prevention are still under study. Better corticosteroid response curves were made due to the fact that an autoimmune reaction causes symptoms and some studies show that early treatment with corticosteroids reduces pain and provides a better prognosis (FEINBERG & REDECKI, 2010; SMITH et al. , 2014), physiotherapy associated with occupational therapy, demonstrated improvement in muscle strength and patient satisfaction in their performance during rehabilitation (JSPEERT et al., 2013) and a new control, an immunoglobulin, improving the prognosis and the residual, focused complaint In the competition of SEPT9 protein involved in proliferation and development of T cells in the peripheral nerves, demonstrated good results in managing pain and recovery of patients (CHUK et al., 2016).

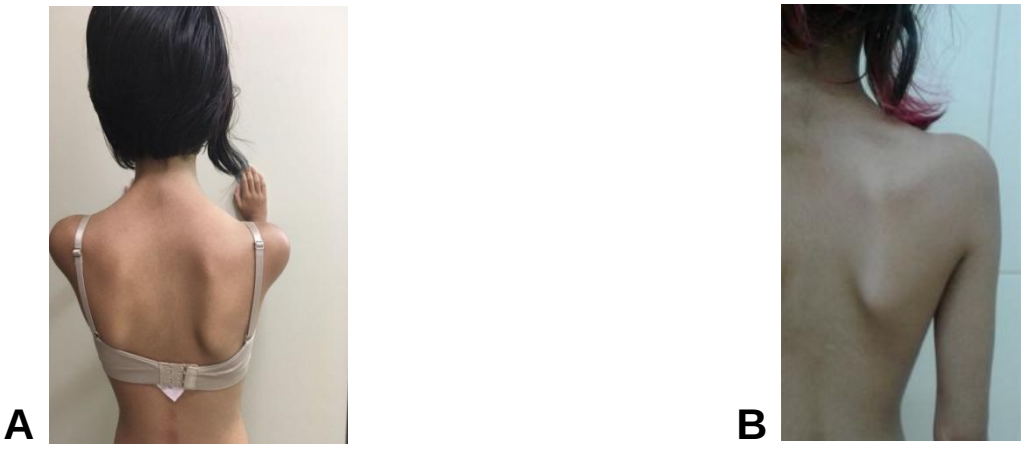


Figure 2: Muscular atrophy of the right shoulder girdle. Image in effect of the first evaluation in A and image 20 days after onset with symptomatic medications in B.

A retrospective study published in the Japanese Society of Internal Medicine showed good results by associating an intravenous immunoglobulin and methylprednisolone pulse therapy, demonstrating an improvement in 9 of 10 patients (NAITO et al., 2012). Pain usually is not effective for the first one - Line analgesics, such as acetaminophen or non-steroidal anti-inflammatory drugs (NSAIDs), and the use of co-analgesics takes a long time to become effective in the acute phase (JEROEN et al. 2016). One of the best levels of pain relief treatment involves the concomitant use of NSAIDs and opioids, demonstrating good pain control in about 60% of individuals versus 31% and 2% of opioids alone and NSAIDs alone, respectively (SMITH et al. ., 2014). Resting and immobilizing the affected limb or extremity during the PTS pain attack phase may help decrease exacerbation. Another important point of view is the association of mental disorders and shoulder pain syndromes, demonstrated in a Canadian article where 18 patients, of 34 evaluated in the tested group, presented a depressive event and another 11 patients presented stress factors, applying changes in the Daily life or serious prognosis in relation to life expectancy. The assessment of these patients included the use of amitriptyline (75 mg / day in divided doses) and lithium carbonate (administered in a dose sufficient to achieve a serum lithium level between 0.5 and 1.0 mEq / l) (TYBER, 1974).

Associating with our case, the loss of a patient's relative might have played a part in triggering the onset of pain, perhaps because of a destruction or condition of mental disorder triggered by the feeling of mourning. Many other types of evaluation and treatment are described in the literature, such as some administrations that use anesthesia, but there is still little evidence for most of them, so they have not been mentioned here. Of course, the etiology can be better understood, in our case the suspect involves the biomechanical trauma due to the school scholarship and upper respiratory tract infection, both of which are mentioned as causes of triggering the PTS attacks (van ALFEN, 2011; Van EIJK, 2016), and also the physiotherapy involved in the second attack. Associated with the

understanding of the etiology comes the development of better treatments and management characteristics to provide patients with pain relief and rapid recovery of muscle atrophy and also other residual complaints The evolution of PTS evolves.

Conclusion

The complexity involving the presentation and progression of PTS and its unusual incidence in medical centers makes its approach complicated and frequently not listed in the physician's differential hypothesis about shoulder damage. Even more when related to a child's involvement as described above, it is more difficult to deal with and get better results. The clinical sense during the assessment of the history and findings on the physical examination is important and necessary, combined with the correct initial approach and the serial follow-up makes an optimal care assessment for the patient. Agreement The patient's mother allowed us to describe and publish her daughter's story, images, exams and diagnosis without objection.

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