

Musculoskeletal changes and pain in Parsonage Turner syndrome patients: integrative review

Alterações musculoesqueléticas e dor em pacientes portadores da síndrome de Parsonage Turner: revisão integrativa

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ABSTRACT

BACKGROUND AND OBJECTIVES: Parsonage Turner Syndrome (PTS) is rare, with an incidence of 2 to 3 cases per 100,000 individuals per year. It's accompanied by intense, self-limiting pain, disappearing after weeks, followed by muscle weakness. The aim of the present study was to describe the musculoskeletal changes, muscle variations, and the pain scenario affected by the syndrome.

METHODS: Integrative review performed in the LILACS, Scielo and Pubmed databases. The inclusion criteria established were case studies, case series, clinical trials and cohort studies in Portuguese, English and Spanish languages, published between 2010 and 2020, which addressed the muscle changes and pain caused by PTS.

RESULTS: Seven scientific articles that met the inclusion criteria were analyzed, with a total sample of 183 patients aged between 7 and 65 years.

CONCLUSION: Generally, patients present alterations of the posterior interosseous nerves, anterior interosseous, axillary, long thoracic and suprascapular, muscular atrophy of the deltoid, supraspinal and infraspinal regions, with pain lasting an average of 15 days in the shoulder and scapular regions.

Keywords: Brachial plexus neuritis, Parsonage-Turner syndrome, Musculoskeletal pain.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A síndrome de Parsonage Turner (SPT) é rara, com incidência de 2 a 3 casos por 100.000 habitantes ao ano. Apresenta-se com dor intensa e auto restritiva, desaparecendo após semanas, seguida de fraqueza muscular. O objetivo deste estudo foi descrever as alterações musculoesqueléticas, variações musculares e quadro doloroso afetados pela síndrome.

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MÉTODOS: Revisão integrativa nas bases de dados LILACS, Scielo e Pubmed. Os critérios de inclusão estabelecidos foram estudos de casos, séries de casos, ensaios clínicos e estudos de coortes nos idiomas português, inglês e espanhol, publicados entre 2010 e 2020, que abordaram as alterações musculares e dor causadas pela SPT.

RESULTADOS: Foram analisados sete artigos científicos que preencheram os critérios de inclusão, com amostra total de 183 pacientes com idade entre 7 e 65 anos de idade.

CONCLUSÃO: Geralmente os pacientes apresentam alterações dos nervos interósseo posterior, interósseo anterior, axilar, torácico longo e supraescapular, atrofia muscular das regiões de deltoide, supraespinhal e infraespinhal, com quadro algico de duração média de 15 dias em região de ombro e escápula.

Descritores: Dor musculoesquelética, Neurite do plexo braquial, Síndrome de Parsonage Turner.

INTRODUCTION

Parsonage Turner Syndrome (PTS), also known as acute idiopathic brachial neuritis or scapular girdle syndrome¹ is a rare and severe syndrome. Spontaneous pain in the scapular girdle region and muscle weakness followed by atrophy are its main features. There is a decrease in functional capacity accompanied by a decline in range of motion, which may cause absence from work activities and daily occupations, also lowering quality of life (QoL).

The annual incidence is 2 to 3 cases per 100,000 inhabitants. Although it can affect both sexes, the higher prevalence is in middle-aged men. The etiology of PTS is unknown, but studies link it to infections, surgeries, hereditary factors, rheumatic diseases, and stressful exercises^{2,3}.

PTS can be diagnosed by nuclear magnetic resonance and electromyography⁴; however, due to their high cost, it's harder for the less economically privileged population to have access to these tests. Thus, the knowledge of the conditions presented is important for the diagnosis, which must be confirmed with these tests. Even so, in some cases the results are not accurate in the first weeks, and it's necessary to repeat the test four weeks after the onset of symptoms⁵.

The clinical scenario is characterized by severe pain with acute onset in the lateral region of the shoulder, self-limited, which may disappear after a few days, and there may be muscle weakness, changes in reflexes, and sensory deficits³. Most patients report difficulty with the lateral abduction movement of the shoulder. The most affected muscles have evidence of liposubstitution and atrophy, which lowers functional capacity⁶.

The syndrome occurs in a chronological order. It begins with pain, which can interfere with the sleep-wake cycle. After three or more weeks of weakness, muscle atrophy begins and lasts for several months, and in some cases may last for years².

It's essential that health professionals know about PTS in order to ensure a more efficient therapeutic follow-up for patients with this syndrome. The professional who has initial contact with a patient with PTS must analyze the alterations caused and give the proper guidance, so that the patient can have a better QoL.

The objective of this study was to analyze the musculoskeletal changes, the main muscles involved and the manifestation of pain.

METHODS

An integrative literature review that followed the steps of determining the subject, establishing the criteria for selecting studies, searching databases, and evaluating the selected articles. The searches were performed in LILACS, Scielo and Pubmed databases. The established criteria were case studies, case series, clinical trials, cohort studies, in Portuguese, English and Spanish, published between 2010 and 2020, and that addressed the muscle changes and pain caused by PTS.

For the search synthesis, descriptors from the Medical Subject Headings (Mesh) platform were used with the following descriptors: "Brachial Plexus Neuritides", "Neuritides Brachial Plexus", "Neuritis, Brachial Plexus", "Girdle Neuropathies Shoulder", "Girdle Neuropathy Shoulder", "Parsonage-Turner Syndrome", "Parsonage Turner Syndrome", "Syndrome Parsonage-Turner", "Neuralgia Amyotrophic", "Amyotrophic Neuralgias", "Neuralgias Amyotrophic", "Amyotrophy Neuralgic", "Amyotrophies Neuralgic", "Neuralgic Amyotrophies", "Neuralgic Amyotrophy", "Amyotrophies Hereditary Neuralgic", "Hereditary Neuralgic Amyotrophies", "Neuralgia Cervicobrachial", "Neuralgias Cervicobrachial". The descriptor search strategies were combined with the Boolean operators AND and OR, according to the database presented in table 1.

Articles were selected by reading the title, abstract, and year of publication. Those that met the inclusion criteria were analyzed in full. In addition, data regarding the type of study, year, author,

population studied, muscles involved, and pain scenario were inserted in a flowchart.

RESULTS

A total of 115 articles were gathered, of which 100 were excluded based on title and abstract. Another 3 were excluded because they were duplicates. Only 12 articles were selected and submitted to a full reading. Later, 5 articles were excluded for not meeting the eligibility criteria, so only 7 articles were included in the review (Figure 1).

The 7 included studies had the participation of 183 individuals, aged between 7 and 65 years old. The largest number of articles was published in 2015. Three are cohort studies, two are prospective clinical trials, and two are case reports. The data on the characteristics of the selected articles are described in table 2.

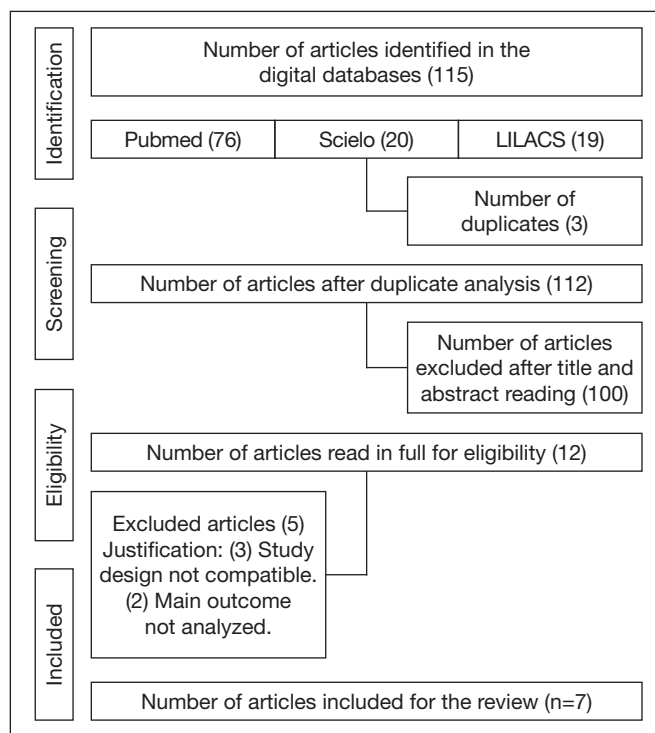


Figure 1. Article selection

Table 2. Characteristics of included studies

Authors	Study design	Objective	Method	Involved muscles and pain scenario	Conclusion
Ibrahim et al. ⁷	Case report	To present an abnormal case of PTS, 10 weeks after rotator cuff tear repair surgery.	A 60-year-old patient presented right upper limb weakness 10 weeks after rotator cuff tear repair surgery.	Weakness in right biceps and brachioradialis with denervation of deltoid, supraspinal, infraspinal and right biceps. Absence of pain.	Recognition of symptom variability allows for greater recognition and better patient care outcomes.
Milner et al. ²	Cohort	Retrospective assessment of the clinical characteristics of all PTS patients seen over 9 years.	Data collected were sex, affected side, laterality, inciting event, clinical presentation, nerve involvement, time spent recovering, recovery extension (n=38).	Patients had muscle weakness; however, there is no report on which muscles were affected. Pain in the shoulder and scapular region.	It's expected that 65% of patients will recover strength level 4 or more in an average period of 10 months after diagnosis.

Continue...

Table 2. Characteristics of included studies – continuation

Authors	Study design	Objective	Method	Involved muscles and pain scenario	Conclusion
Santos et al. ¹	Clinical trial	To describe the clinical, electrophysiological, and imaging findings of PTS and to evaluate the results of conservative treatment.	Eight cases were studied between February 2010 and February 2012, with a minimum time span of one year. All patients answered a clinical questionnaire and underwent functional evaluation by the Constant and Murley score. After clinical suspicion, electromyography was performed to confirm the diagnosis.	Three cases of intense deltoid muscle atrophy, three cases of hypotrophy of the supraspinal and infraspinous. Sudden onset of pain scenario lasting 24 hours or at most 15 days.	Functional recovery was noted in most cases, although muscle strength was not fully restored.
Fransz et al. ⁸	Case report	Describe diagnosis of PTS in a patient two months after initial presentation to the emergency room due to accidental exposure to bloodborne pathogens.	Describe neurological findings and findings of plain radiograph of the right shoulder. Patient was diagnosed with post-vaccine PTS and treatment followed conservatively for 15 months.	The patient presented supraspinal weakness and deltoid muscle atrophy. Pain in the cervical region, irradiating to the right arm.	Conservative treatment is the most recommended. However, almost one third of patients suffer from residual complaints after six years.
Van Alfen, van Eijk and Ennik ⁹	Cohort	To prospectively ascertain the one-year incidence rate of classic neuralgic amyotrophy in a primary care setting.	The study included 14 patients with a classic phenotype. After being included, patients with suspected neuralgic amyotrophy or PTS who had not yet seen a neurologist were taken for neurological evaluation to confirm the diagnosis.	There were no reports of muscle involvement. There was pain in the cervical region, shoulder, and arm.	The conclusion was that the incidence is much higher than previously thought, and a more detailed evaluation in primary care is recommended.
Upadhyaya et al. ⁶	Cohort	To present the distribution and extent of brachial plexus abnormality in known cases of PTS.	The study included 15 patients diagnosed with PTS based on clinical and electrophysiological findings. The results were evaluated by two radiologists performing the analysis and inclusion of the results in an Excel table.	Muscle denervation, edema, fat infiltration in 8 patients, muscle atrophy in the supraspinal, infraspinous, deltoid and pectoralis major. Pain with tingling sensation.	The C5 root is the most commonly affected region. Associated muscle changes were most often seen in the supraspinous and infraspinous.
Charles ¹⁰	Case report	To describe chiropractic treatment in a patient with right arm paralysis and a diagnosis of PTS.	The patient's treatment protocol involved chiropractic manipulation, deep tissue therapy and exercise rehabilitation.	Weakness in pectoralis minor. The patient reported pain in right scalene, pectoralis minor and biceps muscles.	Patients with PTS who do not respond to standard medical or pharmaceutical intervention may benefit from chiropractic treatment using applied kinesiology modalities.

PTS = Parsonage Turner Syndrome.

DISCUSSION

The most common muscle changes detected in individuals with PTS were muscle atrophy, weakness, and muscle denervation, associated with persistent pain. The most affected body regions were the shoulder and scapular girdle. A cohort study including 38 patients found that the initial symptom of PTS is pain, reported by 71% of patients. The most affected regions were shoulder and scapula. Sensory symptoms, such as paresthesia or hypoesthesia, were reported by 89% of patients².

In one study 8 patients had sudden onset of pain, with spontaneous improvement, followed by weakness of the shoulder gir-

dle muscles, which are the main symptoms for the nosological diagnosis of PTS. In all patients, pain persisted for at least 24 hours, lasting at most 15 days, and only after this period muscle involvement was identified¹.

However, atypical presentations may occur, when the classic symptoms are not present, such as the absence of pain from the onset, but presence of muscle weakness and denervation⁷. In this case, the patient had undergone surgery to repair the rotator cuff prior to the syndrome being diagnosed. PTS can modify the patient's pain state, causing changes in the sleep-wake cycle. The painful symptom can remain for up to 4 weeks and, after this, muscle weakness and atrophy can occur in the shoulder and scapular girdle region⁸.

As for the pattern of involvement, most patients will have unilateral involvement with clinical presentation in the unilateral right upper limb with a ratio of 4 to 3 in relation to the left side⁶. Corroborating these findings, in a cohort study that included 38 patients in the age group of 47 years old, the PTS condition was present in the right unilateral limb in 60% of patients².

The most frequent scenario was unilateral, but in some cases it was bilateral. A trial showed that patients aged 29 years old had right-sided dominant limb involvement in 70% of the cases, confirming that age is not a decisive factor in cases of right-sided PTS¹.

The regions most affected by pain and the differences in their manifestations are some of the main characteristics of the syndrome, which makes the clinical diagnosis a challenge. The shoulder and scapula (53%), arm and elbow (13%)² and cervical regions, with irradiation to the upper limbs^{8,9}, were the most affected, which shows variation in the syndrome's pain scenario.

The muscular alterations most associated with the syndrome are related to mobility. According to a cohort study with 15 patients, the individuals reported weakness in shoulder abduction, followed by pain in the arm, cervical spine and shoulder. In 11 of these cases there was evidence of muscle denervation associated with edema, fat infiltration, and atrophy⁶. Eight patients presented muscle alteration in the form of edema, associated with signs of hyperactivity, and the infraspinous and supraspinous muscles were the most affected⁶. A case report showed that PTS compromises functionality due to loss of mobility in the elbow region, forcing continuous maintenance of the flexion position¹⁰.

A clinical trial showed that there is no consensus in the literature about which was the most affected peripheral nerve, which justifies the variety of symptoms and muscles affected by PTS¹. However, in the study that evaluated 8 patients, the long thoracic and suprascapular nerves were the most affected. Therefore, the findings related to muscle denervation, atrophy, weakness in supraspinous and infraspinous muscles are better explained¹. No reports of patients with involvement of the serratus anterior were found. In the article² that evaluated 38 cases of PTS, the most affected nerves were the posterior interosseous (24%), the anterior interosseous (18%) and the axillary (13%); however, this study only reports that the patients presented muscle weakness but doesn't inform which muscles were affected. Deltoid muscle atrophy with a muscular atrophy scenario is the most cited in three studies^{1,6,8}, raising questions about the greater incidence in the axillary nerve.

The conditions of patients with PTS are varied, so diagnosing the syndrome is a challenge¹⁰. In that sense, the knowledge of health professionals about the manifestations of PTS is essential for proper diagnosis and treatment.

The limitations of this study were the methodological quality of the selected articles and the lack of studies with strong evidence on PTS. More studies need to be conducted in order to confirm these data.

CONCLUSION

Generally, patients present alterations of the posterior interosseous nerve, anterior interosseous nerve, axillary nerve, long thoracic and suprascapular nerves, muscular atrophy of the deltoid, supraspinous and infraspinous regions, with a pain scenario lasting an average of 15 days in the shoulder and scapular regions.

AUTHORS' CONTRIBUTIONS

Iago Lisboa Santos

Data Collection, Conceptualization, Project Management, Methodology, Writing - Preparation of the original, Writing - Review and Editing

Vitor Guida Souza

Methodology, Supervision

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