Case Report

Anterior interosseous nerve lesion and distal myoclonus revealing a parsonage turner syndrome associated with hashimoto thyroiditis

Juna Musa\(^a\),* Masum Rahman\(^b\), Abu Bakar Siddik\(^c\), Kristi Saliaj\(^d\), Samar Ikram\(^b\), Ina Kola\(^e\), Alireza Shoushtarizadeh\(^h\), Ali Guy\(^f\), Inva Mamica\(^g\), Abdur Rahman\(^h\), Eram Ahsan\(^h\), Anisa Cobo\(^d\), Ruben Blanco\(^i\)

\(a\) Department of General Surgery, Mayo Clinic, Rochester, MN, USA
\(b\) Department of Neurological Surgery, Mayo Clinic, Rochester, MN, USA
\(c\) Department of Pain medicine, Mayo Clinic, Jacksonville, Fl, USA.
\(d\) University of Medicine, Tirana, Albania
\(e\) Department of Burns and Plastic Surgery, Mother Teresa University hospital center, Tirana, Albania
\(f\) Department of Physical Medicine and Rehabilitation, NY University, NY Medical center, NY, USA
\(g\) Department of Medicine, Life and Care Hospital, Dhaka, Bangladesh
\(h\) Stem Cell Therapy and Skeletal Regeneration Lab, Mayo Clinic, Rochester, MN
\(i\) Centro de Diagnóstico Por Imágenes Clínico Integral (CEDICLIN), Santiago, Dominican Republic

**Abstract**

Parsonage-Turner Syndrome (PTS), also known as brachial neuritis or neuralgic amyotrophy, is a rare disorder affecting 2 to 3 individuals per 100,000 each year. Abrupt onset shoulder pain, followed by motor weakness, paresthesia and hypoesthesia, is usually reported, lasting several months with variable recovery. The etiology of the disease may be idiopathic or triggered by an underlying autoimmune disease in genetically susceptible individuals. Our report addresses a unique case of Parsonage-Turner Syndrome in a patient suffering from concurrent Hashimoto Thyroiditis. A previously healthy 22-year-old female was referred to the Department of Neurology after complaints of sudden-onset motor weakness in her left upper limb. On physical examination, the patient could not make an “Ok sign” with her thumb and distal phalanx or form a complete fist, revealing weakness within the anterior interosseous branch of the median nerve. Further testing with electromyography demonstrated muscular atrophy within the arm’s anterior compartment, forearm, and triceps brachii of the posterior compartment. Additional imaging and physical examination were unremarkable, confirming our diagnosis of PTS. Furthermore, lab reports revealed elevated levels of anti-thyroglobulin and anti-thyroid peroxidase antibodies and our patient was concurrently diagnosed with Hashimoto’s thyroiditis.

*Competing Interests: The authors declare no conflict of interests.
*Corresponding author.

E-mail address: musajuna@gmail.com (J. Musa).

https://doi.org/10.1016/j.radcr.2021.07.067

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This case aims to highlight the rare co-occurrence of Hashimoto’s thyroiditis with Parsonage-Turner Syndrome in an otherwise healthy patient. A 2014 study published by Nugent et al. had also shed light on brachial neuritis in a patient suffering from autoimmune connective tissue disease, and through this case study, we hope to add to the growing literature regarding the correlation between PTS and autoimmune diseases. Symptoms of PTS can easily be misdiagnosed given its similarity to other peripheral neuropathies, and careful assessment and thorough understanding of the disease is required to successfully distinguish it from other neurological pathologies.

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Introduction

Parsonage Turner Syndrome (PTS), otherwise known as Neuralgic Amyotrophy (NA), is a rare neurological condition, affecting primarily the peripheral nervous system (PNS). It consists of two types, a hereditary variant (Hereditary Neuralgic Amyotrophy) and an acquired variant (Idiopathic Neuralgic Amyotrophy or Parsonage Turner Syndrome). The syndrome often presents with a classic triad of symptoms: sudden onset severe pain (usually around the shoulder), dramatic muscle weakness, and wasting (within the upper extremity and ipsilateral chest wall muscles). Alongside these symptoms, there may also be accompanying paresthesia and/or anesthesia. This oftentimes goes unnoticed due to the severe and debilitating pain the patient experiences.

Another classic feature of PTS is that the nerve territory distribution of the pain is often different from that of the paresis. This often leads to misdiagnosis as practitioners fail to fit the patient’s symptoms in a classic neurologic localization paradigm. While about two-thirds of patients affected by NA have classic shoulder involvement, PTS can also affect the peripheral nerves (median, radial, or the lower brachial plexus), sympathetic nervous system (similar to regional pain syndrome), lumbosacral involvement (similar to radiculopathy), or the phrenic nerve (presenting as unexplained dyspnea). Due to the vast phenotypic variability, clinicians have now coined the term Neuralgic Amyotrophy Syndrome that classifies all the acute-onset, painful mono- or multifocal neuropathies with a monophonic course within it.

Unfortunately, PTS is often misdiagnosed as glenohumeral bursitis or as a muscle strain, leading to the patient receiving analgesics that often fail to control their pain. Greater awareness needs to be raised regarding this relatively rare syndrome, that if misdiagnosed, can significantly affect patients’ quality of life.

Case Report

A previously healthy, 22 year-old female was referred to the Department of Neurology, with complaints of episodes of isolated, sudden weakness of the left upper limb. The neurological examination revealed weakness of the distal phalanx of the thumb and middle phalanx of the index finger. The patient was unable to perform the “Ok” sign using the thumb and index finger and was unable to form a fist by completely closing her left hand. (Figs. 1-2)

A decreased grip strength on the left side was also present. The examination showed no sensory deficits. Tendon reflexes were normal in all extremities, with no evidence of muscular wasting. No pathological reflexes or focal

Fig. 1 – The ok sign.

Fig. 2.
neurological signs were present. The rest of the objective examination was normal, with no significant findings. Routine laboratory examinations were within normal limits. Her personal medical history and family history were unremarkable.

An electromyographic (EMG) evaluation of the left upper limb showed neurogenic atrophy of the muscles innervated by the anterior interosseous nerve (AIN); the flexor digitorum profundus pronator quadratus, biceps brachii, triceps brachii, and deltoid muscles. (Figs. 3-6)

The amyotrophy affecting these muscular regions was secondary to Parsonage Turner Syndrome (Idiopathic brachial plexopathy). MRI of the left upper extremity was performed, which showed no abnormalities. (Figs. 7-10)
Strikingly, the patient was found to have elevated levels of both anti-thyroglobulin antibodies, anti-Tg (315 IU/ml) and anti-thyroid peroxidase antibodies, anti-TPO (360 IU/ml). The patient received a diagnosis of Hashimoto’s thyroiditis as well.

To the best of our knowledge, this is the first case report of the simultaneous occurrence of both Parsonage Turner Syndrome and Hashimoto’s thyroiditis.

**Discussion**

Parsonage-Turner Syndrome (PTS) is a rare type of neuropathy, also known as neuralgic amyotrophy. It commonly manifests as a sudden onset of unilateral shoulder pain, often associated with progressive limb weakness, sensory disturbances and muscular wasting [1]. Adults are the most affected, with one study reporting the age of 47 as the average age at diagnosis, while there have been reports of patients ranging from three months old to 75 years of age [1]. The pain is usually continuous, more severe at night and is not caused by a change in posture. The condition is typically self-limiting and persists for 1 to 2 weeks; however, chronic pain has also been reported. The hypoesthesia, paresthesia and muscular atrophy seem to resolve with time. Even though most patients show 80%–90% muscle strength restoration over 2 to 3 years, more than 70% of patients still exhibit residual paresis and exercise intolerance [2,3].
Despite the fact that the etiology and pathophysiology remain ambiguous, PTS has often been identified in a post-operative, post-infectious, and post-vaccination context [1].

Since some PTS variations are inherited, a genetic component has also been proposed. Several antecedent events have been identified, with the most common one being viral infections, followed by recent immunizations [1]. This has lead to the theory that the brachial plexus is damaged either directly by the viral agent or indirectly, by an abnormal immune response to the viral illness or antigen in the vaccination [1].
ory of the autoimmune nature of the condition is in line with our findings, of other autoimmune diseases such as systemic lupus erythematosus (SLE), temporal arteritis and polyarteritis nodosa (PAN), co-existing with PTS [4]. Another theory suggests that sudden pain stems from ischemia triggered by mechanical or inflammatory factors, namely trauma, surgery or inflammatory conditions [1]. This is consistent with the post-operative histopathological examination of nerve biopsies [1]. On rare occasions, no antecedent event can be traced back. Alas, the diagnosis is not straightforward and frequently overlaps with other disorders, including cervical spondylosis, cervical radiculopathy, adhesive capsulitis, and acute calcific tendinitis. The diagnosis is primarily clinical, based on a thorough medical history and pathological findings in the physical examination and electromyographic (EMG) exam. Additional diagnostic work-up, including laboratory and imaging, offer support mainly by ruling out other possible etiologies [5-7]. Therefore, PTS should always be considered in the differential diagnosis of acute-onset shoulder pain.

Ultimately, PTS is a diagnosis of exclusion. It may frequently present with a severe clinical picture associated with normal radiological findings, due to the inadequate sensitivity of common imaging modalities, particularly during the acute phase of the pathology [8-10]. In this context, an in-depth clinical history and physical examination, in tandem with a meticulous EMG evaluation are paramount in establishing a definitive diagnosis of PTS, after ruling out other possible etiologies of acute shoulder pain.

As illustrated by this case report, a thorough physical examination aids in the diagnosis of other potential comorbidities as well, specifically in view of an idiopathic, autoimmune origin.

To date, conservative management with physical therapy has been the treatment of choice [7].

**Conclusion**

In conclusion, PTS is a rare type of peripheral neuropathy that is believed to occur due to external factors, environmental or otherwise, in genetically susceptible individuals. Associations with autoimmune conditions have been previously reported. Nevertheless, to the best of our knowledge, this is the first case report highlighting the concomitant presence of Parsonage Turner Syndrome and Hashimoto’s Thyroiditis. Our patient presented with sudden, isolated weakness of the left upper limb with no motor or sensory disturbances. Clinical and radiologic evaluation established the diagnosis of Parsonage Turner Syndrome. Laboratory examinations revealed the accompanying presence of Hashimoto’s thyroiditis, as well.

It is our hope, this case report helps emphasize the importance of considering Parsonage Turner Syndrome in the differential diagnosis of acute-onset shoulder pain, particularly in patients with pre-existing, underlying autoimmune conditions. Patient consent was obtained for the study.

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