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CASE REPORT

### An unusual cause of shoulder pain and dysfunction of the shoulder girdle: Parsonage–Turner syndrome case report

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Abstract Parsonage-Turner syndrome (PTS) is a rare disorder consisting of a complex aggregate of signs and symptoms with an acute onset of pain followed by muscle weakness in a separate peripheral nerve distribution, dysesthesias, and numbness. Although the etiology of PTS is unclear, it is reported in various clinical situations, including postoperatively, postinfectious, posttraumatic, and postvaccination. We report a case of a 27-year-old man that presented to our outpatient clinic with a throbbing acute shoulder pain and motor weakness over several months that did not improve after 2 years. At the time of consultation, he complained of left-sided neck pain radiating to the left deltoid muscle and axilla as well as left shoulder blade pain with shoulder girdle muscle weakness. Repeated electrodiagnostic studies revealed denervation limited to the supraspinatus and infraspinatus muscles without evidence of cervical radiculopathy. He was diagnosed with PTS. The authors review patient presentation, physical examination, and workup needed for diagnosis of PTS to help physicians diagnose and treat this complex syndrome.

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#### Introduction

Parsonage-Turner Syndrome (PTS) is a rare disease occurring in otherwise healthy individuals, presenting with sudden shoulder pain that may begin rather insidiously but quickly amplifies in severity and intensity. The acute period of pain is replaced over a few days to weeks time with progressive motor weakness, autonomic changes, and sensory abnormalities in varying clinical presentations that typically involve the shoulder girdle musculature and proximal upper limb muscles. PTS evolves to a self-limited disease usually with complete recovery by the end of the third year. It was first described in 1897 by Feinburg [1] who reported a case of unilateral brachial plexus neuritis associated with influenza. Parsonage and Turner in 1948 in The Lancet described 136 cases of the condition as a neuralgic amyotrophy and gave it the eponymous term "shoulder-girdle syndrome" [2].

The condition, also known as neuralgic amyotrophy or brachial neuritis, has been reported in numerous clinical situations that involve some sort of antecedent impact on the patient, whether it be surgical, infectious, traumatic, or even iatrogenic. Nerve biopsies done in this patients show evidence of ischemic changes demonstrated by perineural thickening, neovascularization, and focal fiber loss suggesting an immune pathogenesis [3].

The reported incidence is 1.64 cases per 100,000 people [4] with a higher incidence in men than in women without

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any trend for hand dominance. Patients affected by PTS range from 3 months of age to 75 years with a peak in the third and seventh decades [5].

We present a clinical case of a young patient with disabling shoulder pain, progressive motor weakness of the shoulder girdle muscles, and autonomic impairment of the upper limb that posed a major diagnostic challenge.

#### **Case report**

A 27-year-old male patient without any relevant medical history presented to our outpatient clinic with complaints of left-sided "achy, stabbing, and burning" neck pain that radiated into the axilla and left shoulder and progressive motor shoulder girdle weakness that not only limited his ability to lift his arm but also limited his ability to perform activities of daily living, including disturbance of sleeping pattern.

He was medicated with NSAIDs and scheduled for another appointment.

On his second outpatient, he still presented with complaints of a dull, diffuse pain over the shoulder girdle and showed signs of muscle atrophy of the supraspinatus and infraspinatus muscles (Fig. 1).



Fig. 1 Muscle atrophy of the supraspinatus and infraspinatus

An electromyography (EMG) and magnetic resonance imaging (MRI) were performed 3 months after the onset of symptoms.

EMG showed abnormal motor unit patterns, suggesting a neuropathy mainly in the supraspinatus and infraspinatus muscles. The other muscles of the brachial plexus were normal. Sensory nerve conductions were normal.

MRI of the left shoulder showed diffuse increased T2 signal in the supraspinatus and infraspinatus associated with moderate fatty atrophy (Figs. 2 and 3).

The diagnosis of PTS was established with recommendation for conservative treatment with aggressive pain management and intensive outpatient physical therapy.

#### Discussion

Parsonage–Turner Syndrome is the term given to describe an entity also known as acute brachial neuritis, neuralgic amyotrophy, or brachial neuropathy to name but a few.

It is rare, with an incidence of 1.64 cases per 100,000 population being reported [4]. It appears to affect males more than females, with a peak incidence in patients in their third and seventh decades [6]. There is no relationship to hand dominance, although the condition occurs bilaterally in one third of cases [4, 6].

The precise cause of PTS is unknown with many factors being proposed to cause the neuritis including trauma, infection, heavy exercise, surgical procedures, immunization, and autoimmune conditions [4, 6]. The development of PTS symptoms following surgery can be quite variable occurring within 24 h of the procedure or up to a week or more following surgery. Although postsurgical neurological changes can be iatrogenic, there is no relationship with the surgical technique or anesthesia used [4].

Infection seems to precede the onset of symptoms in up to one quarter of patients, with Tsairis et al. [5] reporting an antecedent flu-like illness. Heavy exercise and surgical procedures are also common triggers of PTS in the literature [7]. A viral etiology has also been proposed, with the Coxsackie B virus implicated in an epidemic reported [8].

Several similarities exist between Parsonage–Turner syndrome and adhesive capsulitis. Both conditions present with severe pain, worse at night, and, initially, are unremitting regardless of position, are idiopathic, have a nonspecific inflammatory component, and will resolve spontaneously with a relatively good long-term prognosis for recovery of function. However, one major difference exists. In patients with PTS, glenohumeral motion is usually completely preserved. This may largely be the result of the different structures involved (capsule vs. nerve) and not a reflection of the etiologic factors that cause the conditions. There are no

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**Fig. 2** MRI of the left shoulder showing diffuse increased T2 signal in the supraspinatus and infraspinatus



current reports of a patient presenting with simultaneous PTS and adhesive capsulitis.

The characteristic pattern of acute pain followed by muscle weakness are generally the clues to the diagnosis of PTS, with confirmation being sought by electromyography. The pain is often referred as a severe ache or throbbing pain radiating from the shoulder distally down the arm or proximally into the neck. These symptoms may last for a few hours or persist for up to 3 or more weeks. The patient will describe weakness after the pain has subsided, with an estimated 70 % of sufferers experiencing weakness 2 weeks after the onset of symptoms [5]. Electromyographic findings are generally found about 3 weeks after the onset of symptoms [7]. As PTS is believed to be an axonal process, the diagnosis is therefore very dependent on the EMG (needle) portion of the electrodiagnostic exam. Widespread denervation is usually seen in involved muscles, and complete denervation is often the case. As PTS is an axonal disorder commonly affecting proximal muscles of the upper limb, motor and sensory nerve conduction velocities and distal latencies routinely tested on the distal upper limb are usually normal. Because of the atypical distribution, electromyographers need to be extremely detail-oriented in the systematic approach to testing specific muscles of the upper limb. Testing of muscles

Fig. 3 MRI of the left shoulder documenting moderate fatty atrophy of the supraspinatus and infraspinatus



that are not routinely examined during EMG studies should be interrogated by needle EMG even when they appear to be clinically asymptomatic.

The most important aspect of evaluating PTS is finding nerve injury in individual muscles. These muscles should not share root innervation, which may imply radiculopathy.

The most frequent patterns of lesion are single and multiple mononeuropathies affecting the proximal musculature involving potentially any part of the brachial plexus [9].

Studies on recovery of PTS based on EMG findings reveal that reinnervation usually begins somewhere between 6 months and 1 year. This time, variable is based on the fact that axonal regeneration and eventual muscle reinnervation are length-dependent processes. The rate of axonal regeneration is classically described as occurring at a rate of 1-4 mm/day.

Although PTS is a clinical diagnosis, EMG is a form of diagnostic testing that can better identify, isolate, and grade severity of denervation and, after a period of time, reinnervation of the involved muscles.

MRI is an important tool for establishing the diagnosis of PTS. MRI excludes causes of shoulder pain such as rotator cuff tear, impingement syndromes, or labral tears. MRI is sensitive for the detection of signal abnormalities in the shoulder girdle musculature that are related to denervation.

MRI also excludes structural abnormalities that may cause similar denervation changes in rotator cuff musculature such as rotator cuff tears or spinoglenoid and suprascapular notch masses.

Intramuscular changes observed in patients with PTS reflect denervation changes and vary with the stage of disease [10].

The mechanism and time course of MRI signal intensity changes in denervated muscle are not understood completely [11]. In the acute phase of denervation, the signal intensity of the muscles may be normal [11]. The earliest detectable change in denervated muscles is a diffuse increased T2-weighted signal, which may occur without a T1-weighted signal change [11]. In the subacute and chronic phases of denervation, T2-weighted signal abnormalities persist and muscular atrophy may develop [11]. Atrophic changes in muscle are reflected by decreased muscular mass and increased intramuscular, linear, T1-weighted signal due to fatty infiltration. MRI intramuscular signal change may revert to normal in several months after the chronic phase [11].

The prognosis of PTS is excellent with one of the largest studies by Tsairis et al. stating that 89 % of patients have fully functional recovery at 3 years [5].

In the earliest stages of PTS, pain management with opiates, NSAIDS, and neuroleptics is the mainstay of treatment. Oral steroids have been recommended, but there is poor literature evidence to support its efficacy [12]. Physical therapy plays an important role in the treatment of PTS.

Modalities such as TENS can help in pain management. A more aggressive therapeutic treatment plan can be instituted once the painful stage has abated. The timing and the role of strengthening exercises are dependent on the degree of muscle denervation, weakness, altered biomechanics, and the premorbid functional level of activity for that patient and should involve the assistance of a physical or occupational therapist [13].

Limited EMG follow-up testing of the involved muscles can be used to demonstrate the extent of reinnervation [14]. This information can be useful in helping determine when muscles can tolerate more aggressive strengthening. Observing functional motion and carefully scrutinizing the biomechanics and the flow of motion can help one determine what type of exercises are appropriate and what levels of aggressive training should be implemented as initiating strengthening of the shoulder girdle when existing muscle denervation can exacerbate the varying muscle strength imbalances that already exist. This alters the biomechanics of the shoulder and establishes a dysfunctional pattern of motion that leads to further injury of the denervated muscle from overstretching and eccentric overload.

#### Conclusion

PTS is a disease that presents acutely with disabling pain and is difficult to diagnose in its acute state. The pain is generally self-limiting, and the natural evolution of PTS leads to a favorable functional outcome. Early diagnosis with a good clinical evaluation, EMG, and relevant imaging (MRI) allow for aggressive pain management, help outline proper therapy, and provide comfort to both patient and physician in establishing a diagnosis.

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethical standard**The authors have written consent of the patient to publish all data pertaining this to case report.

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