CASE REPORT

Atypical presentation of Parsonage-Turner syndrome confounded by surgical rotator cuff injury

Dustin Adam Greenhill, Hesham Abdelfattah, Joseph S Torg, Joseph Milo Sowards

Department of Orthopaedic Surgery and Sports Medicine, Temple University Hospital, Philadelphia, Pennsylvania, USA

Correspondence to
Dr Hesham Abdelfattah,
hesham83@gmail.com

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SUMMARY

Parsonage-Turner syndrome (PTS) is a rare neuropathy that commonly presents as unexpected severe shoulder and arm pain that eventually subsides while weakness or paralysis ensues. During exceptions to this classic presentation, confirming PTS can be challenging. Alternative causes of upper extremity pain may confound the diagnostic algorithm. Moreover, objective findings from necessary diagnostic tests depend on when those tests are performed. We present an atypical onset of PTS, whereby the initial presentation of severe neuropathic pain was preceded by mild shoulder pain that should decrease one's clinical suspicion for PTS. This milder pain coincided with the presence of a rotator cuff injury, whereby surgical intervention preceded impending paralysis and hindered postoperative rehabilitation. Physicians should be aware of the possibility of atypical presentations of PTS in hopes of avoiding either untimely surgery or delays in diagnosis.

BACKGROUND

Parsonage-Turner Syndrome (PTS) commonly presents to a neurology clinic after the onset of unexplained upper extremity pain that precedes impending weakness or paralysis. It less commonly presents to the orthopaedic outpatient setting where concomitant acute surgical pathology may be easily recognised on routine imaging. Alternatively, PTS may develop postoperatively during a time when increased upper extremity pain is expected. This case report is intended to both highlight the possibility of atypical presentations of PTS and also serve as a reminder to practitioners in fields whereby PTS is a relatively uncommon diagnosis that PTS should remain within the differential diagnosis throughout a patient's treatment course.

CASE PRESENTATION

A 57-year-old right-handed man presented to the outpatient orthopaedic clinic 2 weeks after lifting a heavy object overhead with a chief complaint of right shoulder and arm pain. During the initial exam, pain was reproduced with Jobe's supraspinatus test and shoulder abduction but not exacerbated by neck movement. MRI of the shoulder performed after the first outpatient visit revealed a partial-thickness (<50%) articular-sided supraspinatus tear, fluid in the subacromial region and no other abnormalities. There was no evidence of denervation oedema, muscular atrophy or cysts. Cervical spine roentgenograms were unremarkable

and there were no sensory complaints. Pain was documented by the examiner to be more severe than normally attributable to a partial rotator cuff tear. However, after 8 weeks of adequate physical therapy the patient still complained of persistent pain and underwent isolated arthroscopic surgical debridement of the right rotator cuff. The subacromial space was inspected but the suprascapular notch was not approached during the procedure. No perioperative scalene block was performed. The patient's pain did not subside postoperatively. Physical examination 2 weeks after surgery revealed exquisite tenderness over the shoulder and deltoid out of proportion to previous arthroscopic findings, unilaterally diminished deep tendon reflexes of the biceps brachii and marked weakness with both shoulder abduction and external rotation. Electrodiagnostics 3 weeks postoperatively (15 weeks after symptom onset) revealed denervation and reinnervation suggestive of PTS affecting the supraspinatus, infraspinatus, deltoid and biceps. Paraspinal testing was normal. A cervical spine MRI at the same time was unremarkable.

OUTCOME AND FOLLOW-UP

At 6 months, shoulder range of motion was extremely limited. As the severity and distribution of pain steadily improved, aggressive physical therapy was required in attempt to optimise shoulder range of motion during the patient's eventual neurological recovery at 1-year follow-up.

DISCUSSION

PTS is a rare upper extremity neuropathy.¹ The incidence is roughly 1.64 per 100 000 individuals.² It classically presents as localised, severe upper extremity pain corresponding with one or more nerves of the brachial plexus. After several weeks pain is replaced by weakness or paralysis. Electrodiagnostics are typically used to identify denervation. Treatment goals include optimisation of upper extremity motion and strength via physical therapy.³ Corticosteroids are often prescribed to reduce pain but have not been shown to alter the disease course.⁴ Severity and durations of initial pain, neurological deficits and recovery are variable.⁴⁻⁷

The aetiology of PTS is unclear but an inflammatory-immune mechanism is suspected because PTS has been associated with antecedent conditions such as infection, immunisation, operation or trauma.⁸⁻⁹ Moreover, quantities of

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complement-fixing antibodies to peripheral nerve myelin decrease with clinical improvement. Additionally, blood lymphocytes sensitised to brachial plexus nerves in patients diagnosed with PTS have been discovered.^{8–10} Still, haematological inflammatory and immunological markers are generally unremarkable. Both hereditary and idiopathic forms have been described.^{7 11} A mutation of the *SEPT9* gene on chromosome 17q25.3 is associated with the hereditary form, although the mechanism with which this gene mutation causes symptoms is unclear.¹²

Symptoms can occur unilaterally or bilaterally. Pain and subsequent weakness is localised to the distribution of affected peripheral nerves, thus clinical exam should reflect an absence of nerve root involvement. Neck motion should not substantially exacerbate symptoms as much as movement of the affected shoulder or extremity.⁴ In concert with lower motor neuron damage, patients develop diminished deep tendon reflexes and electrodiagnostics demonstrate fibrillation potentials and positive waves. Note that pain is not always restricted to the shoulder girdle and sparse reports have described similar neuropathies of the anterior interosseous nerve, phrenic nerve, lumbosacral plexus or recurrent laryngeal nerve.^{1 11 13}

Common differential diagnoses include rotator cuff pathology, peripheral-nerve compression (including suprascapular neuropathy or damage after arthroscopic surgery), cervical radiculopathy and thoracic outlet syndrome.^{4 14} PTS is usually distinguishable due to a sudden onset of severe pain without preceding trauma or cervical spine involvement.

The presented case study is most concerning because rotator cuff pathology was identified and addressed prior to the presentation of severe neuropathic pain. Given the typical findings of rotator cuff pathology (to include a suggestive MRI), a low clinical suspicion for PTS resulted in arthroscopic shoulder surgery. Yet, the patient's initial complaints did not correlate with the degree of pathology observed during arthroscopy, making us question if this patient's initial complaints were atypical for PTS. It is more likely that the patient in this report either had concomitant disease or developed PTS after surgery. Regardless, surgery led to suboptimal conditions for therapeutic maintenance of shoulder range of motion during the patient's neurological recovery. Van Alfen *et al* noted that out of 246 diagnosed patients, 18.7% experienced pain intermittently for a few weeks before isolating the characteristic continuous severe pain (which they called the 'real attack').^{5 11} Furthermore, pain severity increased in 38.5% of individuals in their study. Martinez-Salio *et al* reported that up to 15% of patients may not report the typical immediate severe pain at the time of diagnosis.¹⁵ Nonetheless, the majority of teaching regarding PTS (especially within the orthopaedic literature) does not warn orthopaedic surgeons about the potential for this atypical onset of pain. In our reported case, it is certainly possible that pain was initially attributed to the rotator cuff tear and surgery preceded the 'real attack' referenced by Van Alfen *et al* and Martinez-Salio *et al*.

Standard limitations of electrodiagnostics and MRI highlight another notable issue with regards to including PTS in the differential among common musculoskeletal shoulder pathology. There is no objective test available to rule out PTS during its acute phase (within 2 to 4 weeks of symptomatic onset). Neither MRI of the brachial plexus nor electrodiagnostics dependably reveal the denervation of affected muscles until after 2 to 4 weeks of symptoms.^{16 17} Initial diagnostic tests, however, are often obtained within this time period to rule out anatomical abnormalities that mimic PTS such as rotator cuff tears and cervical aetiologies. Electromyography is considered most useful after the first 2–4 weeks with diagnostically compatible findings

reported in 96.3% of cases.¹¹ Yet, patients at that point in their disease process may continue to attribute increasing or uncharacteristic pain to their known rotator cuff injury.

It is also worth noting that a delayed or repeated shoulder MRI is more likely to identify potential neuropathic sources of shoulder pain. Due to modern routine fat suppression, T2-weighted MRI images following the acute phase of PTS (at least greater than 4 weeks) may show increased signal intensity in affected muscles suggestive of denervation and oedema.¹⁶ Late in the chronic phase (approximately 1 year after presentation) T1-weighted images may detect fatty infiltration of atrophic muscles.^{16 18} Finally, MRI conducted after reinnervation may reveal little to no abnormalities.¹⁹

Learning points

- This case serves as an important reminder that common shoulder pathology such as rotator cuff tears can mimic Parsonage-Turner syndrome, which is a relatively uncommon diagnosis in the typical orthopaedic outpatient clinic.
- An atypical onset of pain prior to clinically relevant neurological deficits is possible. In this setting, the presence of surgical pathology can lead to diagnostic frustrations and untimely interventions.
- Alternatively, surgery for existing pathology may precede a typical attack of PTS (and impending paralysis) and severely hinder rehabilitation efforts. Postoperative pain should not be confused with the presence of new pathology such as the neuropathic pain associated with PTS.
- Diagnostic tests for PTS are not accurate within the first several weeks of symptomatic onset. In instances where clinical suspicion exists, delaying surgery or repeating diagnostic tests at least 4 weeks after the onset of symptoms is recommended.

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