

Parsonage-Turner Syndrome Masquerading as Shoulder Septic Arthritis: A Curious Case of the Subluxed Shoulder - A Diagnostic Dilemma

Naveen Mathai¹, SashiAier¹, Abhijith S¹, Kiran Acharya¹, Vivek Pandey¹

Abstract

Idiopathic brachial neuritis, also known as Parsonage-Turner syndrome, is a disorder of unknown etiology, with asymmetric involvement of the brachial plexus [1, 2]. It occurs in all age groups but is more common between the third and seventh decade. Men are affected more often than women. Inciting events occurring days before the onset has been reported in 28–83% of the cases in various series [3, 4]. Upper respiratory infection, flu-like illness, immunization, surgery, and emotional stress have been the common triggers. Some incidents occur without a triggering factor. We report a rare case of a middle-aged female with brachial neuritis which masqueraded as septic arthritis of the shoulder.

Keywords: Brachial Neuritis, Parsonage Turner syndrome, Subluxed shoulder

Introduction

Amyotrophic neuralgia of scapular and shoulder region or Parsonage-Turner syndrome (PTS) or idiopathic brachial neuritis was first described by Dreschefeld in two sisters, leading to pain and weakness in the shoulder girdle. However, it was described in detail by Parsonage and Aldren Turner in 136 men in the army in 1948 [1]. There is a rapid onset of pain in shoulder and arm within few days to weeks followed by wasting and weakness of affected areas. Although etiology of PTS remains idiopathic, it is often preceded by immunization, viral fever, or abnormality of the immune system [2,3]. Most of the patients describe additional pain phases in their course. Many patients reported a subsequent persisting musculoskeletal

type pain and associated pathology around the glenohumeral joint.

Case Report

A 48-year-old female presented to us with an acute onset of pain over the right shoulder for 6 days aggravated for a day. She rated her pain as eight on the visual analog scale (VAS) of 0–10. She had a history of fever 3 days before the onset of pain for which she had taken antipyretic prescribed elsewhere. Since 3 days, she was unable to move her right shoulder. There was no history of trauma to the upper limb or any other joint involvement. On examination, the contour of her shoulder was normal. There was a local rise in temperature and shoulder joint was diffusely tender. Both active and passive ranges of movements were very painful and restricted. There was no sensory loss, but the shoulder abduction and elbow flexion were quite weak. There was no weakness in the hand. However, an objective motor power examination around shoulder was not possible due to severe pain. A day before presentation to us, she has undergone magnetic

resonance imaging (MRI) of her shoulder elsewhere which reported an intrasubstance partial supraspinatus tear, moderate effusion in subdeltoid-subacromial effusion, synovial thickening, and mild humeral head subluxation (Fig. 1). Blood investigations revealed a total leukocyte count of $7.8 \times 10^3/\mu\text{L}$. The differential count revealed 71.8% of neutrophil and 19.6% of lymphocytes. Among inflammatory markers, C-reactive protein was 235 mg/dl, and erythrocyte sedimentation rate was 55 mm/hr. Muscle enzymes were also normal. Plain radiograph of shoulder revealed further subluxed glenohumeral joint as compared to MRI (Fig. 2). With severe pain and a history of fever before onset of shoulder pain, very high inflammatory markers, joint effusion, and synovial thickening on MRI, a clinical diagnosis of septic arthritis of the right shoulder was proposed. Blood cultures were sent. Given a provisional diagnosis of septic arthritis, the patient underwent an arthroscopic debridement of the right shoulder joint. However, intraoperatively, there was

¹Department Of Orthopaedics, Kasturba Medical College, MAHE, Manipal, Karnataka

Address of correspondence

Dr. Vivek Pandey,
Associate Professor, Department Of Orthopaedics
Kasturba Medical College, MAHE, Manipal,
Karnataka India- 576104
Email: vivekortho@gmail.com

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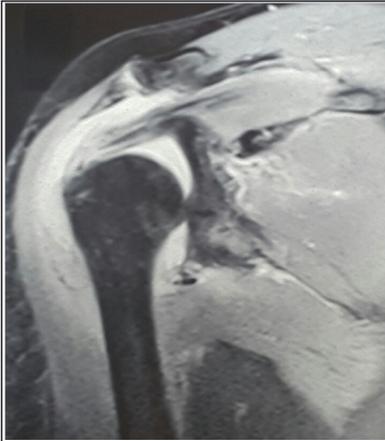


Figure 1: Magnetic resonance imaging of the right shoulder shows effusion in joint and subacromial space with bulky supraspinatus tendon.



Figure 2: Plain X-ray of shoulder joint shows subluxed joint.

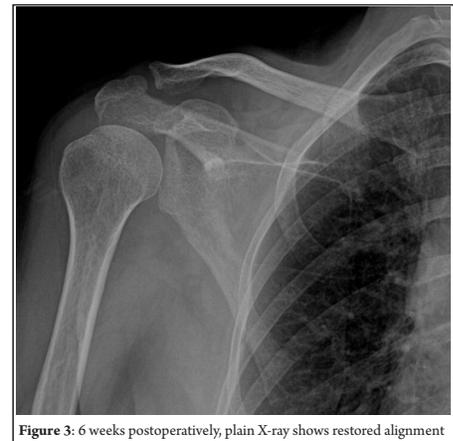


Figure 3: 6 weeks postoperatively, plain X-ray shows restored alignment of the shoulder joint.

minimal fluid in the joint cavity noted with minimal synovial hypertrophy. The fluid obtained was sent for Gram stain and cultures. There was minimal effusion in subacromial space too. Soft tissue subacromial decompression was performed as well. Postoperatively, she was administered intravenous broad-spectrum antibiotic covering gram-positive and negative bacteria (amoxicillin-clavulanic acid and ceftriaxone) as per the opinion of the infectious disease specialist. Despite the removal of the joint and subacromial fluid during arthroscopy, immediate post-operative radiograph revealed a persistent subluxed glenohumeral joint. Further, she had persistent weakness of elbow flexion and shoulder abduction despite a reduction in pain. This raised suspicion of neurological disorder, and hence, neurology consultation was sought. Nerve conduction study revealed significantly reduced compound motor action potential (CMAP) over the axillary and musculocutaneous nerve and mild reduction in CMAP over the suprascapular nerve. The neurologist suggested a provisional diagnosis of PTS and advised oral pregabalin. Since PTS may have an autoimmune origin, levels of rheumatoid and other factors were sought. Entire immune workup was negative except weakly positive anti-telomere CENP antibody.

However, rheumatologist did not consider it to be significant, and treatment mentioned above was continued. Further, gram stain was negative and local tissue, and blood cultures were negative after 72 h of incubation. Intravenous antibiotics were stopped after 7 days, and the patient was given oral antibiotics (amoxicillin-clavulanic acid) for another 4 weeks as we remained in a dilemma about septic arthritis with negative cultures. The patient was started on gentle shoulder rehabilitation which was followed by improvement in pain and movement. Biopsy of local tissue revealed synovitis. The patient was discharged on the 10th day with the advice of medication and physiotherapy. At 1st follow-up after a month, the patient was much better regarding pain (VAS -3) and movement. Active abduction and flexion of the shoulder improved to 0–70°. However, paresis of the deltoid muscle and elbow flexors persisted. The patient was asked to continue physiotherapy and pregabalin for another 2 months. However, the patient did not return for further follow-up. The patient was contacted over the telephone at the end of a year for a review. She reported having continued shoulder rehabilitative exercises at a nearby physiotherapy center. She reported over the phone that she has no pain and has regained full movement and return of strength. We

calculated the subjective questionnaire of Oxford shoulder score and QuickDASH score. The Oxford shoulder scoring was 46 which was a satisfactory and the QuickDASH score was 2.3 which suggested minimal disability and good outcome. Overhead activities were possible for the patient without any discomfort. Carrying heavyweights were also possible.

Discussion

This patient was admitted with a provisional diagnosis of septic arthritis of the shoulder given acute local signs and raised inflammatory markers and subluxed shoulder joint. Radiologically, glenohumeral subluxation in septic arthritis of the shoulder is a well-established fact in literature [4,5]. The probable mechanism may be incompetency of glenohumeral ligaments due to the gradual expansion of joint capsule, as pus accumulates slowly. However, Gram stain and culture and sensitivity revealed absent growth of any organism, and hence, septic arthritis was theoretically ruled out. Further, contrary to the finding of massive intra-articular effusion in septic arthritis, we found little fluid intraoperatively which disputed our provisional diagnosis. Another condition which may present like this is inflammatory myositis. However, muscle enzymes were normal. However, despite having lesser fluid in the joint

and subacromial space during arthroscopy, the post-operative radiograph revealed persistent subluxation of humeral head which drew our attention to some neurological disorder. We suspected underlying brachial neuritis or PTS which is possible after a viral prodrome [2,6]. It is also seen in other immunological conditions such as vaccination, pregnancy, childbirth, or viral infection [7]. PTS could also be of autoimmune origin [8]. Our patient had given a history of fever before the onset of shoulder pain which could lead to PTS. PTS has a varied presentation and can present in orthopaedics, neurology, or general physician outpatient clinic. Majority of them present as sudden onset shooting pain in the upper back which are usually unilateral as seen in our patient. Van Alfen and Van Engelen in their total cases of 246 patients reported a mean duration of severe pain at around 4 weeks [6]. Around 4.9% and 22.7% had a resolution of symptoms in 48 h and 1–7 days, respectively. It is reported that within the 1st week of symptoms (within 24 h in 33% of patients), insidious paresis of the affected nerves branching from the brachial plexus begins. Usually, the upper trunk of brachial plexus is involved, leading to weakness in supraspinatus, infraspinatus, serratus anterior, deltoid, biceps, and triceps [3,9]. However, the pattern of involvement is patchy and asymmetrical. Sensory disturbances are usually seen which are as high as 78.4%. In our patient, there was involvement of deltoid, supraspinatus, and biceps with normal sensations. The gross weakness of shoulder girdle muscle could lead to shoulder subluxation, and that can confuse with rare, but the similar presentation in septic arthritis wherein shoulder may be subluxated with severe pain and effusion [4,5]. An equally confusing and difficult diagnosis would be a rotator cuff injury which may be an

incidental finding in an MRI of a patient with acute severe shoulder pain. In our patient, even though an MRI suggested a supraspinatus partial tear, the absence of any trauma and the presence of glenohumeral subluxation ruled out a rotator cuff pathology contributing such a picture. Another condition, calcific tendinitis can cause severe acute pain in the shoulder. However, there is no paralysis of surrounding muscles, and shoulder is never subluxated. Moreover, X-ray and MRI would confirm the diagnosis of the latter. With a high index of suspicion, the diagnosis of PTS is established with the help of thorough clinical evaluation, nerve conduction velocity (NCV), and electromyography. NCV confirms the “typical neurogenic patchy involvement of the brachial plexus.” Further, detection of autoantibodies (anti-myelin, antiganglioside, or anti-axon) may help in establishing the autoimmune etiology proposed by several authors [10,11,12]. The presence of serum IgM may indicate recent viral infection. Our patient was investigated for the same and reported the following antibody (CENP-B) to be weakly positive. However, rheumatologist concluded this to be of no significance and ruled out systemic lupus erythematosus or other autoimmune disorders in this patient. MRI may detect signal changes in brachial plexus. The treatment of PTS is essentially conservative which comprises medical and rehabilitative measures. The medical management of PTS requires analgesics and long-acting opioids to reduce pain and pregabalin or amitriptyline to reduce neuralgic pain [13,14]. The addition of steroid in early phase may be beneficial in quick recovery [6,15,16,17,18]. Immunoglobulin therapy is recommended if specific autoantibodies are strongly positive [19,20]. Long-term physiotherapy is required to avoid stiffness and return of function. Contrary to all these studies, we did not

start steroid administration in this patient due to the presence of raised inflammatory markers and suspicion of septic arthritis. Nevertheless, she reported early neurological recovery at the end of 6 weeks progressing to full recovery at the end of 1 year. The PTS is a self-limiting disorder. However, 50% of patient may complain of residual pain and weakness for several years. Host et al. reviewed 58 pediatric cases of PTS and concluded that 63% made a full recovery, 25% made a partial, and 13% made no recovery [21]. The natural history of PTS is one of resolution over time, with initial neuropathic pain and paresis followed by the gradual return of muscle strength and function. Historically, the condition has been thought to completely resolve with rare instances of recurrence and residual symptoms [17,22]. The studies reported complete recovery of 89% of patients in 3 years. However, the onset of neurological recovery and the period of recovery vary. Until recent times, it was assumed that PTS recovers uneventfully. However, recently published studies report otherwise [6,23]. A study by Cup et al. suggested that there was a high incidence of residual pain and occasional paresis, which was more than 50% on follow-up after 6–24 months. A similar proportion still had impaired shoulder movements, with more than 80% reporting difficulty in overhead activities, and two-thirds reporting decreased hand strength in the affected extremity [23]. Another author reported that one-third of patients in a series of 89 patients reported significant long-term pain and fatigue and half to two-thirds experienced impairments in daily life [24]. Hence, persistent pain, both neuropathic and musculoskeletal, severe fatigue, and impairments of activities of daily living and are presented in a large proportion of these patients. Residual symptoms are strongly correlated with the altered biomechanics of the

shoulder girdle and the altered movement pattern rather than the neurological insult.

Conclusion

The relevance of this case is in the

puzzling nature of the presentation which posed a diagnostic challenge. PTS generates a diagnostic dilemma in clinicians as varied clinical scenarios masquerade as brachial neuritis which includes cervical radiculopathy, rotator

cuff injury, acute calcific tendonitis, mononeuritis multiplex, and tumors of the brachial plexus. The clinician has to be adept at the differentials in such cases.

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