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Case Report

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A Case with Behçet's Disease Involving Erosive Glenohumeral Joint Arthritis: The Value of Ultrasonography in the Management of Shoulder Pain and Diagnosis of an Erosion

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ABSTRACT

Background: Behçet's disease (BD) is a chronic inflammatory vasculitis with multisystemic involvement. Musculoskeletal manifestations are common but usually present as a non- erosive arthritis affecting peripheral joints. Shoulder arthritis, particularly with erosive changes, is extremely rare in BD and remains poorly documented.

Objectives: This case highlights the importance of considering shoulder involvement in BD, especially when pain persists despite conventional treatments. The presence of glenohumeral joint erosion is particularly unusual in BD, raising diagnostic and therapeutic challenges. US played a pivotal role in detecting early inflammatory changes and guiding the intervention, underlining its value in managing inflammatory shoulder pain.

Methods: We report the case of a 59-year-old woman with a 23-year history of BD, predominantly affecting her skin and mucous membranes, who presented with persistent right shoulder pain with limited range of motion and positive impingement signs.

Results: Standard radiographs were normal, but ultrasonography (US) demonstrated synovitis, inflammatory tenosynovitis, a thickness tendon tear, and an erosion of the humeral head. The patient underwent an ultrasound-guided corticosteroid injection, leading to significant pain relief and functional improvement, followed by a rehabilitation program.

Keywords: Chronic Inflammatory Rheumatism, Shoulder Pain, Behçet's Disease, Ultrasonography, Erosion

Introduction

Behçet's disease (BD) is a chronic, multisystemic inflammatory disorder characterized by recurrent oral and genital ulcers, uveitis, and skin lesions. It is classified as a vasculitis that primarily affects small and large vessels, leading to a wide range of clinical manifestations involving the skin, mucous membranes, eyes, gastrointestinal tract, central nervous system, and musculoskeletal system [1]. While musculoskeletal involvement is observed in BD cases, it typically presents as a non-erosive polyarthritis affecting large joints such as the knees, ankles, wrists, and elbows. In contrast to rheumatoid arthritis,

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BD-related arthritis usually lacks joint erosion, and destructive changes are rare [2-6].

Shoulder arthritis in BD is particularly uncommon, and cases with documented erosive changes remain exceedingly rare in the literature. The glenohumeral joint is infrequently involved in BD, and when affected, the symptoms are often attributed to soft tissue inflammation rather than direct joint damage. The presence of joint erosions raises questions about the underlying inflammatory mechanisms, disease progression, and potential overlap with other inflammatory arthropathies [7-10].

Imaging plays a crucial role in the evaluation of inflammatory joint diseases. While conventional radiography is often the first-line imaging modality, it may fail to detect early structural changes, particularly in cases of mild synovitis or early erosive disease [11,12].

Ultrasonography (US) has emerged as a valuable and widely accessible tool for assessing shoulder pain, offering high-resolution imaging of the joint structures, including the synovium, tendons, and cartilage. Doppler US, in particular, is highly sensitive for detecting active synovitis, inflammatory tenosynovitis, and early bone erosions, making it a key modality in the evaluation of inflammatory arthropathies [13-15]. Additionally, ultrasound-guided interventions enhance the accuracy and efficacy of corticosteroid injections, providing targeted relief for patients with inflammatory shoulder pain [16-18].

Here, we present a unique case of a patient with long-standing Behçet's disease who developed erosive glenohumeral joint arthritis, a rare manifestation of the disease. We emphasize the critical role of ultrasonography in diagnosing shoulder involvement, detecting early erosive changes, and guiding therapeutic interventions. This case highlights the importance of integrating musculoskeletal ultrasound into the routine assessment of shoulder pain in inflammatory diseases, particularly in cases where standard radiographs appear normal.

Case Presentation

A 59-year-old woman with a 23-year history of Behçet's disease, predominantly affecting her skin and mucous membranes, was referred to the Physical Medicine and Pain Clinic due to persistent right shoulder pain of mixed mechanical and inflammatory origin. The pain had been ongoing for two years, resistant to NSAIDs and various analgesics, and had no traumatic origin.

Her medical history includes a non-operated thyroid nodule under surveillance for 12 months, with no history of diabetes or hypertension. Clinical examination of the right shoulder revealed tenderness over the long head of the biceps and supraspinatus tendons, with a VAS pain score of 7/10. Mobility was significantly limited, with forward flexion to 120°, external rotation to 25°, and internal rotation to 30°. Positive clinical tests included the Palm-up test, Job test, and signs of subacromial impingement (Neer's and Yocum's tests). Standard radiographs were normal. Laboratory findings showed an elevated erythrocyte sedimentation rate (40 mm/h), elevated C-reactive protein (10 mg/L), anemia, and thrombocytosis, with no leukocytosis.

Ultrasound imaging of the right shoulder revealed synovial effusion and thickening of the long head of the bicep's tendon sheath with hyperemia on Doppler imaging, consistent with tenosynovitis (Figure 1).

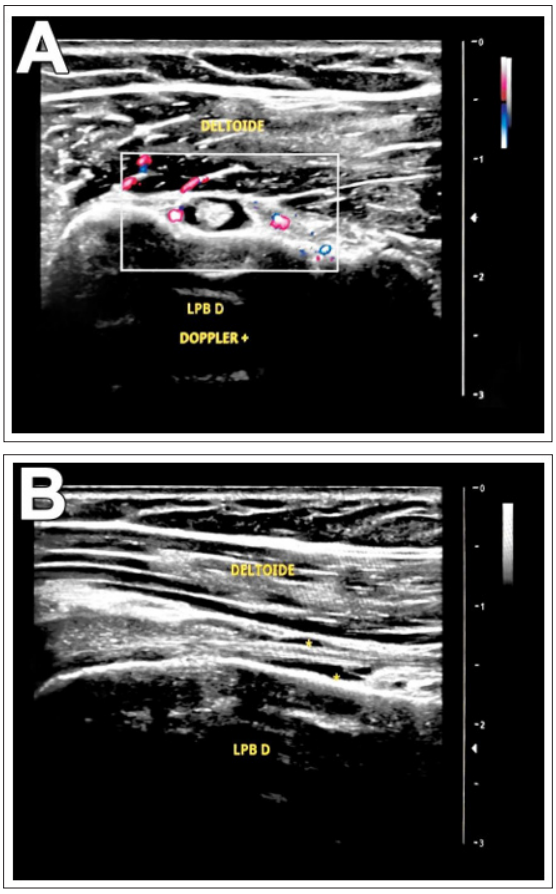
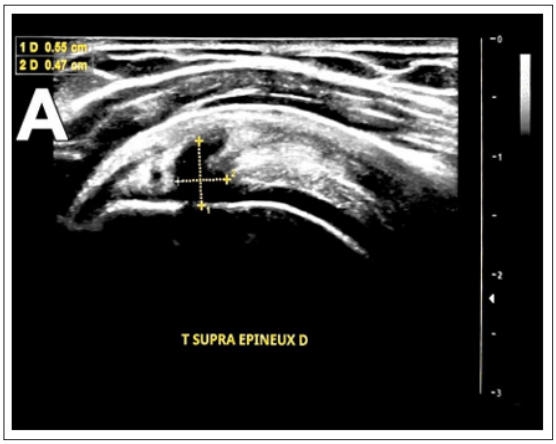


Figure 1: Inflammatory Tenosynovitis of the Right Long Head of the Biceps Tendon.

Axial (A) and longitudinal (B) ultrasound views of the long head of the bicep's tendon. The ultrasound reveals an effusion with synovial thickening of the tendon sheath of the long head of the biceps, along with marked hyperemia on power Doppler (+++).

There was also a partial-thickness tear of the right supraspinatus tendon measuring 5.5 x 4.7 x 4.2 mm, along with a type 1 calcification measuring 3.7 mm in axial section (Figure 2).



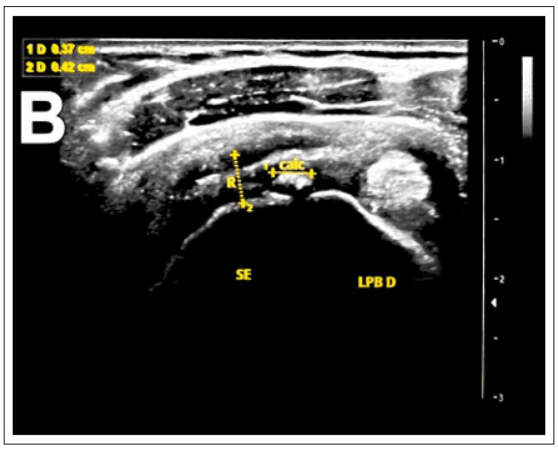


Figure 2: Partial Thickness Tear of the Deep Surface of the Right Supraspinatus Tendon.

Longitudinal (A) and axial (B) ultrasound views of the supraspinatus tendon. The ultrasound shows a focal loss of the fibrillar pattern of the tendon, replaced by a hypoechoic area measuring 5.5 x 4.7 x 4.2 mm, retraction of the ruptured deep fibers, with preservation of the superficial tendon fibers. The ultrasound also reveals a type I enthesis calcification measuring 3.7 mm with a pure posterior acoustic shadow.

The ultrasound examination also revealed a calcifying enthesopathy of the right infraspinatus tendon, characterized by a tendon insertion calcification associated with a cortical erosion of the humeral head measuring 2.5 mm in depth (Figure 3).

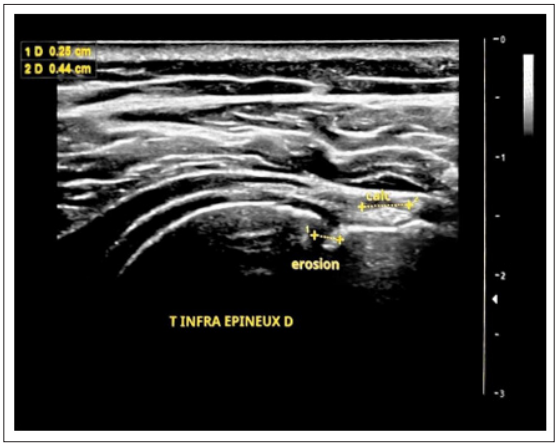


Figure 3: Erosion of the Cortical Bone on the Right Humeral Head. Calcific Enthesopathy of the Right Infraspinatus Tendon.

Longitudinal scan of the right infraspinatus tendon showing a hyperechoic calcification type 3 at the enthesis, without a posterior acoustic shadow measuring 4.4 mm, as well as an erosion of the cortical bone on the right humeral head measuring 2.5 mm.

This finding is particularly unusual in Behçet's disease, where joint involvement is typically non-erosive. An ultrasound-guided corticosteroid injection of 80 mg methylprednisolone acetate (MPA) was administered into the bicipital recess and performed under careful imaging guidance. The procedure was done with the patient in a lateral decubitus position, arm along the body in external rotation, and hand supinated. The puncture is performed with a 50 mm long, 21- gauge needle. The axial ultrasound scan with a superficial linear probe of 8-16 MHz (Samsung HS 40 model 2019) visualizes the tendon of the long head of the biceps in its sheath as well as the bicipital artery located outside, and the bicipital recess is approached in-plane laterally by transfixing the anterior deltoid. The Doppler allowed us to avoid puncturing the bicipital artery (Figure 4).

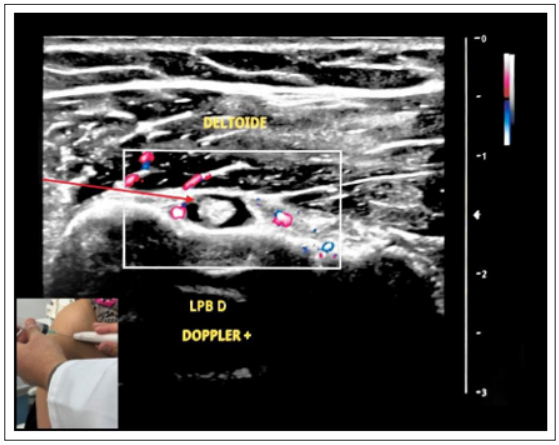


Figure 4: The Right Bicipital Recess Injection Technique.

Axial ultrasound shows the long biceps tendon in its sheath as well as the bicipital artery located outside, we approach the bicipital recess in the needle direction (in-plane) laterally by transfixing the anterior deltoid.

A complementary ultrasound imaging of the left shoulder revealed an asymptomatic calcifying tendinopathy of the supraspinatus tendon (Figure 5) and calcifying enthesopathy of the subscapularis tendon (Figure 6).

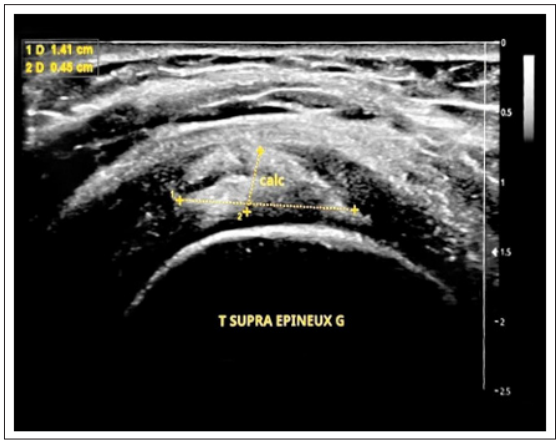


Figure 5: Calcific Tendinopathy of the Left Supraspinatus Tendon.

Longitudinal scan of the left supraspinatus tendon showing a cluster of hyperechoic calcifications type 3 without a posterior acoustic shadow, measuring 4.5 mm x 14.1 mm.

Longitudinal scan (A) of the right subscapular tendon showing no abnormalities. Longitudinal scan (B) of the left subscapular tendon showing hypoechoic remodeling and a fine, arcuate hyperechoic calcification type 3 at the enthesis, without a posterior acoustic shadow, measuring 6.9 mm.

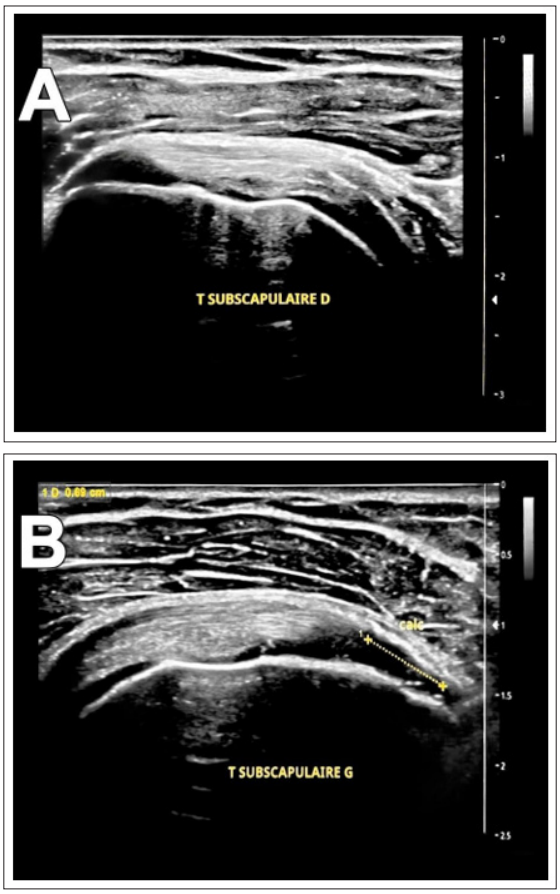


Figure 6: Calcific Enthesopathy of the Left Subscapular Tendon.

One month later, the patient reported a reduction in pain to 2/10, with improved range of motion (forward flexion to 160°, external rotation to 30°, and internal rotation to 40°). Rehabilitation involving 15 physiotherapy sessions, focusing on pain relief, range of motion recovery, and rotator cuff strengthening, was initiated, resulting in further improvement.

Discussion

Behçet's disease (BD) is a chronic, multisystemic inflammatory disorder characterized by recurrent oral and genital ulcers, uveitis, and skin lesions, with variable involvement of the musculoskeletal system. While joint manifestations are common in BD, they usually present as a non-erosive polyarthritis, affecting large joints such as the knees, ankles, wrists, and elbows [1-4]. Joint erosions, however, are exceedingly rare and are generally not associated with BD, distinguishing it from diseases such as rheumatoid arthritis, where erosions are a hallmark feature. The case presented here highlights a unique occurrence of erosive glenohumeral joint arthritis in a patient with long-standing BD, a finding that deviates from the typical non-erosive nature of joint involvement in this disease.

Erosive changes in the glenohumeral joint region are not commonly observed in the literature. Most cases of shoulder arthritis in BD are thought to be the result of soft tissue inflammation rather than direct joint damage. The presence of joint erosions, as seen in our patient, raises important questions regarding the inflammatory mechanisms at play in BD and whether these changes may be indicative of disease progression or an overlap with other inflammatory arthropathies.

Imaging plays a critical role in diagnosing and monitoring joint involvement in inflammatory diseases. Traditional radiography is often the first imaging modality used in clinical practice, but it is limited in detecting early joint changes, particularly in cases where synovitis is mild or where early erosive changes are present. This limitation was evident in our patient, as the initial radiographs did not show any abnormalities, despite the clinical suspicion of joint involvement. This underscores the need for more sensitive imaging techniques to assess joint integrity in the early stages of disease.

In this context, ultrasonography (US) emerges as a highly valuable tool in the evaluation of inflammatory joint diseases [19]. Musculoskeletal ultrasound provides high-resolution images of joint structures, including the synovium, tendons, and cartilage, making it particularly useful in detecting early signs of inflammation and damage. The ability to assess both soft tissue and bone structures in real-time allows for a comprehensive evaluation of joint health [13-15]. In our case, ultrasound revealed several important findings, including synovial effusion and thickening of the long head of the bicep's tendon sheath with marked hyperemia on Doppler imaging, which is characteristic of tenosynovitis. These findings provided clear evidence of active inflammation that was not visible on radiographs.

Moreover, ultrasound also identified a partial-thickness tear of the right supraspinatus tendon and a calcifying enthesopathy of the right infraspinatus tendon, with associated cortical erosion of the right humeral head. The erosion, measuring 2.5 mm, is particularly noteworthy as it is an unusual finding in BD, where joint involvement generally does not include erosions. This calcifying enthesopathy, coupled with the cortical erosion, suggests that there may be more aggressive inflammatory activity occurring in this patient's shoulder, possibly related to prolonged disease activity or an exacerbation of the underlying inflammatory process. The presence of tendon tears and calcifications may also suggest a mechanical component to the patient's symptoms, complicating the clinical picture.

Ultrasound-guided interventions, such as corticosteroid injections, have been shown to be effective in the management of shoulder pain in inflammatory conditions [16-20].

In our case, an 80 mg methylprednisolone acetate injection was administered into the bicipital recess under ultrasound guidance. This approach allowed for precise delivery of the corticosteroid, ensuring that it was injected directly into the site of inflammation, thus maximizing the therapeutic effect while minimizing the risk of complications. The use of ultrasound guidance also provides the advantage of avoiding adjacent structures, such as the bicipital artery, further enhancing the safety and accuracy of the procedure.

One month after the injection, the patient reported significant improvement in both pain and range of motion, with forward flexion increasing from 120° to 160°, external rotation from 25° to 30°, and internal rotation from 30° to 40°. This improvement highlights the effectiveness of ultrasound-guided corticosteroid injections in managing inflammatory shoulder pain, especially in the context of complex conditions like BD, where conventional treatments may not always provide adequate relief [1-18, 20-22].

The intact appearance of the right subscapularis tendon on ultrasound further emphasizes the value of musculoskeletal imaging in differentiating between various tendon pathologies. By providing a clear assessment of tendon integrity, ultrasound can help guide management decisions and prevent unnecessary interventions.

In summary, this case underscores the importance of integrating advanced imaging techniques, such as musculoskeletal ultrasound, into the routine clinical evaluation of patients with inflammatory joint diseases like Behçet's disease. Ultrasound not only aids in the early detection of erosive changes and inflammatory processes but also facilitates targeted therapeutic interventions, improving patient outcomes and reducing the need for more invasive procedures [19,20]. The findings in this case suggest that BD can, in rare instances, present with erosive joint damage, and that the role of musculoskeletal ultrasound in diagnosing and managing such cases is crucial. This case also raises the question of whether aggressive disease activity in BD may predispose patients to more severe musculoskeletal involvement, including erosive changes in the joints [7-10].

Conclusion

This case illustrates an exceptional manifestation of joint involvement in Behçet's disease, characterized by erosive glenohumeral arthritis, whereas the classic joint involvement is usually non-erosive. The use of musculoskeletal ultrasound not only allowed early detection of inflammatory signs and erosions but also precisely guided the therapeutic procedure, resulting in significant clinical improvement. This observation emphasizes the importance of integrating ultrasound in the evaluation of persistent joint pain in patients with Behçet's disease, especially when conventional imaging is normal.

Finally, it highlights the need for further studies to explore the mechanisms underlying these rare erosive presentations and to better understand how they may influence the long-term prognosis and management of patients with Behçet's disease.

Conflicts of Interest/Funding/Acknowledgements: None

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