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ABSTRACT

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Case Report: We present such a case where acute small joint monoarthritis was the presenting symptom in a young male. Detailed history and evaluation led us to diagnose it as the initial peripheral articular manifestation of ankylosing spondylitis.
Conclusion: The role of a detailed history keeping in mind the variety of disorders, both autoimmune and non-autoimmune which can present as such cannot be over emphasized. We reviewed the literature to find that such a manifestation has not been described before. This case challenges the established pattern of presentation of a relatively 'known' entity.

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Keywords: Acute monoarthritis, Ankylosing spondylitis, Psoriatic arthritis, Small joint monoarthritis

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INTRODUCTION

Monoarthritis is a diagnostic challenge presenting not only to the physician or the orthopedician but also to the rheumatologist, as it can be the initial manifestation of many disorders, not all of them autoimmune. The acute onset of monoarthritis though substantially lessening the diagnostic possibilities requires a multi-disciplinary approach to its evaluation. Among the various causes, trauma, septic arthritis and acute crystal arthropathy occupy the place of prominence, while rarer causes include infections such as brucellosis, Lyme disease, leptospirosis and drugs. Besides, many chronic autoimmune disorders can present initially as an acute monoarthritis. However, most of these conditions are known to affect the larger joints predominantly with small joint involvement in a few cases as an additional feature.

Herein, we present a case where the patient presented with acute small joint monoarthritis in the absence of any peripheral large joint affection, and which turned out to be a peripheral articular manifestation of ankylosing spondylitis, a disease not known to present as such. To our knowledge, this manifestation has not been described earlier in literature.

CASE REPORT

A 25-year-old male was referred from the Orthopedics Department of our hospital, where he had presented two weeks before with sudden onset of pain and swelling of the proximal interphalangeal joint of the left middle

finger on waking up in the morning. He had no fever or other constitutional symptoms or history of trauma. He had been evaluated for septic arthritis but the joint fluid aspiration and examination did not reveal any pus. He was prescribed antibiotics empirically on the presumption of septic arthritis along with analgesics. Synovial fluid examination had revealed a straw colored fluid with cell count of 5000/μL with 50% polymorphs, 50% lymphocytes, protein 3.3 g/dL, and glucose 83 mg/dL. Gram staining and cultures of the synovial fluid for bacteria were negative. No crystals could be observed under polarizing microscopy. The blood investigation at the time of presentation revealed hemoglobin 14.4 g/dL (normal range 12.0–15.5 g/dL in male), total leucocyte count (TLC) 8800/μL (normal range 4000–11000/μL), polymorphs 84%, lymphocytes 15%, eosinophil 1%, erythrocyte sedimentation rate (ESR) 74 mm/1st hour by Westergren method (normal range <30 mm/1st hour in males), platelet counts $323x10^3$/μL (normal range $150x10^3$–$450x10^3$/μL), C-reactive protein (CRP) 32 mg/dL (normal range <1 mg/dL), serum urea 32 mg/dL (normal <50 mg/dL), serum creatinine 0.8 mg/dL (normal range <1.2 mg/dL), serum uric acid 4.0 mg/dL (normal range <7 mg/dL), serum aspartate aminotransferase (AST) 34 U/L (normal range <40 U/L), serum alanine aminotransferase (ALT) 27 U/L (normal range <40 U/L), serum alkaline phosphatase 168 U/L (normal <300 U/L). As the workup of septic and crystal causes were negative and routine investigations inconclusive, and there being no improvement in the patient's clinical status with antibiotics, he was referred to our rheumatology clinic for further evaluation.

Review with detailed history revealed that the patient had developed a sudden onset swelling in the left third PIP joint around two weeks back. Histories of trauma to the joint or of insect bite at the site were conspicuous by their absence. The swelling and pain continued to increase over a few days rendering him incapable of flexing the affected finger. There was no history of any discharge from the affected joint. He had no history of any prior gastrointestinal or urinary infection or any skin lesion. No one in the family had any similar problem. The patient, however, mentioned being treated for recurrent redness of the eyes which had been diagnosed by the ophthalmologist as acute anterior uveitis (AAU). He had three documented attacks of AAU starting from 2004 and was treated with local steroids. He had been having low back pain which he had ascribed to his prolonged sitting at his desk-job being a computer professional. However, his low back pain (LBP) was more in the morning, diminishing transiently only when he walked to his office but would recur and persist throughout the day as he performed very little activity. As this pattern was suggestive of an inflammatory origin, we decided to investigate in the light of seronegative spondyloarthritis.

Local examination revealed synovitis of the PIP joint of the left middle finger with flexion deformity (Figure 1). There were no nail signs suggestive of psoriasis. No psoriatic lesions were found in the hidden areas. Systemic examination was remarkably normal. Musculoskeletal examination revealed decreased range of movement of lumbar spine of 3 cm as evidenced by Schober's test.

The repeat blood workup showed raised ESR (54 mm/1st hour) with hemoglobin 14.2 g/dL, TLC 7800/μL, polymorphs 86%, lymphocytes 13%, eosinophils 1%, platelet counts $315x10^3$/μL, serum urea 40 mg/dL, serum creatinine 0.7 mg/dL, serum uric acid 3.9 mg/dL, AST 39 U/L , ALT 33 U/L, ALP 156 U/L. C-reactive protein was elevated 16 mg/L. Rheumatoid factor (RF), antinuclear antibody (ANA), hepatitis B surface antigen, antibody to hepatitis C (IgM), and human immunodeficiency virus (HIV) I & II antibodies were negative. Cultures of blood and urine as well as *Brucella* serology were negative. Urine routine and microscopic examinations were normal. The X-ray of pelvis showed bilateral sacroiliitis (Figure 2). The patient tested positive for HLAB27 by polymerase chain reaction (PCR) technique. The ophthalmologist reviewed the patient and noted changes of irregular pupillary margins of the right pupil, suggestive of past uveitis.

The patient was prescribed oral sulfasalazine in incremental dosage up to 2 g/day along with non-steroidal anti-inflammatory drugs (NSAIDs) (aceclofenac sustained release 200 mg daily) and kept under regular follow-up. The pain, swelling and the flexion deformity of the finger had decreased markedly by 16th week (Figure 3).

Thus with presence of bilateral radiologic sacroiliitis along with HLAB27 positivity and seronegative spondyloarthritis features of uveitis and inflammatory low back pain, the final diagnosis was kept as seronegative spondyloarthritis—ankylosing spondylitis.

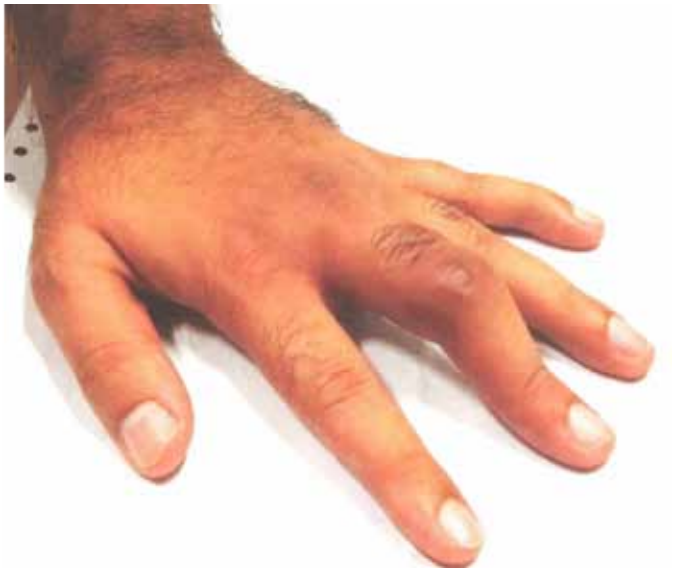

Figure 1: Small joint synovitis of the proximal interphalangeal joint of the middle finger of the left hand with flexion deformity of the joint.

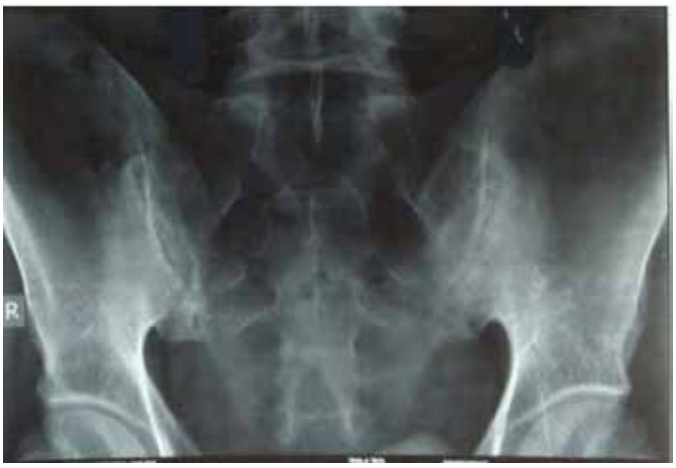

Figure 2: X-ray pelvis of the patient showing bilateral sacroilitis.

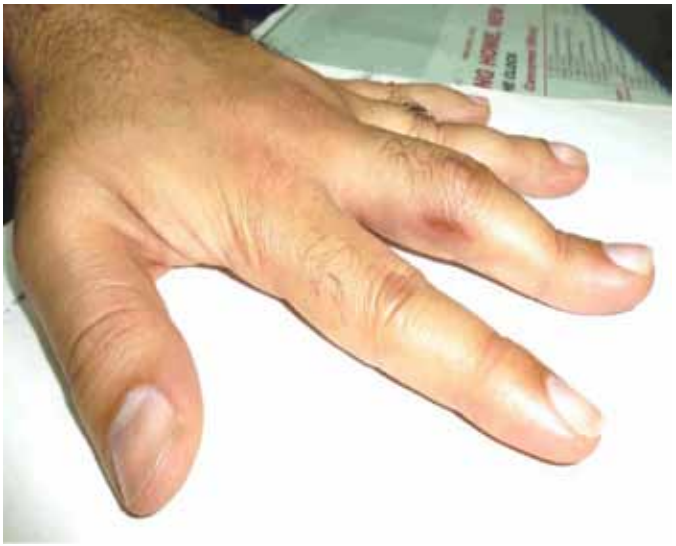

Figure 3: The same joint after four months of therapy with NSAIDs and sulfasalazine. Note the decrease in the degree of flexion deformity.

DISCUSSION

Monoarthritis of sudden onset is a significant diagnostic challenge for the rheumatologist. The importance of a detailed history and physical examination cannot be over-emphasized as diagnostic studies are mostly supportive [1]. The most common differentials of acute monoarthritis are trauma and infection. Infective causes can be directly causative like septic arthritis, Lyme arthritis, plant thorn synovitis, and infective endocarditis, or indirect such as brucellosis and leptospirosis which cause a non-specific reactive arthritis [2]. Other causes like gout, Milwaukee shoulder, rapidly destructive articular disease, amyloid arthropathy, hemophilic arthropathy, primary synovial osteochondromatosis, pigmented villonodular synovitis, neuropathic arthropathy, and foreign-body synovitis, loose bodies, and ischemic necrosis are also fairly common [3, 4]. These disorders have many common and overlapping clinical features. Septic arthritis because of associated morbidity and mortality is the most serious and should be dealt as an emergency.

Inflammatory monoarthritis of 'autoimmune' origin can be (a) common acute monoarthritis which include crystal arthritis and reactive arthritis, (b) acute monoarthritis of polyarthritic conditions like psoriatic arthritis, enteropathic arthritis, rheumatoid arthritis, palindromic rheumatism and juvenile idiopathic arthritis (JIA), and (c) acute monoarthritis of systemic disease such as lupus, vasculitis, Henoch-Schönlein purpura, Behçet's disease, and relapsing polychondritis [4].

Detailed history of the patient and examination of the synovial fluid ruled out trauma, infections, crystal arthritis, and other causes of non-septic arthritis enumerated above, thus narrowing the possibilities to either one of the inflammatory monoarthritis or mono-arthritic presentation of polyarticular disease. The age of the patient, history of inflammatory low back pain, documented recurrent uveitis, bilaterally symmetrical radiological sacroiliitis, raised CRP and positive HLAB27 confirmed the diagnosis to be spondyloarthritis.

The constellation of inflammatory low back ache (ILBP), HLAB27 positivity, uveitis and radiological sacroilitis, and peripheral joint involvement can be found in ankylosing spondylitis as well as psoriatic arthritis, both being subsets of seronegative spondyloarthritis. In contrast to ankylosing spondylitis, sacroilitis in psoriatic arthritis is usually unilateral or asymmetrical. Also, involvement of small joints in psoriatic arthritis is usually associated with nail changes in three-fourths of the cases [5], and can be seen in the polyarticular and the distal interphalangeal (DIP) involvement modes of presentation. However, small joint affection apart from wrists has not been described in ankylosing spondylitis [6, 7], although peripheral joint affection can be found in up to 30% of patients [8]. Besides the nail changes, psoriatic arthritis is also characterized by tenosynovitis and dactylitis. Patients with psoriatic arthritis usually have concurrent psoriasis in most cases, or its history either in the patient or in the first-degree relatives. Although 10–15% of cases may present with arthritis before onset of psoriasis, these patients will usually have the nail changes of psoriasis [7]. Hence nail changes are to be expected in psoriatic arthritis presenting as acute monoarthritis.

We faced the dilemma whether to ascribe the small joint affection to psoriatic arthritis subset or to ankylosing spondylitis which has not been described before. Our patient fulfilled both the 1984 modified New York criteria [9] for ankylosing spondylitis and the new ASAS [10] criteria for axial spondyloarthritis but not the CASPER criteria for psoriatic arthritis [11]. Hence, it is our contention that acute onset PIP monoarthritis in our patient is a component of ankylosing spondylitis; a pattern which has not been described earlier in literature.

CONCLUSION

We conclude that acute onset small joint monoarthritis can be a feature of ankylosing spondylitis, however rare. The importance of a detailed history cannot be overemphasized especially in eliciting the occurrence of seronegative spondyloarthritis features, which can present at different periods of time to different specialties, and hence are likely to be missed.

KEY MESSAGE

Apart from trauma, sepsis and crystals, the clinician must always keep in mind the other causes for which a detailed history is essential. Rarely, ankylosing spondylitis can also present as acute onset small joint monoarthritis.

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Author Contributions

Deepak Rath – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Bijit Kumar Kundu – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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