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Case Report Open Access

An atypical presentation of axial spondyloarthritis with severe acute low back pain: A case report

Leyla Bagheri[1], Maryam S.Fakhri B.[2,3], Fatemeh Rashidi[2], Shila Aghayani[4]

[1]Department of Internal Medicine, Shahid Modarres Hospital, Shahid Beheshti University of Medical Science, Tehran, Iran. [2]School of Medicine, Tehran University of Medical Science, Tehran, Iran. [3]Department of Internal Medicine, Imam Khomeini Hospital Complex, Tehran University of Medical Science, Tehran, Iran. [4]Department of Rheumatology, Imam Khomeini Hospital Complex, School of Medicine, Tehran University of Medical Science, Tehran, Iran.

Axial spondyloarthritis (axSpA) represents a chronic inflammatory condition primarily impacting axial joints, characterized by sacroiliitis and spondylitis. In axSpA patients, low back pain (LBP) typically assumes a chronic and inflammatory nature, where diagnostic delays can precipitate disability. However, atypical axSpA presentations pose challenges for early diagnosis and management. In the current study, we detail the case of a 39-year-old male presenting with acute, severe LBP and bilateral sacroiliitis. Initial treatment with non-steroidal anti-inflammatory drugs (NSAIDs), the standard first-line therapy, was discontinued due to severe gastrointestinal bleeding. Following a bone biopsy that excluded alternative etiologies, axSpA was confirmed based on diagnostic criteria. Elevated inflammatory markers, contraindications to NSAIDs, and magnetic resonance imaging findings prompted the initiation of anti-tumor necrosis factor (TNF) therapy. Remarkably, a marked improvement was observed in less than six weeks post anti-TNF therapy commencement. This case underscores the significance of recognizing atypical axSpA presentations and underscores the potential for swift and robust responses to anti-TNF agents. Optimal patient outcomes hinge upon effective disease pattern recognition and treatment selection.

Keywords: *ankylosing spondylitis; axial spondyloarthritis; acute low back pain; anti-TNF; adalimumab*

Introduction

Axial spondyloarthritis (axSpA) is a chronic inflammatory disorder primarily impacting the axial joints, characterized by sacroiliitis and spondylitis [1, 2]. The predominant manifestation of low back pain (LBP) in axSpA is typically chronic and inflammatory, significantly contributing to the disability experienced by affected individuals [3]. While subacute LBP is feasible, acute LBP is relatively uncommon in axSpA [3]. Acute LBP typically endures for less than 6 weeks, whereas subacute LBP persists for 6 to 12 weeks, and chronic LBP lasts beyond 12 weeks [4]. Hyperacute pain refers to an abbreviated period of intense discomfort lasting 24 to 48 hours, distinguished by symptoms that provoke immobilization and hindered motion due to severe pain and spasms [4].

Despite the characteristic spinal sclerosis known as "Bamboo Spine," individuals affected by axSpA demonstrate varying degrees of

*Corresponding author: Shila Aghayani, Assistant Professor of Rheumatology, Department of Internal Medicine, Department of Rheumatology, Imam khomeini hospital, Tehran University of Medical Sciences Tehran, Iran; Tel: +989183883084; E-mail: Shila.aghayani@yahoo.com

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osteopenia and osteoporosis, rendering fractures a conceivable etiology for acute low back pain. Consequently, a comprehensive investigation to exclude such fractures is paramount [5]. Radiographic assessments are warranted to delineate ankylosing spondylitis (AS)-related changes, as delineated in the New York Classification Criteria for AS and the more recent ASAS classification criteria introduced by the Assessment of Spondyloarthritis International Society [6]. These evaluations also aid in discerning alternative sources of non-specific LBP, necessitating consideration of differential diagnoses encompassing infection, neoplasms, osteoporosis, fractures, inflammatory processes, radicular syndromes, and cauda equina syndrome. We present a case of a 39-year-old male with axSpA who presented atypically with acute LBP and exhibited a rapid response to anti-tumor necrosis factor (TNF) agents.

Case Presentation

A 39-year-old male presented at the emergency department with a chief complaint of severe acute LBP, persisting for the past ten days. Remarkably, he could recall the precise time, location, and onset of the pain. Initially localized to the lower back, the pain extended towards the hip region. Initially unilateral, the pain became bilateral after approximately a week. The patient reported experiencing about an hour of morning stiffness and noted that physical activity exacerbated the pain to the extent that maintaining a standing position for routine activities, including walking, was challenging. Notably, there were no accompanying symptoms such as fever, diaphoresis, abdominal pain, or weight loss. The patient denied any significant past medical history, consumption of unpasteurized dairy products, recent travel, traumatic incidents, or smoking. Furthermore, his family history was negative for rheumatic disorders or malignancies. Regarding his medication history, he had intermittently used non-steroidal anti-inflammatory drugs (NSAIDs).

On physical examination, vertebral alignment appeared to be within normal limits, albeit with mild kyphosis noted due to a painless posture. Evidence of sacroiliac joint involvement was observed, supported by positive findings on tests including the Flexion, ABduction, External Rotation (FABER) test, compression test, distraction test, and Gaenslen test. There was no discernible tenderness over the vertebral spine upon percussion, and the neurological examination yielded normal results. Deep tendon reflexes were all 2+ and within normal limits. An eye examination conducted by an ophthalmologist revealed no abnormalities. Cervical and lumbosacral X-rays exhibited the absence of pathological changes, including the lack of lytic or sclerotic lesions and fractures. Vertebral alignment was deemed normal.

Subsequently, to evaluate the possibility of intra-abdominal/intra-pelvic lesions and malignancies, an abdominopelvic computed tomography (CT) scan with intravenous contrast was conducted. Furthermore, lumbosacral magnetic resonance imaging (MRI) revealed discernible structural impairments characterized by joint erosions and sclerosis, as depicted in Figures 1 and 2. Notably, abnormal signals were observed bilaterally in the sacroiliac joints, with predominance on the right side. Additionally, bone marrow edema was noted in the sacrum and iliac bones. Findings from a bone biopsy further confirmed the diagnosis of sacroiliitis while simultaneously excluding alternative differential diagnoses.

During hospitalization, the patient experienced massive gastrointestinal bleeding (GIB) while consuming the inflammatory dosage of NSAIDs (naproxen 500 mg/BD). Endoscopic and colonoscopic examinations were performed to pinpoint the source of this bleeding. An adherent clot was identified in the bulb region, prompting consideration for endoscopic intervention and necessitating the discontinuation of NSAID administration due to contraindications. A comprehensive investigation was initiated to elucidate the underlying etiology of the atypical LBP, with a focus on excluding malignancies and infections such as brucellosis and lymphoma. Various assessments, including Wright, Coombs Wright, and 2ME tests, yielded negative results. Similarly, both the purified

protein derivative (PPD) test and interferon-gamma release assay (IGRA) returned negative outcomes. Furthermore, HLA-B27 was found to be positive.

Given the presence of inflammatory LBP, elevated erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) levels, along with corroborative MRI findings, and the exclusion of alternative differential diagnoses, including infections, malignancies, and fractures, as well as the contraindication of NSAID use due to gastrointestinal bleeding, treatment with Adalimumab 40 mg subcutaneously every other week was initiated. After the administration of four doses of Adalimumab, a significant alleviation of pain was observed. This response was evaluated using the Visual Analog Scale (VAS) pain scoring criteria, demonstrating substantial improvement in both subjective and objective measures of pain relief. Disease activity was evaluated using the Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) score both before and after the treatment intervention, as illustrated in Table1. Additionally, the patient was monitored for one month in the clinic, during which no complaints of low back pain were reported, and laboratory markers returned to within the normal range.

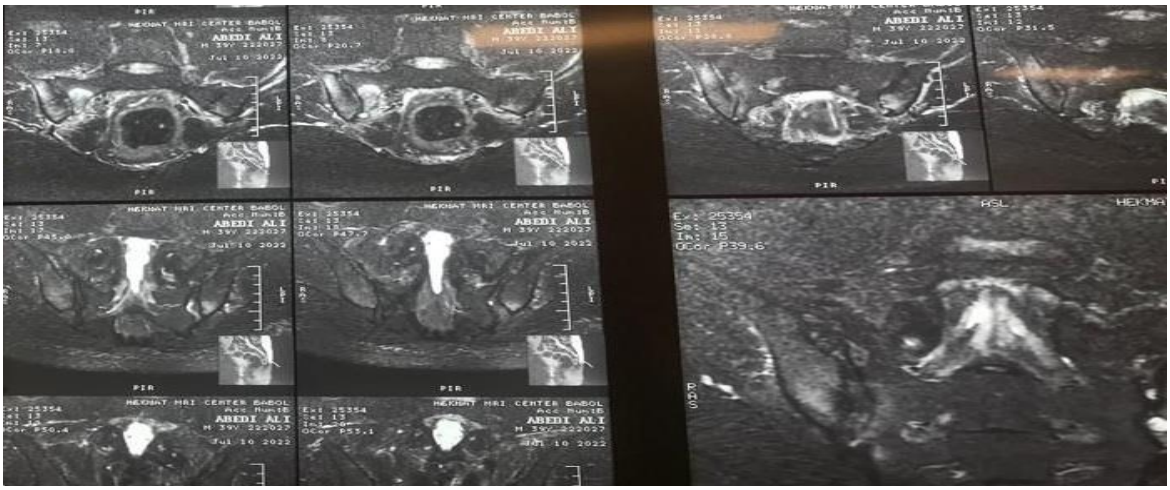

Figure 1. Bilateral sacroiliitis in Flair MRI

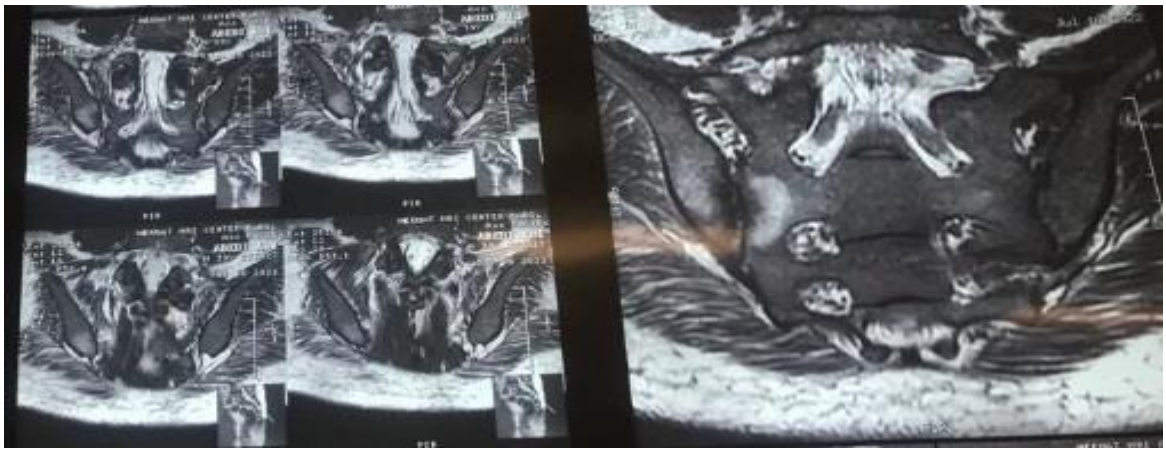

Figure 2. Bilateral Sacroiliitis in T1 MRI

Table 1: BASDAI score of the patient before and after 6 weeks of treatment initiation.

BASDAI questions	Before treatment	After treatment
1. How would you describe the overall level of fatigue/tiredness you have experienced?	7	2
2. How would you describe the overall level of AS neck, back, or hip pain you have had?	10	3
3. How would you describe the overall level of pain/swelling in joints other than neck, back, hips you have had?	10	1
4. How would you describe the level of discomfort you have had from an area tender to touch or pressure?	10	1
5. How would you describe the level of morning stiffness you have had from the time you wake up?	8	2
6. How long does your morning stiffness last from the time you wake up?	9	1
Patient's BASDAI score	**9.1**	**1.7**

Discussion

axSpA is a chronic inflammatory condition categorized by spondylitis and enthesitis, with frequent involvement of peripheral joints, primarily affecting young males. Diagnosis is typically established through clinical manifestations supplemented by laboratory findings such as elevated levels of ESR, CRP, and the presence of the HLA-B27 antigen [7-9]. In the general population, these patients commonly present with chronic LBP, which often requires a significant duration from initial suspicion to definitive diagnosis [10]. However, understanding atypical presentations is crucial for clinicians, whether through encountering various patients or learning from the experiences of colleagues, as early diagnosis remains a focus of clinical efforts [11, 12].

In this study, we introduce a clinical case involving a 39-year-old patient who presented with acute LBP accompanied by bilateral sacroiliitis evident on MRI imaging. To exclude other potential causes of severe pain, including malignancy and infection, a bone biopsy was performed. Consequently, the definitive diagnosis of axSpA was established, followed by the assessment of disease activity utilizing both the Ankylosing Spondylitis Disease Activity Score (ASDAS) and the Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) [13-16]. Following the initiation of NSAIDs as the primary therapeutic approach, the adverse effects associated with these agents became apparent [17-21]. GI bleeding occurred, prompting the need for further investigations to exclude other potential causes and to control bleeding if necessary, leading to the performance of endoscopy and colonoscopy. Subsequently, second-line therapy was initiated. While NSAIDs can effectively prevent disease progression and alleviate symptoms, as well as modify radiographic spinal features, they carry considerations and contraindications, such as the risk of GI bleeding [22, 23]. This decision is consistent with previous research indicating that TNF-α blockers are effective therapeutic options, particularly when NSAIDs are contraindicated or produce an inadequate response [24-27].

As supported by previous investigations, the positive response to TNF inhibitors depends on various factors, including disease characteristics, individual patient attributes, and variables elucidated in previous literature [22, 28]. Moreover, an earlier diagnosis is associated with an increased likelihood of favorable treatment outcomes [28, 29].

The central issue in this case revolves around predicting the treatment response to anti-TNF agents over approximately 12 weeks. A remarkable improvement was observed within less than six weeks following the initiation of anti-TNF therapy, as evaluated using metrics such as the ASDAS and BASDAI. In cases where an adequate response remained elusive after more than 12 weeks of treatment, consideration for alternative biological therapies was warranted. Intriguingly, Macfarlane et al. reported a median duration of 14 weeks (interquartile range 12–17) for treatment response, with variable response rates ranging between 33% and 52% [30, 31].

Despite several scoring systems developed and utilized over time, rheumatologists' clinical judgment remains pivotal in selecting treatment options. This case report highlights a patient with an unusual presentation of ankylosing spondylitis manifestations and a rapid response to treatment, aiming to assist clinicians in refining their ability to recognize disease patterns.

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Conflict of Interests

The authors declare no conflict of interest regarding the publication of this paper.

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