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Sacroiliac Manifestations in Adult-Onset Still's Disease: A Case Report

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1. Abstract
Adult-onset Still's disease (AOSD) is a rare systemic disease characterized by the classic triad of fever, arthralgias/arthritis, and skin rash. Joint involvement is often polyarticular and destructive in the long term. Involvement of the sacroiliac (SI) joints is traditionally considered rare and minimally symptomatic. We report the case of a 45-year-old male patient, an insulin-dependent diabetic, followed for AOSD, who developed severe and chronic bilateral sacroiliitis, confirmed by magnetic resonance imaging (MRI), highlighting the potential for axial spondylarthropathy-like involvement in this disease.

2. Keywords
Adult-Onset Still's Disease, Sacroiliitis, MRI, Chronic Low Back Pain.

3. Introduction
Adult-onset Still's disease (AOSD) is a multi-systemic inflammatory disorder of unknown etiology. Its diagnosis is based on clinical and biological criteria (Yamaguchi or Fautrel criteria: Table 1) [1]. While joint involvement is a diagnostic cornerstone, often manifesting as non-erosive symmetric polyarthritis in the early stages, its progression can lead to chronic joint destruction, similar to rheumatoid arthritis [2]. Involvement of the sacroiliac (SI) joints, although described, remains an infrequent and often subclinical manifestation. We present here an original case of an insulin-dependent diabetic patient with AOSD and active, severe bilateral sacroiliitis, documented by MRI, complicating the picture of mixed low back pain.

Table 1: Classification Criteria Proposed by Yamaguchi (1992)

Number of criteria required : ** ≥ 5 criteria including at least 2 major criteria, with no exclusion criteria.

Major Criteria:

- Fever ≥ 39°C (102.2°F), lasting for at least one week.
- Arthralgia, lasting for at least two weeks.
- Typical macular or maculo papular, salmon-pink, non-pruritic rash, usually appearing with fever.
- Leukocytosis ≥ 10,000/mm³, with at least 80% granulocytes.

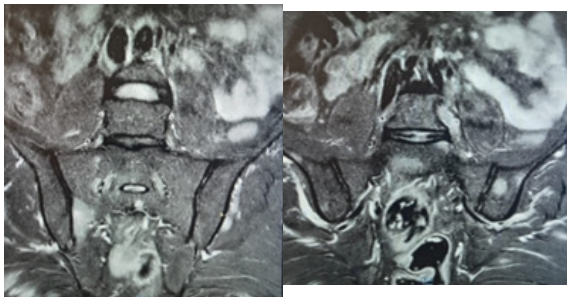
Minor Criteria:

- Sore throat.
- Lymphadenopathy and/or splenomegaly, recently discovered clinically or by ultrasound.
- Liver dysfunction, elevated transaminases or LDH, not attributable to drug reactions or allergy.
- Negative rheumatoid factor and negative antinuclear antibodies.

Exclusion Criteria:

- Infections, particularly sepsis and infectious mononucleosis.
- Malignancies, particularly lymphomas.
- Inflammatory rheumatic diseases, particularly polyarteritis nodosa and rheumatoid vasculitis.

4. Case Presentation
A 45-year-old male patient, followed for uncontrolled type 1 diabetes on insulin therapy, was admitted to the rheumatology department for a flare of his Still's disease, previously diagnosed based on hectic fever, a suggestive skin rash, polyarthritis, neutrophilic leukocytosis, and major hyperferritinemia. During follow-up, the patient developed chronic low back pain of a mixed nature: mechanical upon movement and inflammatory (nocturnal, with morning stiffness > 30 minutes). Lumbosacral magnetic resonance imaging (MRI) was performed (Figure 1 and 2). Sequences using STIR (Short Tau Inversion Recovery), which is sensitive to inflammatory edema, showed the presence of multiple bilateral hyperintense areas affecting both sides (iliac and sacral) of both sacroiliac joints, suggestive of active and severe sacroiliitis. No advanced ankylosing or erosive lesions were noted.



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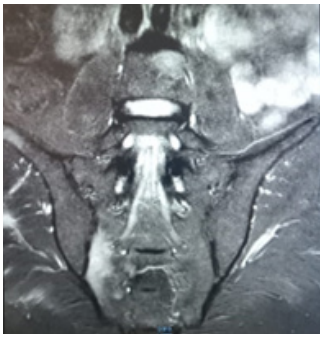

Figure 1: MRI STIR sequence showing hyperintense areas and nodular lesions on both sides of the sacroiliac joints.

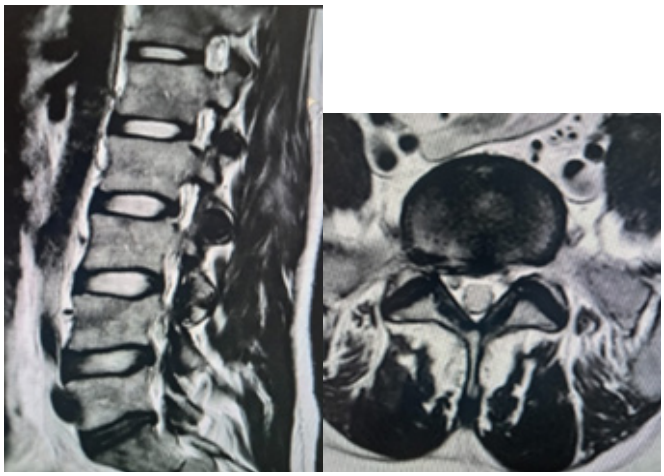

Figure 2: Sagittal and axial T2-weighted MRI showing L5-S1 disc disease, with a right paramedian and foraminal disc herniation.

5. Discussion

Adult-onset Still's disease is a rare systemic inflammatory disease of unknown origin. It manifests with a classic triad of symptoms: high daily fever, salmon-colored skin rash, and joint pain (arthralgia). Articular manifestations are at the forefront of the disease. In the initial phase, it is often a fleeting arthritis affecting multiple joints (polyarthritis), primarily the knees, wrists, and ankles. The pain can be intense, limiting movement. In nearly half of the patients, the disease evolves into a chronic articular form. This phase can lead to irreversible destruction of cartilage and bone, particularly in the wrists and metacarpophalangeal joints of the hands, sometimes mimicking severe rheumatoid arthritis. Ankylosis can occur, leading to significant functional disability and requiring aggressive background treatment. Management aims to control inflammation and preserve joint function.

This case is remarkable in several respects. First, it objectively documents, via MRI, severe bilateral inflammatory involvement of the SI joints in AOSD. While older studies based on standard radiography estimated the prevalence of sacroiliitis between 5 and 17% of cases [3], the use of MRI, a much more sensitive technique for detecting active inflammation (subchondral bone edema), suggests that this involvement may be underestimated.

The mixed low back pain in our patient is also intriguing. The inflammatory component perfectly matches the sacroiliitis revealed. The mechanical component could be multifactorial, potentially influenced by age, overweight, or a history of lumbar strain, although a link with associated degenerative disc disease cannot be ruled out.

The potential role of diabetes, as a pro-inflammatory state, in modulating the severity of joint involvement in AOSD remains an open question. No data in the literature establishes a direct link between diabetes and the prevalence of sacroiliitis in AOSD. It is probably an independent comorbidity in this case.

Finally, this case highlights the importance of a rigorous investigation of low back pain in patients with AOSD. Faced with suggestive symptoms, even mixed ones, performing an MRI of the sacroiliac joints should be considered, as it can reveal inflammatory activity requiring adjustment of background therapy, as was the case for our patient [4].

6. Conclusion

This case report reminds us that sacroiliac involvement in adult-onset Still's disease should not be considered anecdotal. It can be severe, active on MRI, and significantly contribute to chronic low back pain symptoms. The use of MRI, the examination of choice for detecting early inflammation, is justified in cases of clinical suspicion. Better recognition of this entity could allow for more aggressive and early therapeutic management, potentially protective against long-term joint destruction.

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