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A Case of SAPHO Syndrome

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Abstract: SAPHO syndrome is a rare chronic inflammatory disorder characterized by synovitis, acne, pustulosis, hyperostosis, and osteitis, which is often misdiagnosed due to its heterogeneous clinical manifestations. We report a 54-year-old male patient presenting with low back pain radiating to both lower limbs for 4 days and a 10-year history of palmoplantar pustulosis. Physical examination revealed scattered erythema, scaling, pustules, and crusts on both lower limbs and feet. Laboratory tests showed no specific abnormalities. Imaging examinations demonstrated sternoclavicular and sternocostal arthritis, lumbar disc bulging, spinal stenosis, and degenerative changes. Whole body bone scintigraphy revealed increased radiotracer uptake at the bilateral proximal clavicles, upper sternum, and L3 spinous process, consistent with the characteristic bull's head sign, confirming the diagnosis of SAPHO syndrome. The patient received integrated traditional Chinese and Western medicine therapy, including oral herbal decoction, external herbal application, fire acupuncture, nonsteroidal anti inflammatory drugs, and topical dermatological agents. After treatment, the patient's low back and lower limb pain was significantly relieved, and plantar lesions improved. This case highlights that whole body bone scintigraphy is crucial for early diagnosis in patients with sequentially occurring cutaneous and osteoarticular manifestations, which can effectively reduce misdiagnosis and improve clinical outcomes.

1. Patient Information

A 54-year-old male presented with low back pain radiating to both lower limbs for 4 days.

1.1. Chief Complaint

Low back pain accompanied by bilateral lower limb pain for 4 days.

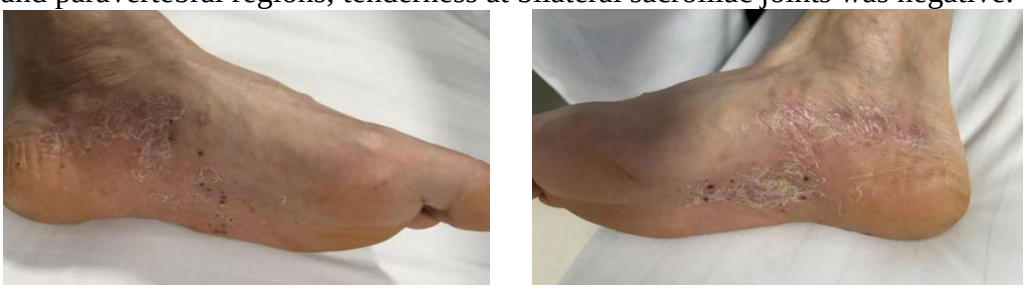
1.2. Present and Past Medical History

Four days prior to admission, the patient developed low back pain without obvious, radiating from both buttocks to the lateral aspect of the lower legs. Oral analgesics provided unsatisfactory

pain relief, and the patient presented to the orthopedic department. The patient had a 10-year history of palmoplantar pustulosis. He was previously treated with adalimumab at a tertiary hospital but developed drug eruption after two injections and discontinued therapy without further systematic management. There was no family history of similar diseases.

1.3. Physical Examination

Scattered erythema with white scaling, pustules, and crusts were noted on both lower limbs and feet (Figures 1). No nail pitting was observed. The lumbar spine showed no obvious scoliosis, but lumbar flexion was limited. Tenderness and percussion pain were positive at the L4–S1 spinous processes and paravertebral regions; tenderness at bilateral sacroiliac joints was negative.



(a) Left (b) Right

Figure 1: Pustules, crusts, partial confluence, desiccation and scaling were observed on both plantar surfaces. No exudation or discharge was noted locally.

1.4. Auxiliary Examinations

Mild abnormalities were detected in blood routine, liver function, electrolytes, and blood lipids. Renal function, thyroid function, ESR, CRP, coagulation function, D-dimer, HLA-B27, rheumatoid factor, T-SPOT, immunoglobulins, complement, AFP, and CEA were all negative. Chest CT demonstrated bilateral sternoclavicular and first sternocostal arthritis (Figure 2), suspicious for SAPHO syndrome. Lumbar CT revealed L1–S1 disc bulging, L4/5 spinal canal and bilateral intervertebral foraminal stenosis, L2–L5 vertebral endplate degeneration, lumbar degenerative changes, and L4/5 facet hyperplasia. Lumbar radiography showed lumbar hyperosteogeny with mild pseudospondylolisthesis of the L4 vertebra (Figure 3). Sacroiliac joint CT showed articular sclerosis, irregularity, and narrowed joint spaces, consistent with bilateral sacroiliac joint degeneration (Figure 4). Whole body bone scintigraphy showed abnormal radiotracer uptake at the bilateral proximal clavicles, upper sternum, and L3 spinous process (Figure 5). Combined with clinical history, SAPHO syndrome was highly suspected.

Figure 2: Chest CT showed sclerosis and destruction of the first sternocostal joints and sternoclavicular joints.

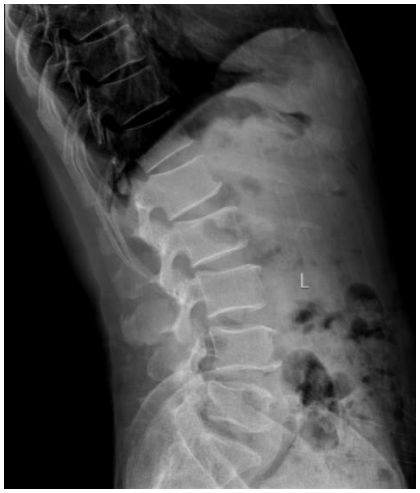


Figure 3: Lumbar radiograph revealed lumbar hyperosteogeny with mild pseudospondylolisthesis of the L4 vertebra.

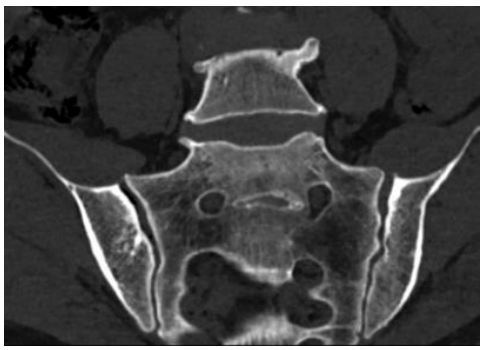


Figure 4: Sacroiliac joint CT demonstrated articular sclerosis, irregularity and narrowing of the sacroiliac joint spaces.

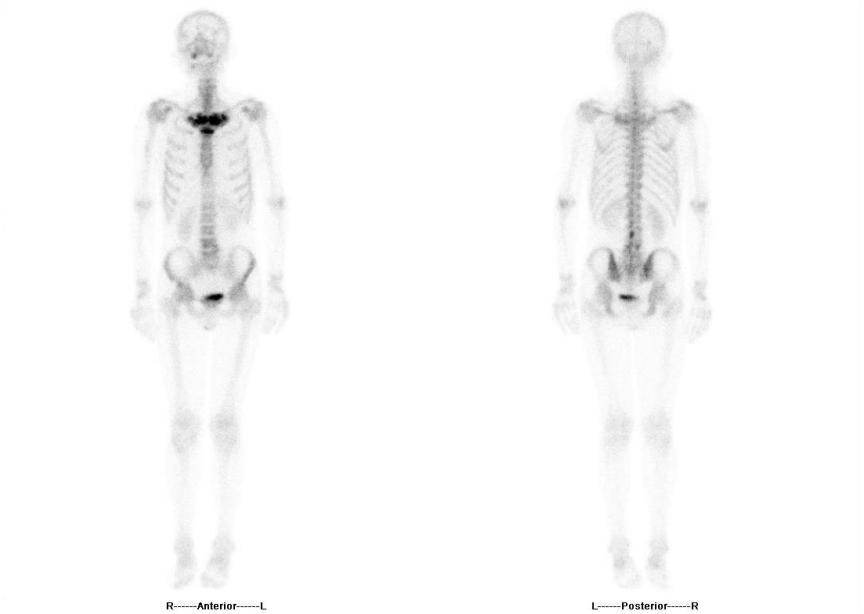


Figure 5: Whole body bone scintigraphy showed clear skeletal visualization. Abnormal increased radiotracer uptake was noted at the bilateral proximal clavicles, upper sternum and L3 spinous process, consistent with the bull's head sign.

1.5. Clinical Diagnosis

Western Medicine Diagnosis:

- SAPHO syndrome
- Palmoplantar pustulosis
- Lumbar spinal stenosis

Traditional Chinese Medicine Diagnosis: Lumbar spondylopathy (cold dampness obstruction syndrome)

1.6. Treatment and Outcome

(1) Traditional Chinese Medicine Treatment

A herbal decoction was prescribed to warm meridians, dispel cold, and unblock collaterals: Coix seed 30 g, Phellodendron chinense 12 g, wine processed Achyranthes bidentata 30 g, Armeniacae semen 12 g, Arecae semen 12 g, Amomum tsao ko 12 g, Magnolia officinalis 9 g, Chuanxiong 12 g, Pinellia ternata 15 g, Stachyurus chinensis 6 g, Talcum 18 g, Atractylodes lancea 12 g, Licorice 9 g, Cinnamon twig 12 g, Chaenomeles speciosa 20 g. The decoction was taken orally, one dose daily, divided into morning and evening. External herbal patches and hot compress were applied to the lower back. Fire acupuncture was performed on plantar lesions to puncture pustules.

(2) Western Medicine Treatment

Anti inflammatory analgesic and neuroprotective therapy was administered. Oral diclofenac sustained release capsules 0.1 g once daily were prescribed. After dermatology consultation, calcipotriol ointment and mometasone furoate cream were applied topically to plantar lesions twice daily.

After treatment, the patient's low back and lower limb pain was significantly relieved compared with admission, and plantar lesions were alleviated. The patient remains under close follow-up.

2. Discussion

2.1. Disease Overview

SAPHO syndrome is a rare chronic immune mediated inflammatory disease characterized by synovitis, acne, pustulosis, hyperostosis, and osteitis. Its etiology remains unclear, with infection, immune dysregulation, and genetic factors considered potential pathogenic mechanisms[1-2]. The annual prevalence in Caucasians is approximately 0.01%, with a predilection for middle-aged women aged 30–50 years[3-4]. Clinical manifestations include osteoarticular and cutaneous lesions, with osteoarticular involvement as the core feature, occurring with or without skin manifestations. Osteoarticular lesions include synovitis, hyperostosis, and osteitis, most frequently affecting the anterior chest wall (65%–90%), followed by the axial skeleton, long bones, and peripheral joints[5]. Cutaneous manifestations are predominantly palmoplantar pustulosis and severe acne, with less common presentations including pyoderma gangrenosum and Sweet syndrome[6]. This patient presented with low back pain radiating to both lower limbs, limited lumbar flexion, tenderness and percussion pain at L4–S1, and typical palmoplantar pustulosis, consistent with the clinical features of SAPHO syndrome.

2.2. Diagnosis

The concept of SAPHO syndrome was first proposed by Chamot et al. in 1987[7]. The 2012 Nguyen diagnostic criteria are widely accepted:①osteoarticular manifestations plus acne fulminans

or hidradenitis suppurativa; ② osteoarticular manifestations plus palmoplantar pustulosis; ③ hyperostosis (anterior chest wall, extremities, spine) with or without cutaneous lesions;④chronic recurrent multifocal osteomyelitis involving axial or peripheral bones with or without cutaneous lesions. Diagnosis is confirmed by meeting any one criterion; this patient satisfied criterion 2.

Diagnostic delay is a prominent feature of SAPHO syndrome. Epidemiological studies report an average diagnostic delay of (3.8±5.3) years, with patients consulting (5.7±3.4) physicians and undergoing multiple imaging examinations before confirmation[8]. In this case, osteoarticular symptoms occurred 10 years after cutaneous manifestations, further complicating early diagnosis.

2.3. Examinations

SAPHO syndrome lacks specific laboratory biomarkers; ESR and CRP may be elevated, while HLA-B27, rheumatoid factor, and autoantibodies are typically negative[9-10]. Imaging is critical for diagnosis due to nonspecific clinical symptoms and laboratory findings. Whole body bone scintigraphy exhibits the highest sensitivity for early detection of bone lesions, superior to radiography, CT, and MRI[11]. The bull's head sign is a pathognomonic imaging feature of sternoclavicular involvement in SAPHO syndrome[12]. Radiography demonstrates hyperosteogeny or spinal ankylosis; CT comprehensively evaluates advanced hyperostosis, hypertrophy, and sclerosis; MRI identifies early bone marrow edema and active inflammatory lesions[13]. The characteristic bull's head sign on whole body bone scintigraphy was the key diagnostic finding in this case.

2.4. Differential Diagnosis

2.4.1. Ankylosing Spondylitis

Ankylosing spondylitis commonly involves the sacroiliac joints and spine, presenting with chronic low back pain and morning stiffness, mimicking SAPHO syndrome. However, HLA-B27 is usually positive, and sacroiliac joints show erosive destruction, narrowed spaces, and ankylosis, whereas SAPHO syndrome is characterized by hyperostosis and sclerosis without erosive changes. Differentiation is achieved via HLA-B27 testing, inflammatory markers, and sacroiliac joint imaging[13].

2.4.2. Lumbar Disc Herniation

Lumbar disc herniation is prevalent in middle-aged individuals, presenting with low back pain radiating to the lower limbs, numbness, and weakness, similar to this patient's symptoms. However, it is pathologically characterized by disc herniation and nerve root compression, with pain exacerbated by specific postures. Lumbar MRI clearly delineates disc herniation and nerve compression. SAPHO syndrome spinal involvement manifests as vertebral endplate inflammation, facet hyperplasia, and hyperostosis, with secondary nerve compression and characteristic scintigraphic uptake, facilitating differentiation[14].

2.4.3. Mediastinal Tumor

Chronic SAPHO syndrome may cause severe anterior chest wall hyperostosis and fusion, compressing adjacent vessels and nerves, resulting in chest and upper limb pain and swelling ("chest wall syndrome"), which may be misdiagnosed as mediastinal tumor. Mediastinal tumors present with chest pain, dyspnea, and cough, with visible masses on chest CT. SAPHO syndrome lesions are confined to the chest wall bones and joints without mediastinal masses, enabling

differentiation via chest CT and whole body bone scintigraphy[6,8].

2.5. Treatment and Prognosis

There is no standardized treatment for SAPHO syndrome, with therapeutic goals focused on relieving symptoms and preserving functional mobility. First-line therapy includes nonsteroidal anti inflammatory drugs and analgesics. Systemic corticosteroids, conventional synthetic disease modifying antirheumatic drugs, TNF α/IL 1 targeted biologics, and bisphosphonates are used in clinical practice, though responses vary among patients[12]. Integrated traditional Chinese and Western medicine serves as an effective adjunctive approach. Traditional Chinese medicine exerts benefits in pain relief, skin lesion improvement, and reduction of Western medicine adverse effects, but is insufficient as monotherapy, requiring multimodal combined and individualized regimens. This patient achieved significant symptom relief with oral herbal medicine combined with nonsteroidal anti inflammatory drugs, fire acupuncture, and topical dermatological agents. SAPHO syndrome follows a benign chronic course with a favorable prognosis, and most patients achieve sustained symptom relief and reduced recurrence with standardized treatment.

3. Conclusion

SAPHO syndrome is a rare disease with heterogeneous clinical manifestations, and cutaneous and osteoarticular symptoms often occur sequentially. This patient's 10-year delay in osteoarticular involvement highlights the diagnostic challenges posed by the disease's heterogeneity. Whole body bone scintigraphy demonstrating the bull's head sign provides a critical diagnostic marker. Integrated traditional Chinese and Western medicine effectively relieved pain and improved skin lesions in this case. Clinicians should enhance awareness of SAPHO syndrome. For patients presenting with unexplained bone pain and characteristic cutaneous lesions, early whole body bone scintigraphy is recommended to minimize misdiagnosis, facilitate early diagnosis and individualized treatment, and optimize patient prognosis.

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