Fractures in SAPHO Syndrome: A Systematic Review of Clinical Features, Imaging Findings, and Management

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Introduction:
SAPHO syndrome (synovitis, acne, pustulosis, hyperostosis and osteotitis) is a rare autoinflammatory disorder characterized by a combination of cutaneous and osteoarticular manifestations. Fractures in SAPHO patients are underrecognized and may be misdiagnosed as infectious or neoplastic lesions. This systematic review is the first to synthesize fracture characteristics, associated clinical features, and management strategies in SAPHO, with the goal of improving recognition among orthopedic surgeons.

Methods:
We conducted a systematic search of the literature using the terms “SAPHO”, “synovitis acne pustulosis hyperostosis and osteomyelitis”, or “acquired hyperostosis syndrome” and “fracture”. The databases MEDLINE, Embase, Cochrane Central Register of Controlled Trials, CINAHL, and Web of Science were searched. Two independent reviewers, B.W.G and A.H., screened studies and included all studies with original clinical data of patients with SAPHO syndrome that have evidence of bone fracture. Abstracts, reviews, commentaries, studies without original data, and studies not in English were excluded. A third reviewer, S.W.C., independently settled all conflicts in the screening process. 13 case reports describing 13 unique patients were produced. All reviewers independently extracted demographic and clinical data from included studies relevant to SAPHO syndrome and bone fractures. All screening and extraction was conducted using the Covidence online software.

Results:
Among the 13 patients, the majority were male (61.5%) and over 50 years of age (53.8%). Fractures occurred most commonly in the spine, clavicle, and ribs, and were frequently atraumatic or pathological. Palmoplantar pustulosis was the most prevalent dermatologic feature (61.5%). Imaging often revealed hyperostosis, marrow edema, or cortical erosion. Biopsies showed sterile chronic osteitis in most cases. Treatment included NSAIDs, DMARDs, bisphosphonates, antibiotics, and JAK inhibitors. Surgical intervention was required in 46% of patients.

Discussion:
Fractures in SAPHO syndrome are a rare but serious complication, most often reported in older adults but occasionally occurs in children. Recognition is challenging due to overlap with infection, malignancy, and other bone disorders, though characteristic dermatologic signs such as palmoplantar pustulosis can provide important diagnostic clues. This study was limited by the small sample size and collection from case reports, which reflects the rarity of SAPHO and the recognition of its manifestations. The data came from case reports that have various levels of detail on treatment plans and reporting. However, there was a consistent presentation across these reports that suggest clinical patterns worth further investigating.

Significance/Clinical Relevance:
Fractures in SAPHO are frequently mistaken for malignancy, infection, or trauma, leading to delays and unnecessary interventions. The high prevalence of palmoplantar pustulosis offers a simple clinical clue, underscoring the need for orthopedic surgeons to consider SAPHO in the differential for atypical fractures.