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MANDIBULAR MANIFESTATIONS OF SAPHO SYNDROME. A SYSTEMATIC REVIEW

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ABSTRACT

Background: SAPHO syndrome is a rare, long-lasting inflammatory disease. While it usually affects the chest wall and spine, it attacks the lower jaw in about 10% of patients. Because its symptoms closely mimic regular tooth infections, it frequently leads to wrong diagnoses and delayed care.
Aim: This study aims to provide a clear, detailed review of SAPHO syndrome in the jaw based on 22 scientific reports. The goal is to describe clinical signs, imaging results, and treatment options, and to compare modern medicines with major surgeries.
Results: The most common signs are recurrent severe jaw pain, facial swelling, and severe trismus. Without treatment, the jaw joint can completely fuse or melt away. Early diagnosis requires advanced scans: MRI with a STIR setting is the best tool to find early bone fluid (edema), while CT scans show a characteristic "onion-skin" bone pattern. Traditional treatments like antibiotics offer only short-term relief. Today, modern targeted drugs—such as biologic agents, bisphosphonates, and Yunke (99Tc-MDP)—are used to successfully stop bone damage and calm the immune system.
Conclusion: The treatment of SAPHO syndrome in the jaw has shifted from major bone surgery to smart, targeted medicines. Early diagnosis using MRI is critical to avoid pulling healthy teeth. Surgery should only be used to rebuild the face or free a stuck joint after the medicine has stopped the active disease.

KEYWORDS

SAPHO Syndrome; Mandible; Temporomandibular Joint; Osteitis; Hyperostosis; Misdiagnosis; Targeted Therapy

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Introduction

SAPHO syndrome is a rare, long-lasting disease that causes inflammation. Doctors first gave it this name in 1987 [1]. The name stands for a specific mix of skin, joint, and bone problems [2]. This disease usually affects the chest wall and the spine. However, it also attacks the lower jaw in about ten percent of all patients [3]. Because the disease is so rare and looks like other common problems, it is very hard for dentists and jaw surgeons to diagnose it correctly [4].

Aim of the Study

The main goal of this study is to provide a clear and detailed review of SAPHO syndrome in the jaw. We aim to describe the clinical signs, imaging results, and treatment options using twenty-two scientific reports. By putting this information together, we hope to help doctors spot the symptoms early and avoid wrong diagnoses. Another goal is to compare how well modern medicines work compared to major surgeries. Ultimately, this review wants to give doctors a clear, evidence-based plan to manage this disease and help patients feel better.

Materials and methods

The systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2020 guidelines. Eligible studies were English-language primary original investigations (case reports, case series or observational studies) that reported the diagnosis or treatment of jaw osteitis in patients with clinically diagnosed SAPHO syndrome. A comprehensive search was performed in PubMed and Scopus using the query ("SAPHO" OR "SAPHO syndrome" OR "synovitis, acne, pustulosis, hyperostosis, and osteitis") AND ("jaw" OR "mandible" OR "maxilla" OR "mandibular" OR "maxillary" OR "jaw osteitis" OR "gnathic") AND ("diagnosis" OR "treatment" OR "therapy" OR

"management"), yielding 144 records; searches closed on 13rd April 2026. After removal of duplicates (n=82), the remaining 62 titles/abstracts were screened.

Twenty-two full texts were retrieved, and 18 studies met the inclusion criteria for qualitative synthesis. The PRISMA flow chart is presented in Fig. 1. All screening steps were undertaken independently by two reviewers (M.M. and P.S.) with disagreements resolved by consensus.

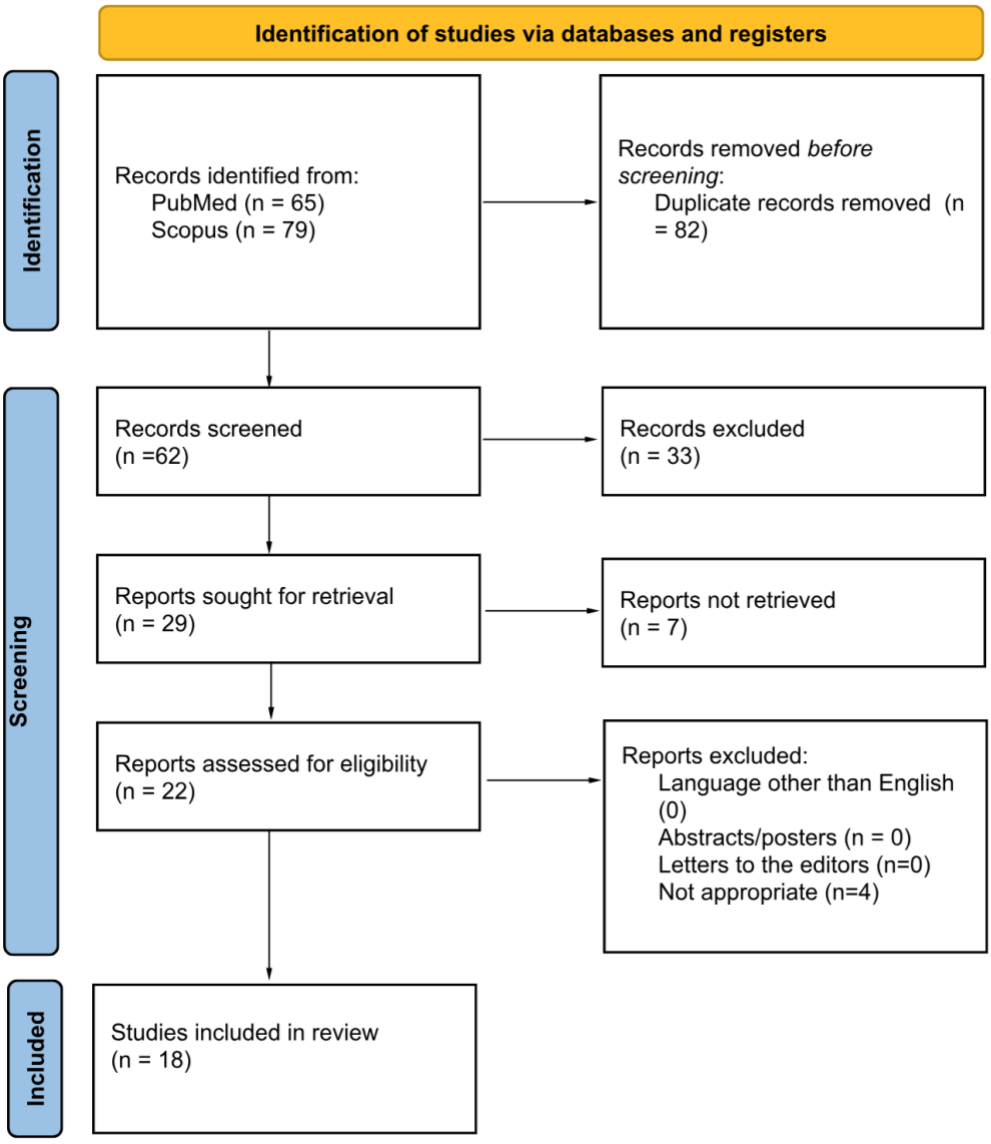

Fig. 1. *PRISMA flow chart of the study selection process.*

Results

Patients with SAPHO syndrome in the jaw often have symptoms that look just like regular tooth infections. This similarity often leads to wrong diagnoses at first, which delays the right treatment [5]. A study looking back at seventeen patients showed that severe jaw pain and facial swelling that keep coming back are the most common signs [6]. For many patients, this hardening and swelling in the jaw bone is the very first sign of a larger immune system problem that might later affect other bones [7]. Patients often feel a constant, dull ache in the jaw joint area. Over time, they also develop severe trismus, which means they cannot open their mouths to eat normally [8]. If the inflammation is not treated, it can cause the jaw joint to completely fuse and get stuck [9]. To fix this severe physical block and let the patient open their mouth again, surgeons sometimes have to cut out parts of the jaw joint [10].

Sometimes, the damage to the jaw joint is so fast and severe that it looks like bone cancer. Doctors have reported cases where the top part of the jaw joint completely melts away, and the swelling spreads deep into

the face [11]. In very rare and scary cases, the bone inflammation can spread from the jaw directly into the ear bone, which can cause sudden hearing loss [12].

Detailed Diagnosis and Imaging: Experts agree that finding SAPHO syndrome early is the most important step. Because regular X-rays often look normal at the start, doctors must use advanced scans. Experts suggest that specialized imaging should be started as soon as possible to avoid pulling out healthy teeth by mistake [13]. Magnetic resonance imaging, or MRI, is the best test because it uses a specific setting called STIR. This setting is very sensitive because it can see "bone marrow edema," which is basically fluid and swelling trapped inside the hard bone [14, 15]. Another useful test is Computed Tomography (CT), which shows the physical changes in the bone structure. In SAPHO patients, CT scans often show an "onion-skin" pattern, where the bone grows in layers as it tries to heal from constant inflammation [16]. Finally, doctors use bone scintigraphy (a whole-body scan) to see if the inflammation has spread to other areas, like the ribs or spine, which confirms that the problem is not just in the jaw [17].

Detailed Medical Treatment: Treatment has moved away from simple painkillers and towards drugs that change how the immune system behaves. In the past, doctors tried regular drugs like antibiotics and steroids, but these only give short-term relief [15, 16]. Modern medicine now focuses on advanced drugs that target the immune system directly. For example, special drugs called biologic agents, like anti-TNF therapy, have been very successful in stopping jaw inflammation [17]. Also, giving patients a specific medicine through an IV, known as Yunke (99Tc-MDP), quickly reduces severe bone pain and calms the immune system [18]. Other studies show that strong bone-building drugs, called bisphosphonates (like zoledronic acid), work very well to stop bone damage by blocking the cells that destroy bone tissue [19]. A comprehensive summary of the key findings from the reviewed literature regarding clinical symptoms, imaging diagnostics, and treatment strategies (References 5-22) is presented in **Table 1**.

Table 1. Summary of key scientific reports on mandibular manifestations of SAPHO syndrome (References 5-22).

Study (First Author, Year)	Country	Study Design	Sample Size (N)	Main Parameters / Interventions	Key Findings / Outcomes
Kikuchi (2018)	Japan	Case Report	1	Differential diagnosis of jaw pain	Mandibular symptoms closely mimic dental infections, leading to frequent misdiagnosis.
Cai et al. (2025)	China	Case Series	17	Clinical presentation analysis	Recurrent severe jaw pain and facial swelling are the most dominant clinical signs.
Parlier-Cuau (2014)	France	Observational	N/A	Bone hardening patterns	Mandibular osteitis can be the first indicator of a systemic immune disorder.
Kotaki et al. (2020)	Japan	Clinical Study	N/A	TMJ symptoms in SAPHO	Chronic dull pain in the TMJ often leads to progressive and severe trismus.
Müller-Richter (2009)	Germany	Case Report	1	TMJ complications	Advanced inflammation can result in complete joint ankylosis (fusion).
Utumi et al. (2008)	Brazil	Case Report	1	Surgical management	Severe mechanical joint blocks may require bilateral condyle and coronoid removal.
Kodama et al. (2013)	Japan	Case Report	1	Condylar resorption	Aggressive bone loss in the condyle can radiologically mimic malignant tumors.
Marsot-Dupuch (1999)	France	Case Report	1	Spread of inflammation	Jaw inflammation can extend to the temporal bone, causing sudden hearing loss.
Huang et al. (2025)	China	Imaging Study	22	CT diagnostic features	CT scans show a characteristic "onion-skin" pattern, which is vital for early diagnosis.
Baba et al. (2016)	Japan	Imaging Study	N/A	MRI STIR utility	MRI with STIR sequences is the gold standard for detecting early bone marrow edema.

Study (First Author, Year)	Country	Study Design	Sample Size (N)	Main Parameters / Interventions	Key Findings / Outcomes
Roldán et al. (2001)	Germany	Case Series	4	Traditional drug efficacy	Conventional oral antibiotics and steroids provide only temporary and incomplete relief.
Deng et al. (2012)	China	Clinical Study	N/A	Treatment failure analysis	Standard infection treatments often fail, causing months of unnecessary suffering.
Marí et al. (2014)	Spain	Clinical Study	N/A	Biologic therapy	Biologic agents (anti-TNF) effectively halt bone inflammation and induce remission.
Shao et al. (2020)	China	Clinical Trial	N/A	99Tc-MDP (Yunke) effect	IV Yunke treatment significantly reduces bone pain and calms the immune system.
Li et al. (2019)	China	Retrospective	31	Bisphosphonate therapy	Zoledronic acid blocks bone-destroying cells and effectively relieves clinical symptoms.
Liu et al. (2023)	China	Comparative Study	N/A	Surgery vs. Medication	Radical bone surgery is ineffective without systemic medical control of the disease.
Delrieu et al. (2023)	France	Case Series	N/A	Microvascular reconstruction	Immediate jaw reconstruction is necessary if functional surgery is required.
Mori et al. (2024)	Japan	Case Report	1	JAK inhibitor therapy	Janus kinase inhibitors show promise for severe cases resistant to other medicines.

Abbreviations: CT: Computed Tomography; **IV**: Intravenous; **JAK**: Janus Kinase; **MDP**: Methylene Diphosphonate; **MRI**: Magnetic Resonance Imaging; **N**: Number of patients/sample size; **N/A**: Not Available/Not Applicable; **SAPHO**: Synovitis, Acne, Pustulosis, Hyperostosis, and Osteitis; **STIR**: Short Tau Inversion Recovery; **TMJ**: Temporomandibular Joint; **TNF**: Tumor Necrosis Factor.

Discussion on Management Strategies

The way doctors treat SAPHO syndrome in the jaw has changed a lot. In the past, surgeons tried to cut out the diseased bone. However, these major surgeries often failed because the inflammation comes from a body-wide immune problem, not just a local infection. A very important case study showed that even if a surgeon removes the entire lower jaw, the pain might continue if the patient does not get the right medicine for their immune system [20]. Because of this, if surgery is absolutely necessary, doctors must immediately rebuild the jaw to protect the patient's quality of life [21]. Recently, new immune-targeting pills called Janus kinase inhibitors have shown amazing results in patients who did not get better with surgery. This proves that the future of treating this disease relies on smart medicines [22]. Today, surgery is only used as a helpful tool to fix stuck joints, not as a cure for the disease itself.

Limitations

While this review covers a lot of information, there are a few limits. First, because SAPHO syndrome is so rare, most research papers only look at one patient or a small group of patients. The lack of large studies makes it hard to create rules that work for everyone. Also, different studies use different tests and treatments, which makes it hard to compare their results directly. Finally, many reports do not follow the patients for a long time. This missing long-term data makes it hard to know if the jaw pain comes back years after treatment.

Future Perspectives

Looking ahead, doctors around the world need to work together on larger studies. Creating a global list of patients with jaw SAPHO syndrome could solve the problem of small study sizes. Future research should also try to understand the exact genes and cells that cause this immune problem, which could lead to treatments made specifically for each person. The great early results from new biologic drugs and pills should be tested more in strict medical trials. Also, creating standard step-by-step guides for dentists, joint specialists, and bone doctors will help patients get the right diagnosis and care much faster.

Conclusions

The treatment of mandibular SAPHO syndrome requires a major change in medical thinking. Because this disease is easily mistaken for common dental problems, educating dentists and doctors is essential to prevent unnecessary tooth extractions and delayed care. Our findings show that early detection using advanced imaging is the most crucial step in diagnosing the condition. Furthermore, research clearly shows a shift from aggressive bone surgeries to targeted medicines as the main standard of care. Today, surgeries are no longer viewed as a cure for the disease itself. Instead, they are a final option for restoring jaw function- such as fixing a fused joint and should only be performed once the active inflammation is controlled with medication. Ultimately, a team-based approach focused on early medical treatment offers the best chance to preserve jaw function and improve the patient's quality of life.

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