RESEARCH PAPER

Refractory Severe Ankylosing Spondylitis With Total Temporomandibular Joint Ankylosis And Latent Tuberculosis

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Abstract

Background: Ankylosing spondylitis (AS) typically affects the axial skeleton. Progression to complete bony ankylosis of the temporomandibular joint (TMJ) is an extremely rare and debilitating manifestation that leads to severe functional impairment and malnutrition. This case highlights the complexity of managing "end-stage" AS with high disease activity and comorbid latent tuberculosis.

Case Presentation: A 31-year-old male presented with a 13-year history of progressive generalized stiffness, eventually becoming bedridden. He exhibited total jaw fixation due to bilateral TMJ ankylosis, limiting intake to soft liquids. Imaging revealed a "bamboo spine," complete sacroiliac fusion, and Sawhney Stage IV TMJ ankylosis. Despite previous conventional treatments, disease activity remained high (ASDAS-CRP > 3.5). The patient also tested positive for latent tuberculosis (IGRA+). He was treated with the IL-17A inhibitor Secukinumab (150 mg monthly) alongside Isoniazid prophylaxis. Over seven months, the patient showed significant reduction in pain scores and inflammatory markers (CRP decreased from baseline to 2.87 mg/L), achieving stabilized disease activity without tuberculosis reactivation.

Conclusions: Severe TMJ ankylosis is a rare but catastrophic complication of AS. This case demonstrates that IL-17A inhibitors are effective in controlling refractory inflammation even in advanced structural stages. Furthermore, it confirms the safety of Secukinumab in patients with latent tuberculosis when combined with appropriate prophylaxis.

Keywords: Ankylosing spondylitis; Temporomandibular joint ankylosis; Secukinumab; IL-17A inhibitor; Latent tuberculosis; Bamboo spine

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Background

Ankylosing spondylitis (AS) is a chronic systemic inflammatory rheumatic disease and a hallmark of axial spondyloarthritis (axSpA) [1]. It predominantly targets the sacroiliac joints and the spine, leading to characteristic syndesmophyte formation, bony fusion, and significant functional impairment [2]. While axial skeleton involvement is the primary feature, peripheral joint manifestations are frequently observed. However, involvement of the temporomandibular joint (TMJ) remains a rare and often underdiagnosed complication [3]. Although minor TMJ symptoms, such as joint clicking or mild pain, are reported in some AS cohorts, progression to complete bony ankylosis is an extreme manifestation that can lead to catastrophic failure of oral function and severe malnutrition [4], [5].

The pathogenesis of AS involves a complex interplay of genetic factors, most notably the HLA-B27 antigen, and inflammatory cytokines such as tumor necrosis factor-alpha (TNF-α) and interleukin-17A (IL-17A) [6]. IL-17A, in particular, has been identified as a key driver of both the inflammatory response and the pathologic new bone formation that leads to ankylosis [7]. In advanced or refractory cases, where conventional synthetic disease-modifying antirheumatic drugs (csDMARDs) and non-steroidal anti-inflammatory drugs (NSAIDs) fail to control disease activity, biological therapy becomes essential [8].

Literature regarding total TMJ ankylosis in AS is limited. Previous reports have documented various degrees of involvement; for instance, de Andrade Freitas Oliveira et al. (2013) reported a 31-year-old male with a 16-year history of AS who developed unilateral TMJ ankylosis and severe trismus [9]. More recently, Kojima et al. (2024) described a 53-year-old patient whose initial presentation of AS was fibrotic TMJ ankylosis, while Jayaprakash et al. (2024) emphasized that the risk of TMJ cartilage and disc degeneration increases significantly with disease duration [5], [10]. These cases collectively highlight the rarity of TMJ fusion in AS and the necessity of advanced imaging for diagnosis and surgical intervention for functional restoration.

This case report presents a unique clinical challenge: a 31-year-old male with severe, "end-stage" AS characterized by total, bilateral TMJ ankylosis and generalized skeletal fusion. The management of this case is further complicated by comorbid latent tuberculosis (LTB), a significant hurdle in endemic regions when initiating biological therapy. This report aims to highlight the efficacy and safety of IL-17A

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inhibition (Secukinumab) in managing high disease activity in advanced AS and to discuss the necessity of a multidisciplinary approach in restoring function in patients with extensive structural damage.

Case Presentation

A 31-year-old male, Mr. W, presented to Dr. Soetomo General Hospital with a chief complaint of generalized body stiffness. The stiffness had progressed over 13 years and significantly worsened in the four months prior to admission. The symptoms initially began 15 years ago as bilateral knee and hip pain that gradually spread to the entire body. The patient reported inflammatory back pain characteristics, including morning stiffness that improved with physical activity. Over a decade, the progressive ankylosis moved from the knees to the spine, neck, and eventually the jaw. Consequently, the patient had been bedridden and unable to walk for the past 12 years. Furthermore, severe temporomandibular joint (TMJ) involvement prevented him from opening his mouth, limiting his nutritional intake to soft porridge consumed through the gaps between his teeth. He had been diagnosed with Ankylosing Spondylitis (AS) at a regional hospital five months prior. There was no history of diabetes, hypertension, or cardiovascular, renal, or cerebrovascular diseases. His current medications included Sulfasalazine 500 mg, Meloxicam, folic acid, calcium, and vitamin B.

Physical examination revealed a patient in stable general condition with a Glasgow Coma Scale of 15 (E4V5M6). Vital signs were within normal limits (BP 103/58 mmHg, HR 85 bpm, RR 20 bpm, Temp 36.7°C). Head and neck examination showed no conjunctival pallor, icterus, or lymphadenopathy. Cardiopulmonary and abdominal examinations were unremarkable. Musculoskeletal assessment demonstrated extensive stiffness and pain in the cervicothoracic, lumbosacral, bilateral hip, and temporomandibular joints. Symmetrical ankylosis was noted, although no active joint swelling, subcutaneous nodules, or subluxations were found.

Laboratory investigations revealed an elevated Erythrocyte Sedimentation Rate (ESR) of 43 mm/h, indicating active inflammation. Hematological and biochemical profiles, including renal and liver functions, were largely within normal limits, except for a slightly elevated SGPT (58 U/L) and mild hyponatremia (133 mEq/L). Notably, the Interferon-Gamma Release Assay (IGRA) was positive, suggesting latent tuberculosis (LTB) infection, while screenings for HBsAg, anti-HCV, and anti-HIV were non-reactive.

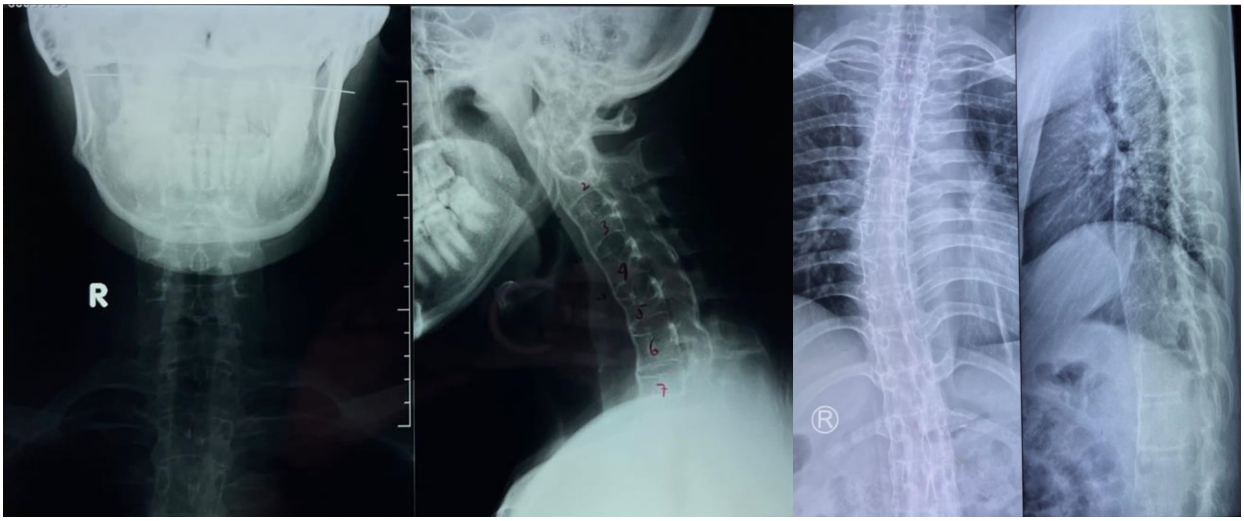

Figure 1. Cervicothoracic X-rays

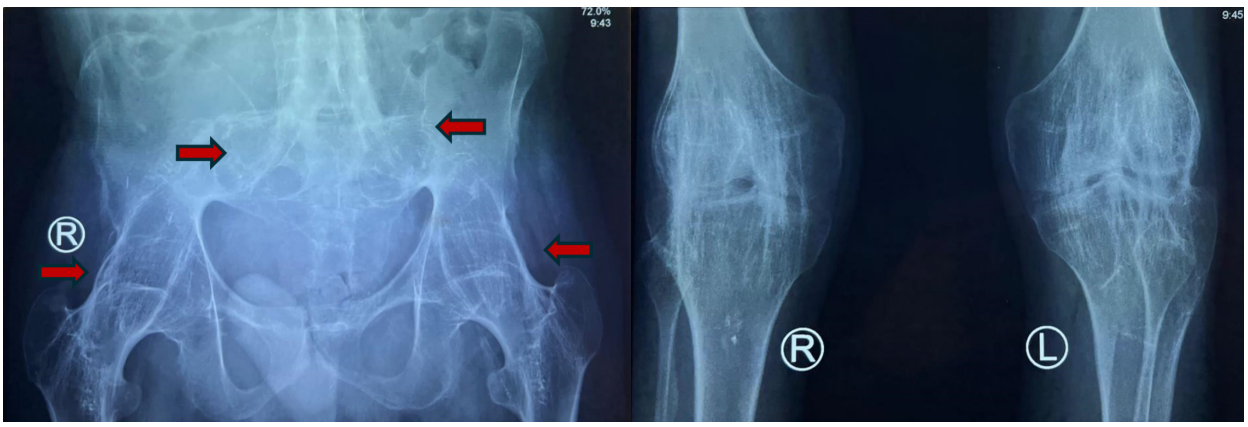

Figure 2. Pelvic and knee X-ray

Radiological imaging provided hallmark evidence of advanced disease. Cervicothoracic X-rays showed ossification of the anterior and posterior longitudinal ligaments (C2–C7) and bilateral symmetric syndesmophytes, resulting in the classic "bamboo spine" appearance (Figure.1). Pelvic X-rays revealed complete fusion of the bilateral sacroiliac joints (Grade IV sacroiliitis) and ankylosis of the bilateral hip joints. Knee radiographs showed extensive femorotibial and femoropatellar ankylosis (Figure 2). MRI of the abdomen and pelvis confirmed sacroiliac and hip fusion with fatty marrow changes, signifying disuse osteoporosis and fatty muscle infiltration. A non-contrast head CT scan with 3D reconstruction confirmed bridging ankylosis between the right mandibular ramus and zygomatic arch (Sawhney Type IV) and left TMJ fusion (Sawhney Type II) (Figure 3).

The patient was diagnosed with Severe Ankylosing Spondylitis with TMJ Ankylosis, refractory to NSAIDs and conventional Synthetic Disease-Modifying Antirheumatic Drugs (csDMARDs), with very high disease activity. He was hospitalized for the initiation of Secukinumab (an anti-IL-17A monoclonal antibody). The initial management included a high-calorie, high-protein diet (2100 kcal/day), Secukinumab 150 mg subcutaneously, and the continuation of Sulfasalazine and Meloxicam. Due to the positive IGRA, Isoniazid (INH) 300 mg daily was initiated as prophylaxis for latent TB.

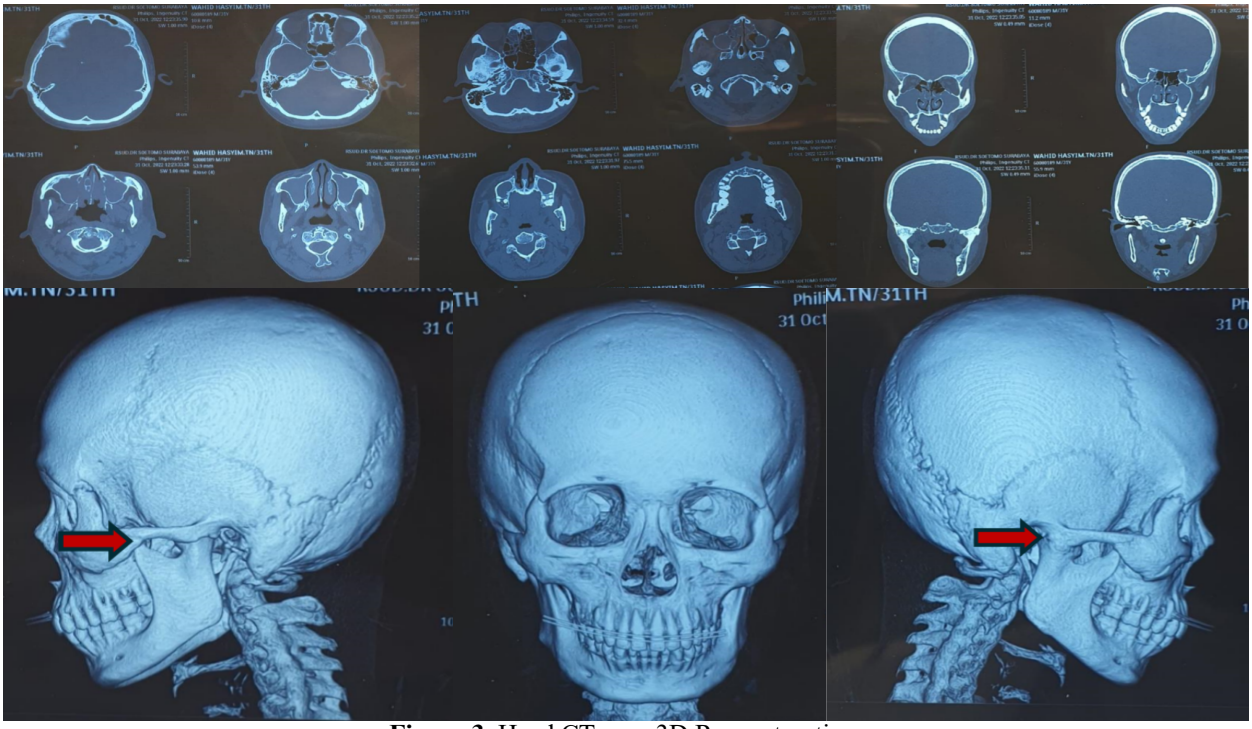

Figure 3. Head CT scan 3D Reconstruction

Following the first injection in June, the patient reported a reduction in joint pain (NRS 6-7). However, further Secukinumab dosing was briefly synchronized with LTB treatment protocols. By July, after the second injection, his inflammatory markers improved (ESR 5, CRP 2.38), though ASDAS scores remained high (ASDAS-CRP 4.3). Throughout the subsequent months (August to December), the patient received monthly Secukinumab injections (third through sixth doses). Despite persistent TMJ stiffness, he reported a significant decrease in tailbone and shoulder pain. In February, after the seventh injection, the patient's pain significantly subsided (NRS 4-5), and by March, his inflammatory markers were stabilized (ESR 41, CRP 2.87) with an ASDAS-CRP of 2.8.

At the final evaluation, the patient demonstrated clinical improvement in AS-related pain. For long-term management, he was transitioned to oral Sulfasalazine (3000 mg/day) and Diclofenac. To address the functional limitation of the jaw and improve quality of life, the patient was referred to the Plastic Surgery department for consideration of TMJ ankylosis release surgery.

Discussion

Ankylosing spondylitis (AS) is characterized by chronic inflammation that leads to syndesmophyte formation and spinal ankylosis [11]. However, the progression to complete, bilateral bony ankylosis of the temporomandibular joint (TMJ) represents an extreme and rare phenotype [4]. This case provides a unique clinical perspective on managing "end-stage" AS manifestations where mechanical failure and high systemic inflammatory activity coexist.

The rarity of Sawhney Stage IV TMJ ankylosis in AS cannot be overstated [12]. While axial involvement is the hallmark of the disease, the TMJ is often "the forgotten joint" in rheumatological assessments [13]. In this patient, the replacement of the right TMJ with a solid bony bridge (as confirmed by 3D-CT) led to severe trismus, preventing basic oral functions. This mirrors the

"bamboo spine" pathology but in a location that directly threatens nutritional status [14]. Literature suggests that early detection of TMJ tenderness or reduced mouth opening is critical, as once bony fusion occurs, pharmacological intervention becomes secondary to surgical salvage [4].

A pivotal discussion point in this case is the management of refractory disease activity in the presence of latent tuberculosis (LTB). The patient's ASDAS-CRP score of >3.5 indicated "Very High Disease Activity" despite previous csDMARD therapy. The initiation of Secukinumab, a human IL-17A inhibitor, was a strategic choice. Recent evidence, including the MAXIMISE trial, has highlighted the efficacy of IL-17A inhibition in treating both axial and peripheral manifestations of spondyloarthritis [15]. Pathophysiologically, IL-17A is a key driver of the "bone-remodeling paradox" in AS—promoting both erosive inflammation and pathologic new bone formation [16]. By targeting this pathway, Secukinumab addresses the core mechanism of syndesmophyte progression more directly than traditional therapies [16], [17].

Furthermore, the safety of biologics regarding TB reactivation is a paramount concern in endemic regions like Indonesia [18]. Unlike TNF-α inhibitors, which are known to disrupt granuloma stability, IL-17A inhibitors have shown a lower propensity for TB reactivation in clinical registries [19], [20]. The successful co-administration of Isoniazid (INH) prophylaxis with Secukinumab in this patient reinforces the safety profile of IL-17A inhibitors, provided that rigorous screening protocols (such as IGRA) are followed [21].

Finally, this case underscores the necessity of a "Total Patient" management approach [22]. While Secukinumab successfully reduced systemic inflammatory markers (ESR and CRP) and pain, the established bony ankylosis remains a permanent structural defect [23]. The transition from medical stabilization to surgical referral for TMJ release (gap arthroplasty) exemplifies the multidisciplinary collaboration required to restore quality of life in advanced AS [24].

This case highlights a rare and severe presentation of Ankylosing Spondylitis involving total TMJ ankylosis. It demonstrates that IL-17A inhibitors, such as Secukinumab, are effective and safe for managing high disease activity, even in patients with latent tuberculosis. However, the restoration of mechanical function in "burnt-out" joints requires early multidisciplinary intervention, combining biological therapy with specialized surgical procedures [22], [23].

Conclusion

This case highlights a rare and severe manifestation of ankylosing spondylitis with total bilateral temporomandibular joint ankylosis and generalized skeletal fusion, resulting in profound functional impairment. Secukinumab, administered with isoniazid prophylaxis in the setting of latent tuberculosis, was effective in reducing inflammatory activity and pain without evidence of tuberculosis reactivation, supporting the potential safety and utility of IL-17A inhibition in complex refractory cases. However, established bony ankylosis remained irreversible, emphasizing that restoration of function requires timely recognition of temporomandibular joint involvement and a multidisciplinary approach integrating biologic therapy with surgical intervention.

LIST OF ABBREVIATIONS

- AS: Ankylosing Spondylitis
- ASDAS: Ankylosing Spondylitis Disease Activity Score
- CRP: C-Reactive Protein
- CT: Computed Tomography
- ESR: Erythrocyte Sedimentation Rate
- IGRA: Interferon-Gamma Release Assay
- IL-17A: Interleukin-17A
- INH: Isoniazid
- MRI: Magnetic Resonance Imaging
- NSAIDs: Non-Steroidal Anti-Inflammatory Drugs
- TMJ: Temporomandibular Joint

DECLARATIONS

Ethics approval and consent to participate: Not applicable.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Availability of data and material: All data generated or analysed during this study are included in this published article.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: RTH diagnosed the patient and was a major contributor in writing the manuscript. LDR supervised the clinical management and revised the manuscript for intellectual content. All authors read and approved the final manuscript.

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Refractory Severe Ankylosing Spondylitis With Total Temporomandibular Joint Ankylosis And Latent Tuberculosis 1, Mar. 2016, doi: 10.24198/pjd.vol28no1.13518.