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ANKYLOSING SPONDYLITIS ASSOCIATED WITH CRANIOMANDIBULAR DISORDER—A COMBINED ORTHODONTIC AND PROSTHODONTIC THERAPEUTIC APPROACH

Ankylosing spondylitis is a disease that causes inflammatory changes of the involved joints. Although the initial clinical signs are pain and discomfort, synovial changes progressively involve all the axial joints, including the temporomandibular joint (TMJ). Eventually, bony alterations develop (condylar erosions, flattening, sclerosis) that affect the position of the condyle, the superior joint space, and the range of movements. These symptoms correlate with the severity of the disease. Besides physiotherapy and surgery, no dental rehabilitation has been reported for these patients. This report of a female patient with ankylosing spondylitis and a TMJ disorder emphasizes dental rehabilitation. The aim of the splint, orthodontic, and prosthodontic treatment was to relieve the subjective symptoms through establishing a stable optimum occlusion. Anamnestic, laboratory, and clinical findings including pre- and postradiographic examination records are presented. World J Orthod 2009;10:371–377.

Key words: *ankylosing spondylitis, craniomandibular disorders, rehabilitation*

Ankylosing spondylitis is a chronic inflammatory disorder of the axial skeleton, sacroiliac, and large peripheral joints.[1] The clinical signs vary from mild mobility limitation to total ankylosis and can be compounded by extra-articular manifestations.[2]

Ankylosing spondylitis has a strong association with psoriatic arthritis, a subset of reactive arthritis, and arthritis/sacroiliitis in inflammatory bowel diseases.[3] It occurs in 0.2% of the population with a male/female ratio of 2.5 to 5.0:1.0. Ankylosing spondylitis typically presents in young adulthood, but symptoms may arise in adolescence or earlier.[4]

The etiology of ankylosing spondylitis is suspected to be an interrelation between HLA genes, sex factors, and environmental agents.[5] A familial clustering exists, with Class I HLA-B27 antigen of the major histocompatibility complex (MHC) being the major allele in 95% of affected persons. Many candidate gene loci for the susceptibility of this disease have been suggested, but no evidence of linkage is yet confirmed.[6] The cytokine tumor necrosis factor alpha (TNF-α) may also be involved.[7] Trauma and bacterial infections act as triggering factors, which disturb the immune system.[8] The 60kDa heat shock protein of Klebsiella Pneumoniae plus its homology in sequence with other enterobacteria indicate their involvement in ankylosing spondylitis.[9] As for HLA-B27–negative families, a hypothesis is that genes for psoriasis or other alleles of the MHC may be etiologically important.[6,10]

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Table 1 Laboratory findings in patients with ankylosing spondylitis showing which components are elevated, present, or absent

Elevated	Present	Absent
ESR	HLA-B27	RA factor
CRP	Normochromic normocytic anemia	Autoantibodies
Akaline phosphatase	Leukocytosis	
IgA, IgG		

The inflammatory process begins at the ligament, tendon, and joint capsule attachments to bone—the entheses. This comprises an agglomeration of chronic inflammatory cells and bone erosion. Later, features of healing by fibrosis, sclerosis of the underlying bone, and new bone formation in the adjacent ligaments can be observed.[11] In case of the vertebral column, the new bone, termed syndesmophyte, forms bridges both laterally and anteriorly to vertebral bodies. This leads to a fusion of the spine, thus converting it into a rigid structure, which is vulnerable to fracture.[12]

The onset of this disease is insidious, often beginning in the sacroiliac region and spreading upward to the thoracic and cervical spine. A suspicious symptom of ankylosing spondylitis is lower back pain that persists longer than 3 months, worsening by inactivity but improving with exercise. In time, flexion and rotation are inhibited, rigidity becomes extreme (ankylosis), and the cervical spine is curved while chest expansion is limited and respiration impaired.[13,14] Additional symptoms are low-grade fever, fatigue, tachycardia, and anorexia responsible for loss of weight and muscle.[15]

Peripheral asymmetric oligoarthropathy[16] and joint synovitis is seen in 30% of patients. Systemic manifestations also include ocular lesions (anterior uveitis and iridocyclitis) and cardiovascular affections (aortitis, mitral valve involvement, and pericarditis). Rare complications are pulmonary fibrosis, neurologic disorders, and secondary amyloidosis.[17–19] The progress of ankylosing spondylitis can be relentless or can cease at any stage.[15] Laboratory findings support the inflammatory nature of the disease (Table 1).[3,20]

Diagnosis of ankylosing spondylitis is based on a patient's symptoms: a family history of iritis, psoriasis, or spondyloarthropathies; and physical examination that demonstrates a decreased range of spinal motion in all directions.[14] Laboratory findings verify the diagnosis; however, radiographic examination is helpful. The main laboratory finding that characterizes ankylosing spondylitis is the presence of HLA-B27, which is present in 40% to 95% of all patients.[21] The radiologic findings of ankylosing spondylitis usually include symmetric sacroiliac joint space narrowing and blurring of joint margins, subchondral sclerosis, squaring of vertebral bodies with paravertebral syndesmophytes, general erosions, and sclerosis of ligamentous attachments. These pathologic findings progressively lead to the characteristic ankylosed "bamboo spine."[22,23]

Treatment relies on daily physical therapy, including stretching, back extension, and breathing exercises. Medications such as NSAIDs suppress articular inflammation, pain, and muscle spasms; new NSAIDs, known as COX-2, have even fewer adverse effects. Sulfasalazine, glucocorticoids, narcotics, analgesics, or muscle relaxants are also beneficial.[24–27] Anti-TNF-α agents, such as infliximab[28,29] and radium-224 in intravenous injections,[25] seem promising. If ankylosing spondylitis leads to immobility, surgical treatment can be indicated. Radiotherapy is recommended as a last resort due to the risk of developing acute myelogenous leukemia.[30,31]

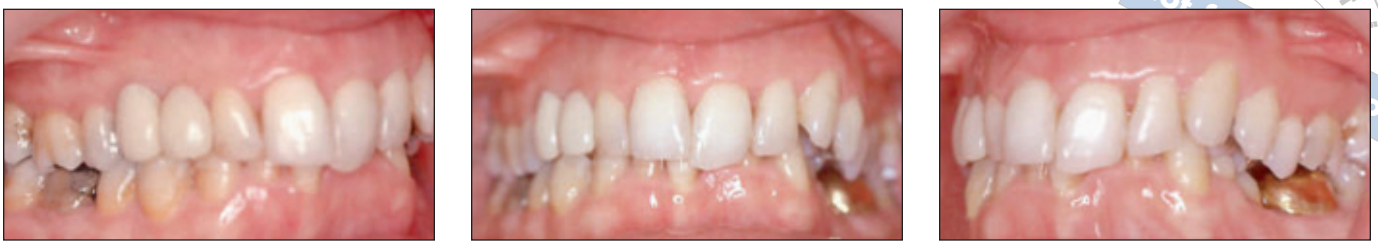

Fig 1 Patient's initial intraoral situation: Increased overbite, crowding, and buccal nonocclusion of the maxillary left first and second premolars.

TMJ INVOLVEMENT

Ankylosing spondylitis has a tendency to affect fibrocartilaginous structures such as the TMJ. In fact, TMJ affections occur in 11% to 35% of all ankylosing spondylitis patients. The variation in frequency can be explained by the type of study, the examined population, and the tools used to assess TMJ affection.[32–35] The most common subjective symptoms are difficulties in wide mouth-opening, TMJ crepitus, pain, stiffness, swelling, and headaches.[36,37] Computed tomography (CT) and magnetic resonance imaging (MRI) images show temporal flattening, abnormal condylar shape, erosions, sclerosis, disc alterations, osteophytes and even total ankylosis of the joint.[38,39] TMJ treatment includes physiotherapy and occasionally moist heat and ultrasound application.[40] In acute phases of TMJ arthritis, glucocorticoid injections are indicated. To prevent muscle hyperactivity and articular strain due to bruxism, occlusal splints can be inserted, but in cases of obvious occlusal interferences, occlusal adjustment or prosthetic reconstruction could be necessary. Orofacial pain can be treated with transcutaneous nerve simulation. When the TMJ is ankylosed, surgical interventions such as arthroplasty and synovectomy with discectomy are indicated. If not, a joint prosthesis becomes necessary.[37,41]

PATIENT REPORT

A 36-year-old female presented with bilateral pain in her masticatory muscles and both TMJs, headaches localized in the temporal and iliac region, and a feeling of fatigue during speech. She reported an earlier diagnosis of scoliosis, iritis, and an operation on the right inner ear due to an angiotumor of the tympanon. General symptoms such as malaise or pain throughout all joints, especially with humid weather, were also present. The clinical examination revealed tenderness to palpation of the right TMJ as of her right sternocleidomastoid and posterior belly of digastric muscles. In addition, swelling of the ankle joints with persistent depression upon pressure and pain upon pressure on the distal and proximal interphalangeal joints were noticed. Maximum mouth-opening amounted to 49 mm, right/left lateral excursion to 12 mm/13 mm, and protrusion to 12 mm.

She had an Angle Class I molar and canine relationship and a deep curve of Spee on her left side (Fig 1). The maxillary and mandibular anterior teeth were crowded, and the overbite increased so that the crown of the mandibular left central incisor was covered by its antagonist by 110%. The mandibular left first and second premolars were severely lingually inclined, thus leading to a buccal nonocclusion of their antagonists.

Laboratory findings revealed that rheumatoid factor was negative, while the HLA-B27 antigen was positive (Table 1). The diagnosis of ankylosing spondylitis was verified by a rheumatologist. The specific differential diagnosis was based primarily on the laboratory findings and only secondarily on the rheumatologic symptoms.

The aim of the dental rehabilitation was to relieve the subjective symptoms by aligning all teeth optimally and establishing an ideal stable occlusion. This was

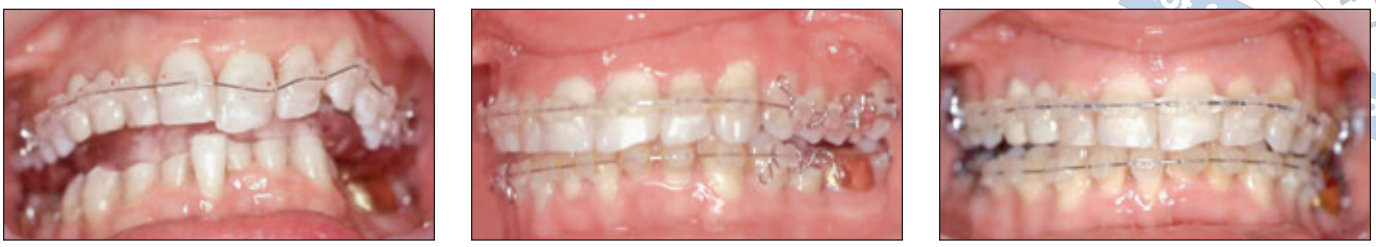

Fig 2 Intraoral situation with a mandibular stabilization splint in place at the initiation of orthodontic treatment.

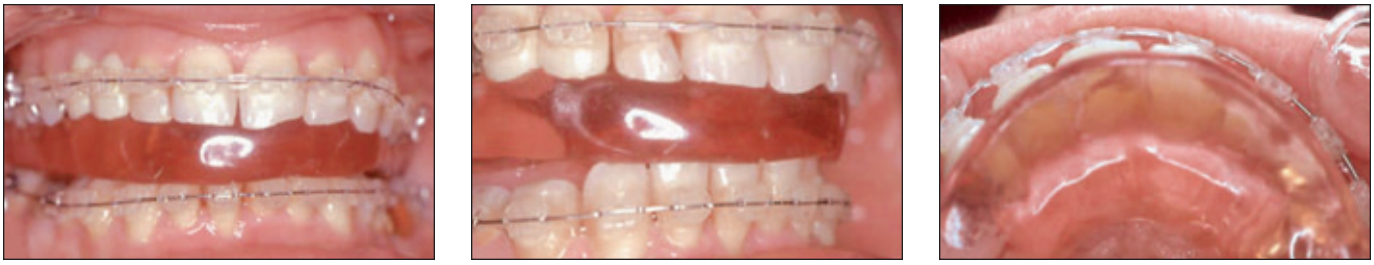

Fig 3 Introaral situation after placement of an anterior repositioning splint.

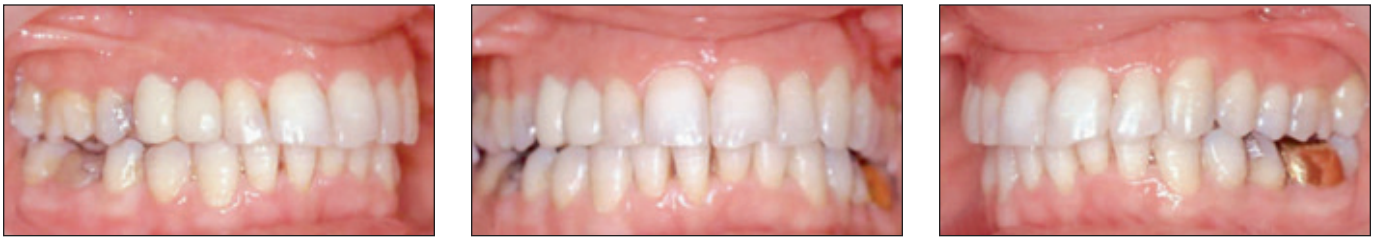

Fig 4 Patient's final occlusion after orthodontic therapy with reduced overbite, eliminated crowding, and corrected nonocclusion of the mandibular left first and second premolars.

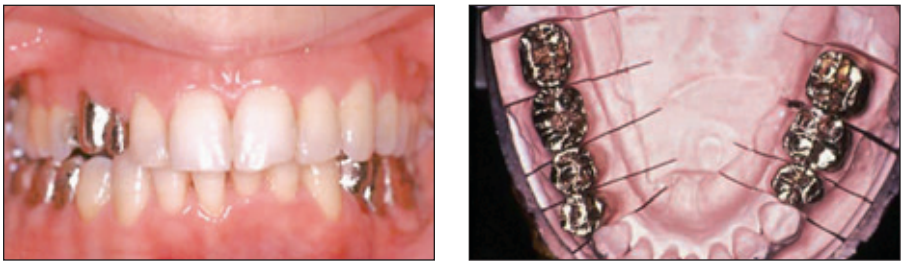

Fig 5 Clinical try in of the cast copings, including the maxillary right first premolar and a cantilever for the maxillary right canine; porcelain-fused-to-metal crowns with metal surfaces restored the posterior teeth.

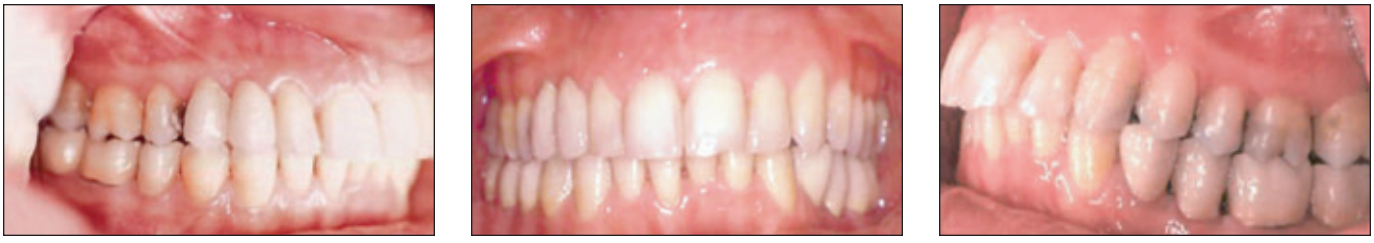

Fig 6 Intraoral situation at the completion of the prosthetic reconstruction.

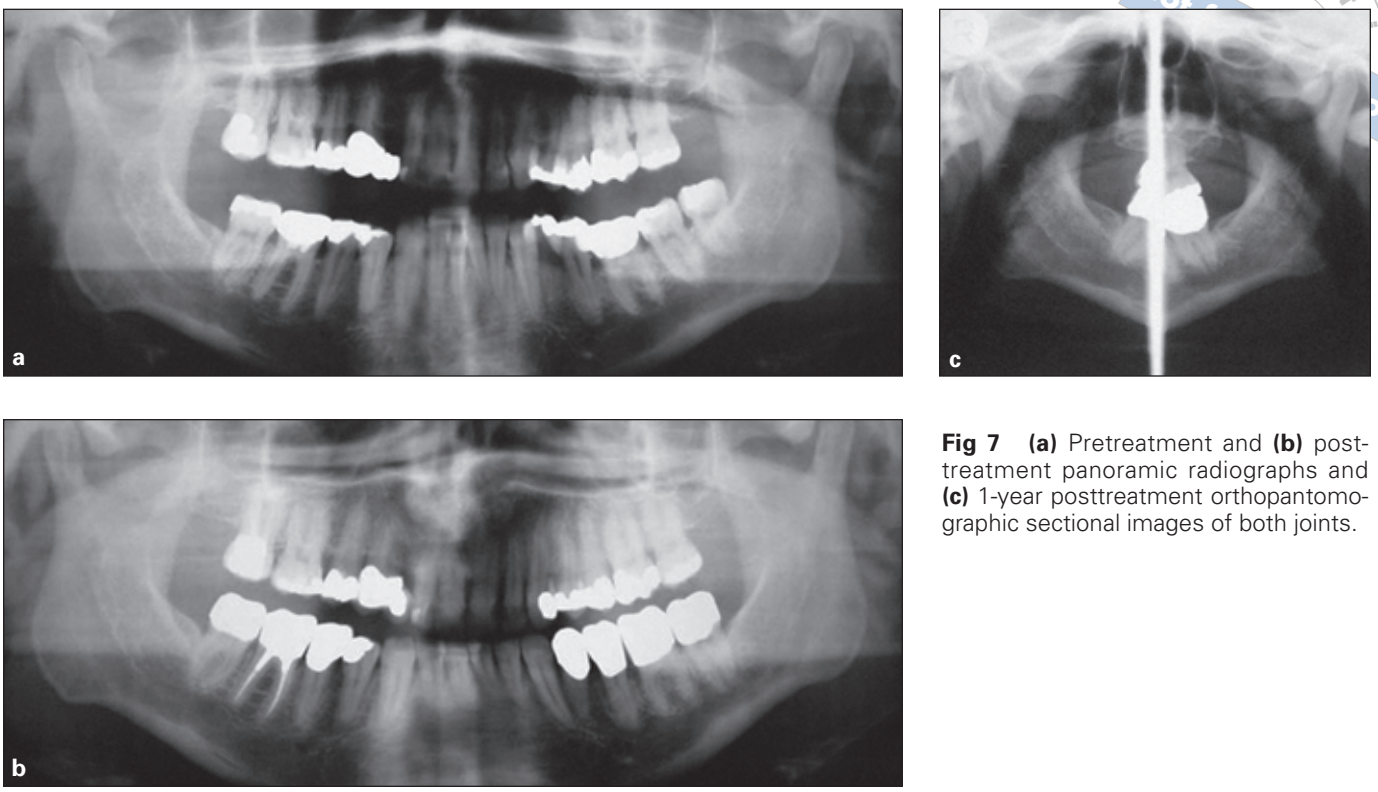

Fig 7 **(a)** Pretreatment and **(b)** posttreatment panoramic radiographs and **(c)** 1-year posttreatment orthopantomographic sectional images of both joints.

achieved by a combination of kinesio- and splint therapy, followed by orthodontic and prosthodontic treatment. Kinesiotherapy for the head and neck was initiated to diminish the chance of TMJ ankylosis and relax the masticatory muscles.

The full-coverage stabilization mandibular splint aimed at a load reduction of the TMJ, especially during mastication, thus reducing the sensitivity of the joints and masticatory muscles (Fig 2). At the same time, the splint raised the bite, which helped to correct the lingual nonocclusion of the mandibular left first and second premolars. Subsequently, to relocate both condyles in a more physiologic position and further unload the TMJ, an anterior repositioning splint with a significant increase of the vertical dimension (8 to 9 mm) was inserted (Fig 3). With the accompanying orthodontic treatment, the buccal nonocclusion was corrected and the crowding alleviated, which led to desensitization of both the joints and muscles.

Finally, upon completion of orthodontic treatment (Fig 4), the patient received a prosthetic rehabilitation of her posterior teeth (Fig 5). The prosthetic rehabilitation was thought to further stabilize the patient's occlusion, developing almost pure canine guidance (the maxillary right first premolar was included because the maxillary right canine was a cantilever). Also, occlusal interferences were eliminated, masticatory function improved, and esthetics enhanced by replacing the unattractive prosthetic work (Fig 6).

The combined orthodontic and prosthetic therapy was succeeded by periodical examinations for a period of 2 years. These follow-ups included both clinical and radiologic examinations.

All therapeutic efforts resulted in the elimination of the clinical symptoms including reduction of the patient's headaches and fatigue during speech. The relocated condyles maintained their position without any evidence of deterioration 2 years posttreatment as revealed radiographically (Fig 7). With minor occlusal refinements, the patient remained asymptomatic 8 years after the completion of treatment.

CONCLUSION

In a case of ankylosing spondylitis, it was possible by combined orthodontic and prosthodontic treatment to relieve subjective symptoms through realignment of elements of the stomatognathic system, to achieve optimum tooth and arch positions and relationships, and to develp a permanently functional and stable occlusal scheme.

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