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Research Paper

Tmj Ankylosis In Still's Disease – A Case Report

[*]Dr. Pravin Patel[1],Dr. Anil Managutti[2],Dr. Shailesh Menat[3], Dr. Jigar Tandel[1], Dr. Jigar Patel[4]

[1]*Post graduate student, Department of Oral and Maxillofacial Surgery, Narsinhbhai Patel Dental College & Hospital, Sankalchand Patel University, Visnagar, Gujarat*
[2]*Professor & HOD, Department of Oral and Maxillofacial Surgery, Narsinhbhai Patel Dental College & Hospital, Sankalchand Patel University, Visnagar, Gujarat*
[3]*Professor, Department of Oral and Maxillofacial Surgery, Narsinhbhai Patel Dental College & Hospital, Sankalchand Patel University, Visnagar, Gujarat*
[4]*Private practitioner*
**Corresponding author: *Dr. Pravin Patel*

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ABSTRACT: *Rheumatoid arthritis (RA) is a chronic inflammatory disease characterized by joint swelling, joint tenderness, and destruction of synovial joints, leading to severe disability and premature mortality. TMJ complaints are present in about more than 50% of patients of RA. TMJ is usually among the last joint to be involved and is associated with many clinical signs and symptoms of which pain is a major problem later leading to inflammation, limited movements, swelling (joint stiffness) and muscle spasm. If it occurs in early age it may result in mandibular growth disturbance, facial deformity, and ankylosis and in adult these can vary from mild joint stiffness to total joint disruption with occlusal-facial deformity. The diagnosis and management of TMJ involvement in RA is exclusionary based on history, physical findings, radiographic study, and lab testing. Hence a multidisciplinary approach is necessary. The present paper reports a case of RA with bilateral TMJ involvement with its classical radiographic findings.*

Keywords: *Arthritis, Juvenile idiopathic athtritis, Still's disease, TMJ ankylosis*

I. INTRODUCTION

Juvenile idiopathic arthritis (JIA) is the most common autoimmune autoinflammatory musculoskeletal disease in children all over world, manifesting in girls more frequently than male 3:1.[1] It is defined as persistent arthritis for more than 6 weeks with an onset at younger than 16 years of age, after excluding other causes of joint inflammation.[2] The etiology is not clear but appears to be multifactorial and may be related to genetic factors associated with trigger events such as psychological stress, trauma, abnormal levels of hormones, or infections. All other joints can be affected in JIA including the temporomandibular(TMJ). TMJ involvement was first reported in 1897 by Still when he described chronic arthritis in childhood. The reported frequency of TMJ affection varied in the literature depending on the population investigated, the subtypes of JIA represented and the method by which TMJ disease is diagnosed. In all subtypes of JIA, one or both TMJs can be affected and may even be the initial joint involved.[4]

The disease is associated with many clinical signs and symptoms such as pain, joint stiffness, difficulties in mouth opening and open bite. In severe cases of temporomandibular joint disorders (TMD), masticatory movement may be hampered, as complications of arthritis TMJ ankylosis can also occur. Complete clinical examination along with radiological examination is required for the diagnosis of TMJ ankylosis. Cone beam computed tomography [CBCT] or CT scan is most useful radiograph for diagnosis and treatment planning of TMJ ankylosis.[6] This article describes case report of bilateral TMJ ankylosis diagnosis and surgical management.

II. CASE REPORT

A 17 year old male patient came with the chief complain of inability to open his mouth since 2 years. Patient had history of PUO (pyrexia of Unknown Origin) since April 2011. He had hypochromic, microcytic anemia with low S.iron suggestive of iron deficiency with elevated WBC and mild degree thrombocytosis with no abnormal cells in peripheral smear. His ESR was high(>150), while biochemical parameters were within normal limits except LDH which was mildly elevated. His Ig profile was normal. His RA and ANA were negative. Bone marrow reviewed did not show any evidence of hematological malignancy. Extensive work up for fever done outside (Leptospira, Brecellosis, CT abdomen, ASO, Blood culture, Urine cultures) were negative. X-ray chest was normal. LAP score was normal. 2-D echo did not show any evidence of vegetation. In light of these findings and history of recurrent fever, fleeting joint pain with high ESR, he was diagnosed as systemic onset juvenile rheumatoid arthritis by rheumatologist in a year of December 2011. After that patient was on ayurvedic medicines for the fever.

Since 2011 he had pain in the TMJ region followed by slowly decrease in mouth opening. When he visited our department he did not have fever and other join pain. The maximal inter-incisal mouth opening distance was 2 mm and no movement felt on bilateral TMJ region. No obvious facial deformity was present. No any other body joints were affected.Three different types of imaging diagnosis were performed, Orthopantomogram, Cone beam computed tomography (CBCT) and hand wrist X-ray. OPG(Fig.1) shows radio opaque mass on bilateral TMJ region, coronal image of CBCT(Fig.2) shows complete destruction of joint space and complete bony fusion of joint on medial and lateral side. No appreciable changes in hand wrist X-ray (Fig-3).Bilateral gap arthroplasty of TMJ and placement of interpositional material (Temoralis myofascial flap) was planned under general anesthesia.Fiber optic intubation was done, preauricular incision was made and bilateral region gap arthroplasty(Fig.4) done and 43 mm interincisal mouth opening(Fig.5) was achieved, temporomyofacial flap(Fig.6) was obtained and sutured with 4.0 Vicryl on medial side of the joint to prevent the reankylosis. Layer by layer closure was done. Mouth opening exercise was started after 7 days. Postoperative healing was good and patient was kept on regular follow up and there was no postoperative complication seen.

III. DISCUSSION

Juvenile idiopathic arthritis (JIA) is the most common autoimmune autoinflammatory musculoskeletal disease in children.[1,4]

Systemic onset juvenile arthritis(formerly called Still's disease or systemic onset juvenile rheumatoid arthritis) is a subset of juvenile idiopathic arthritis (JIA). The course and prognosis of systemic onset juvenile arthritis are highly variable. The typical child with systemic onset juvenile arthritis experiences a four to six month period of spiking fevers and rash, with varying degrees of arthralgia and arthritis. This is often followed by a relative quiescence of the systemic manifestations. Arthritis resolves completely in approximately 40 to 50 % of patients. The child who appears well after six months has a substantial probability of remaining well, whereas the child who continues to have active disease (eg. Fevers, arthritis, elevated platelet count, and continued requirement for glucocorticoid therapy) three to six months after diagnosis typically has a more difficult course. In our case patient had active disease and had typical feature of arthritis that is destruction of joints (TMJ).By definition, children with JIA have disease onset prior to age 16 years, and present with joint pain, stiffness and swelling that persists for longer than 6 weeks. Without appropriate treatment, JIA may result in devastating consequences. Children may experience permanent disability from joint destruction, growth deformities or blindness (from chronic uveitis associated with JIA). In the case of the systemic-onset form of JIA, untreated disease may even result in multiple organ failure and death.[6] In our case patient had destruction of both TM joints which lead to ankylosis.

The International League of Associations for Rheumatology (ILAR) has concluded that the term ''juvenile idiopathic arthritis'' describes seven subtypes of arthritis according to the clinical features during the first 6 months of disease: oligoarticular JIA, polyarticular JIA, both rheumatoid factors positive and negative, systemic JIA, juvenile psoriatic arthritis, enthesitis-related arthritis, and undifferentiated arthritis.[1]

Rheumatoid Arthritis (RA) may have states, of which one is active (when the tissue is inflamed) and other is inactive (remission), when inflammation decreases. During inactive state of RA patient doesn't feel any symptoms of the disease. But when disease relapses, symptoms start appearing (flare). RA progresses in a symmetrical pattern. multiple joints of both the sides of the body are affected simultaneously. Morning stiffness (generally occurs for one hour), fatigue, loss of energy, lack of appetite and low grade fever are the common symptoms associated with this disease. Joints of wrists, fingers, knees, feet and ankles are most commonly affected. Aching and stiffness of joints as well as muscles also takes place. Joints frequently become red, swollen, painful and tender. Loss of cartilage, erosion and weakness of the bones and joint deformity are other consequences of RA. RA is known as juvenile arthritis when it occurs below the age of 16. Limping, irritability, crying and poor appetite are the symptoms of juvenile arthritis, which are seen among children.[3]

Approximately 50 percent of all cases of rheumatoid arthritis show involvement of the temporomandibular joint out of that 30 percent may exhibit chronic involvement with some limitation of motion and rarely ankylosis. Involvement of the temporomandibular joint also may precipitate a change in the occlusion of the teeth and progressive anterior open-bite. Osteoporosis, limitation of condylar movement, and marginal condylar irregularities are other signs of temporomandibular joint involvement. Ultimately ankylosis may occur when the temporomandibular joint is involved by rheumatoid arthritis during childhood; there is accentuation of the antegonial notch, the height of the ramus is reduced and the body is shortened. In children, the temporomandibular joint may be the first one to be involved.[4]

In the present case, there was no associated family history. The clinical findings in the TMJ affected with JIA are similar to those described for other joints, that is, pain, swelling, movement impairment and crepitation. Malocclusion of the teeth and anterior open bite may occur in advanced stages.[6] The joints are tender upon pressure, and there is morning stiffness usually seen and decreased masticatory force.[7]

In children, it may disturb in mandibular growth, facial deformity and ankylosis of joint. TMJ ankylosis secondary to JIA is generally found in the later stages of the disease, but it is a rare finding.[4,8]

Diagnosis of TMJ ankylosis with JIA is based on history, physical examination, radiological examination and laboratory investigation. Certain abnormal blood antibodies are frequently found in patients with JIA like rheumatoid factor(RF) and raised ESR values. Other confirmatory tests like citrulline antibody and CRP test are recommended in doubtful cases, [1-3] our case showed an elevated CRP level.The radiographic changes of TMJ includes flattening of the mandibular head, cortical erosion, gradual decrease in joint space due to granulation, deossification, pencil head or spiked deformity of the condylar head.[6,8]More recently, CBCT has been the object of study for TMJ examination by various authors who consider it the best recommended imaging for the degenerative diseases of TMJ.[10] In our case CBCT and OPG showed bilateral TMJ ankylosis.

The involvement of TMJ in JIA usually correlates well with the radiographic damage of the joints in the hands and feet. The present case TMJ was primarily involved there is a no appreciable changes in hand wrist radiograph (Fig-7).A 7-step Kaban protocol has been developed for the treatment of TMJ ankylosis: 1) aggressive resection of the ankylotic segment, 2) ipsilateral coronoidectomy, 3) contralateral coronoidectomy when necessary, 4) lining of the joint with temporalis fascia or cartilage, 5) reconstruction of the ramus with a CCG, 6) rigid fixation of the graft and, 7) early mobilization and aggressive physiotherapy.[12,13] In our case bilateral gap arthroplasty and inter positional temporomyofacial flap placement was carried out.

IV. CONCLUSION

Diagnosis of still's disease is very important and needs medical treatment, in case of active disease involving TMJ and causing TMJ ankylosis requires gap arthroplasty and inter-position of the temporalis myofacial flap, myofacial flap has superior results than any other non-vascular interpositional graft and reconstructive materials in terms of functional stability of joint, recurrence and economical which proves its versatility in the management of TMJ ankylosis.

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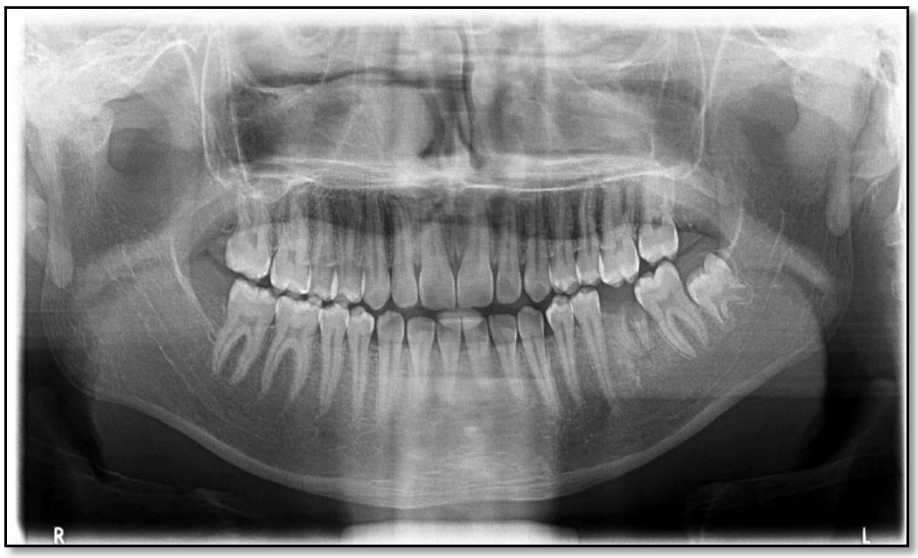

Fig 1 : Pre-op OPG shows radio opaque mass on bilateral TMJ region

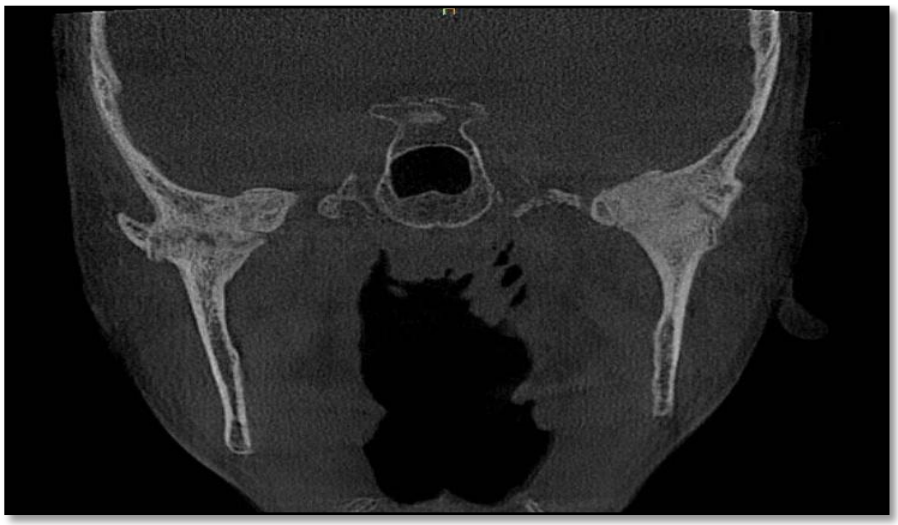

Fig 2 : Coronal image of CBCT shows complete destruction of joint space and complete bony fusion of joint on medial and lateral side

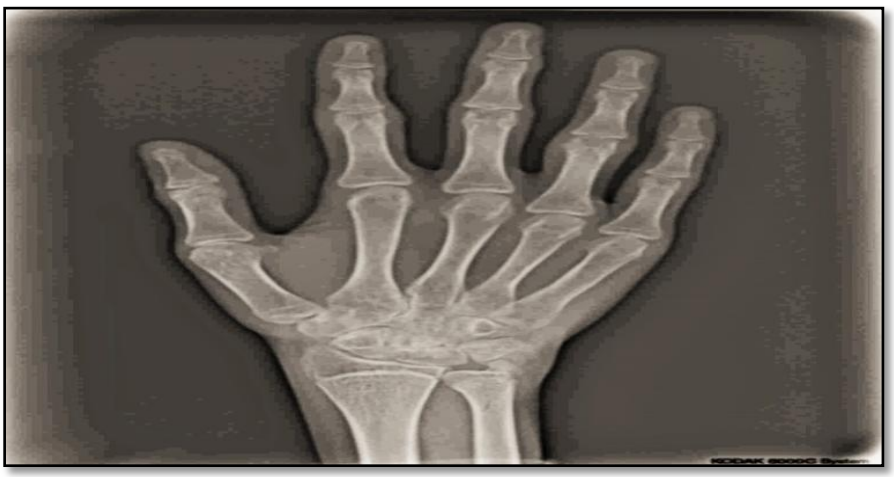

Fig 3 : No appreciable changes in hand wrist X-ray

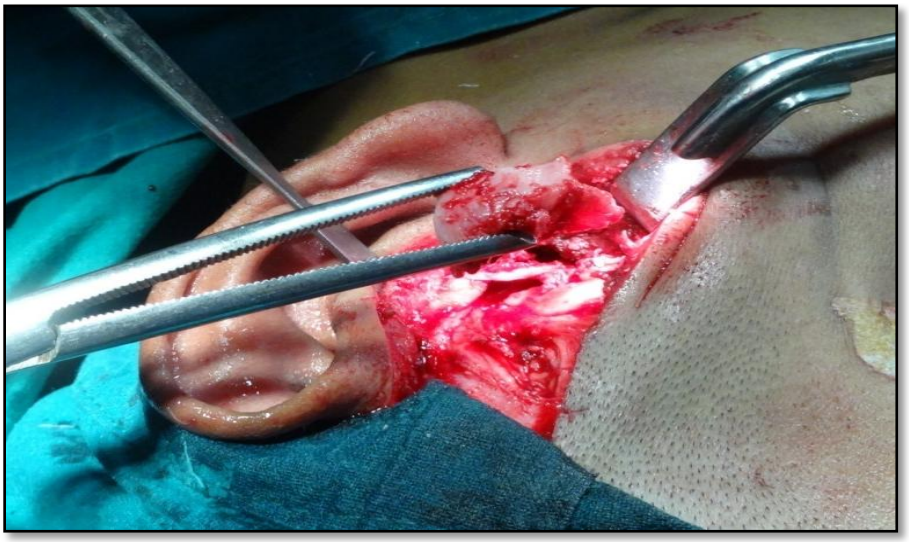

Fig 4 : Gap arthroplasty

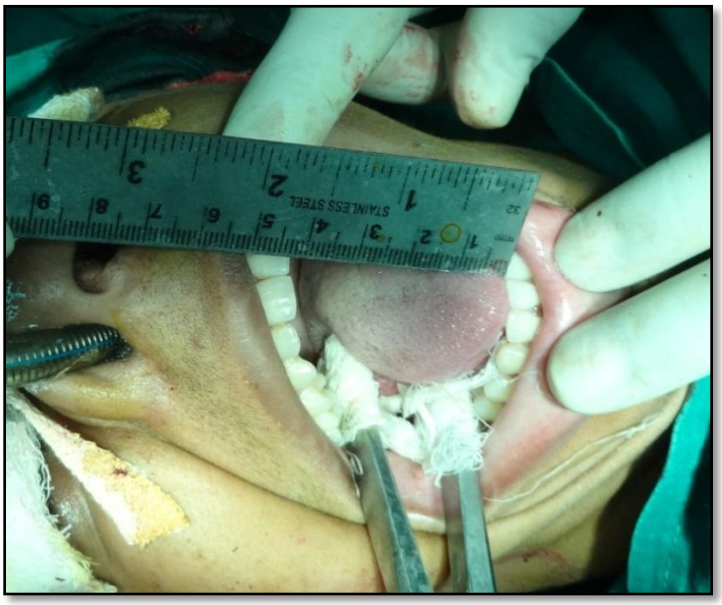

Fig 5 : 43 mm interincisal mouth opening was achieved intraoperatively

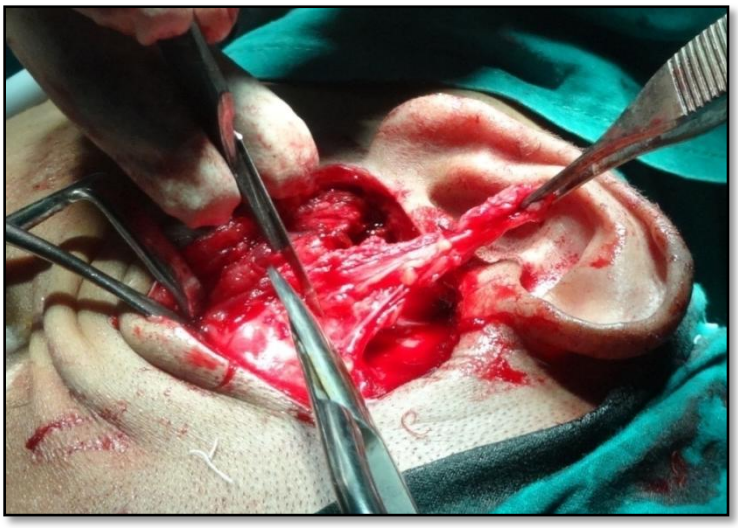

Fig 6 : Temporomyofacial flap was obtained

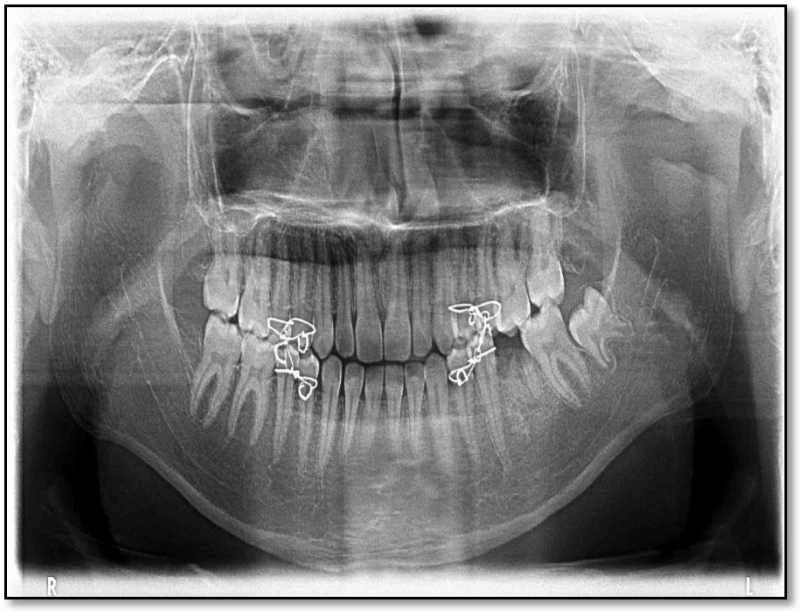

Fig. 7 : Postoperative OPG

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