

Concomitant Temporomandibular Joint Ankylosis with Kyphoscoliosis: A Rare Case Report

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ABSTRACT

Background: Kyphoscoliosis is known as the musculoskeletal disorder with an abnormal curvature of the spine. Concomitant TMJ ankylosis and kyphoscoliosis is a rare occurrence which affected the quality of life of the patient in terms of curved body posture, impaired speech clarity, reduced food intake and deviated facial symmetry. Hereby we are presenting with a rare case of a 13 years old child of idiopathic congenital kyphoscoliosis reported with chief complain of limited mouth opening (hypomobility). The hypomobility of mandible was diagnosed as the temporomandibular joint (TMJ) ankylosis. Surgical intervention was the treatment of choice. Ankylotic bone-fibrous mass was excised followed by interposition arthroplasty under general anaesthesia. The initial maximal incisal opening (MIO) was < 03 mm and reached up to 33 mm. Deviated facial symmetry, food intake, and speech clarity have been improving significantly over the months. However, congenital kyphoscoliosis and delayed treatment severely affected the quality of life.

Keywords: Kyphoscoliosis; temporomandibular joint (TMJ) ankylosis; gap arthroplasty; interpositional Arthroplasty.

INTRODUCTION

Ankylosis of temporomandibular joint is an osseous or fibrous fusion of the disc-condyle complex to the temporal articular surface results in hypomobility of the TMJs, which affects the quality of life in terms of impairment of speech, reduced food intake, deviated facial symmetry, dental malocclusion etc. The degree of hypomobility and disabling condition is directly related to the severity of the above sequels. The TMJ ankylosis treatment in medical practice has been continuing for the last 200 years with several surgical and management practices. However, the current improvement of surgical outcome has been critically evaluated and reviewed by several authors time and again^{1,2,3}. The ankylosed TMJ can be classified as partial or

complete based on the degree of mobility and intracapsular or extracapsular as per the fusion site. Involvement of tissues like fibrosis, osseous, or both (fibro-osseous) is also referred to different types of ankylosis⁴. Vasconcelos *et al.* in their review described four types of TMJ ankylosis involving bony fusion and fibrous adhesion around the joint⁵. Pseudo ankylosis may also be seen due to coronoid process hyperplasia, malformation of zygomatic or maxillary bone at the extra-articular site. There are reports of pseudo-TMJ ankylosis due to scoliosis where two or more cervical vertebrae are fused⁶. Restricted mouth opening and change of range of motion related to the temporomandibular joint are also correlated with scoliosis in literatures⁷. TMJ

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Ankylosis may be congenital or acquired. The nonspecific trauma such as forceps delivery, blow to the chin, the condylar fracture is reported in a paediatric patient more than the true congenital one. Infectious and inflammatory diseases like rheumatoid arthritis, septic arthritis, otitis media, mastoiditis, parotitis, osteomyelitis, osteoarthritis, tonsillitis etc. are also considered as the potential etiological factors for acquired TMJ ankylosis in children^{2,8}. Although TMJ ankylosis is mostly manifested in childhood, the adult occurrences are not uncommon. Several hospital-based studies indicate the high prevalence of TMJ ankylosis in developing countries, including India among the population of poor socio-economic status⁹. The present study of TMJ ankylosis is a rare case as a concomitant to kyphoscoliosis.

CASE REPORT

A 13 yr old scolio-kypho male child (Fig 1) was presented at Hi-Tech Dental College and Hospital with a chief complaint of limited mouth opening (hypomobility), which was progressive since last 8 years (Fig 2). The hypomobility impaired the speech and affected the food intake up to a considerable extent. Parent revealed his history of spontaneous and natural vaginal delivery as a breech baby at 36 weeks of gestation with congenital kyphoscoliosis. There was no other family history of congenital scoliosis or the TMJ ankylosis. The patient was the elder child of two siblings, and his parents are non-consanguineous. The child had achieved all his milestones with slow progression. Parents narrated the history of recurrent infection in his right ear. The family had a poor social, economic background.

External examination elicited a compressed and twisted heap, the facial asymmetry with short lower facial height, convex right cheek and ipsilateral deviation of the chin (Fig 2). Intraoral examination revealed the dental malocclusion with a maximal incisal opening (MIO) of < 3 mm. Lateral open bite with left occlusal cant towards the right side was also recorded. Radiographic investigations included orthopantomogram (OPG), AP/Lat views of the hip joint and AP/Lat view of the spine. The orthopantomogram revealed incomplete condyle formation, non-radiographic joint space and a prominent antegonial notch on the right side (Fig 3). Kyphoscoliosis was clearly marked with the shallow acetabulum, resorption of femoral heads

migrated (left>right) femur and spina bifida at L4-L5 level. CBC report revealed sub-optimal values for Hb, HCT, MCV and MCH.

With sceptical aetiology, the child was diagnosed as a subject of temporomandibular joint (TMJ) ankylosis. Surgical intervention was the treatment of choice to make the TMJ functional. Correction of ankylosis to restore mouth opening, impaired speech and deviated facial symmetry were the objectives of the treatment with prognostic prevention of recurrence. Intervention such as gap arthroplasty, interposition arthroplasty, post-surgery physiotherapy, and correction of malocclusion were some of the main treatment plans. Written consent was taken from the patient and his guardian for the surgical procedure to prevail.

The surgical site of the patient was demarcated at the right temporal and preauricular region. Surgery was performed under general anaesthesia. Administration of anaesthesia for a patient with minimal mouth opening is always challenging. Intubation with a tracheostomy was done to secure the airways. The ankylotic site was accessed by Popowich & Crane's modification of Alkayat & Bramley's preauricular incision procedure¹⁰. The flap was then raised up in a downward and forward direction along the external auricular cartilage to reach the zygomatic arch (Fig 4). Care was taken to prevent the injury of the zygomatic branch of the facial nerve. The ankylosed part was exposed, and the Ankylotic bone- fibrous mass was excised by osteotomy (using bone chisel and mallet to prevent injuries to the internal maxillary artery or pterygoid plexus of veins) (Fig 5). In addition to the ipsilateral coronoidectomy contralateral intra-oral coronoidectomy was also done at left side. Thus by gap arthroplasty, a gap of 15mm was created between the right glenoid fossa and the mandible. However, the objective was achieved as an interoperative active mouth opening over 40 mm. The interposition arthroplasty was done where temporalis fascia was harvested and pushed over zygomatic arch into the temporomandibular joint. This was intended to maintain the vertical height of ramus and to prevent a recurrence. The flap was secured using sutures with the medial tissues using 3-0 vicryl. Post-operation schedule of physiotherapy for TMJ to enhance and maintain the

MIO was recommended. On the next day of the

operation,

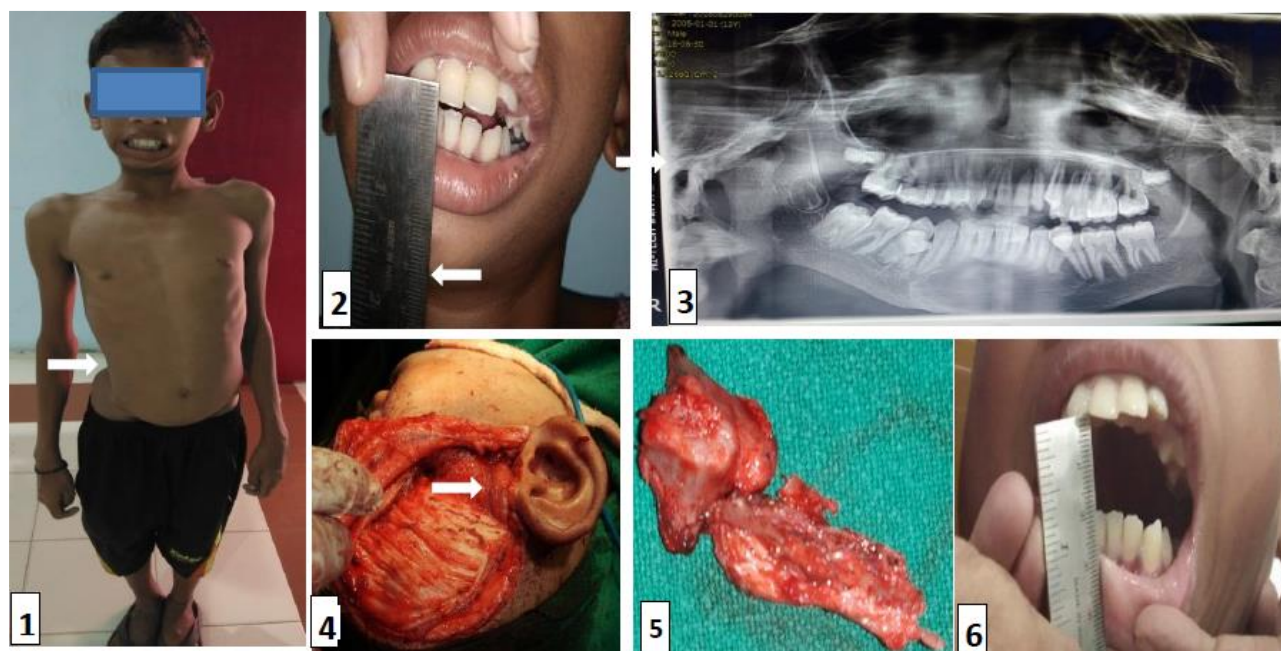


Fig 1: Kyphoscoliosis condition (arrow), Fig 2: Deviated chin to ipsilateral to TMJ ankylosis and Initial MIO < 3mm (arrow), Fig 3: Orthopantomogram showing TMJ ankylosis (arrow), Fig 4: Temporalis muscle interpositional arthroplasty (arrow), Fig 5: Ankylosed mass excised, Fig 6: MIO > 20mm during follow up months.

a mild exercise of the jaw was started with chewing gums and fingers. Latter on the systematic exercise of the jaw using wooden spatula blocks of increasing thickness was practised. Post-operatively follow-up recorded 28 mm and 33 mm MIO after one and three months of surgical intervention, respectively (Fig 6). The patient was referred to the department of orthodontics for occlusal correction and to the department of otolaryngology to treat the chronic otitis media. The patient was followed up, and both verbal and written consent was taken for the use of his clinical data and images for academic purpose only.

DISCUSSION

Kyphoscoliosis is known as the musculoskeletal disorder with an abnormal curvature of the spine. Scoliosis with the temporomandibular disorder have been reported and addressed with surgical and or aggressive physiotherapy ^{6,7}. Concomitant TMJ ankylosis and kyphoscoliosis is a rare occurrence which affected the quality of life of the present patient in terms of curved body posture, impaired speech clarity, reduced food intake and deviated facial symmetry. Kyphoscoliosis of the subject is congenital, which made us skeptical about

identifying the aetiology of TMJ ankylosis. Rayan et al³, in a recent review, described the TMJ ankylosis occurrence as congenital (neonatal) and pediatric expression. In the congenital anomaly cases, they attributed the aetiology of neuromuscular disorder during embryonic life. They described difficult forceps delivery as the acquired neonatal trauma rather than the true congenital. As the subject was delivered as a breach baby, it did not require forceps for the purpose. Hence he might have acquired TMJ ankylosis during his early pediatric period. No doubt the baby was born as congenital kyphoscoliotic, but congenital and neonatal etiologies for TMJ ankylosis are not seen. Local or systemic infection can cause TMJ ankylosis. Several reports and reviews considered infection as one of the prime aetiology of TMJ ankylosis^{11,12}. Those studies described that inflammation and infection might be due to otitis media, mastoiditis, osteomyelitis of mandible or from some systemic diseases through the blood. A recent report revealed that inadequate treatment for staphylococcus infection of some systemic disease caused TMJ ankylosis in three years she child ¹³. In the present study, the medical history of chronic fever and otitis media with delayed or inadequate

treatment during his early childhood might have caused the ankylosis in temporomandibular joints. Effective surgical intervention and post-surgical management reduce the chances of reankylosis. GA is a short duration and low-cost routine type of TMJ surgery where the ankylotic mass is removed, creating a gap of at least 10 mm between ascending ramus and temporal bone. The surgery may also extend to ipsilateral and or contralateral coronoidectomy. In a meta-analysis study, Junil et al¹⁴ reported the greater possibilities of reankylosis in GA than the IA. In another case study of fifty patients, Maqsood et al.¹⁵ applied codylectomy followed by temporalis muscle flap interpositioning and recorded an average MIO of 31.3 mm on the fifth day of post-op follow up. In the present study codylectomy with ipsilateral and contralateral coronoidectomy could make a gap of 15mm between the right glenoid fossa and the mandible. The GA was followed by IA and MIO was recorded up to 33 mm during 3rd month follow-up. However, postoperative recurrence of TMJ ankylosis are mostly reported when the ankylotic mass is not properly excised, or the patient does not have adequate physiotherapy^{5,9}. In scoliosis, the malalignment of pelvic and lumbar bones affect the thoracic bones and spinal muscle performance which may cause hypomobility of TMJ up to some extent^{6,7}. In this context, kyphoscoliosis of the present study might have contributed for TMJ hypomobility up to some extent. Poor socio-economic status is well reflected on TMJ ankylosis in many reviews mainly due to non-access to medical treatment or for delayed treatment¹³. A similar situation could have delayed the present subject up to 13 years of age.

CONCLUSION

This case revealed that chronic otitis media and scoliosis might have contributed to TMJ ankylosis fully or partially. However, the appropriate surgical interventions could restore facial symmetry deviation, normal food intake and speech clarity and improved the quality of life of the subject.

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

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