Case Report

Unilateral temporomandibular joint ankylosis with contralateral aplasia

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ABSTRACT

The temporomandibular joint (TMJ) is considered as one of the complex joints of the human body. Mandibular condylitis is distinguished from condylar aplasia by its non-association with aural/facial anomalies, and also as normal development appears to proceed until the lytic event occurs. It is further distinguished from primary and secondary condylar hypoplasia by the following: Absence of condyle rather than it being small, the normal development appears to proceed until the lytic event occurs, and its non-association with aural/facial anomalies or temporomandibular ankylosis. In the present report, a patient with a unilateral missing mandibular condyle with contralateral TMJ ankylosis is presented and the treatment is outlined and discussed.

Key words: Ankylosis, condyle, temporomandibular

INTRODUCTION

The temporomandibular joint (TMJ) is considered as one of the complex joints of the human body. The initial functions of TMJ start at the 20th week of the fetal stage, when mouth opening movements appear. The development process is not completed until the 12th year of life. Varying degrees of condylar hypoplasia, from minimal to complete absence named as condylar aplasia, may occur due to abnormal development and growth of TMJ. The most common causes of condylar alterations are inflammatory process in the area, rheumatoid arthritis, and radiotherapy. The parathyroid hormone – related protein also affects the bone formation and chondrocyte differentiation and, consequently, the condyle formation. Condylar resorption, or condylysis, can be defined as progressive alteration of condylar shape and decrease in mass. As a result, most patients exhibit a decrease in posterior face height, retrognathism, and progressive anterior open bite with clockwise rotation of the mandible. Although the cause is unknown, condylar resorption has been associated with rheumatoid arthritis, systemic lupus erythematosus, steroid usage, trauma, neoplasia, orthodontic treatment, and orthognathic surgery. In most cases, however, there is no identifiable precipitating event and hence the term idiopathic condylar resorption. This condition appears to have a predilection for females in the age range of 15-35 years with preexisting TMJ dysfunction and high mandibular plane angle. The condition is usually bilateral. Bone scintigraphy of the mandibular condyles may be used to evaluate ongoing resorption. Pigeon breast [pectus carinatum (PC)] is a protrusion deformity of the chest which is not noticed until the child is a year or more of age. It then continues to increase for a variable length of time, often over a period of years, and may reach its maximum in the teens. The deformity presents typical progressive growth, and can be accompanied or not by cardiorespiratory symptoms. Here, we present a patient with a unilateral missing mandibular condyle with contralateral TMJ ankylosis, and outline the treatment and discuss it along with DentaScan findings.
**Case Report**

**History with clinical and radiographic examination**

A 6-year-old female patient reported to our outpatient department with the complaints of nil mouth opening, facial disfigurement, and night episodes of respiratory distress. On examination, the patient had no movements of the right TMJ, with pronounced digonial notches on either side or reduced chin [Figure 1]. Systemically, the patient had all the signs and symptoms of obstructive sleep apnea syndrome (OSAS) with PC of the chest [Figures 2 and 3]. The patient’s chief complaint was respiratory distress with facial asymmetry, which was first noticed in childhood and gradually progressed. Panoramic examination revealed that the right mandibular condyle exhibited ankylotic changes, while there was complete absence of the left condylar head and the ramus terminated relatively obliquely below the level of the sigmoid notch [Figure 4]. For obtaining further information, the patient was submitted to a DentaScan evaluation. The images revealed the complete absence of the left condylar head and neck [Figure 5]. On evaluating the patient for excluding the syndromes, the chest exhibited PC (pigeon breast) with shallow breathing and chest expansions. PA chest revealed a barrel-shaped chest with normal lung and heart fields. The patient was further evaluated for episodes of apnea and snoring.

**Treatment**

Preparations were made for emergency access to the airway. IV line was established and the patient was sedated for blind intubation. After successful blind nasotracheal intubation from the left nostril, access to bony chunk on the right TMJ region was made with the help of AlKayat–Bramley incision and the bony chunk was relieved. Intraoperative mouth opening of 3.5 cm was achieved and the gap was interpositioned with temporal fascia. Layer wise closure was done. Post-surgical active jaw physiotherapy was started after 48 h of surgery. Mouth opening was maintained at 3.2 cm after 15 days of follow-up.

**Discussion**

Atypical mandibular condyles are generally classified in terms of aplasia, condylysis, hypoplasia, hyperplasia, and bifold condyle. The most common etiologic factors affecting the condylar region are trauma during growth, inflammatory conditions like juvenile arthritis, and...
In our case, besides aplasia of the mandibular condyle, there were no other commonly found syndromes or soft tissue manifestations. Condylar aplasia of developmental origin occurs due to defects in the first branchial arch.\[^{16}\]

In 2001, Katsavrias et al. conducted a study on the growth of the articular eminence height during the postnatal craniofacial growth period of 90 skulls at different ages and concluded that the articular eminence height grows at a very high rate until the age of 7 years (the period of deciduous dentition), almost stops at the age of 11 years (the period of mixed dentition), and acquires the rest of its height by the age of 20 years.\[^{18}\] If a condyle is present, a well-defined fossa and articular eminence should be present. So, in the case of absence of a condyle, those adjacent structures should also be absent. Total absence of the condyle points to an earlier period of condyle formation. In our case, there was no radiologically visible articular fossa or eminence suggesting incomplete development. This may constitute evidence that the defect originated during the prenatal or perinatal period of the patient’s life.\[^{11}\]

Atypical mandibular condyles can also be seen in connection with different syndromes of the head and neck. In these cases, there will, as a rule, also be soft tissue manifestations. In our case, there were no anomalies affecting either of the ear, eyelid, or any other soft tissue structures.

Atypical mandibular condyles are present in several syndromes, as observed in hemifacial microsomia, Goldenhar syndrome, Treacher Collins syndrome, Proteus syndrome, and auriculocondylar syndrome. But the condyle aplasia is extremely rare when not connected to any syndrome. No clinical or radiographic evidence favors any syndromic association in our case. The concept of mandibular condylysis was first introduced by Rabey\[^{11}\] who defined it as an acquired absence of the mandibular condyles, unassociated with TMJ ankylosis or aural/facial anomalies. He distinguished it from condylar aplasia and primary and secondary condylysis by the absence of condyle rather than it being small, and also, as normal development appeared to proceed until the lytic event occurred. The concept of secondary condylar hypoplasia versus condylysis does not appear to warrant their classification as separate entities, since they are merely degrees of destruction along a continuous spectrum. Condylar hypoplasia is defined by apnea–hypopnea index (the total number of episodes of apnea and hypopnea per hour of sleep), obstructive sleep apnea syndrome (OSAS, the syndrome characterized by episodes of upper airway obstruction during sleep, and nocturnal hypoxemia. It is defined by apnea–hypopnea index (the total number of episodes of apnea and hypopnea per hour of sleep), or respiratory disturbance index, of five or higher in association with excessive daytime somnolence.\[^{21}\]

In our case, patient had nil mouth opening and anterior deep bite due to contralateral TMJ ankylosis. This case is unique in that it has unilateral TMJ ankylosis and...
contralateral TMJ aplasia without any syndromic features in other soft and hard tissues. The timing and regimen of surgery is still an issue to be resolved. A costochondral rib graft can be used to help establish an active growth center. Condylectomy and reconstruction with either autogenous materials, for example, sternoclavicular grafts, or alloplastic materials, represent other treatment modalities. Early and aggressive surgery is the essential part of the treatment.

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How to cite this article: Singh N, Pal US, Mohammad S, Singh RK, Mehta G, Makadia HS. Unilateral temporomandibular joint ankylosis with contralateral aplasia. Natl J Maxillofac Surg 2013;4:256-9.

Source of Support: Nil. Conflict of Interest: None declared.