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 condylysis 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.

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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.[1] 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.[2] The parathyroid hormone – related protein also affects the bone formation and chondrocyte differentiation and, consequently, the condyle formation.[3,4] 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 erythematosis, steroid usage, trauma, neoplasia, orthodontic treatment, and orthognathic surgery.[5-10] In most cases, however, there is no identifiable precipitating event,[11] 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.[12,13] Bone scintigraphy of the mandibular condyles may be used to evaluate ongoing resorption.[14] 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.[15] 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.

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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 antigonial 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.

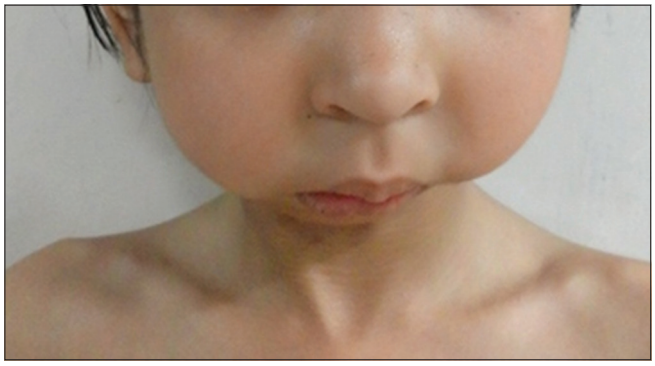

Figure 1: Frontal profile view showing retruded chin

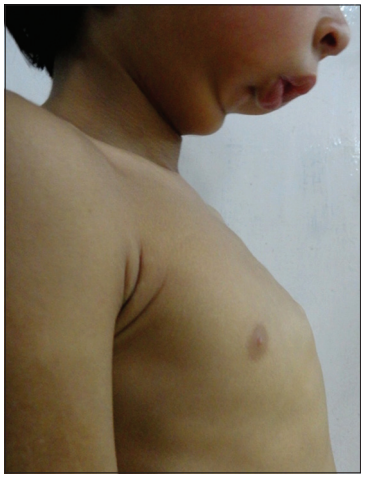

Figure 3: Side view of chest showing prominent sternum

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 bifid condyle. The most common etiologic factors affecting the condylar region are trauma during growth, inflammatory conditions like juvenile arthritis, and

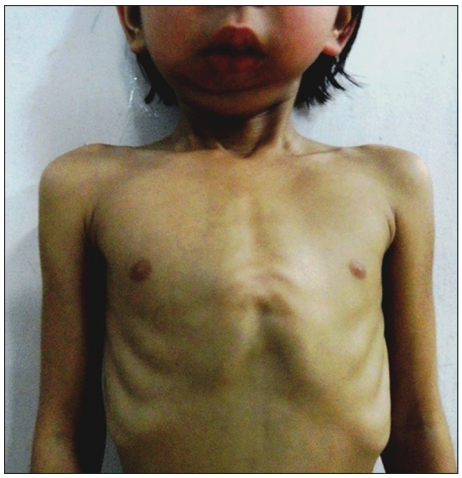

Figure 2: Frontal view of chest showing prominent upper sternum

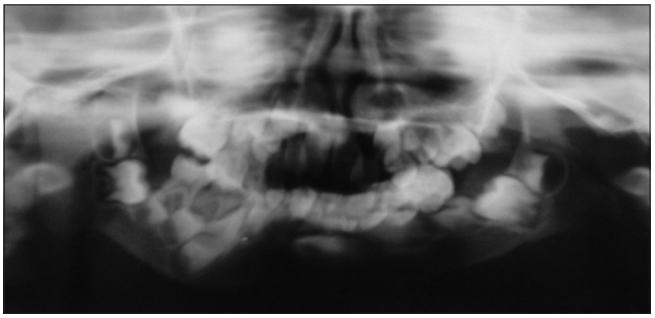

Figure 4: OPG showing absence of condylar head and neck with ankylotic changes on the contralateral side

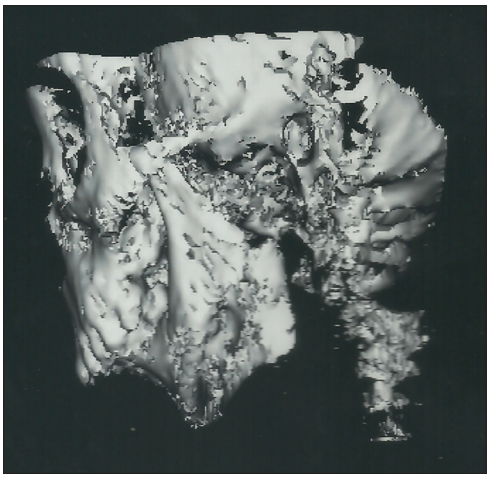

Figure 5: Denta scan (CBCT) of the patient showing absence of condylar process

radiotherapy. In the case of trauma, it must have happened before the age of 2 years.[16] The condylar anomalies may also be caused by any unknown cause, parathyroid hormone–related protein deficiency, or may be associated with any syndromic condition. Our case had no history of trauma or infection. Santos *et al*. reported a case of aplasia of the mandibular condyle with unknown etiology and claimed it to be of developmental origin.[17] Prowler *et al*. reported a case with condylar hypoplasia and claimed it to have an acquired origin because the condition obviously started after the age of 6.[18] 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.[19] 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.[1]

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 condylar hypoplasia 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. Condylysis would appear to be an appropriate term for use in cases where there is an acquired "lysis" of the mandibular condyle, whether or not there is destruction to the point of complete absence. Due to the aplasia of condyle of one side and ankylosis of the contralateral side, the growth of the mandible is affected leading to micrognathia and retruded chin, thus reducing the tongue space and pharyngeal spaces resulting in episodes of apnea. The episodes of apnea are aggravated by the simultaneous presence of PC. People with PC usually develop normal hearts and lungs, but the deformity may prevent these from functioning optimally. In moderate to severe cases of PC, the chest wall is rigidly held in an outward position. Thus, respiration is inefficient and the individual needs to use the diaphragm and accessory muscles for respiration, rather than normal chest muscles, during strenuous exercise. This negatively affects gas exchange and causes a decrease in stamina. The PC deformity is not rare, affecting 1 in 1000 adolescents. Symptoms result from associated diseases, such as bronchitis and bronchial asthma, or emotional disorders, caused by the unaesthetic appearance of the chest wall.[20] The OSAS is a potentially disabling condition characterized by excessive daytime sleepiness, disruptive snoring, repeated 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.

References

1. Speber GH. Temporomandibular joint. In: Speber GH. Craniofacial development. Hamilton: BC Deker; 2001. p. 139-43.
2. Neville BW, Damm DD, Allen CM, Bouquot JE. Developmental defects of the oral and maxillofacial region. In: Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 2nd ed. Philadelphia: WB Saunders; 2002. p. 17-8.
3. Shibata S, Suda N, Fukada K, Ohyama K, Yamashita Y, Hammond VE. Mandibular coronoid process in parathyroid hormone-related protein-deficient mice shows ectopic cartilage formation accompanied by abnormal bone modeling. Anat Embryol (Berl) 2003;207:35-44.
4. Ishii-Suzuki M, Suda N, Yamazaki K, Kuroda T, Senior PV, Beck F, *et al.* Differential responses to parathyroid hormone-related protein (PTHrP) deficiency in the various craniofacial cartilages. Anat Rec 1999;255:452-7.
5. Ogden GR. Complete resorption of the mandibular condyles in rheumatoid arthritis. Br Dent J 1986;160:95-7.
6. Ramon Y, Samra H, Oberman M. Mandibular condylosis and apertognathia as presenting symptoms in progressive systemic sclerosis (scleroderma). Pattern of mandibular bony lesions and atrophy of masticatory muscles in PSS, presumably caused by affected muscular arteries. Oral Surg Oral Med Oral Pathol 1987;63:269-74.
7. Iizuka T, Lindqvist C, Hallikainen D, Mikkonen P, Paukku P. Severe bone resorption and osteoarthrosis after miniplate fixation of high condylar fractures. A clinical and radiologic study of thirteen patients. Oral Surg Oral Med Oral Pathol 1991;72:400-7.
8. Lindqvist C, Söderholm AL, Hallikainen D, Sjövall L. Erosion and heterotopic bone formation after alloplastic temporomandibular joint reconstruction. J Oral Maxillofac Surg 1992;50:942-9.
9. Philips RM, Bell WH. Atrophy of mandibular condyles after sagittal ramus split osteotomy: Report of case. J Oral Surg 1978;36:45-9.
10. Bouwman JP, Kerstens HC, Tuinzing DB. Condylar resorption in orthognathic surgery. The role of intermaxillary fixation. Oral Surg Oral Med Oral Pathol 1994;78:138-41.
11. Rabey GP. Bilateral mandibular condylysis: A morphanalytic diagnosis. Br J Oral Maxillofac Surg 1977;15:121-34.
12. Moore KE, Gooris PJ, Stoelinga PJ. The contributing role of condylar resorption to skeletal relapse following mandibular advancement surgery: Report of five cases. J Oral Maxillofac Surg 1991;49:448-60.
13. Kerstens HC, Tuinzing DB, Golding RP, van der Kwast WA. Condylar atrophy and osteoarthrosis after bimaxillary surgery. Oral Surg Oral Med Oral Pathol 1990;69:274-80.
14. Pogrel MA, Kopf J, Dodson TR, Hattner R, Kaban LB. A comparison of single-photon emission computed tomography and planar imaging for quantitative skeletal scintigraphy of the mandibular condyle. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1995;80:226-31.
15. Fonkalsrud EW, Anselmo DM. Less extensive techniques for repair of pectus carinatum: The undertreated chest deformity. J Am Coll Surg 2004;198:898-905.
16. Kaneyama K, Segami N, Hatta T. Congenital deformities and developmental abnormalities of the mandibular condyle in the temporomandibular joint. Congenital Anomalies 2008;48:118-25.
17. Santos KC, Dutra ME, Costa C, Lascala CA, de Oliviera JX. Aplasia of mandibular condyle: Case report. Dentomaxillofac Radiol 2007;36:420-2.
18. Prowler JR, Glassman S. Agenesis of the mandibular condyles; diagnostic findings and treatment of deformity by polyethylene implant. Oral Surg Oral Med Oral Pathol 1954;7:133-9.
19. Katsavrias EG, Dibbets JM. The growth of articular eminence height during craniofacial growth period. Cranio 2001;19:13-20.
20. Coelho Mde S, Guimarães Pde S. Pectus carinatum, J Bras Pneumol 2007;33:463-74.
21. Casale M, Pappacena M, Rinaldi V, Bressi F, Baptista P, Salvinelli F. Obstructive sleep apnea syndrome: From phenotype to genetic basis. Curr Genomics 2009;10:119-26.

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