International Journal of Innovative Research In Medical Science (IJIRMS)
Volume 01, Issue 07, Sept-2016, ISSN No. – 2455-8737
Available Online at – www.ijirms.in

Condylar Agenesis: A Rare Case Report

Shakti Agarwal[1], Kamini Kiran[*2], Shailesh Kumar[3], Rameshwar Singh[4], Gourab Das[5]

[1]MDS, Department of Oral Medicine and Radiology, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

[*2]MDS, Department of Oral and Maxillofacial Pathology & Microbiology, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

[3]MDS, Department of Oral and Maxillofacial Surgery, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

[4]Professor, Department of Prosthodontics Crown Bridge and Implantology, Azamgardh Dental College, Uttar Pradesh, India

[5]PG Student, Department Of Oral and Maxillofacial Surgery, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

Abstract:

Condylar agenesis is a rare case involving development, growth and aesthetic of face as well as dentition development and occlusal problems. In the absence of any long term study and shortage of subject material, management protocol of problems associated with agenesis of condyle vary from institute to institute and there is no internationally accepted protocol for the same. We are presenting a case of unilateral condylar agenesis with severe malocclusion that has never been published before. Subject in discussion is a 25 year old boy with agenesis of left condyle and perfectly normal right side condyle resulting in deviation towards left side of face.

Keywords: *- Condylar agenesis, condyle, unilateral.*

Introduction

The temporomandibular joint (TMJ) is a ginglymus diarthrodial joint, one of the most complex joints of the human body which is capable of both rotational and translatory movements. It consists of the mandibular condyle and the articular eminence of the temporal bone. The condyle is very important because it controls the expression of mandibular growth. Like other diarthrodial joints, TMJ lags morphologically during prenatal life behind other synovial joints in both in its progress and in timing of its appearance, so that at birth the joint is still largely underdeveloped. The TMJ first appears in the 8th week of gestation, when two separate areas of mesenchymal blastemas appear near the eventual location of the mandibular condyle and glenoid fossa [1, 2]. At approximately the 10th gestational week bone and cartilage are first seen in the mandibular condyle. First condylar blastema developed from which the mandibular condyle cartilage. Next is the temporal blastema, which eventually forms the articular surface of the temporal component and the structures of the upper portion of the joint. By the 12th week the mandibular condyle and temporal blastemas begin their growth at relatively distant sites; they then move towards each other as the joint develops. At time of birth, the articular surfaces of both the mandibular condyle and temporal bones are covered with fibrous connective tissue. Later, this tissue is slowly converted to fibrocartilage as the fossa deepens and the mandibular condyle develops under functional influences [3, 4]. Growth disturbances such as aplasia, hypoplasia or agenesis of the mandibular condyle in the development of mandibular condyle may occur in uterus late in the first trimester. Underdevelopment or defective formation of condyle may be congenital or acquired. Condylar agenesis is congenital non formation of condyle while hypoplasia is poor development. Although both are distinct entity yet wrongly but very often these terms are used interchangeably. It is difficult to ascertain whether non formation of condyle is congenital or due to condylar mal development in adult age group. During our search we were unable to find any substantial study over the topic.

History

A 25 year old male presented in O.P.D with complain of facial asymmetry which was increasing with age (Fig-1(a,b)). Patient had history of normal birth with no history of trauma to facial region later in life. There was no history of any hormonal disturbance or premature birth. Any relevant positive family history was absent. He had absolutely no problem with mastication and was unaware of his severe malocclusion.

Corresponding Author - Kamini Kiran, - MDS, Department of Oral and Maxillofacial Pathology & Microbiology, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

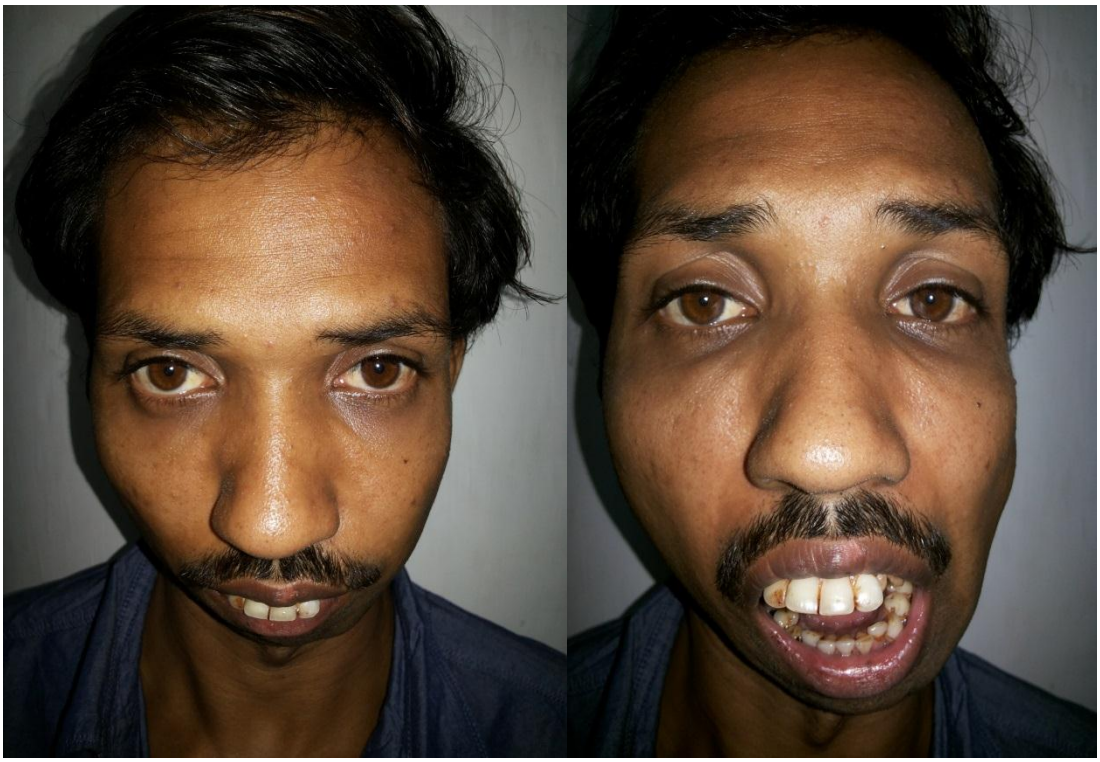

Fig. 1(a) **fig. 1(b)**

Clinical Examination

General physical examination had no significant finding related to case. On local examination- deviation towards left was obvious, maxillary anterior teeth were protruded, midline shifted, chin retruded and molars were in class 2 malocclusion with lingual tilt giving a bird face appearance. Absence of normal condylar contour on palpation of left side condylar region. Further examination confirmed absence of lateral pterygoid as well as temporalis and poorly developed masseter with prominent antigonial notch on left side. Patient had restricted side to side movements but mouth opening and protrusion were with in normal range (of course with deviation). Though there was no abnormal finding related to zygoma, ear or frontal bone. Presence of autoimmune disease like rheumatism or scleroderma was also ruled out.

Radiological Findings

OPG shows missing condyle and underdeveloped ascending ramus on left side, Glenoid fossa was not developed on the left side(Fig-2).
PA view shows deviation of chin to left (Fig-3).

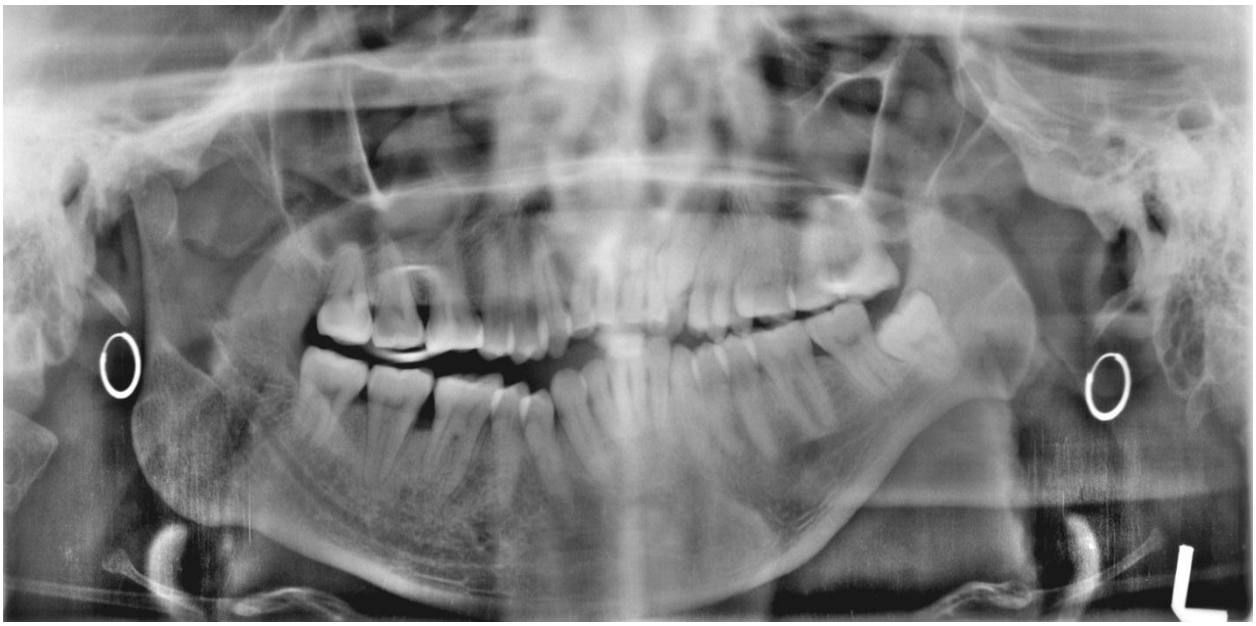

Fig. 3

Corresponding Author - Kamini Kiran, - MDS, Department of Oral and Maxillofacial Pathology & Microbiology, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

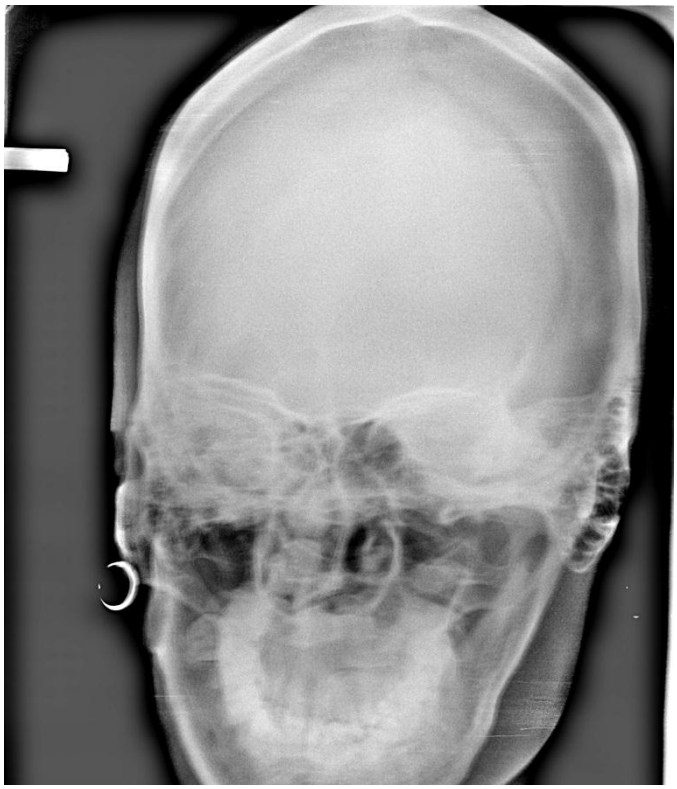

Fig. 4

Identification of Problem and Management

Patient was advised complete systemic evaluation and referred to general medicine, cardiology, ophthalmology, ENT, and orthopedics to rule out any syndromes. Medical evaluation revealed no abnormalities. Based on the clinical and radiographic findings, a final diagnosis of non syndromic agenesis of the left condyle was given. Patient was referred to oral surgeon and orthodontist for the best possible treatment.

Discussion

There has been sporadic case reports of condylar agenesis but to the best of our knowledge and there is no substantial long term study with reliable data in literature to describe any surgical treatment. More often condylar agenesis reports have been related to Goldenhar's syndrome (hemifacial microsomia) and there has been no concrete explanation of why condylar agenesis takes place. Etiology varies from trauma during delivery to genetic aberrations to disturbance in mesenchymal cells from which condyle get formed. Difference between condylar agenesis and hypoplasia is that in agenesis there is no condyle or articular disc present right since birth while in hypoplasia condyle is rudimentary or maldeveloped may be due to genetic or acquired factors. Another term condylolysis means loss of condylar structure after its complete formation and is completely different from previous two. Main aim in an adult case of condylar agenesis is to establish a stable esthetic and functional rehabilitation.

The congenital deformities and developmental abnormalities of the mandibular condyle can be classified as hypoplasia or aplasia, hyperplasia, and bifidity. Hypoplasia or aplasia of the mandibular condyle indicates underdevelopment or non-development associated mainly with various craniofacial abnormalities. These may be either congenital or acquired [5]. Primary (or Congenital) condylar hypoplasia is characterized by unilateral or bilateral underdevelopment of the mandibular condyle and usually occurs as a part of some systemic condition originating in the first and second branchial arches, such as Mandibulofacial dysostosis (Treacher Collins syndrome), Hemifacial microsomia (first and second branchial arch syndrome), Oculoauriculovertebral syndrome (Goldenhar syndrome), Oculomandibulodyscephaly (Hallermann- Streiff syndrome), Hurler's syndrome, Proteus syndrome, Morquio syndrome and Auriculocondylar syndrome [5]. As a rule, in each of these conditions some soft tissue manifestations accompany the condylar agenesis and/or condylar malformations. Development may be arrested in the secondary (or Acquired) condylar hypoplasia is due to injury to condyle during active growth. The most common causes are mechanical injury, such as infection of the joint itself or the middle ear, trauma (before the age of 2), radiotherapy, childhood rheumatoid arthritis, and parathyroid hormone-related protein deficiency which affect bone formation and chondrocyte differentiation [5]. Any defined etiology is not confirmed by several authors for the mandibular deficiency. Aplasia of the mandibular condyle without any other facial

Corresponding Author - Kamini Kiran, - MDS, Department of Oral and Maxillofacial Pathology & Microbiology, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India

malformations is an extremely rare condition. The cases of nonsyndromic mandibular condyle aplasia have been previously reported by Krogstad , Prowler and Glossman , Akihiko et al. , Santos et al., Bowden Jr. and Kohn, Canger and Celenk and so forth. At about the 8th week of conception the TMJ develops from initially widely separated temporal and condylar blastema. By about the 20th week of intrauterine life eventually they grow towards each other and ossify to form a functional joint [5]. In our case, total absence of the condyle and glenoid fossa on the left side constitute an evidence that the defect originated in the prenatal period. Most of the time it is treated by multimode with the help of oral surgeon, general surgeon, plastic surgeon, and orthodontist. The treatment could then be a costochondral graft transplant, preferably before the growth spurt, orthognathic surgery at the end of the growth period, or both. Krogstad reported that effective results were obtained through the application of a form of orthodontic activator which aimed to swing the mandible to the unaffected side and promote formation of a mandibular condyle. Surgery is often required, but the timing and regimen of this choice is still an issue to be resolved.

Conclusion

In conclusion we report a rare case of total condylar agenesis on the left side not related to any clear pathological disorder. This case of unknown etiology was thoroughly examined; based on clinical and radiographic findings, we suggest that this case is of congenital origin. Non syndromic condylar agenesis is exceedingly very rare condition and very few case reports are published till date. In this context, our case is an important addition to the literature. Early detection and prompt treatment are imperative to restore esthetics and thus provide psychologic benefit to these patients.

References

[1] D. Buchbinder andA. S.Kaplan, “Biology,” in *Temporomandibular, Disorders Diagnosis and Treatment*, A. S. Kaplan and L. A. Assael, Eds., pp. 11–23, Saunders, Philadelphia, Pa, USA, 1991.

[2] J. W. Choi, J. T. Kim, J. H. Park et al., “gp130 is important for the normal morphogenesis ofMeckel’s cartilage and subsequent mandibular development,” *Experimental&MolecularMedicine*, vol. 39, pp. 295–303, 2007.

[3] J. F. Cleall, “Postnatal craniofacial growth and development,” in *Oral and Maxillofacial Surgery Volume One*, D. M. Laskin, Ed., pp. 70–107, Mosby, St Louis, Mo, USA, 1980.

[4] S. Pruzansky, “Postnatal development of craniofacial malformations,” *Journal of Dental Research*, vol. 47, no. 6, p. 936, 1968.

[5] K. Kaneyama, N. Segami, and T. Hatta, “Congenital deformities and developmental abnormalities of the mandibular condyle in the temporomandibular joint,” *Congenital Anomalies*, vol. 48, no. 3, pp. 118–125, 2008.
Case Reports in Dentistry 5

Corresponding Author - Kamini Kiran, - MDS, Department of Oral and Maxillofacial Pathology & Microbiology, Sardar Patel Post Graduate Institute of Dental and Medical Sciences, Uttar Pradesh, India