Case Report
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Symptomatic Bilateral Bifid Mandibular Condyle and Oligodontia in a Non-syndromic Patient - A Rare Case Report and Review of Literature

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Abstract

Bifidism of mandibular condyle is an uncommon anomaly of unknown etiology and uncertain pathogenesis. Usually it is asymptomatic and detected incidentally on a panoramic radiograph. However, it may be symptomatic where panoramic radiograph should be supplemented with computed tomography or magnetic resonance if surgical treatment is planned. The literature has reviewed many cases of bifid condyle mostly unilateral. The association of other dental abnormalities associated with bifid condyle has not been reported. We present a rare case of bilateral bifid condyle along with anterior disc displacement with reduction which also showed multiple missing teeth and retained deciduous teeth. This case report signifies the importance of both developmental and occlusal factors as an etiology for symptomatic temporomandibular joint.

Keywords: Bifid Condyle, Cone-beam Computed Tomography, Mandibular Condyle, Oligodontia, Temporomandibular Joint

1. Introduction

The Bifid mandibular condyle (BMC) also called as the 'bilobed condyle' or 'double headed condyle' is a rare anomaly, most commonly reported as an incidental finding. The first description of bifid condyle was given by Hrdlicka in 1941after he identified the same in 21 cadaveric skulls[1]. Since then it has been reported many times in dried skulls[2–5]. The first report of its occurrence in living individual was made by Schier in 1948[6] which was unilateral BMC. Bilateral Bifid Mandibular Condyle (BBMC) is even rarer. The first BBMC in living individual was reported by Shaber in 1987[7]. A literature search was carried out using PUBMED database with the keywords "bifid condyle", "bifid mandibular condyle", "bilateral bifid mandibular condyle". A total of 176 articles were found including case reports and studies and of that only bilateral bifid mandibular condyle cases or studies were selected. A total of 85 cases have been reported till now (Table 1) but the number of reported cases continue to accumulate due to introduction of advanced imaging modalities. Despite this, it is still considered to be as uncommon entity due to its asymptomatic clinical course and its incidental diagnosis on panoramic radiograph.

The pathogenesis of bifid condyle is broadly categorized into traumatic and developmental theories. The literature shows reports of BBMC in association with other developmental abnormalities[8–10]. However, BBMC in association with oligodontia has not been reported.

This case report describes symptomatic BBMC in association with oligodontia in a non-syndromic patient and reviews the possible etiopatho genesis along with a detailed review of literature.

2. Case Report

A male patient aged 35yrs reported to dental hospital with the complaint of left preauricular pain since 8 months. The pain was spontaneous in onset, progressive, moderate in intensity, continuous and was more pronounced on left side. It was radiating to the head, aggravated on chewing

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Table 1. Review of literature of (BBMC) bilateral bifid mandibular condyle

S. No.	Name of author/ Year	No. of cases/BBMC
1	Shaber et al.[7] 1987	1
2	Mc Cormick et al.[30] 1989	3
3	Kellenberger et al.[31] 1996	2
4	Stefanou et al.[32] 1998	4
5	Preda L et al.[33] 1999	1
6	Ghigi et al.[34] 2001	2
7	Artvinli and Kansu et al.[35] 2003	1(1 condyle bifid and other trifid)
8	Antoniades et al.[18] 2004	1
9	S. Alpasalan et al.[36] 2004	1
10	Espinosa-Femenia M et al.[37] 2006	1
11	Aggarwal et al.[38] 2006	1
12	Ackigoz et al.[39] 2006	1
13	Dennison et al.[16] 2008	1
14	Menezes AV et al.[11] 2008	2
15	Rehman TA et al.[8] 2009	4 cases with TMJ ankylosis
16	O Miloglu et al.[12] 2010	8
17	Gunduz et al.[40] 2010	1
18	Balaji SM[9] 2010	4 cases with TMj ankylosis
19	Almasan OC et al.[41] 2011	1
20	Melo S et al.[42] 2011	1
21	Sahman et al.[13] 2011	27
22	Tejasvi et al.[43] 2011	1
23	Tutar O et al.[44] 2012	1
24	Sahman et al.[14] 2012	3
25	Tanner JM et al.[45] 2012	1
26	Cho BH et al.[15] 2013	7
27	Jha et al.[46] 2013	1
28	Haghnegahdar AA et al.[47] 2014	3
	Total	85

and relieved on taking analgesic, anti-inflammatory medication. There was no history of trauma. On examination, the facial profile was straight. The mouth opening was reduced to 19mm. There was deflection of mandible towards the left side on opening with bilateral reciprocal click. Bilaterally lateral and posterior poles of condyle were palpable on extra-auricular and intra-auricular examination of TMJ respectively with tenderness present on left side (Visual analog scale 6). Bilateral lateral pterygoid muscles were tender on palpation.

Intraoral examination revealed missing 14,34,35,45, retained 52,53,55,63,64,65 and 75. The teeth 53,63 and 75 were carious. The maxillary anteriors were retroclined with reduced overjet and increased overbite resulting in traumatic anterior bite. (Figure 1) The molar relation was Class I. The clinical diagnosis was anterior disc displacement with reduction on left side.



Figure 1: Intraoral photograph showing mixed dentition and traumatic anterior occlusion.

Orthopantomogram (OPG) was advised which showed the absence of 12,13,14,15,23,24,25, 34 and 35. It also showed bilaterally bilobed condyle with a classical heart shape appreciated on the left side. Other appreciable features included haziness of right maxillary sinus (Figure 2).

The patient was advised cone beam computed tomography which confirmed the classical heart shape of bifid condyle bilaterally in cross sectional slicing (when the long axis was directed along the long axis of condylar head) along with the dental findings seen in OPG. The two condylar heads were appreciable in mediolateral direction separated by a groove in antero-posterior direction. The CBCT also confirmed the right maxillary sinusitis (Figure 3). The final diagnosis was anterior disc



Figure 2: Panoramic image showing the bilateral bilobed condyle and multiple retained deciduous teeth and oligodontia.

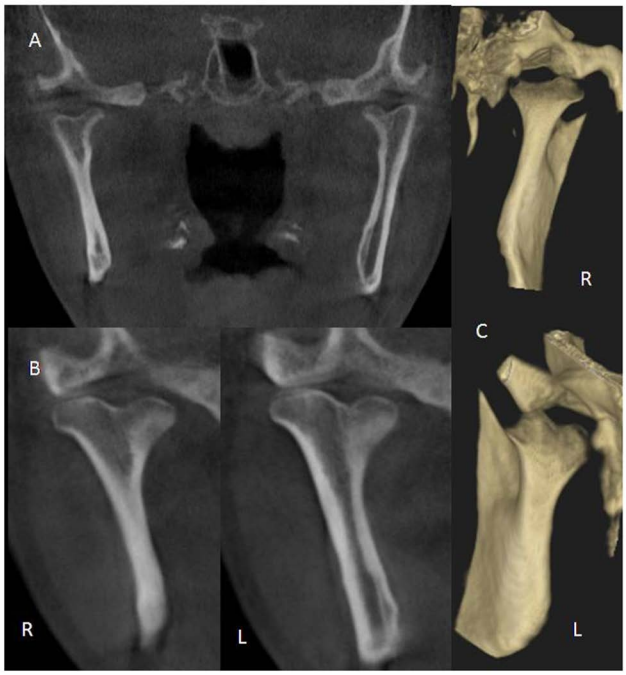


Figure 3: A)Coronal section showing the BBMC
B)Cross-sectional slicing showing beautifully heart shaped BBMC
C)3-D reconstruction showing BBMC

displacement with reduction secondary to bifid condyle and malocclusion.

A treatment plan was formulated which included anti-inflammatory agents, muscle relaxants and physiotherapy including transcutaneous electric nerve stimulation (TENS) therapy for 20 mins daily at low-energy and continuous mode. Patient was also advised hot and cold compressions for 15 mins daily at home. The restoration of carious teeth, extraction of retained teeth, replacement of missing teeth and correction of anterior occlusal abnormalities was advised. After 4 weeks of pharmacotherapy and physical treatment, the VAS reduced from 6 to 2 and mouth opening increased from 19 to 25 mm. Orthodontic treatment is still continuing and the patient is on continued follow up.

3. Discussion

Bilateral duplication of mandibular condyle is very rare. Very few epidemiological studies have been carried out to determine the prevalence of this anomaly and all showed varied results. In living subjects, Menezes et al.[11] reported 0.004% of BBMC cases in Brazilian population in 2008 while Miloglu et al.[12] reported prevalence of 0.07% in Turkish population in 2010 and 0.14% was shown by Sahman et al.[13] in Turkish population in 2011. In 2012, Sahman et al.[14] reported the prevalence of 0.54% of BBMC in Turkish population and 0.1% prevalence was found in 2013 by Cho et al.[15] in Korean population. The increased prevalence in succeeding years could be due to the introduction of advanced imaging modalities.

The term 'bifid' literally means "cleft into two parts". Hence, it shows duplication of condyle with the presence of two condylar heads usually located over a single neck. The occurrence of two condylar heads with separate neck is extremely rare. Dennison[16] suggested that bifid condyle should be referred for only those cases where 2 condyles appear in anterior and posterior part of sagittal plane.

The etio-pathogenesis of bifid condyle is uncertain. Several theories have been stated which are broadly categorized as traumatic and developmental. The most widely accepted theory is the traumatic one. Bifid condyles have been reported subsequent to condylar fracture[17,18]. Minor trauma to the growth centre and deficient remodelling after fracture can produce bifid condyles[19]. Any rupture of the vessel in the cartilage in early postnatal life due to mechanical injury might impair the ossification process with resultant formation of bifid condyle. Hotz[20], on observing stages of remodelling after fracture reported that the fractured condyle never forms a callus but a new condyle forms in position with resorption of the fractured process but till what age new condyle can form is unclear. So, early childhood fractures in general may lead to formation of bifid condyle which is generally unilateral. Also, trauma may not be present in every case. Fracture of condylar head could result in medio-lateral as well as antero-posterior orientation of condyles as shown by Yao et al.[21]. However, it is usually considered

that a sagittal split with antero-posterior orientation is traumatic in origin whereas medio-lateral orientation is developmental in origin. In the present case, there was no history of trauma and the condyles were oriented medio-laterally thus favouring a developmental cause.

In the developmental etiology various processes have been postulated. Alteration in the cartilage[22] or in the position of fibroblastic cells around the disc surface[23] could affect the bone remodeling process developing a bifid condyle. The persistence of fibrovascular septa (that appears in the condylar cartilage at 20 weeks intrauterine) till the second year of life might impair ossification[24]. Gundlach et al.[5] reported that teratogenic agents and misdirection of muscle fibres may affect the bone formation such that bony spikes may form at the area of muscle insertion and that bifid condyle is the result of abnormal spiking due to muscle insertion.

Quale and Adams[25] reported that endocrine disturbances, nutritional deficiency, infection, irradiation during the development of condyle could result in bifidism.

Though many cases of BMC have been reported in association with TMJ ankylosis[8,9] which is a developmental anomaly, the association with other anomalies is sparse. One case has been reported with unilateral condylar hyperplasia[26]. Two cases have been reported with hemifacial microsomia[10]. One case of bifid condyle has been reported with asymmetric contralateral ascending ramus of the mandible, supernumerary nipples (polythelia), supernumerary-rudimentary postaxial sixth fingers and clinodactyly[27]. These associations could be genetically related. But all these concomitant abnormalities have been seen with unilateral BMC. Oligodontia in association with BMC has never been reported. The present case to the best of our knowledge is the first reported case of bilateral bifid mandibular condyle in association with oligodontia in a non-syndromic patient.

The term 'hypodontia' refers to the phenomenon of congenitally missing teeth that are fewer than six teeth. Congenital absence of more than six teeth is defined as 'oligodontia' and the complete absence of teeth is defined as 'anodontia'.

Hypodontia/oligodontia can be non-syndromic, where it is an independent congenital oral trait, or syndromic, where it is acquired as part of a specific disease. The non-syndromic form can be sporadic or familial.

It has been shown that the genes involved in odontogenesis may have an effect on the other systems or organs of the body. The signaling molecules Bmp-2 and Bmp-4 are expressed during odontogenesis and they are also expressed at the site of endochondral bone ossification[28]. So, it could be that abnormality in the similar factors responsible for development have lead to concomitant expression. We hypothesize that there may be genetic effect that led to simultaneous occurrence of two abnormalities but genetic studies are required to confirm this hypothesis.

No other appreciable abnormality was detected on physical examination and during detailed systemic examination of the patient. Since, a syndrome is considered to be a group of signs and symptoms that occur together and characterize a particular abnormality, we referred this to be non-syndromic.

The normal width of the condyle in frontal, sagittal and horizontal plane is 18.7-21.8mm, 5.5-16mm and 9.8-10.1 mm respectively[29]. The condyles in our case were 23.5 mm & 23.1 mm in frontal plane, 11.6 mm and 11.1 mm in sagittal plane and 7.9 mm & 8.4 mm in horizontal plane at maximum width for right and left condyles respectively. The bifidism in this case was mainly seen in the posterior surface and it was not seen through and through till the anterior surface with a distance of 2 mm and 2.6 mm remaining from the anterior surface for right and left condyles respectively. The medial part of the bilateral bifid condyles was thinner as compared to the lateral part (Figure 4).

The duplication of two condylar heads is appreciated by a groove or rift which can be distinct or indistinct. The indistinct ones can be classified as cleft, notch or gap as suggested by Dennison[16] or as partial bifid condyle as suggested by Sahman et al.[13]. In our case, the bifidism was distinct with a depth of 2.8mm bilaterally from the highest points on the articulating surfaces of two heads (Figure 5). The condyles normally are rotated on the condylar neck with posterior inclination of medial pole at an angle of 15°-33° with the frontal plane. In our patient, the inclination was 24° and 27° in the right and left side respectively. The more the angle, the better will be the visualization in the sagittal plane as seen in the panoramic image where the bifidism on the left is well appreciable as compared with the right. The posteriorly

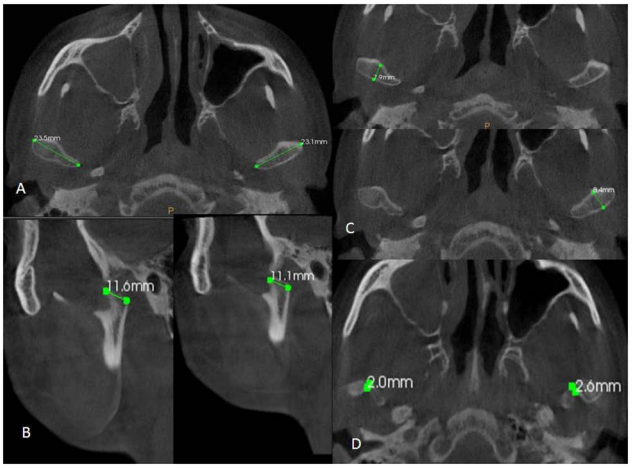


Figure 4: A)Axialsection showing condylar dimensions in frontal plane
B)Sagittal section showing dimensions in sagittal plane
C)lAxial section showing condylar dimensions in horizontal plane
D)Distance between bifidism and anterior surface

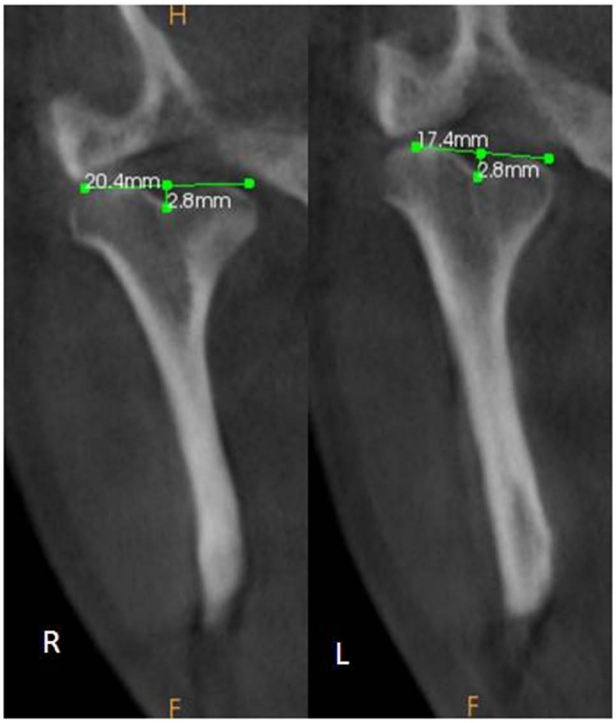


Figure 5: The depth of bifidism of BBMC from the highest points on condylar surface.

inclined condyles made an angle of 127° as compared to the normal angle of 150°-170° (Figure 6).

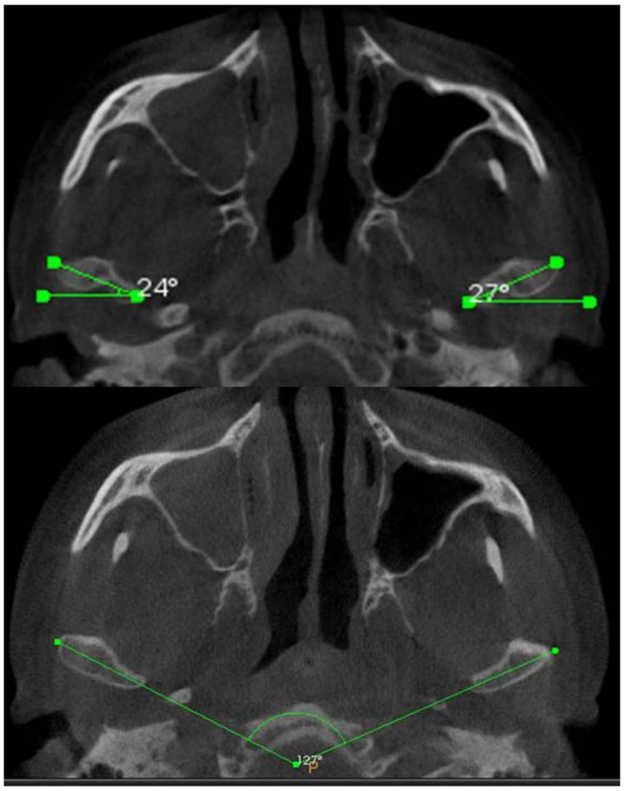


Figure 6: The inclination of BBMC with the frontal plane and the horizontal condylar angle.

Lund et al.[19] reported that if the formation of BMC occurs before the formation of condylar centre at 10th week when the lower meniscal surface is formed, two articular surfaces on the glenoid fossa may be seen. In contrast in BMC of traumatic origin, only a single glenoid fossa is seen. On reviewing the embryology of temporomandibular joint, the articular fossa is the first structure to become recognizable at 7-8 weeks and begins to ossify at 10-11th week[29]. The condyle, at first cartilaginous, develops between 10th and 11th week from an accumulation of mesenchymal cells lateral to Meckel's cartilage. Endochondral ossification then progresses apically, creating a bony fusion with the body of the mandible. The duplication of articular fossa can be best appreciated in archaeological specimens. In our case, duplication of glenoid fossa was not appreciated on CBCT but there was no history of trauma. Hence, most likely it could be developmental in origin.

Most of the cases of BBMC reviewed in the literature are asymptomatic. However, the patient in our case was symptomatic. The underlying cause for symptomatic nature could be the traumatic anterior deep bite present which led to disc derangement. The unilateral symptom presentation could be the result of excellent adaptive and remodelling capacity of this ginglymo-diarthroidal joint. Literature suggests that symptomatic cases should undergo surgical intervention. However, the patient was not willing for the same so it was managed conservatively by anti-inflammatory agents, muscle relaxants and physiotherapy.

BMC may be associated with anomalous morphology of articular disc as shown by Shriki et al.[10] where meniscus was seen T shaped on Magnetic Resonance Imaging (MRI). MRI study of the disc could also explain why symptoms were present on only one side while bifidism was present on bilateral condyles. But the unwillingness of the patient for surgery didn't allow us to subject the patient for MR due to ethical reasons.

The strengths of the case was a detailed insight we achieved by examination and the use of CBCT derived images in multiple planes, which has lead to derivation of possible etiology and understanding the morphology of anomalous condyle. However, due to lack of surgical treatment only symptomatic pain relief was achieved with little improvement in mouth opening. Moreover, the orthodontic treatment was not completed and there was a lack of long term follow up.

In conclusion, BBMC is a rare developmental anomaly and its association with other developmental disorders is scantily reported. The present case is the first reported case of symptomatic bilateral bifid mandibular condyle in association with oligodontia and 86th reported case of BBMC but will be the first richly illustrated CBCT case study showing all the morphological deviations in dimensions.

4. Potential Areas of Conflict

The authors declare that there are no potential areas of conflict.

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