

CASE REPORT

Bilateral condylar anomaly: A case report and review

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ABSTRACT

A case report of trifold and aplastic condyle in a 26-year-old female has been discussed. A marked oval radiolucency in the head of right condyle and absence of left condyle was observed on a panoramic radiograph. Diagnosis of trifold condyle was established and confirmed using computed tomography. Magnetic resonance images showed degenerative changes of the disc.

Key words: Aplasia, bifid condyle, temporomandibular joint disorder, transmigration, trifold condyle

INTRODUCTION

The temporomandibular joint (TMJ) is a complex skeletal structure which is essential for normal functioning of the jaw. There are many causes of various unilateral and bilateral growth disturbances of the mandibular condyle and its related structures.^[1] Aplasia and trifidity are two different anomalies affecting mandibular condyle. Bifidity and trifidity are rather an uncommon condition and is characterized by duplicity of the mandibular condyle.^[2] Aplasia of the mandibular condyle indicates nondevelopment of condyle associated mainly with various craniofacial abnormalities.^[1] The manifestation of both together is rare and has not been reported previously in the literature. We describe a unique case of morphological variations of the condyle in the same person. Various etiologies for morphological variations/defects of the mandibular condyle have been discussed. Furthermore, a modification of the classification for condylar morphological variations has been proposed.

CASE REPORT

A 26-year-old female patient visited the dental clinic for orthodontic correction of her teeth. Medical and family history was unremarkable. Clinical examination revealed mild facial asymmetry, micrognathia and deviation of the mandible to left side with 37 mm interincisal distance on opening. Intraoral examination revealed missing upper right

second premolar and lower right canine. Lower left, the first molar was malformed.

Her panoramic radiograph [Figure 1] showed a marked oval radiolucency in the head of right condyle and absence of left condyle. The growth of body and ramus of the mandible were adequate on the right side but showed narrowing of the ramus width and deep ante gonial notch of the left side. Transmigration of the right mandibular canine-oriented horizontally below the apices of the lower contralateral first molar was also noticed. A three-dimensional (3D) computed tomography (CT) reconstruction demonstrated a complete absence of the left condyle with a rudimentary glenoid fossa [Figure 2]. Right condyle was trifold [Figure 3] with the fossa shaped accordingly to accommodate the trifold condyle. Sagittal magnetic resonance (MR) sections of the right condyle showed divisions of the condylar head with mild degenerative changes in the disc [Figure 4]. Orthodontic treatment and orthognathic surgery including a costochondral rib graft were planned to improve function and esthetics of the patient. As the patient was not willing for treatment, we have not performed any treatment for her.

DISCUSSION

TMJ is a complex skeletal joint which is essential for normal functioning of the jaw. Disturbance in growth affecting

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Figure 1: Panoramic radiograph showing oval radiolucency in right condyle, absence of left condyle and transmigration of lower right canine

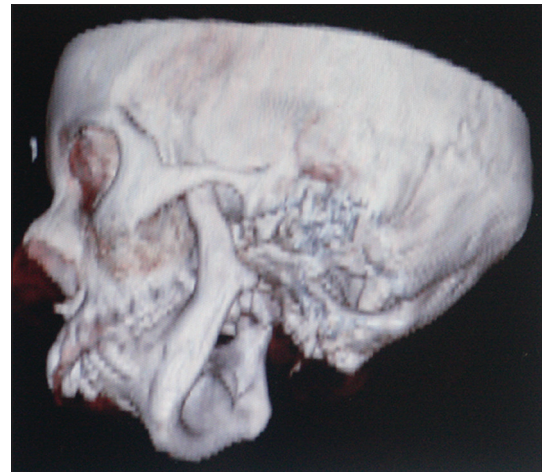


Figure 2: Three-dimensional computed tomography reconstruction demonstrating absence of left condyle with rudimentary glenoid fossa

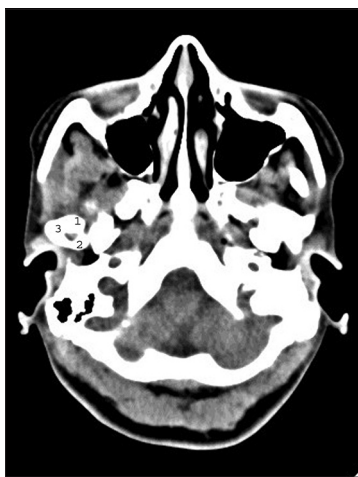


Figure 3: Axial computed tomography view through the level of condylar head demonstrating trifid right condyle with anterior (1), medial (2) and lateral (3) condylar heads

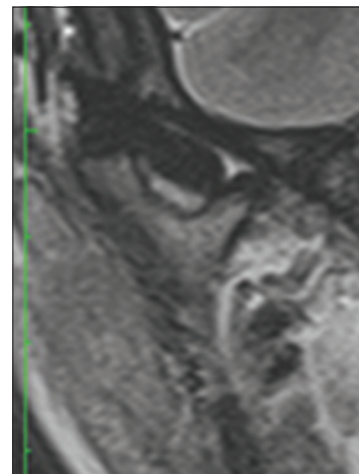


Figure 4: Sagittal magnetic resonance section of right condyle showing divisions of condylar head and degenerative changes in disc

TMJ results in facial abnormalities. Such disturbances may occur congenitally/developmentally or during postnatal development (acquired).^[1]

Congenital deformities and developmental abnormalities of the mandibular condyle are classified as (1) hypoplasia or aplasia; (2) hyperplasia; and (3) bifidity.^[1] Bifidity reports are scanty in literature, whereas trifid^[2-8] and tetrafid^[9] reports are extremely rare. The current classification does not accommodate condyles with trifid/tetrafid cases. Hence, it necessitates further modification of current classification to include cases with more than one condylar head as polyfidity instead of bifidity. In the present case, there was trifidity in right condyle and aplasia in the left side.

Specific stage wise growth and development process of TMJ and condylar cartilage in intrauterine life is well described but their mechanistic basis is poorly understood.^[10] Various theories have been proposed regarding etiology and pathogenesis of bifid mandibular condyle [Table 1] and

aplasia [Table 2]. Etiologies for trifid and tetrafid could be similar to that of bifid.

Aplasia of mandibular condyle is a rare anomaly which may be either congenital or acquired. Congenital aplasia occurs as one of the several facial manifestations in various syndromes [Table 2]. In these syndromes, as a rule, there will be other soft tissue or skeletal malformations/findings involving external or internal ear, temporal bone, zygoma, the angle of the mouth and the soft tissue environment.^[1]

In this case, other manifestations of previously mentioned syndromes were absent. Aplasia of mandibular condyle without other facial malformations is extremely rare but has been reported previously.^[19-23] A rare case of bilateral agenesis of mandibular condyle has been reported by Akihiko *et al.*^[24]

Acquired condylar aplasia may be caused during active growth by local factors such as mechanical trauma, infection of TMJ and middle ear, inflammatory process in the area,

Table 1: Etiologies of bifid mandibular condyle

Persistence of fibrous tissue septum in the midzone of the mandibular condyle ^[11,12]
Obstruction of blood supply to condylar cartilage (would affect the remodeling process leading to bifid condyle) ^[13]
Trauma, repeated microtrauma, malocclusion, radiotherapy, nutritional deficiency, endocrine disorders, genetics and infections ^[14]
Association with syndromic anomalies (hemifacial microsomia) ^[15]
After condylectomy or after fracture of the mandibular condyle insufficient capacity to remodel the mandibular condyle ^[16]
Teratogenic effects ^[17]
Forceps delivery (dislocation of joint integrity due to birth trauma) (rupture of septal blood vessels-hematoma) ^[18]

Table 2: Etiologies of aplasia

Acquired aplasia: Caused by local factors as mechanical trauma during active growth, infection of temporomandibular joint and middle ear, inflammatory process in the area, rheumatoid arthritis or irradiation ^[19]
Congenital aplasia: Occurs as one of the several facial manifestations of syndrome (hemifacial microsomia, Treacher Collins syndrome, Goldenhar syndrome, Oculomandibulo dyscephaly, Hurler's syndrome, Proteus syndrome, Morquio syndrome, Auriculocondylar syndrome) ^[19]
Nonsyndromic ^[19]

rheumatoid arthritis or irradiation [Table 2]. In the present case, history of trauma or any of the above-listed etiologies were absent. Etiology for condylar aplasia in the present case, therefore, remains unknown, but could be a developmental defect.

Condylar development starts with ecto-mesenchymal condensation of condylar blastemas at 7–8 weeks of intrauterine life and definitive condylar cartilage appears at 10 weeks. Thus, if aplasia of the condyle occurred as a developmental defect, it probably happened before the 10th week postconception, when the mandibular condylar cartilage would have started its development.^[19]

Manifestation and severity of the congenital abnormality may depend on the alteration of gene expression profiles.^[25] Thus the role of signaling proteins such as Indian hedgehog (Ihh),^[1] fibroblastic growth factor (FGF),^[26] bone morphogenic proteins, Spry1 and Spry2 (sprouty genes),^[27] Shh, Runx2, parathormone-related protein^[1] are indispensable for condylar development. (e.g., absence of Ihh causes condylar growth deficiency).

Proper signals (tissue interaction) from developing condyle are required for glenoid fossa development. In the absence of mandibular condyle, development of articular fossa will be initiated but fails to sustain and subsequently will regress leading to an ill-defined/rudimentary fossa.^[27] In the present case, glenoid fossa was rudimentary which would imply that the defect was initiated prenatally.

N-sulfotransferase (Ndst1) dependent heparan sulfation has been found to be critical for TMJ development. Severely affected Ndst1 mutation affects the onset of condylar development at early stages of embryogenesis, leading to its absence. In mildly affected mutations condylar growth plate is found to be dysfunctional, showing a wider distribution of Ihh activity and ectopic ossification along condylar lateral borders.^[28] Thus, we can assume that ectopic ossification may be the cause for trifid condyle on the left side. However, Ndst1 mutation was not evaluated in this case.

Various theories have been proposed regarding etiology of bifid/trifid mandibular condyle. Some authors agree to the opinion that trauma involving TMJ results in bifid/trifid condyles. Five out of seven reported cases of trifidity have reported a history of trauma.^[3-9]

However, in the present case, we cannot rule out the role of muscle as an etiologic factor for trifid development. As there is aplasia of condyle on one side, the stability of the TMJ might have been maintained by the tripod effect of the contralateral condyle. In normal individuals stability of the joint is maintained by constant activity of muscles. During 8–10 weeks of intrauterine life lateral pterygoid muscle is attached to condylar blastema tissue which guides conical condyle to form TMJ.^[29] The compensatory orientation of these muscle fibers to maintain tripod effect for joint stability might have led to trifid condyles. Gundlach *et al.* postulated that misdirection of muscle fibers leads to bony spikes formation in the area of fiber attachment, further development of which leads to the bifid/trifid condyle.^[17]

Bifid mandibular condyle usually occurs without significant complaints or clinical features, such as pain or restricted movement; its diagnosis usually depends on radiological rather than clinical evidence. Although it is an incidental finding during the routine panoramic radiographic examination, these radiographs are not efficient because of poor visualization of structures that make up the joint and different degrees of distortion.^[30] In this case, trifidity was not discernible on a panoramic radiograph.

Other imaging techniques like MR image and 3D CT can provide additional information regarding such condition. In the present case, both sagittal CT view and 3D reconstruction demonstrated trifidity.

Trifid condyles are accidentally diagnosed during a routine examination. In cases, without a history of trauma genetic screening may be useful in finding the etiology. Trifid condyles are not associated with any form and function problems. In cases with form and function problem, rehabilitation would be beneficiary.

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Conflicts of interest

There are no conflicts of interest.

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