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The bipartite condyle: A unique rare case report

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Abstract
Double headed condyle or bifid mandibular condyle is rare anatomic anomaly that can result from congenital malformation, tumor, trauma or infection. This is an extremely rare disorder characterised by a duplication of the head of the mandibular condyle. We hereby report a case of double headed mandibular condyle with a separate neck which was diagnosed using computed tomography scan.
Methodology: A 33 year old male patient presented with an alleged history of assault. CT faciomaxillary with 3D reconstruction revealed right coronoid process fracture and symphysis fracture and an incidental finding of a double condylar head with a separate neck on the right side. The fractures were fixed under general anaesthesia. The patient was followed up for 3 months. CT DICOM files were made followed by a stereolithographic model for the intricate understanding of this rare anomaly.
Results: The shape of the main condylar head was conical with mild resorptive changes. The accessory condylar head branched from the neck of the main condylar head anteromedially with a separate neck. On regular follow up of three months, the patient had absolutely no complain of TMJ Pain and had normal TMJ movements bilaterally in all directions.
Conclusion: Being one of the unique cases, this case is one of its kind since it exhibits one of the rarest morphological variant of the condylar heads. Treatment for such cases isn't a compulsion however long term follow up is a must.

Keywords: Double headed condyle, mandible, bifid condyle, TMJ, anatomical variation, trauma

1. Introduction
The double-headed condyle or bifid mandibular condyle (BMC) is signified by the duplicity of the head of the mandibular condyle. Hrdlička was the first one to report double condyles in 21 skull specimens in the Smithsonian Institution's collection of dried skulls in 1941 [1]. This has been the most richly illustrated description of this anomaly. In 1948, Schier reported this anomaly in the living population for the very first time [2].
Usually diagnosed as an incidental finding during routine radiographic examinations, this anomaly typically has no distinct clinical symptoms. Some circumstances such as genetic tendency, trauma, infection, teratogenic drug use and exposure to radiation may be responsible for these variations, although the exact aetiology is unknown [3].
This is a unique case report of double headed mandibular condyle with a separate neck which was diagnosed using computed tomography scan.

2. Case presentation
A 33 year old male patient reported to the Department of Oral and Maxillofacial Surgery with an alleged history of assault. Patient gave a history of one episode of ear bleed on the right side although no active bleed was present at the time of admission.
Clinical examination revealed a diffuse swelling in the right preauricular region extending upto the lower border of the mandible. There was a laceration present on the chin measuring 3cm X 1.5cm. The mouth opening was approximately 2cm. Palpation revealed tenderness in the right preauricular region. Intraorally there was a laceration present on the ventral surface of the tongue measuring 1.5cm X 0.5cm. Coleman's sign was present and the occlusion was deranged bilaterally. Segmental mobility was present in between 41 and 31.

CT faciomaxillary with 3D reconstruction was done which revealed right coronoid process fracture and symphysis fracture and an incidental finding of a double condylar head with a separate neck on the right side. Open reduction and internal fixation was done under general anaesthesia for the symphysis fracture and the retrieval of the fractured coronoid process was done.

The patient was recalled after 3 months for routine follow up and had absolutely no complain of TMJ Pain and had normal TMJ movements bilaterally in all directions. CT DICOM files were made following which a stereolithographic model was prepared for the intricate understanding of this rare anomaly.

The shape of the main condylar head was conical, the superior surface being irregular with mild resorptive changes. The distance of the main condylar head mediolaterally was 15mm and anteroposteriorly 10mm. The shape of the accessory condylar head was that of an inverted cone and the superior surface showed mild resorptive changes. The distance of the accessory condylar head mediolaterally was 10mm and anteroposteriorly 12mm. The accessory condylar head branched from the neck of the main condylar head anteromedially with a separate neck. The neck of the accessory condylar head arose approximately at a distance of 10.6mm from the lingula posterosuperiorly and at a distance of 13mm from the mandibular foramen. The distance between the main condylar head and accessory condylar head was approximately 4.6mm and the angulation in between them was approximately 49.6 degrees.

3. Discussion

Briefing the developmental process, the ossification of the lower jaw occurs in the membrane that overlies Meckel's cartilage, from the forepart of this cartilage, and from some accessory cartilages of later development. One of these accessory cartilages forms the condyle and a narrow wedge like the ramus, but no separate center of ossification is present. The ossifying process spreads through the membrane and invades the cartilage. Although reason behind the doubling of the condyle is unexplainable; yet it would seem that the cause must lie here. This possibly must consist of a phenomenon that leads to the doubling of the condylar head and even the neck, though this is rarely observed. In case the ossification proceeds from before backward, i.e. from the body of the ramus to the summit of the condyle, and if an early blood vessel of some size ran once through the cartilage from before backward (or reverse) at or near the middle of what would be the neck of the condyle, then conceivably the obstruction produced by this blood vessel might, as it occasionally does elsewhere, cause a doubling of the condyle to develop. Occasionally, these hindrance by a blood vessel with the ossification of a part happens with marked results in the basilar process of the occipital, leading to the characteristic uni-or bi-lateral fissure in the sides of the process; which might be observed in other parts of the skull and skeleton. Although, any evidence yet of such a blood vessel in the condyle or the sub condylar part (neck) of the lower jaw has not been documented, such vessels either disappear in the process of growth or their grow is so insignificant that they escape attention. In the absence of any phylogenetic connection of the formation, it would seem that the assumption of some mechanical interference with the ossification of the part concerned may be the only remaining possibility [1].

Temporomandibular Joint (TMJ) is a freely movable articulation between squamous portion of the temporal bone at the base of the skull and the condyle of the mandible. The condyle is considered very special because the expression of the mandibular growth is provided by mandibular condyle [4].

The variation in mandibular condyle is observed both in size and shape. The condyle is roughly ovoid in outline when viewed from above. It is 15mm to 20 mm side to side and 8mm to 10 mm from front to back. A great variation in the size and shape of the components of the temporomandibular joint is observed. According to a common assumption, the normal condylar head must have a convex configuration throughout and that symmetry should exist between contralateral sides in the same individual. The normal condylar morphology can have variation which is observed with gender, age, facial type, occlusal force, malocclusion type, functional load and between right and left sides. The most prevalent morphologic changes are detected in the TMJ of elderly persons attributing to the onset of joint degeneration. The reason behind these morphological changes may be based on simple developmental variability as well as remodelling of condyle for accommodation of developmental variations, malocclusion, trauma and other developmental abnormalities and diseases [4].

In 1961, Yale *et al*. [5] was the first one to report about the different shapes of mandibular condyle. Initially, they classified the condylar head on the basis of the superior view into three categories namely convex, concave and flat, however later on they simplified it into four categories namely convex, rounded shape, flattened and angled. Variability of the condyle can be related to the inclination of the condylar head. And shape variability of the fossa is related to inclination of the eminence and fossa height.

Major change in condylar size during growth was noticed in mediolateral dimension as compared to the antero-posterior dimension. There have been various morphological variations reported such as bifid, trifid, tetrafid [6] condyle and even double headed condyles. The terms “bifid” and “trifid” were derived from the Latin word meaning a cleft into two and three parts. Talking about the bifid condyle, its articulating surfaces are divided by a groove and can be oriented mediolaterally or anteroposteriorly. This condylar splitting can range from a shallow groove to two distinct condyles with a separate neck. There is no age predilection as far as the unilateral bifid condyle is concerned. The age of the patients can range from 3 to 67 years (mean age, 35 years) and the male/female ratio being approximately 1.5:1. The anomaly is frequently unilateral but it may occur on both sides apparently without any marked predilection for any one side. However, it appear to involve the left side more than the right side.[6] Predisposing factors of unilateral bifidism includes trauma, such as condylar fractures, birth trauma, or surgical condylectomy, whereas bilateral bifidism can result from a primary aberrancy of embryological or postnatal development [6].

There is a considerable variation in the degree of condylar head separation, with few cases having only a 1-2-mm indentation in the condylar head, while others having complete separation of the heads. There are few reported cases of complete condylar head separation [7]. Duplication of the mandibular condyle is exceptionally rare. Szentpetery *et al*. [8] conducted a study on 1,882 cadaveric skulls (2077 condyles) and found the incidence of this anomaly to be 0.48%.

Two different theories have been postulated to account for the pathogenesis of the mandibular condyle duplication. According to the Blackwood’s postulate, a band of vascular or

fibrous tissue should be identified between the two condylar heads on MR imaging [9, 10]. This hypothesis can be contradicted by the lack of MR imaging visualisation of a fibrous band or of an abnormal vascular structure. However, the presence of fibrous or vascular tissue might have been there in the embryonic period before the ossification of the mandibular condyle but regressed later in the development. Hence, to concise Blackwood and Moffett [11] suggested that a retained fibrous septum or a vascular structure leads to the impedition of the ossification of the mandible, hence leading to the splitting of the condyle into two heads.

Contrastingly, Thomason and Yusuf [12] have suggested prior trauma as a cause. However, the duplication has been described in patients without any antecedent identifiable traumatic event which was similar to our case who did not give any history of prior trauma. The differentiating factors here is the orientation of the mandibular head. Developmental cause might be the reason behind the mediolateral orientation. However, in case of a sagittal split with anteroposterior orientation an antecedent identifiable, traumatic event can be the probable reason. However, Yao *et al*. [13] demonstrated experimentally that a fracture of the mandibular condyle could result in mediolateral as well as anteroposterior orientation of the mandibular condyles.

In our case, though the patient did not give any history of trauma priorly, the shape of condyle somewhat indicated towards trauma being a reason for this anomaly. On observing the condylar surfaces, the irregular resorptive changes visualized could be reasoned with trauma, as mentioned by Thomason & Yusuf [12] and Yao *et al*. [13]. But since the patient denies any history of trauma priorly, this might be a developmental anomaly as described by Blackwood [9] and Moffett [11].

Post Traumatic BMC involves relatively specific factors that cause a double-headed mandibular condyle. According to Szentpetery *et al*. [8] the emergence of a BMC and its symptoms were influenced by the patient's age, the type of injury i.e. (direct or indirect, high or low fracture), the extent of damage to the joint structures (disc, capsule and articulate surfaces), hemarthrosis and the presence or absence of inflammation.

The direction of the fractured condylar fragment is affected by the lateral pterygoid muscle, which is an important factor in BMC formation. The minimum requirement for the formation of BMC is met when the muscle force is adequate to dislocate the condylar head. The ability for the formation of the new condylar head at the original site and insufficient remodeling capacity of the fractured condylar head must happen simultaneously. The relation in between the remodelling ability to that of age, sex or race is uncertain. Another important aspect is the vector of the fractured condyle because it regulates the arrangement of the BMC [14].

Condylar abnormality may be genetic in origin, or acquired. According to Quayle *et al*. [15], it is possible that the abnormality reported could have a genetic basis, either inherited or by virtue of a mutation. However, there was no known history of condylar problems in any of the patient's relatives and although an isolated TMJ abnormality or genetic origin is conceivable, there were no other abnormalities such as deafness, abnormal external ear or facial asymmetry to support a genetic aetiology in his report. Non-genetic congenital causes could include nutritional deficiency or drug induced mandibular dysostosis. Acquired condylar growth abnormalities may occur due to a variety of local causes such as trauma, infection or following irradiation. Alternatively, systemic factors including endocrine and dietary may be involved. None of these factors were known to be contributory in the case reported in the study which was similar to our case.

3.1 Tables and Figures

Table 1: Sizes and Shapes of Condyle in Various Conditions [4]

1. Developmental defects	▪ Condylar hyperplasia ▪ Condylar hypoplasia ▪ Agenesis ▪ Bifid condyle
2. Syndromes	▪ Hemifacial Microsomia ▪ Treacher Collins Syndrome ▪ Hallermann-Steiff Syndrome ▪ Pierre Robin Syndrome ▪ Oculo mandibulo dyscephaly ▪ Progeria
3. Degenerative joint disease	
4. Inflammatory/infectious diseases	▪ Rheumatoid Arthitis ▪ Psoriatic Arthitis ▪ Septic Arthitis
5. Cysts of TMJ	▪ Aneurysmal Bone Cyst ▪ Simple bone cyst ▪ Ganglion cysts and synovial cysts
6. Tumours of the TMJ	▪ Osteoma ▪ Osteochondroma ▪ Chondroblastoma ▪ Osteosarcoma ▪ Ewing's sarcoma
7. Metabolic Disease	▪ Gout
8. Endocrine Disturbances	▪ Gigantism and Acromegaly ▪ Hypothyroidism & Hypopituitarism
9. Trauma	
10. Radiation	

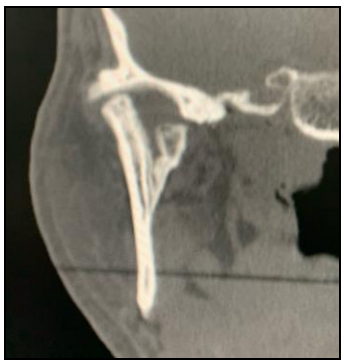

Fig 1: CT-Coronal section showing double condylar head with a separate neck

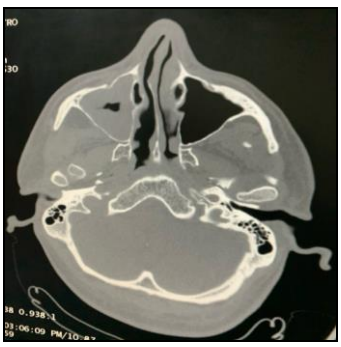

Fig 2: CT-Axial section showing 2 condylar heads

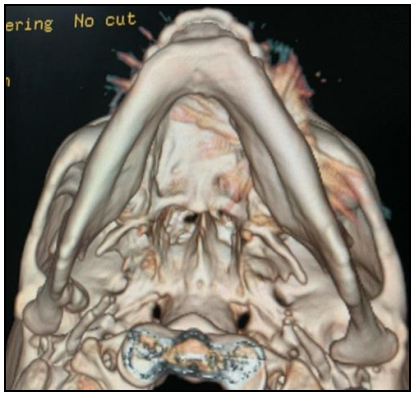

Fig 3: CT-3D Reconstruction showing 2 condylar heads

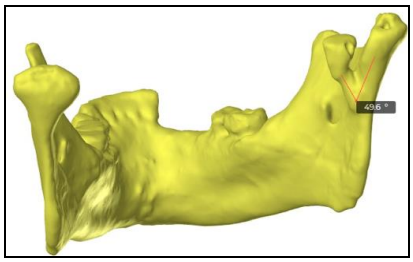

Fig 4: VSP showing Angulation between the main and the accessory condylar neck (49.6°)

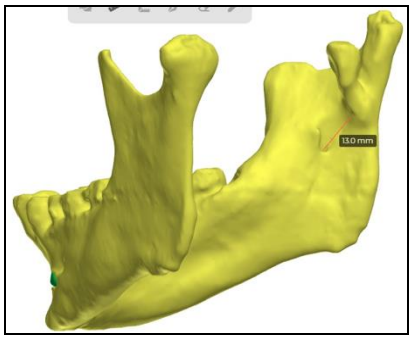

Fig 5: VSP showing distance between the mandibular foramen and neck of the accessory condylar head (13mm)

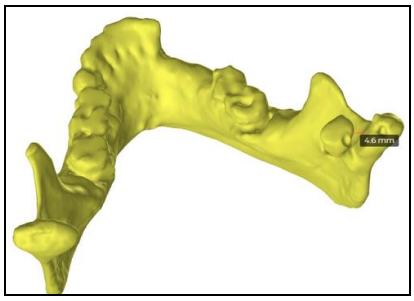

Fig 6: VSP showing distance between the main and the accessory condylar head (4.6mm)

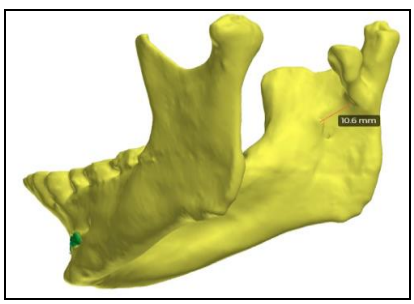

Fig 7: VSP showing distance between the lingula and the accessory condylar neck (10.6mm)

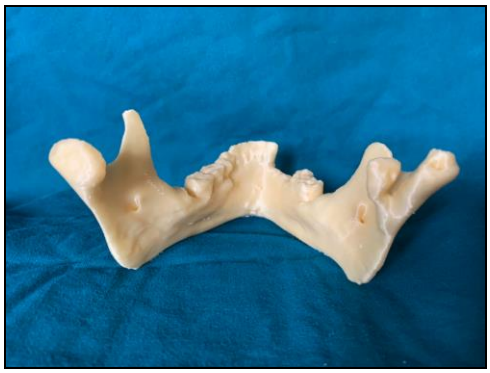

Fig 8: Stereolithographic model showing double condylar head with a separate neck- Posterior view

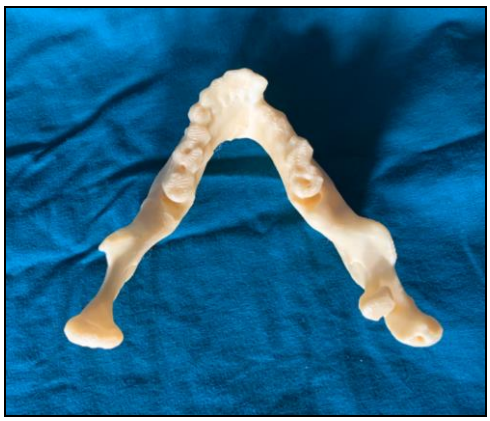

Fig 8: Stereolithographic model showing double condylar head with a separate neck-Superior view

4. Conclusion

We believe, this case report is definitely one of its kind since it exhibits one of the rarest morphological variant of the condylar heads. Such morphological alterations regarding mandibular condyle may be encountered during routine examinations. Clinicians should be aware of these conditions and advanced imaging modalities should be applied in order to avoid possible misleading interpretations. In general, treatment isn't a compulsion owing to a lack of clinical symptoms, but proper long-term follow-up and information to the patients about this condylar variation is a must.

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