Bifid mandibular condyle with ankylosis in a 3-year-old child: A rare presentation and review

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Abstract

Bifid mandibular condyle with ankylosis is an extremely rare condition and may arise as a developmental or traumatic defect. It may be associated with ankylosis. We here report a case of unilateral bifid mandibular condyle with ankylosis in a 3-year-old child. This is the youngest patient reported with the condition making it one of its first kind in the literature. The detailed description of the case, its radiological findings, and the literature on bifid condyles are reviewed.

Keywords: Ankylosis, bifid condyle, temporomandibular joint

Bifid condyle, as the name suggests, is the duplicity of the head of the mandibular condyle. There are many probable etiologies of the condition though none of them have been confirmed yet. It can be due to misdirected muscle fibers getting interposed during the developmental period.[1] The fibrous septa of vascular bodies can also separate the condyle into two different heads.[2] Blood supply itself can obstruct the condylar head, create a groove, and thus lead to a bifid head.[3] All kinds of trauma such as traumatic delivery with the use of forceps, and other several prenatal and postnatal trauma (surgical, accidental) in the condylar region could also be a possible etiology.[4] Fracture, dislocation, or other disruption of joint integrity can lead to mandibular remodeling, resulting in the formation of a secondary functional condyle. The genetic, endocrinological, nutritional disorders, infection, irradiation, or pharmacological origin has also been speculated.[5,6] Many bifid condyles develop in cases with insufficient remodeling capacity.

Due to its asymptomatic nature, the diagnosis of a bifid condyle usually relies on radiological rather than clinical evidence. In living patients, it is an incidental finding on radiograph. Radiographs including orthopantomogram are good screening methods for the condition but a three-dimensional CT scan is the best for determining condylar deformity.

The treatment of a bifid condyle depends upon several factors like the age of the patient, the function, the esthetic need, the maintenance of growth in the case of children, the presence or absence of ankylosis, and the signs and symptoms the patient has presented with. Asymptomatic cases are usually not intervened clinically or surgically. As we still do not have sufficient data to assess the long-term effects of this anomaly on the function of the temporomandibular joint, the standardized treatment plan formulation remains a task to be accomplished in future. Many patients are followed up without intervention and surgical interventions are usually done after the completion of growth. Initially the patient can be treated conservatively with muscle relaxants, analgesics, and occlusal splints. Later, if signs and symptoms persist and if facial esthetic is at compromise, a combined orthodontic and surgical treatment can be done. When associated with ankylosis, three basic techniques by Manganello-Souza and Mariani[7] can be undertaken, namely, (1) gap arthroplasty, (2) interpositional arthroplasty, and (3) joint reconstruction with grafts, all following the Kaban’s protocol.

Case Report

Children are gifts of god but at the same time not all children are equally gifted. Though bitter, there lies the reality. Following is the case report of a child who made us realize the same.

A 3-year-old child visited the Department of Pedodontics and Preventive Dentistry along with his parents with the chief complaint of not being able to chew food and open mouth completely. The father and mother were the informers. Further inquiry revealed the notice of the condition of the child by parents as early as 1 year of age when the upper jaw started becoming prominent as the child grew. After the eruption of anterior teeth in both upper and lower archs, they found constant increase in the overjet. Even with the eruption of all the milk teeth, the child was unable to chew food. As the patient tried chewing, there was escape of food from the anterior area. Along with it, the mouth opening also gradually started decreasing. Any incidence of trauma in the head and neck region was not in the notice of the parents.

As the child is unable to chew properly, he is being fed on
mashed food. There is a psychological impact on the child due to his condition. At that tender age, he is aware that some problem exists in his mouth. So, whenever he is free, he is found to be playing with toothbrush and toothpaste. Sometimes, the brushing frequency exceeds even five times.

Past medical history suggested recurrent episodes of upper respiratory tract infection. Also, the parents had been taking him for homeopathy in the hope that his mouth opening will improve. Prenatal history was not significant. The delivery of the child was by cesarian section. The birth weight of the child was 3.7 kg. Postnataally, there was a history of intermittent fever 4 days after the delivery which lasted for 15 days. Definitive diagnosis of the fever could not be made at that time. The child was hospitalized and intravenous drugs were used.

On general examination, the patient was cooperative, well oriented to time, place, and person, and happily sitting semisupine on the dental chair and smiling.

On extraoral examination [Figures 1 and 2], the face was asymmetrical with a convex profile. Bird facies appearance was prominent. There was increased fullness on the left side and the lower jaw deviated toward the same side on mouth opening. Lips were incompetent with existing lip trap and limited mouth opening (15 mm). On palpation, temporomandibular joint movement was asynchronous. Movement was reduced and did not follow smooth path on the left side compared to the right side which shows normal movement. However, no crepitus was heard.

On intraoral soft tissue examination, mild marginal gingival inflammation with respect to upper anteriors was present which could be attributed to his ineffective lip seal. However, local deposits were found to be scanty. The palatal vault was high arched. Hard tissue examination revealed the presence of all primary teeth which were nonspaced. Lower incisors were supra-erupted. There was increased overjet (11 mm) and overbite (7 mm). The primary second molar relationship was distal step terminal plane bilaterally. The reduced mouth opening and deviation of jaw toward the left was prominent while doing intraoral examination.

Due to patient’s uncooperativeness, clear images in orthopantomogram could not be obtained. So, to go ahead with further investigation, CT scan was done under deep sedation with syrup Pedichloryl, 10 ml, 15 min before the procedure. 3D reconstructed images were also obtained. The left-sided ramus was short. The coronoid process was normal and the condylar process was obscured by the zygomatic process likely because of hypoplastic nature and ankylosis. The left mandibular notch was prominent [Figure 3]. The axial section showed hypoplastic and bifid left condyle. Soft tissue density was seen in both the maxillary and the ethmoidal sinuses suggestive of sinusitis [Figure 4]. The sagittal section revealed normal condylar process and glenoid fossa on the right side. The left side was bifid and hypoplastic. Left articular eminence was thickened and sclerosed [Figure 5]. The coronal section demonstrated left bifid condyle with ankylosis of the lateral head with articular eminence [Figure 6]. The lateral head was larger than the medial head. The left glenoid fossa was shallow and showed remodeling with mild sclerosis. The left ramus was short compared to the right suggestive of hypoplasia [Figure 7]. Rest of the visualized bones were normal in all the thin sections.

This gave us a final diagnosis of the bifid left mandibular condyle with ankylosis of the lateral head. A treatment plan has been formulated as left sided condylectomy with constochondral graft reconstruction under general anesthesia. After several rounds of discussion and consultation with the plastic surgeon and the pediatrician, owing to the tender age of the patient and the need for immediate postoperative rigid

Figure 1: Frontal view of the face. Note the lip trap and deviation of chin toward the left

Figure 2: Profile view of the face. Note the retrognathic chin giving bird facies appearance
Figure 3: 3D reconstructed images showing a normal right mandibular condyle, coronoid process, and ramus. The left-sided ramus is short. The coronoid process is normal and the condylar process is obscured by the zygomatic process likely because of the hypoplastic nature and ankylosis. The left mandibular notch is prominent.

Figure 4: Right side shows a normal condylar process. The left side is hypoplastic and bifid. Soft tissue density is seen in both the maxillary and the ethmoidal sinuses suggestive of sinusitis.

Figure 5: Right side shows a normal condylar process and glenoid fossa. The left side is bifid and shows a hypoplastic and bifid condyle. There is thickened and sclerosed left articular eminence.

Figure 6: Left bifid condyle with ankylosis of the posterior process with the articular eminence: the lateral head is larger than the medial head. The left ramus is shorter compared to the right.

Figure 7: Right side shows the normal mandibular condyle. The left side shows a hypoplastic and bifid condyle. The left glenoid fossa is shallow and shows remodeling with mild sclerosis. The left ramus is short compared to the right. Rest of the visualized bones are normal.

fixation, early mobilization, and aggressive physiotherapy for functional rehabilitation, the patient has been kept under regular follow-up and surgical intervention has been decided to be undertaken later.

Discussion

Bifid mandibular condyles were first described by Hrdlicka\(^3\) in 1941. He reported 20 cases from skeletal specimens collected from different parts of the world. It was first reported in a living person by Schier\(^8\) in 1948. The number of living cases reported since 1941 to 2000 is 28, and the total cadaver cases are 32.\(^9\) From 2000 to 2009, further 13 new cases have been reported including 2 cadavers.\(^10\) So, including our case report, the total number of bifid condylar heads reported till now is 74 cases among cadaver specimens and live patients. A total of 76% of the patients with diagnosed bifid mandibular condyles were asymptomatic.\(^10\) In 24% of the cases, the signs and symptoms associated were usually related with previous trauma. Complaints if any are mainly
limited to pain, headache, clicking sensation, and limited mouth opening if associated with ankylosis.

The rarity of this skeletal anomaly can be attributed to many cases being underreported due to its asymptomatic nature. It can be found both unilaterally or bilaterally but the bilateral presentation is uncommon, the ratio being 3:1.[11] If we include the cadaveric specimens, the ratio rises to 4.6:1.[11] It has no predilection for age, sex, or race. The left side has been seen to be more commonly involved than the right side. In unilateral cases, the ratio of left to right is 2:1.[10] The bifidism can be either mediolateral or anteroposterior. Such orientation of the mandibular head has been used as a differentiating factor whether the defect is developmental or traumatic.[9] Mediolateral orientation results from a developmental cause and the anteroposterior split usually results from traumatic event. In our case, we have found mediolateral presentation which further supports the developmental etiology of the condylar deformity. From our search of the literature, we found three cases of trifid condyles[9] and one of them has been attributed to childhood trauma.

To our knowledge, this patient is the youngest one to have been diagnosed with the condition. As the signs were prominent right at the age of 1 year, the radiological findings might have been obtained at that age if timely investigation was done. The other case in Ramos et al.[12] involved an 8-year-old child with an unspecified history of trauma who complained of pain while chewing. Her CT scan showed a bifid left condyle with the lateral head articulating with the temporal bone. The underdeveloped lateral head was surgically removed. Also, this case reported in our department is the seventh case of bifidism associated with ankylosis. The literature shows only six cases till now.[13,5,11,14,15] The incidence of a bifid condyle may be a lot more than perceived till now not only due to its asymptomatic nature but also to limited epidemiological studies in this field.

Reports of a bifid mandibular condyle are mainly concentrated on those cases with a history of trauma and those without. Many have associated it with trauma. Poswillo[16] suggested that a bifid condylar head may develop after remodeling of a condylar head fracture. In patients with incomplete remodeling, a defective resorption of the smaller fragment is demonstrated which leads to bifidism. From our case, we believe that the bifid mandibular condyle was a developmental anomaly. As the condyle was ankylosed, the cartilage could not become an effective growth center leading to hypoplasia. The mandibular remodeling also depends upon the activity of the musculature attached to it. In our case, the constant feeding of the child on soft diet may have compromised this activity too.

If we go to the developmental origin of the mandible, it develops between the fifth and seventh week of gestation from the mesenchymal tissue of the first branchial arch. It consists of Meckel’s cartilage that appears bilaterally below the oral pit. A rapid proliferation of this mesenchymal tissue in paired primordia results in thickening of the cartilage which extends from the midline to the posterior otic capsule. As the intramembranous ossification takes place, the cartilage begins to disappear. The middle ear bones (stapes, malleus, and incus) develop at the dorsal end of Meckel’s cartilage. The posterior part of the mandible ossifies by endochondral development. This creates the functional articulation of the jaw at about 20 weeks. For rest of the gestational period, the body of the mandible and the condyle forms angulation. The angle and ramus both undergo series of remodeling lengthening the mandible. The height of the ramus increases by growth at the head of the condyles. After birth, the bilateral halves of the mandible fuse at the midline symphysis. Till first birthday of the child, growth at the midline suture continues. The initial appearance of portions of the temporomandibular joint and fossa are visualized around week 12 in fetal development and the joint is formed at week 20.

As this particular anatomical variation may mimic a fracture or tumor, knowledge and awareness about this entity is important for a general dental practitioner[17] and a further study with a long-term follow-up is the demand of the time.

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