Bifid Mandibular Condyle: Case Report and Etiological Considerations

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Abstract

Bifid mandibular condyle, usually diagnosed on routine radiographic examination, is described in the literature as a rare entity. Its cause is controversial, and it has no predilection by sex or ethnic background. Dental professionals should have some knowledge of this anatomic abnormality, as well as its implications for function and appropriate treatment modalities, so that they can be alert to this potential diagnosis. This paper reports an unusual case of bifid mandibular condyle with possible traumatic cause, with emphasis on the radiographic and tomographic findings.

MeSH Key Words: *child; jaw abnormalities/radiography; mandibular condyle/abnormalities*

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The bifid mandibular condyle (BMC) represents a rare developmental anomaly first described in 1941,[1] and only a few cases have been reported since. Several factors have been cited as possible causes of BMC, including condylar fracture, developmental anomalies, perinatal trauma, teratogenic embryopathy and surgical condylectomy.[2,3] It usually affects only one condyle, and the observation of bilateral BMC is exceptionally rare.[4] BMC is usually identified as an incidental finding on routine radiographic examination.[5] This paper reports a clinical case of unilateral BMC possibly resulting from trauma, with emphasis on causative factors, diagnosis, radiographic and tomographic features, and management.

Case Report

A 4-year-old white boy was admitted for clinical evaluation 4 days after experiencing facial trauma in a bicycle accident. Facial asymmetry was observed on admission, with midline deviation to the right side associated with restriction of jaw opening and pain (**Table 1**). The child had not previously had any serious disease, and there was no significant family history of disease or anomaly. Dental panoramic radiography revealed intracapsular fracture of the right mandibular condylar head (**Fig. 1**). Initial treatment consisted of nonsurgical reduction (closed technique) of the condylar fracture under general anesthesia (via nasotracheal intubation) and maxillomandibular fixation. During the procedure, condylar repositioning was difficult even with use of a muscular relaxant. The patient was discharged from hospital 2 days after the procedure. Orthodontic therapy with fixed appliances and rubber bands was initiated to guide mandibular functional movements. The patient subsequently underwent clinical and radiographic follow-up every 6 months.

During the routine follow-up examination 2 years after the initial nonsurgical treatment, more severe facial asymmetry and midline deviation to the right side were noticed (**Table 1**), although the patient reported no pain. Radiographic examination at this time revealed changes in the shape and size of the right condyle as well as the collateral mandibular ramus and chin (**Fig. 2**).

The patient continued with orthodontic therapy, and at the 4-year follow-up the clinical findings were as follows (**Table 1**): jaw opening of 35 mm; severe facial asymmetry and midline deviation to the right side; painless, asymptomatic temporomandibular joints (TMJ); and preservation of mandibular function.

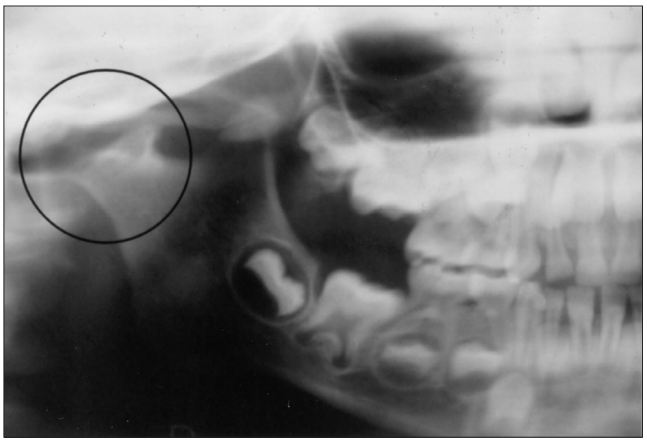

Figure 1: *Detail of dental panoramic radiograph obtained during the initial presentation shows fracture of the right condylar head.*

Figure 2: *Axial radiograph obtained at the 2-year follow-up. Alterations in the right condyle are indicated by morphological changes and deviation of the contralateral mandibular ramus and chin.*

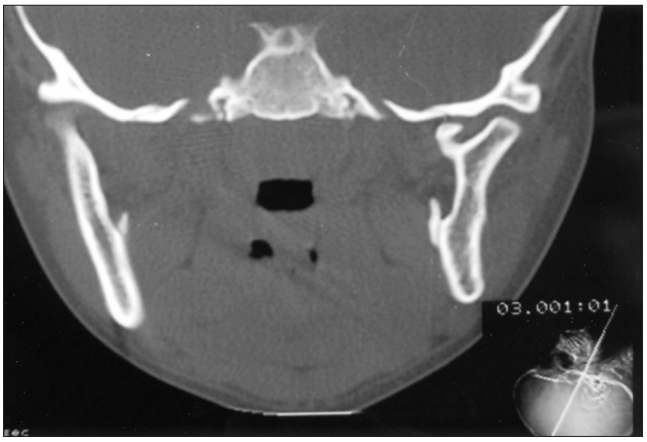

Figure 3: *Coronal computed tomography section with X, Y and Z axes, obtained at the 4-year follow-up, indicates bifid mandibular condyle on the right side.*

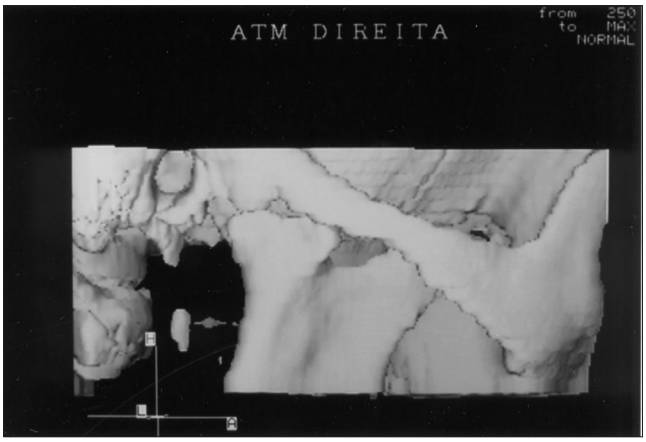

Figure 4a: *Three-dimensional reconstruction of the temporomandibular joint, based on helical computed tomography (performed at the 4-year follow-up). This image shows the affected side.*

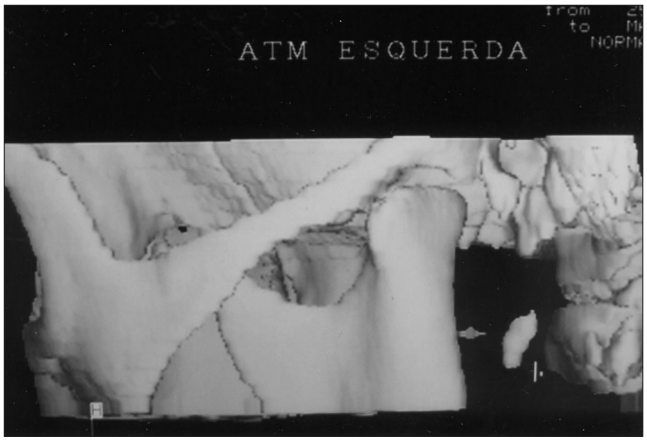

Figure 4b: *Three-dimensional reconstruction of the temporomandibular joint, based on helical computed tomography performed at the 4-year follow-up. This image shows the normal side.*

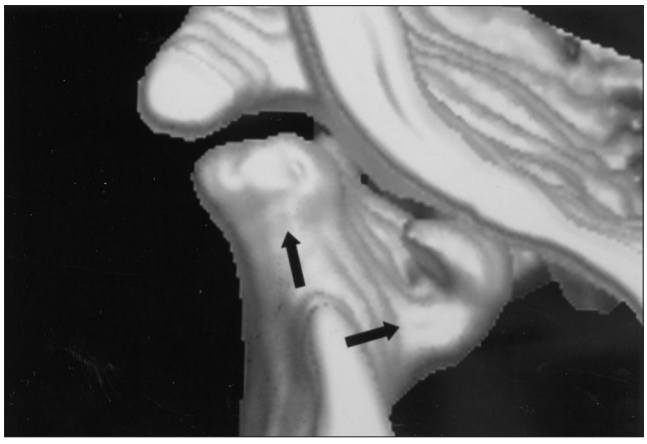

Figure 5: *Three-dimensional reconstruction based on helical computed tomography (performed at the 4-year follow-up) indicates the presence of a bifid mandibular condyle. The medial (left) and lateral (right) condylar heads are evident.*

Table 1 **Clinical data for a child with bifid mandibular condyle who experienced facial trauma at 4 years of age**

Time of evaluation	Clinical data		
	Facial asymmetry	Midline deviation	Jaw opening
Initial presentation	Yes	2 mm	40 mm
2-year follow-up	Yes	4 mm	36 mm
4-year follow-up	Yes	8 mm	35 mm

Unexpectedly, radiographic and tomographic examination at the same time clearly showed development of BCM on the right side, which led to the suggestion of a possible correlation with the previous condylar fracture (**Figs. 3** to **5**). Helical tomography and 3-dimensional (3D) reconstruction were performed at the Radiology Institute of Natal (Natal-RN, Brazil) with an Elscint Twin-Flash device (Elscint, Haifa, Israel). This scanning was performed in the axial plane according to usual TMJ protocols. The patient continues to receive regular follow-up. Once his facial growth has stopped, he will undergo surgical treatment consisting of maxillomandibular osteotomies and further orthodontic treatment.

Discussion

The first description of BMC was published by Hrdlicka,[1] who found 27 cases of this anomaly while analyzing male and female dried human skulls in a Smithsonian Institution collection. Since then, only a few cases have been reported in living human beings. Szentpétery and others[6] examined a total of 1,882 skulls in 1990 and identified 7 with signs of BMC; in 2 cases, 1 mandible had bilateral signs of grooving. According to Cowan and Ferguson,[5] at least 36 clinical cases of BMC had been reported up to 1997. MEDLINE was used to search the medical literature (from January 1998 through October 2002) for English-language case reports of BMC in living subjects. Only 2 additional papers, reporting a total of 5 cases, were identified, which yields a total of 41 cases (see **Table 2** at http://www.cda-adc.ca/jcda/vol-70/issue-3/158.html). These data indicate the rarity of this condition.

BMC has been reported in patients of various ethnic backgrounds and both sexes.[4,18] According to Loh and Yeo,[18] 67% of patients with BMC had no complaints related to the affected condyles: the condition was detected as an incidental finding during dental radiographic examination. Nonetheless, nonspecific symptoms have been detected in some cases, such as general pain, swelling, articular clicking and, occasionally, limited oral opening and deviation toward the affected side.[17,21]

The etiopathogenesis of BMC remains unknown, despite the various factors that have been suggested as possible causes.[5,15,22] For example, Blackwood[23] stated that the condylar cartilage, during the early stages of its development, is divided by well-vascularized fibrous septa and suggested that persistence of such a septum, in exaggerated form, within the growing cartilage might lead to an error in development that would in turn give rise to the bifid condition. This author also mentioned rupture of septal blood vessels as another possible cause of BMC. This theory might explain how forceps delivery, if it caused hematoma, could lead to BMC formation. However, Gundlach and others[15] found no evidence of persistent septa in the cases of BMC that they examined.

It must be stressed that the mandibular condyle region is a crucial centre of facial growth. Thus, injuries during childhood and puberty could lead to condylar malformations including BMC and severe facial asymmetry.[20] Post-fracture healing and remodelling of the mandibular condyle, if they involve lateral and medial fragments, have also been linked to development of ankylosis or BMC.[24] Szentpétery and others[6] stated that the site of the fracture and, most probably, its relation to the insertion of the lateral pterygoid muscle may determine future development of a normal or bifid condyle.

A possible genetic origin of this abnormality has also been suggested, and some authors have speculated that it occurs secondary to a variety of factors such as endocrine disturbances, exposure to teratogenic substances, nutritional deficiencies, infection and radiation.[25,26] Support for the latter suggestions comes from the work of Gundlach,[22] who experimentally induced bifid condyles in animals by injecting teratogenic substances such as *N*-methyl-*N*-nitrosourea and formhydroxamic acid in different concentrations at various stages of pregnancy. In addition, some reports of BMC have established a relation between the origin of the condition and the glenoid fossa: in developmental BMC there is a separate glenoid fossa for each of the 2 parts, whereas in traumatic BMC there is only one glenoid fossa.[6,9]

The reported existence of 2 different patterns of condyle bifidism might be related to distinct causes for each type. Thus, it is postulated that the anteroposterior pattern results from facial trauma during childhood, and the mediolateral form can be linked with persistence of the fibrous septa at the condylar cartilage.[6] Although this model may fit in the majority of cases, Cowan and Ferguson[5] gave several examples of BMC that did not fit this profile. Moreover, the degree of splitting ranges from a shallow groove on the condyle to 2 distinct condyles with a separate neck; the orientation of the head may be mediolateral (coronal) or anteroposterior (sagittal).[6,20]

Advances in dental science in the past few years have included the development and improvement of new diagnostic techniques that allow easier and earlier detection

of pathological conditions in the maxillofacial region. In this context, advances in radiographic and imaging methods have led to various options for visualizing the TMJ.

In general, the first diagnostic radiographic survey performed for examination of condyle disturbances is dental panoramic radiography, because of its wide availability and low cost.[21] However, computed tomography (CT) is undoubtedly the best choice for TMJ examination because it allows bilateral visualization without osseous superpositioning. Helical CT is now the state-of-the-art method for TMJ visualization, especially after facial trauma.[20] Helical CT, which is based on the acquisition of multiple continuous slices of the anatomic site of interest, has several advantages, such as complete recording of the area with a single scan, short examination times (which makes it especially suitable for children), lower radiation dose and better image quality.[20,27] With this technique it is possible to evaluate both quantitatively and qualitatively the effects of trauma on condyle size and shape, sclerosis and cortical irregularities, joint position, neck length, condylar and intercondylar angle, flattening of the articular eminence and depth of the glenoid fossa.[20,24] Three-dimensional reconstruction can then be used to assess condylar shape (including deformity) and neo-arthrosis or pseudo-arthrosis formation, as well as hypoplastic and hyperplastic changes at the condyle.[20] Furthermore, 3D reconstruction allows more accurate evaluation of condylar morphology than 2D tomographic images.[27]

The long-term functional effects of BMC are not well known because the literature on this subject is scant. Some authors have suggested that sagittal fracture may cause bifidism of the condyle.[6,24] Wu and others[19] demonstrated that surgically induced fracture of the mandibular condyle could produce secondary trauma to the disc and the glenoid fossa, with resultant osteoarthrosis of the injured TMJ. Thus, BMC linked to mandibular fractures could lead to the alterations described.

According to García-Gonzáles and others,[21] appropriate treatment for BMC depends on the symptoms. Asymptomatic cases do not require any treatment, although long-term follow-up is necessary. Patients with internal articular derangement should be treated with occlusal splints and arthroscopic surgery.[21] Condylectomy and arthroplasty for functional repair is recommended in cases with associated articular ankylosis; success depends on correct excision of the affected area.[17]

In the case described here, it is believed that the bifid condyle is related to the mandibular trauma suffered during childhood. The long-term treatment options are costochondral graft or osteogenic distraction. At the time of the future surgical treatment, maxillomandibular osteotomies will be performed. At present, the patient is only undergoing regular follow-up, because facial growth is incomplete and the success of surgical treatment at this time would be limited. Two major factors must be considered in determining the appropriate time for surgery: functional and esthetic considerations. If there are no problems with either maxillomandibular function or esthetic appearance, surgery should be postponed until the patient's facial growth has stopped. In the case described here, it has been possible to postpone surgical treatment because maxillomandibular function has been preserved. Although there is some facial asymmetry, the patient has no complaints in this regard. The precise orthodontic treatment cannot be predicted, but it will be designed to align the level of the dental arches. ◆

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Table 2 Summary of literature reports of bifid mandibular condyle[a]

Reference	No. of cases	Age (years)	Sex	Ethnicity	Side	Symptoms	Imaging technique
Hrdlicka[1]	27[b]	NA	NA	NA	NA	NA	NA
Schier[7]	1	NA	NA	NA	NA	Luxation	NA
Honée and Bloem[8]	1[b]	NA	NA	NA	NA	NA	NA
Stadnicki[9]	1	3	F	White	Left	Limitation of mouth opening	Laminagraphy, lateral oblique view
Lysell and Oberg[10]	1[b]	21	F	NA	Right	Pain in TMJ	Dental panoramic
Farmand[11]	1[b]	45	M	NA	Left	Painless circumscribed swelling in TMJ area	Dental panoramic, lateral view
Forman and Smith[12]	2	28	M	White	Left	Asymptomatic	Dental panoramic, reverse Towne tomography
		30	M	White	Left	Asymptomatic	Dental panoramic, transpharyngeal TMJ
Balciunas[13]	1	67	F	Black	Left	Asymptomatic	Dental panoramic
Shaber[14]	1	26	F	White	Bilateral	Asymptomatic	Dental panoramic, Towne tomography
Gundlach and others[15]	5	23	M	NA	Left	Opening click	Dental panoramic, saggital tomography
		NA	NA	NA	Left	Asymptomatic	Saggital tomography
		NA	NA	NA	Left	NA	Dental panoramic
		NA	NA	NA	NA	NA	Dental panoramic, saggital tomography
		NA	NA	NA	Right[b]	NA	NA
McCormick and others[16]	3	38	M	White	Bilateral	Asymptomatic	Dental panoramic
		61	F	White	Bilateral	Loud pop bilaterally	Dental panoramic
		50	M	White	Bilateral	Bilateral pop at completion of mouth opening	Dental panoramic
To[17]	1	39	M	NA	Left	Limitation of mouth opening	Dental panoramic
Loh and Yeo[18]	4	24	M	Chinese	Left	Asymptomatic	Dental panoramic
		21	M	Chinese	Right	Asymptomatic	Dental panoramic, transorbital TMJ
		27	F	Malay	Right	Clicking and pain in TMJ	Dental panoramic, transorbital TMJ
		NA	NA	NA	Right[b]	NA	NA
Szentpétery and others[6]	7[b]	NA	5F	NA	5 right	NA	NA
		NA	2M		2 left	NA	NA
Wu and others[19]	2	21	M	NA	Right	NA	NA
		23	M	NA	Right	NA	NA
Kahl and others[20]	1	14	F	NA	Right	NA	CT
Cowan and Ferguson[5]	1	24	F	NA	Left	Limitation of mouth opening	Dental panoramic, transpharyngeal TMJ, CT

NA = not available, F = female, M = male, TMJ = temporomandibular joint, CT = computed tomography

[a]*Adapted from Loh and Yeo.*[18]

[b]*Study in skulls or cadaver.*

Table 2 **Summary of literature reports of bifid mandibular condyle[a] (continued)**

Reference	No. of cases	Age (years)	Sex	Ethnicity	Side	Symptoms	Imaging technique
Stefanou and others[4]	4	55	F	White	Bilateral	Asymptomatic	Dental panoramic, open and closed TMJ lateral view
		47	M	White	Bilateral	Asymptomatic	Dental panoramic, open and closed TMJ lateral view
		39	F	White	Bilateral	Asymptomatic	Dental panoramic, open and closed TMJ lateral view
		69	F	White	Bilateral	Pain	Dental panoramic, open and closed TMJ lateral view
García-González and others[21]	1	63	M	NA	Left	Limitation of mouth opening	Dental panoramic, lateral tomography, CT

NA = not available, F = female, M = male, TMJ = temporomandibular joint, CT = computed tomography

[a]*Adapted from Loh and Yeo.*[18]

[b]*Study in skulls or cadaver.*