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Bilateral bifid condyles: a rare etiology of temporomandibular joint disorders

Short title:

Bilateral bifid condyles

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Abstract (250 words)

Background: Bifid mandibular condyle (BMC) is a rare etiology of temporomandibular joint (TMJ) disorders characterized by a duplication of the head of the mandibular condyle.

Case report: The authors report the case of a 20-year-old patient complaining of a painful and clicking TMJ and mandibular hypomobility which had been progressing for several months. Radiological investigations (dental panoramic radiograph and X-ray CT scan) revealed right and left abnormalities of the TMJ due to bilateral BMC requiring surgical management.

Conclusion: Despite a prevalence of 0.31% to 1.82% and the controversies surrounding its pathophysiology, maxillofacial surgeons should be aware of BMC to avoid misdiagnosis related to the clinical presentation (pain, clicking, hypomobility, or ankylosis) and provide adequate management.

Keywords:

temporomandibular joint / bifid mandibular condyle / temporomandibular dysfunction / management

Text

1. Introduction

Described for the first time by Hrdlicka [1] as a condylar split or groove of variable depth, biffid mandibular condyle (BMC) is an extremely rare temporomandibular joint (TMJ) disorder with a prevalence of 0.018% to 1.82% [2,3]. BMC can be asymptomatic or present non-specific clinical signs and symptoms such as pain, swelling, noise, hypomobile joint blockage, deflection, joint luxation, or even ankylosis [4]. The diagnosis of BMC is most often made after an incidental finding on a panoramic radiograph during a routine exam and confirmed by computed tomography (CT), considered as the gold standard for confirmation of its diagnosis [5-7].

2. Case Report

The authors report the case of a 20-year-old female patient, with no relevant past medical or surgical history, coming to the maxillofacial department complaining of chronic pain from both TMJ, with progressive worsening in the previous months without any objective triggering factor. The clinical examination revealed pain upon TMJ palpation, predominantly on the right side, with associated audible clicking as well as a limited mouth opening measuring 21 mm, a right lateral deviation, and inability to perform mandibular propulsion (Figure 1). To rule out an organic etiology, a panoramic radiograph (Figure 2a) was performed and showed an atypical deformity of the left mandibular condyle requiring additional investigation. A dynamic X-ray CT scan with open and closed mouth positions confirmed the diagnosis of bilateral BMC with mediolateral orientation of the condyle head

and a predominant TMJ abnormality on the right side explaining the clinical features (Figure 2b and 2c).

Considering the progressive worsening of symptoms (pain, limited mouth opening on the right side, and ankylosis), the authors decided to perform a bilateral arthrotomy with the resection of lateral articular condyle surfaces with Piezotome surgery through a preauricular bilateral approach (Figure 3). The TMJ exploration revealed a healthy disc, with no degenerative lesions or visible perforations. Pathology findings of the resected lateral joint surfaces were in favor of the excision of healthy condylar heads without suspicion of malignancy, with bone spans and cartilage surfaces demonstrating completely normal architecture and thickness.

At one month post surgery, the clinical examination showed an improvement in mouth opening (measuring 3 cm), restoration of mandibular propulsion, and correction of deviation during mouth opening on the right side. After 12 months post operative and 9 months of active rehabilitation, the patient no longer reported pain, clicking, or limitation of the mouth opening (measuring more than 5 cm) (**Figure 4**).

3. Discussion

BMC is a rare etiology of TMJ disorders [2,3], and awareness and knowledge about this condition is essential for better understanding of its pathophysiological process and its potential progression. This will allow clinicians to establish an appropriate therapeutic management system. The increasing number of papers on BMC bears witness to its importance and confirms the difficulty in determining its etiopathogenesis and impact.

Although BMC can be unilateral or bilateral (ratio 2:1), in 1990 Szenpetery et al. [8] suggested that the anteroposterior position of the condyles most likely results from early childhood fractures, while the mediolateral condyle orientation was associated with a non-traumatic etiology, probably due to persistent developmental septa. However, the description of post-traumatic mediolateral BMC clearly questions this theory. In a recent meta-analysis, Guven [9] identified two types of BMC. The author proposed differentiating between a type I non-traumatic, non-symptomatic mediolateral BMC from a type II with a history of mandibular trauma to the anteroposterior or mediolateral BMC. This study subdivided type II into 2 groups: a) with a Y-shaped condyle secondary to an intracapsular or vertical condyle trauma; and b) with two separate and anteroposteriorly located condyles, where the formation of the anteroposterior condyle might be due to insufficient remodeling of the ramus/condyle of the displaced or dislocated subcondylar fracture resulting from force exerted by the lateral pterygoid muscle.

However, for Dennison et al. [10], rather than the orientation of the articular surfaces of the BMC, clinical aspects should be favored. They preferred to refer to "condylar notching", for asymptomatic BMC with a history of incidental discovery, and "true" BMC with a clinical impact: pain, clicking, limited freedom of mandibular movement, in some cases progressing to "true" mandibular ankylosis.

This classification is more relevant, as it makes it possible to guide the therapeutic strategy which favors foregoing therapeutic treatment and instead recommends regular monitoring in the case of asymptomatic BMC. Stepwise management is recommended in the case of "true" BMC guided by the clinical symptoms. Thus, symptomatic treatment with non-steroidal anti-inflammatory drugs, analgesics, physiotherapy, or an occlusal splint is recommended as a conservative approach [11, 12] in case of pain, creaking, or swelling. For hypomobility, severe pain, or ankylosis, surgical treatment [13-15] (arthroscopy, arthrotomy, or

condylectomy with reconstruction with TMJ prosthesis or costal graft) is described, with the aim of restoring function. In this case, the severe symptomatology and mandibular hypomobility make it difficult to envisage functional treatment. Thus, in the current study, the authors opted for bilateral arthrotomy of the lateral articular surfaces.

The choice of the preauricular pretragal approach may be opposed to other condylar approaches (submandibular, retroauricular, or endobuccal). Nevertheless, it offers the advantage of complete exposure of the TMJ with minimal esthetic sequelae and no risk of secondary ear canal stenosis. Its use requires careful dissection in the auricular perichondral plane until contact with the zygomatic arch, then moving towards the TMJ. This dissection under the plane of the facial nerve limits the risk of nerve damage. It is nevertheless important to control the force exerted on the parotid tissue to avoid any stretching of the nerve branches.

4. Conclusion

Bifid condyle is a relatively rare condition whose prevalence is increasing, and it requires clinicians to rule out a probable misdiagnosis and understand its real pathophysiology. In these cases, it seems important to propose a precise examination and to complete the assessment with an X-ray CT scan. Regarding its management:

- In the event of incidental discovery, regular monitoring is required to ensure appropriate early management.
- In cases with a clinical impact, bilateral arthrotomy using the preauricular approach is a functional and esthetic surgical alternative provided that a rigorous active physiotherapy plan is followed.

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Captions to illustrations

Figure 1: Clinical view of mandibular hypomobility and deviation at the mouth opening on the right side.

Figure 2: (a) Panoramic radiograph and (b-c) 3D volume rendering reconstruction of the X-ray CT scan of the patient confirming the right (white arrow) and left (black arrow) bifid mandibular condyles.

Figure 3: 3D volume rendering reconstruction of the postoperative X-ray CT scan of the patient showing the lateral arthrotomy of the lateral head of the (a) right (white arrow) and (b) left condyle (black arrow).

Figure 4: Clinical view at 12 months post surgery and after 9 months of active physiotherapy showing an oral opening measuring more than 5 cm, the correction of the lateral deviation, and a discreet preauricular scar.

Conflict of interest statement:

The authors declare that they have no competing interest.

Financial disclosure:

None

Human rights statements and informed consent:

The patient's informed consent has been obtained for the use of his photos and personal data.

Keywords:

temporomandibular joint / bifid mandibular condyle / temporomandibular dysfunction / management

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