Rare disease

Bifid mandibular condyle

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Summary

A 20-year-old young male patient presented with limited mouth opening and cosmetic disfigurement since childhood. On examination, marked mandibular hypoplasia resulting in a convex facial profile was noted. Further radiographic investigation using orthopantomograph and CT scan with three-dimensional reconstruction revealed bifid mandibular condyle of the right side.

BACKGROUND

This is a unique case of bifid mandibular condyle (BMC) of the right side reported in a young patient, which brings together the fields of oral medicine and radiology, oral surgery and orthodontics.

Although this condition is rare, an increased number of cases have been diagnosed in the last two decades, with the advent of imaging techniques like CT, three-dimensional imaging, as well as improved quality of radiological images. The purpose of this article is to report a unique case of unilateral bifid condyle.

CASE PRESENTATION

A 20-year-old male patient reported to the Department of Oral Medicine and Radiology, complaining of pain in relation to upper anterior teeth after a fall 2 years back. The patient also revealed a history of limited mouth opening and cosmetic disfigurement since birth. On clinical examination fracture in relation to 11, 12 and 21 were observed.

Further examination of the head and neck region revealed a marked mandibular micrognathia and a convex profile (figure 1). Neither the patient nor his parents reported any history of childhood trauma. Also, no history of snoring or drug intake was given. The patient had no difficulty in speech or swallowing. Mouth opening was limited to 27 mm and lateral jaw movements were restricted. The patient's left eye was blind since birth. The patient reported no pain in response to palpation of the masticatory muscles or the temporomandibular joint (TMJ). No clicking sounds were detected on manipulation of the TMJ.

A provisional diagnosis of Elli's class III fracture was made in relation to 11, 12 and 21.

A secondary provisional diagnosis of bilateral intra-articular TMJ ankylosis was made based on the clinical findings.

INVESTIGATIONS

The patient was further subjected to radiographic examination. The intraoral periapical radiograph confirmed Elli's grade III fracture in relation to 11, 12 and 21.

Radiological investigation of the TMJ yielded very interesting results. The panoramic radiograph suggested a possible bifid morphology of the right mandibular condyle

which was further confirmed using axial and coronal CT. Three-dimensional (3D) reconstruction of the CT revealed the distinct medial and lateral heads. The left condyle, however, presented a slightly bulbous appearance than normal.

Diagnosis of BMC was confirmed using OPG, CT and MRI. 3D CT images provided a vital role in the diagnosis. MRI was done out of interest and showed bilobed appearance of the condyle (figures 2–6).

DIFFERENTIAL DIAGNOSIS

- Myospasm could be suspected as it presents with trismus and limitation of jaw movements, but the

Figure 1 Profile photo of the patient showing mandibular growth deficiency.

Figure 2 OPG suggesting bifidism of the right mandibular condyle.

absence of masticatory muscle tenderness or dysfunction and the lack of any trigger points eliminate the condition. Moreover, marked mandibular hypoplasia was a very contrasting feature.

- ▶ Osteoarthritis could also be considered as it also presents with limited jaw movements and trismus, but it is usually seen in the elderly and more frequently in women. Pain due to synovial inflammation is also a feature of arthritis which was absent in this case.
- ▶ A diagnosis of capsulitis/synovitis could be made, as the limitation of jaw movements is usually seen in this condition, but the lack of continuous pain that exacerbates during function, clearly rules out the condition.
- ▶ Rheumatoid or septic arthritis could be a likely diagnosis as it is also associated with trismus, but the absence of any bacterial infection or inflammation involving multiple joints easily separates out the condition.

TREATMENT

The patient was advised to undergo a combined orthodontic-surgical treatment for the correction of facial skeletal anomaly and the rehabilitation of his mouth opening and mandibular functional movements.

Figure 3 CT image showing clefting of the right condyle.

Figure 4 3D reconstructed CT images showing the 2 lobes of the right condyle.

OUTCOME AND FOLLOW-UP

However, the patient was not willing to undergo surgery.

DISCUSSION

BMC is a rather uncommon entity with a very unclear aetiopathology. It may be either developmental, or secondary to trauma.^{1 2} It is usually diagnosed during routine radiographic examination or after physical examination of

Figure 5 3D reconstructed CT images showing the 2 lobes of the right condyle.

Figure 6 MRI suggesting bifid right condyle.

a patient referred for TMJ disorders, pain and trismus. From the first description of BMC in a patient in 1948, until 2010, researchers have reported a total of 131 cases in living people.³ The fact that the condition is usually asymptomatic makes it more difficult to diagnose.

The term 'bifid' is derived from the Latin word '*bifidus*' which means cleft into two separate parts.³ The BMC or double-headed condyle is considered to be a rare pathology of the TMJ characterised by the duplicity of the head of the mandibular condyle. The articulating surfaces of the bifid condyle are divided by a groove and can be oriented mediolaterally or anteroposteriorly. The anomaly may occur on both sides but is frequently unilateral, apparently without any marked predilection for any one side. The condylar splitting ranges from a shallow groove to two distinct condyles with a separate neck.^{4 5}

In 1941, Hrdlicka first described BMC in mandibles from dissected human skulls in Smithsonian Institution, Washington. He had reported 21 cases of which 18 were unilateral and 3, bilateral.^{1 3 6-11} He defined the morphological anomaly as a mandibular condyle split or divided into two parts by a sulcus or groove of variable depth.³ The first report on this condition in a living individual was made in 1948 by Schier.^{1 8 9 12} According to Cowan and Ferguson, at least 36 clinical cases of BMC had been reported up to 1997.¹³ This high frequency suggests that BMC is a more frequent condition than is presently perceived. Menezes *et al*¹⁴ examined 50 080 panoramic radiographs between 1999 and 2006 and reported only nine bifid condyle cases. Subsequently, in 2010, Miloglu *et al*¹⁵ examined 10 200 panoramic radiographs in a Turkish population and reported 32 BMC. The study of 1882 cadaveric skulls by Szentpetery *et al* in 1990 found the incidence of this anomaly to be 0.48%.^{16 17}

Although the exact aetiology is not known, various theories have been suggested. It is said that it may be a developmental abnormality or secondary to trauma.^{1 8} Although it is not confirmed, infection, irradiation, exposure to teratogens, genetic discrepancy, endocrinal, pharmacological and nutritional disorders may also play a vital role.^{1 3} Hrdlicka postulated that obstructed blood supply to the condyle during its development caused the division of the condyle. Blackwood suggested the persistence of a

fibrous septa (seen during early stages of development of the condyle) in the exaggerated form within the growing cartilage, might lead to an error in the development, that would in turn give rise to the bifidism of the condyle.^{3 17} Tejasvi *et al* in the literature review stated that bifidism of condyle, could be due to trauma as per Walker's experiments on monkeys.⁷ Poswillo *et al* stated that bifidism occurs as a result of changes in the position or form of the disc, leading to the formation of intra-articular septa across the joint space.

Gundlach *et al* had experimentally induced bifid condyles in rats by injecting teratogenic substances like *N*-methyl nitrosurea in various concentrations.¹⁸ He believed that bifid condyle was caused by a combination of teratogenic agents and the misdirection of the muscle fibres, which then influence bone formation. Another theory postulates trauma as the cause with dislocation of joint integrity due to birth trauma, condylar fractures or surgical condylectomy.¹ The site of the fracture and its relation to the insertion of the lateral pterygoid muscles are factors determining the future development of bifid condyle.¹

Other authors proposed that trauma involving TMJ or micro trauma due to malocclusion might cause bifid condyle (Artvinli & Kansu; Heskert *et al*; de Sales *et al*, 2004; Fuentes *et al*, 2009).¹⁰ According to the current English literature, history of condylar trauma is evident in approximately 25% of all cases reported.^{19 20} It has been postulated that anteroposterior splitting of the condyle occurs in patients due to trauma while mediolateral splitting is usually developmental in origin.¹ Yao *et al* however demonstrated experimentally that a fracture of the mandibular condyle could result in mediolateral as well as anteroposterior orientation of mandibular condyles.¹⁷

Majority of cases are detected during routine radiographic examination. In most cases, patients have no symptoms. However, it may be associated with pain, swelling, trismus, facial asymmetry, restricted mouth opening and most commonly TMJ clicking.^{1 21 22} In this case, restricted mouth opening was a marked symptom.

Treatment in BMC cases is usually conservative and similar to the treatment for the closely associated TMJ pain dysfunction syndrome (analgesics, anti-inflammatory agents, muscle relaxants, physiotherapy and splint) (Quayle *et al*²²). Surgical treatment is described in TMJ ankylosis that used to appear in BMC secondary to trauma (Corchero-Martin *et al*, 2005).²³

CONCLUSION

BMC is a rare anatomic anomaly causing division of the mandibular condyle by a sulcus or groove of variable depth. It may manifest in different forms, with bilobular being the most common. Investigators have suggested that a congenital origin is the primary cause, although trauma may alter condylar growth, resulting in an anatomical defect. As no history of trauma was revealed, our case may be developmental in aetiology. The diagnosis of BMC usually rests on radiological, rather than clinical evidence. Due to advanced imaging modalities, many more cases will be highlighted in future and hence, dental professionals should have appropriate knowledge about its anatomical abnormality, as well as its implications, for a potential diagnosis.

Learning points

- ▶ Bifid mandibular condyle (BMC) can be considered as a rare but definite cause for trismus.
- ▶ CT scan is the best modality for detection of BMC because it allows detailed evaluation of condylar morphology.^{11 24}
- ▶ Overlapping of structures can hide the bifid appearance in an OPG.

Competing interests None.

Patient consent Obtained.

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