

Mandibular Coronoid Aplasia with Condylar Hypoplasia: A Very Rare Case with the Review of Literature

Kondiler Hipoplazi ile Birlikte Mandibular Koronoid Aplazi: Literatür Taraması ile Çok Nadir Bir Olgu

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ABSTRACT Mandibular coronoid aplasia is defined as the absence of the coronoid process and hypoplasia is defined as defective or underdeveloped coronoid process. Hypoplasia of the mandibular condyle means that the condyle is smaller than normal. Both of these situations are very rare in the literature. In the literature, condylar hypoplasia is usually associated with coronoid hyperplasia and is seen together. The case report we prepared is a case of unilateral coronoid aplasia with condylar hypoplasia, which is not associated with the syndrome, and is the second case in the literature in this way. In the clinical examination of a 38-year-old female patient, a decrease in the right vertical face height and a deviation to the right at the chin tip were observed. As a result of radiological evaluation, right coronoid process aplasia and right condyle hypoplasia were observed.

Keywords: Mandible; temporomandibular joint; condylar hypoplasia; coronoid aplasia

ÖZET Mandibular koronoid aplazi; koronoid çıkıntının mevcut olmaması, hipoplazi ise kusurlu veya az gelişmiş olması olarak tanımlanır. Mandibular kondilin hipoplazisi ise kondilin normalden küçük boyutta olmasıdır. Literatürde bu 2 durum çok nadirdir. Literatürde kondiler hipoplazi genellikle koronoid hiperplazi ile ilişkilidir ve beraber görülür. Hazırladığımız vaka raporu, sendromla ilişkili olmayan, kondiler hipoplazi ile birlikte görülen, tek taraflı koronoid aplazi olgusudur ve literatürde bu şekilde görülen 2. vakadır. Otuz sekiz yaşında olan kadın hastanın klinik muayenesinde, sağ dikey yüz yüksekliğinde azalma, çene ucunda sağa sapma izlendi. Radyolojik değerlendirme sonucunda, sağ koronoid çıkıntı aplazisi ve sağ kondil hipoplazisi izlendi.

Anahtar Kelimeler: Mandibula; temporomandibular eklem; kondiler hipoplazi; koronoid aplazi

Mandibular coronoid aplasia is defined as the absence of the coronoid process and hypoplasia is described as defective or underdeveloped coronoid process. It is a very rare developmental anomaly of the oral and maxillofacial region.¹ While changes in temporal muscle activity contribute to the development of the condyle and coronoid process. The increase or decrease in the activity of temporal muscle may result in hyperplasia, hypoplasia or aplasia of bone.² Increased temporal muscle activity causes coronoid hyperplasia.³ However, decreased temporal muscle activity may be observed in coronoid hypoplasia, and unchanged temporal muscle activity may accompany

the condition.^{2,4,5} Due to the low number of coronoid hypoplasia cases no consensus could be reached on its etiology. Coronoid hypoplasia can be seen together with the syndrome or can occur alone.

According to our literature research; this is the sixth case of mandibular coronoid hypoplasia documented in the literature. Four of the other cases are bilateral coronoid hypoplasia.^{1,4,6,7} Only one case is unilateral coronoid hypoplasia.⁸ Our case is the second unilateral coronoid aplasia case reported so far. In this report; A case of coronoid aplasia with condylar hypoplasia of a 38-year-old female patient not associated with any syndrome will be presented.

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CASE REPORT

A 38-year-old female patient was admitted to the oral, dental and maxillofacial radiology department of our faculty with complaints of facial asymmetry. In her anamnesis, it was learned that she did not have any systemic disease, had no trauma, did not have a similar situation in her family, and had not received any surgical treatment before.

An informed consent form was obtained from the patient.

In extraoral examination; a decrease in vertical face height was observed on the right side of the patient. The chin tip had deviated to the right (Figure 1). Apart from these, no facial syndromic abnormality was observed. In the intraoral examination, the patient was in the centric position, while the teeth on the right side were in occlusion, there was an open bite on the left side. Maximum mouth opening was within normal limits (40 mm) with deflection to the right in the midline (10 mm) (Figure 1). When we examined the panoramic radiograph of the patient, the right condyle was hypoplastic (white arrow), and the right coronoid process was aplasic (black arrow) (Figure 2).

3D cone beam computed tomography (CBCT) was taken from the patient. According to the coronal, sagittal and axial sections the right mandibular condyle was hypoplastic and the right coronoid process was aplasic (Figure 3).

Condyle dimensions were measured in the mesiodistal and anteroposterior directions in the axial

sections. The right condyle was measured 13.4x5.7 mm and the left condyle was measured 19.9x8.3 mm (Figure 3).

On panoramic radiography; linear measurements were made by determining various anatomical points. Co, O1, O2, A line, B line, condyle height (CH), ramus height (RH) were determined according to Habets et al. (Figure 4).⁹

Linear measurements made are shown (Figure 5).

CH: Distance between Co-O1

RH: Distance between O1-O2

Measurements were performed on both the right and left sides. The right condyle and ramus were shorter than the left.

The asymmetry indices were determined using the formula developed by Habets et al.⁹

Asymmetry Index (AI)=[(Right-Left)/(Right+Left)]×100

In addition, the gonial angle is one of the important parameters that gives an idea about the vertical parameters and symmetry of the facial skeleton. For this reason, an artificial cephalometric was obtained from the patient's CBCT images, and right and left gonial angles were measured on this image. The gonial angle was measured by taking the tangent to the posterior border of the ramus and tangent to the lower border of the mandible on lateral cephalogram.¹⁰ Right gonial angle value was 134.5; left gonial angle value was 129.2 in 3D cephalometric images (Figure 6).



FIGURE 1: Showing extraoral and intraoral findings.

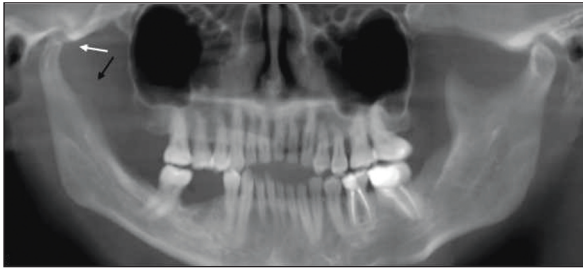


FIGURE 2: Panoramic radiograph of the patient. Right condyle hypoplastic (white arrow), Right coronoid process aplasia (black arrow).

Various treatment approaches were recommended to our patient. Multimodal treatment was suggested with the help of oral, dental and maxillo-facial surgeons, general surgeons, plastic surgeons and orthodontists.

DISCUSSION

Coronoid aplasia is a rare condition and its etiology is unclear.^{1,2} In 1978, Gorlin and Langer noticed that

the coronoid process was hypoplastic in a patient on routine radiographic examination. The same patient had rounded mandible angles, cystic formations in the mandibular molar region and ramus region, and cystic formations in the lower part. The second and third molars had not formed. The condition of this patient was reported as Melnick-Needles syndrome characterized by osteodysplasia.⁴ Our case is a case of unilateral coronoid aplasia not associated with any syndrome.

Of the 5 patients in the literature, 3 are female and 2 are male.^{1,4,6-8} Our patient is a female.

Four of previous 5 cases were bilateral coronoid hypoplasia.^{1,4,6,7} One was a case of unilateral coronoid hypoplasia.⁸ In our patient, only the right condyle is hypoplastic and coronoid aplasia, and the left side has a normal appearance. This means that our patient is the second case of unilateral coronoid aplasia in the literature.

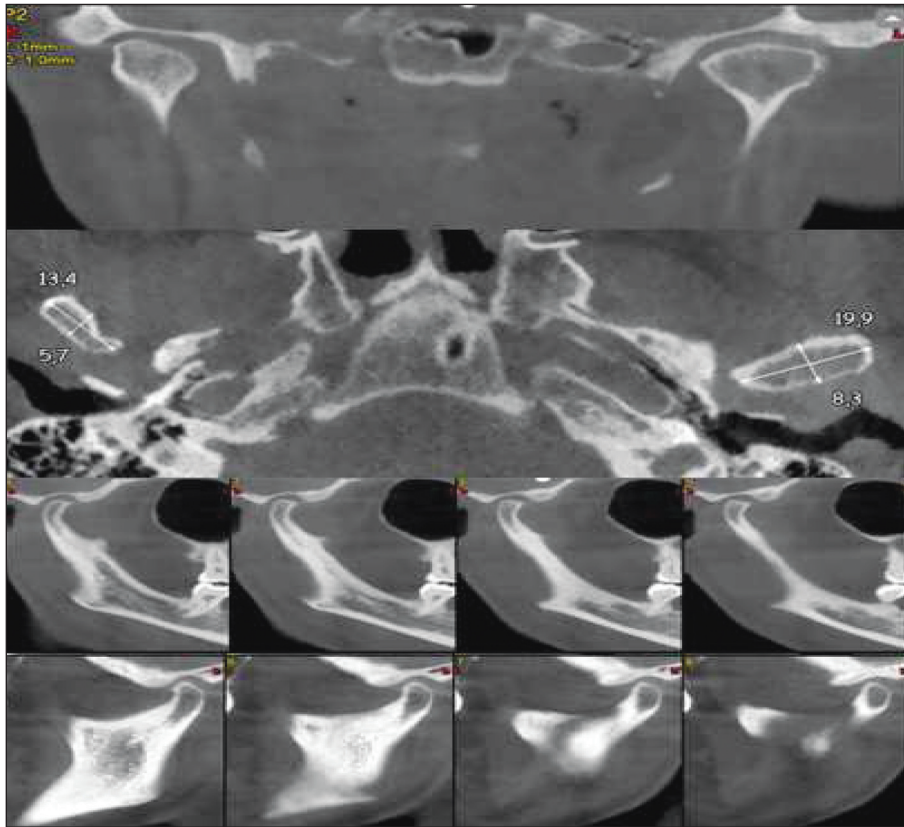


FIGURE 3: Cone beam computed tomography axial, coronal ve sagittal sections; right condyle hypoplastic, right coronoid process aplasia.

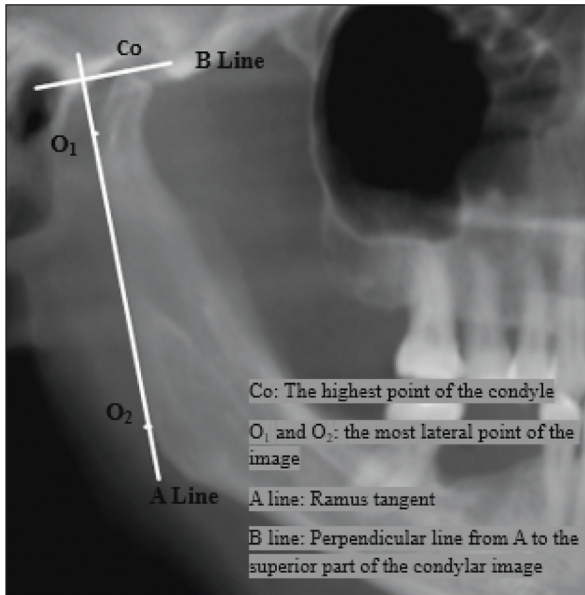


FIGURE 4: Panoramic image; anatomical points and definitions.

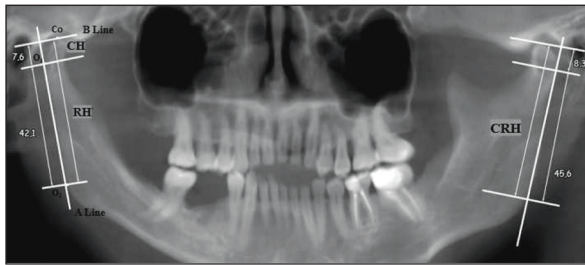


FIGURE 5: Panoramic image; linear measurements.
 CH: Condyle height; RH: Ramus height.

Condylar hypoplasia is a rare anomaly known as condylar underdevelopment. It may be seen as congenital or acquired.¹¹

Many scientists such as Petrovic et al., Björk and Skieller demonstrated that the condyle supported mandibular development in their studies.^{12,13} Any disruption in the growth of the condyle will also result in a reduction in the vertical dimension of the face.¹⁴ This pathology creates a classic “bird face” appearance when seen on the condyles on both sides, and creates asymmetry when seen on one side. In our case, there is an asymmetry. In the measurements we made on the CBCT sections, the height and width of the right condyle are less than the left. Likewise, the right RH is less than the left. Accordingly, the patient has an asymmetry where the chin tip clearly deviates to the right. In this case report, clinical and radiographic features; are similar to the cases discussed in the literature.^{1,4,6-8}

Differently from other cases in the literature; we made various measurements on the sections we obtained from the CBCT images of our patient.

According to the measurements made on panoramic radiograph obtained from CBCT, the right condyle and ramus are shorter than the left (Figure 5). The patient’s AI was calculated by using the CH and RH values.

$$AI = \frac{(\text{Right} - \text{Left})}{(\text{Right} + \text{Left})} \times 100 \quad (9)$$

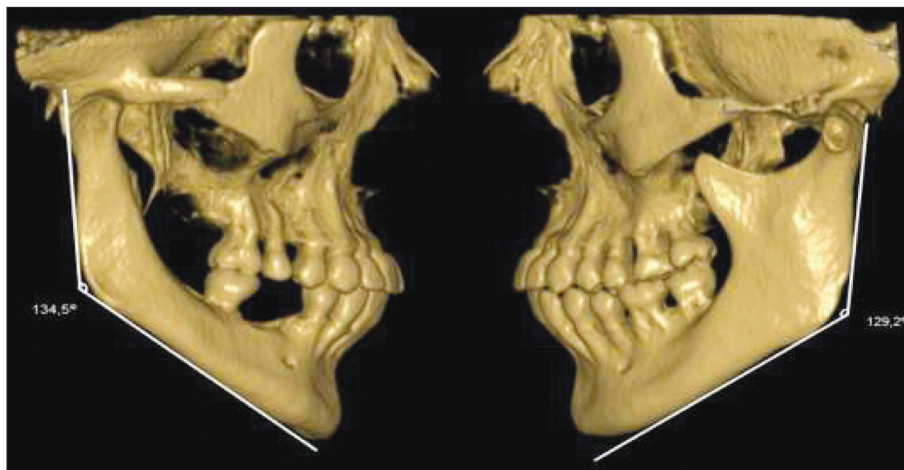


FIGURE 6: Cone beam computed tomography 3D image; right and left gonial angle measurement.

Condyl AI: $[(7.6-8.3)/(7.6+8.3)] \times 100 = 4.40$

Ramus AI: $[(42.1-45.6)/(42.1+45.6)] \times 100 = 3.99$

According to the formula developed by Habets et al., when AI > 3%, it indicates the presence of asymmetry.⁹ In our patient, AI was greater than 3% in both condyle values and ramus values.

In addition, gonial angle is one of the important parameters giving an indication about the vertical parameters and symmetry of the facial skeleton.¹⁰ The right gonial angle of this patient is larger than the left (Figure 6). The reason for this may be that the height of the right ramus is less than the left. This difference; supports the patient's asymmetry.

This case is a unique case of unilateral and ipsilateral coronoid and condylar hypoplasia. These patients need to be carefully distinguished from syndromes and other conditions of similar appear-

ance. In addition, the appropriate treatment modality should be planned for both functional activity and aesthetic appearance.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

All authors contributed equally while this study preparing.

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