



## Mandibular coronoid process tumor resembling a mandibular condyle: A case report



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### ABSTRACT

Abnormal elongation of mandibular coronoid process, often defined as coronoid hyperplasia, is a rare condition, which is frequently associated with limited mouth opening. In some cases, the enlarged coronoid pushes the zygoma forward causing facial asymmetry. This case report describes a 16-year-old boy whose chief complaint was a progressive difficulty and deviation in mouth opening, together with a deformity appearing at maximum opening at the zygomatic area. The diagnosis was Unilateral Accessory Mandibular Condyle at coronoid process, without reduction of the mouth opening capacity. A coronoidectomy was carried out by means of piezoelectric surgery, instead of a coronoidotomy which is usually performed in these cases, due to a suspect of ramus neoplasm.

### 1. Introduction

Accessory Mandibular Condyle is a rare anomaly that sometimes arises from the mandibular coronoid process, much like a coronoid process hyperplasia [1]. This condition usually causes slow and progressive reduction of mouth opening [2,3] because of the impingement of the enlarged coronoid processes against the medial surfaces of the body of the zygomatic bones. It generally occurs at puberty and continues over the years. It can be associated with facial asymmetry.

Etiology includes temporalis muscle hyperactivity, trauma and neoplasia, but in many cases there is no clearly identifiable pathogenesis. Some authors suggested that the coronoid reactive elongation depends on the increased pull of the temporalis muscle [4,5], or that bony hypertrophy of the coronoid process and mandibular angle could be the result of a prolonged and significant contraction of the masticatory muscles [6]. Unilateral Coronoid Hyperplasia can also be the consequence of a trauma or a pathologic condition such as a

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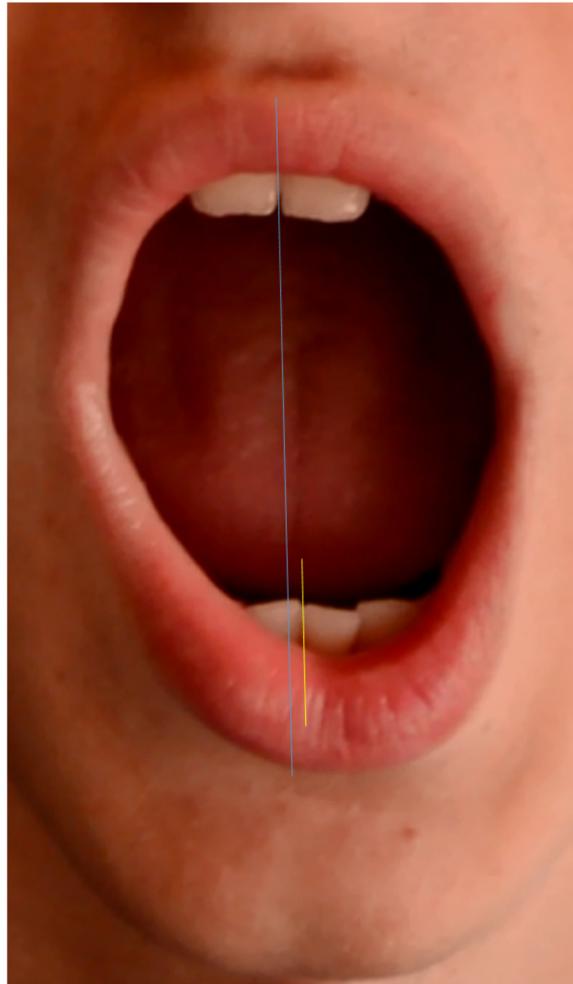


Fig. 1. Patients light deviation in opening pattern.

neoplasm (eg. Osteochondroma) [7,8]. A pseudo-joint between the coronoid process and the zygomatic arch due to an osteochondroma was first described by Jacob in 1899 [9]. The differential diagnosis of coronoid hyperplasia includes all causes of ankylosis and pseudo ankylosis of the temporomandibular joint, in which an overgrowth of the coronoid process causes a pseudo-joint formation with the zygomatic arch [7,10–12].

The diagnosis can be made with panoramic x-ray imaging, which is a widely available screening modality, or via 3D computed tomography [13–15]).

As Mulder reported in a systematic review there is no significant incidence and prevalence regarding Coronoid Process Hyperplasia (CPH) [16]. The mean age of patients affected by CPH is 23 years and the age distribution has a peak between 15 and 19 years. Usually there is a time frame of approximately 7 years between disease onset and diagnosis. The condition can be uni or bilateral, with the bilateral form occurring 4.1 times more frequently than the unilateral one, and with a male-to-female ratio of 3.3 to 1 [16,17].

Several surgical approaches have been proposed such as extraoral/intraoral coronoidectomy along with a histopathological examination in order to determine the nature of the specimen. As Mulder reported in his review the therapeutic approach was a coronoidectomy in 84% of the cases [16].

This is a report of a case with non-neoplastic accessory mandibular condyle located at the coronoid process without limitation of mouth opening, treated with transoral piezoelectric surgery.

## 2. Case report

A 16-year old boy was referred to the Section of Temporomandibular Disorders and Orofacial Pain of the University of Naples Federico II, complaining about a progressive difficulty and deviation in mouth opening, with a slight face deformity appearing during maximum opening at the zygomatic area.

Two years prior to his first examination, the patient had a facial trauma and referred for a progressive development of a mandibular



Fig. 2. First visit panoramic x-ray.



Fig. 3. 3D VR Image frontal open.



Fig. 4. 3D VR Image submental open.

deviation during mouth opening. The mandibular range of motion was as follows: active mouth opening 60mm, passive opening 62 mm, right lateral movement 11 mm, left lateral movement 13 mm, protrusion 6 mm. The patient did not report any masticatory muscle pain, nor TMJ pain or facial pain, both spontaneous and at palpation. The opening pattern showed an uncorrected deviation of about 2 mm towards the left side (Fig. 1). A slight facial asymmetry, with swelling at the right zygomatic region, appeared at maximum opening.

During all mandibular movements, a clicking noise was reported by the patient and recorded during clinical examination on the right side, and a small bump in the right malar zone was observed during opening. Intraoral examination showed an Angle class I occlusion. The patient sometimes complained of low back pain. During clinical examination, an overall body hyperlaxity was evident with a Beighton index of 9. There was no facial or trigeminal nerve involvement and no history of TMJ open locks or dislocations.

Panoramic x-ray, prescribed during a previous consultation one month earlier (Fig. 2), revealed a quite normal shape of the right coronoid process, and normal-appearing condyles located in the glenoid fossa. In order to better evaluate the morphology of the coronoid process, a coronal and axial computed tomography (CT) was requested. CT confirmed a large bony overgrowth arising from the

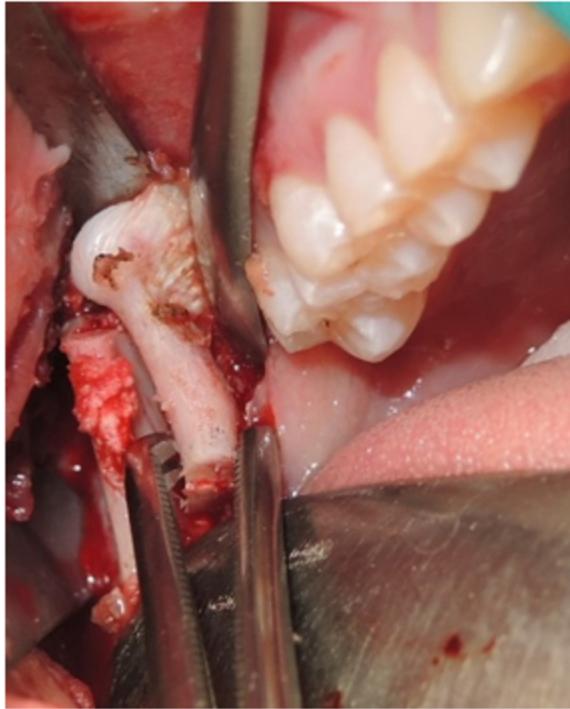


Fig. 5. Condylar accessory head at coronoid process removing.

right coronoid process. This atypical right coronoid process was projected laterally from the mandibular ramus with an angle of about 30° (Figs. 3 and 4). This part of the bone was morphologically similar to a condylar head, without any impingement with the zygomatic arch. In open mouth position, a hemifacial soft tissue displacement was observed, due to the right coronoid process atypical growth. This deformity was not observed in closed mouth position.

The patient and his parents were informed about the diagnosis and walked through the surgical treatment protocol, including the excision of the accessory condyle.

### 3. Surgical procedure

Based on clinical and radiographic images, a provisional diagnosis of Coronoid Hyperplasia was made. Treatment protocol included resection of the accessory bone with a piezoelectric device.

The patient was placed in a supine position under general anesthesia with naso-tracheal intubation. After infiltration of local anesthetic and vasoconstrictor in the upper right hemimandibular vestibular fornix region, a cold knife incision of the mucosa anterior to the ramus was performed. The periosteum was then elevated from the anterior border of the ramus to the outer surface of the middle part of the ramus, upward to the lingual and lateral surface of the coronoid process, 1 cm above the sigmoid notch.

Temporalis muscle attachment was partly preserved. A retractor was inserted from the outer side of the ramus and a malleable retractor was placed lingually to protect the soft tissues and the inferior neurovascular bundle when cutting the coronoid process.

Piezoelectric surgery was performed with an osteotomy from the sigmoid notch to the anterior border of the ramus, parallel to the inferior border of the mandible. The removed bony specimen appeared very similar to a condylar head and was covered by a white and smooth surface (Figs. 5 and 6). Extraoral pressure dressing was placed over the skin above the ramus to prevent severe swelling after surgery. Histology revealed a non-neoplastic fibrous perichondrium covering a hyaline cartilage cap in continuity with the periosteum of the underlying bone (Fig. 7a and b). A diagnosis of osteochondrogenous exostosis was made.

### 4. Post-surgical management

No significant differences in the patient aspect were highlighted before and after surgery. In the early postoperative period the patient's maximum mouth opening (MMO) was reduced. Home exercises were therefore prescribed after surgery [18,19].

After 1 month of exercises, MMO gradually improved from 27mm to 38mm. After 6 months, the active mouth opening was 51mm and the passive one 53 mm, without any difficulty in opening/closing pattern.

A control CT scan was performed four months after surgery and no osteo-muscular alterations were detected. Histology revealed an osteochondrogenous exostosis (Fig. 7a and b), in agreement with the clinical diagnosis of coronoid hyperplasia, like in Jacob's disease.



Fig. 6. Bony specimen.

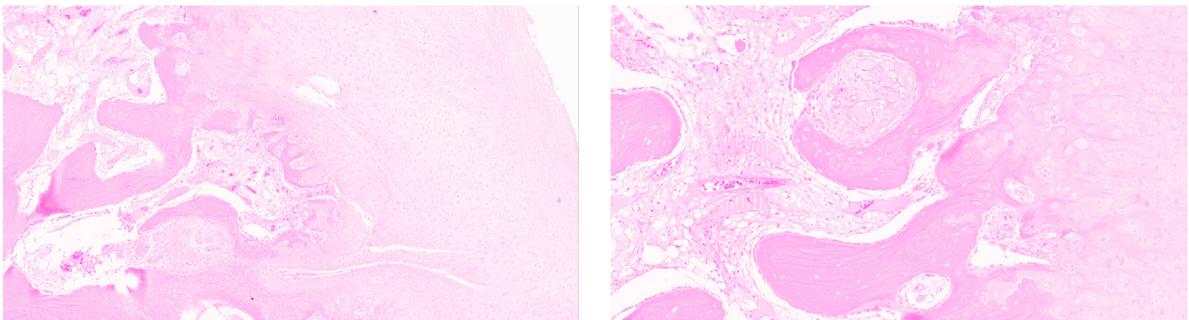


Fig. 7. a,b Histological appearance of the lesion (low and high power magnification): the perichondrium covers the cartilage cap that merges into the periosteum of the underlying bone (a: haematoxylin and eosin stain, original magnification x 40; b: haematoxylin and eosin stain, original magnification x80).

## 5. Discussion

Mandibular development abnormalities, like CPH and accessory condyle, are rare disorders of uncertain etiology that can occur in both unilateral and bilateral forms. Although CPH has been considered an uncommon condition, its actual incidence is expected to be higher than that reported in the past, taking into account the existence of many unreported and/or undiagnosed cases. Most common sign, in unilateral or bilateral cases, is a facial asymmetry with a deformation of the zygomatic arch, accompanied by a slow and progressive reduction in mouth opening with no reported pain [16,17].

In the case here reported the patient was concerned about progressive deviation during the mouth opening pattern, with a slight face deformity at the zygomatic area, and without ankylosis between the ramus and the arch, neither any reduction of mouth opening.

CPH can be easily diagnosed if the patient presents a limited mouth opening [14,16,20,21]. Indeed, the CPH diagnostic criteria according to the expanded taxonomy for temporomandibular disorders [15], include a progressive enlargement of the coronoid process that impedes mandibular opening due to the zygomatic process mechanical obstacle. The patient usually reports a history of jaw opening limitation. During clinical examination there should be a reduction of active and passive maximum jaw opening, imaging (panoramic x-rays or CT/CBCT) should show an elongated coronoid process which comes into close contact with the posterior aspect of the zygomatic process of the maxilla during opening.

The novelty of this case is that the patient did not present any reduction in mouth opening and the diagnosis was not made at an early stage. Thus in the patient previous panoramic x-ray, a quite normal shape of the right coronoid process was evident, although it was not clearly represented.

The very special shape of coronoid process, as shown in the CT images, was defined as accessory condyle [1], since it was covered with cartilage, as reported by the histopathological report. The cartilaginous component was a typically condylar characteristic, which does not normally belong to the coronoid process.

The histological report could be consistent with a Jacob's disease [13,22,9]. The coronoid shape, his relationship with zygomatic

arch and the absence of a joint with the arch, and the coronoid appearance to a mandibular condyle, like a duplication of a portion of the facial skeleton [23] induced us to name this clinical condition as an accessory condyle on coronoid process.

Hyperplastic coronoid process without impingement to the zygomatic bone could be followed through a slight elongation [14], but causal relationship of the morphological changes is uncertain. In this case it could be speculated that the previous trauma lead to the unexpected elongation and atypical shape of the coronoid process.

The aim of a coronoidectomy, which removes the coronoid process completely, is to relieve any interference between an elongated coronoid process and the zygoma, or an articulation between them, as well as to prevent the upper displaced coronoid process from interfering with the movement of the mandible by contacting the upper part of the ramus. However, coronoidectomy needs wider tissue reflection, which causes more tissue damage and creates more scarring after surgery, and which may cause trismus to occur again. It is usually difficult to perform coronoidectomy if articulation of the coronoid process and zygoma exists. Some surgeons approach this by extraoral approach to ensure complete coronoidectomy combined with myotomy, fasciotomy, and zygomatic osteotomy.

Coronoidotomy was conventionally used [24] for CPH treatment with good results, frequently creating a gap by upward displacement of the coronoid process after coronoidotomy, so as to prevent reunion of the two bone segments, and avoid probable relapse. The gap coronoidotomy can also be applied [25] when separation between coronoid process and the zygoma is difficult, or when complete removal of the prominent deformed coronoid process is very difficult.

In the present case the coronoid process was spontaneously elongated with a curve that allowed it to exceed the zygomatic arch, thus avoiding the impaction during mouth opening.

A piezoelectric surgery, was adopted, proceeding to a coronoidectomy, instead of the coronoidotomy, that was normally used in these cases, due to a suspect of ramus neoplasm [26–34].

## 6. Conclusion

An accessory condyle originating from the coronoid process is the atypical clinical condition of the presented case, in which the patient did not report any significant facial deformity or limitation in mouth opening.

This condition is quite different with respect to the majority of coronoid process hyperplasia cases reported in the literature. These are normally characterized by a marked opening mouth limitation and a clear lateral deviation of the mandible during its opening movement.

A coronoidectomy was performed to remove the pathological process with the histological diagnosis of osseocartilaginous exostosis and no recurrence has occurred. .

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