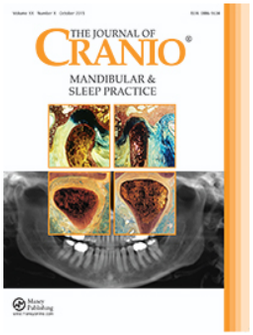

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CASE REPORT

Chronic mandibular hypomobility related to a unilateral accessory condyle. A case report

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ABSTRACT

Background: Chronic mandibular hypomobility is characterized by a long-standing limitation of the mouth opening related to multiple etiologies, including elongation of the coronoid apophysis. Unlike the most frequent pathologies that cause coronoid elongation, such as hyperplasia, osteoma (OM), and osteochondroma (OC), the accessory mandibular condyle (AMC) is a rare entity.
Clinical Presentation: The AMC shows a configuration similar to a normal mandibular condyle with an articular surface covered by fibrocartilage that articulates with the temporal wall of the zygomatic bone, and histologically, does not show neoplastic growth. The patient was treated with a coronoidectomy, removing the coronoid apophysis as well as the AMC.
Clinical Relevance: This case report presents a case of an AMC to describe the clinical, imaging, surgical, and histological characteristics, establishing its differential diagnosis with hyperplasia, OM, and OC of the coronoid apophysis.

ARTICLE HISTORY

KEYWORDS
Mandibular condyle; coronoid apophysis; accessory condyle

Introduction

Chronic mandibular hypomobility (CMH) is characterized by a generally long-standing limitation of the mouth opening and may be produced by multiple etiologies, such as mandibular trismus of muscular origin, arthropathies of the temporomandibular joint (TMJ), congenital malformations, and mechanical problems caused by the enlargement of the coronoid apophysis (CA) [1–3]. The latter can be unilateral or bilateral [4–6] and has been related to hyperplasia, hypertrophy, exostosis, osteoma (OM), osteochondroma (OC), deformation, elongation, and Jacob's disease [4–7].

In addition, the accessory mandibular condyle (AMC), described by Peacock et al. [8] in 2011, is considered to be an extremely rare entity that causes CMH. Its etiology is unknown, and it originates outside the limits of the mandibular fossa. It consists of a structure similar to a condyle, without evidence of neoplastic growth and articulates with the temporal surface of the zygomatic bone, allowing limited rotational movements [8].

In order to establish an adequate differential diagnosis among the diverse causes of CMH and its etiology, it is necessary to carry out a thorough anamnesis, including investigating previous traumas in the region, the presence of any systemic disease, capsulitis, disc adhesion or displacement, synovitis, retrodiscitis, infectious pathologies, or medical procedures in the TMJ, such as arthroscopies, infiltrations, or open surgery [9–11], among others. Moreover, imaging methods are essential in the case's analysis with limited painless mouth opening. In particular, computed tomography (CT) allows the visualization of deformity of the tissues, size of the lesion, shape, composition, location, and relationship with the adjacent structures [2,3,5–7]; the histopathological findings are necessary for a definitive diagnosis.

This article aimed to describe the clinical, imaging, surgical, and histological characteristics of an AMC associated to a CMH, as well as to establish its differential characteristics with the hyperplasia of the CA, OM, and OC.

Case report

A 52-year-old female presented with a long-standing impairment of mouth opening ability to a degree that it formed a hindrance for intake of solid food, for speech, and oral hygiene. The impairment had evolved progressively during the last three decades. The patient experienced moderate discomfort over her left zygomatic arch on mouth opening concomitant with a tingling sensation

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in the infra-orbital area, but beyond that, no pain. She had no recollection or knowledge of any trauma to the maxillofacial region. The patient also mentioned having been diagnosed with TMJ dysfunction and contracture of the masticatory muscles, initiating a treatment of forced mouth opening under local anesthesia, and multiple infiltrations (triamcinolone acetonide) in the left TMJ, without results. At maximum mouth opening, inter-incisal distance measured 12 mm (Figure 1a), and the mandibular midline deflected to the left. Laterotrusive mandibular movement was impaired to the right.

Apart from an asymmetric lower skeletal rim of the nasal aperture and a difference in size of the maxillary sinuses, the left sinus being smaller than the right, CT measurements revealed no asymmetry of the facial skeleton, nor was there any asymmetry seen in the face or lateral external photos (Figure 1b). An intraoral examination revealed a partial permanent dentition, gingivitis at multiple locations, and caries cavities in molars of all four quadrants.

The CT examination disclosed an elongation of the left CA combined with a volumetric increase forming an anterior mushroom-like structure (Figure 2a), which articulated against the dorsal face of the zygomatic bone (Figure 2b). The bone trabeculae were coarse and sparse with large inter-trabecular spaces. The density within different inter-trabecular spaces varied in attenuation with values of Hounsfield Units (HU) ranging from 633.7 to 208.9, which excluded a pervading content of bone marrow, since HU for bone marrow would be below 400. Hence, some inter-trabecular spaces comprised soft tissue that was denser than bone marrow. A volumetric CT reconstruction of the upper third of the left mandibular ramus depicted the increased CA and its resemblance with the mandibular condyle (Figure 2c,d).

Proton density-weighted magnetic resonance (MR) imaging in maximum intercuspation position was acquired in the sagittal plane to assess the position of the articular disc in both TMJs. According to Isberg´s [12] disc displacement classification, under normal conditions, when the mandible is at rest, the intermediate band of the articular disc is located between the anterior aspect of the condyle and the posterior aspect of the articular tubercle, so that the posterior band is more or less close to the 12 o'clock position relative to the mandibular fossa, and the anterior band is placed in front of the condyle. In the case presented here, the intermediate band of the articular disc was observed without contact with the anterior aspect, so that there was considered to be a partial anterior displacement in both TMJs (Figure 3a,b). T2-weighted MR images taken from the dynamic sequence showed an extensively wider insertion of the temporal muscle onto the left CA compared with the right side (Figure 3c,d)).

Surgical treatment was performed under general anesthesia. A circumvestibular approach was applied with caudal extension toward the mandibular ramus in order to execute a sub-periosteal dissection exposing the left maxillary zygomatic arch and the anterior rim of the mandibular ascending ramus. At this point, the mandible was manually guided to perform a laterotrusive mouth opening, which allowed detection of a protuberance covered with fibrous tissue and thus, mimicking a capsule (Figure 4a,b). An incision was made in the fibrous tissue followed by a blunt dissection. An apophysectomy was performed, including the elongated section of the CA. During the apophysectomy,

Figure 1. Clinical examination of the patient. A. Limited mouth opening of 12 mm. B. Frontal extraoral view where facial asymmetry is not evident.

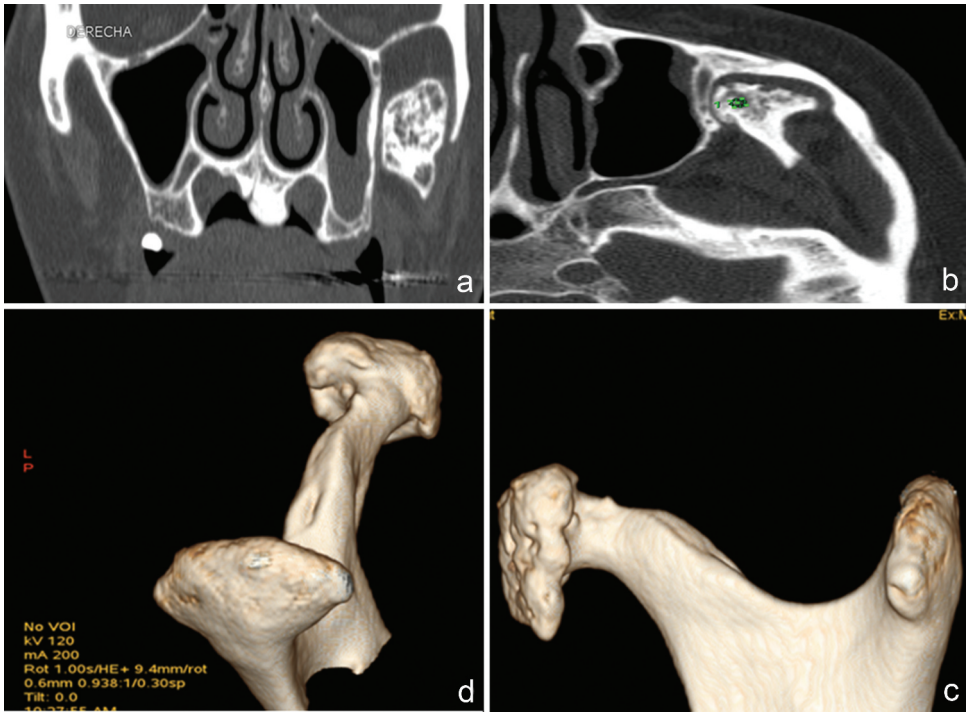

Figure 2. A. Computed tomography coronal view shows an elongation of the left coronoid apophysis combined with a volumetric increase forming an anterior mushroom-like structure and a decrease in the image corresponding to the ipsilateral maxillary sinus. B. Axial view shows the slight depression of the posterior area of the zygomatic bone related to the enlarged coronoid apophysis: C. Posteroanterior and lateral (D) views of the coronoid apophysis and left condyle that shows the similarity between the structures.

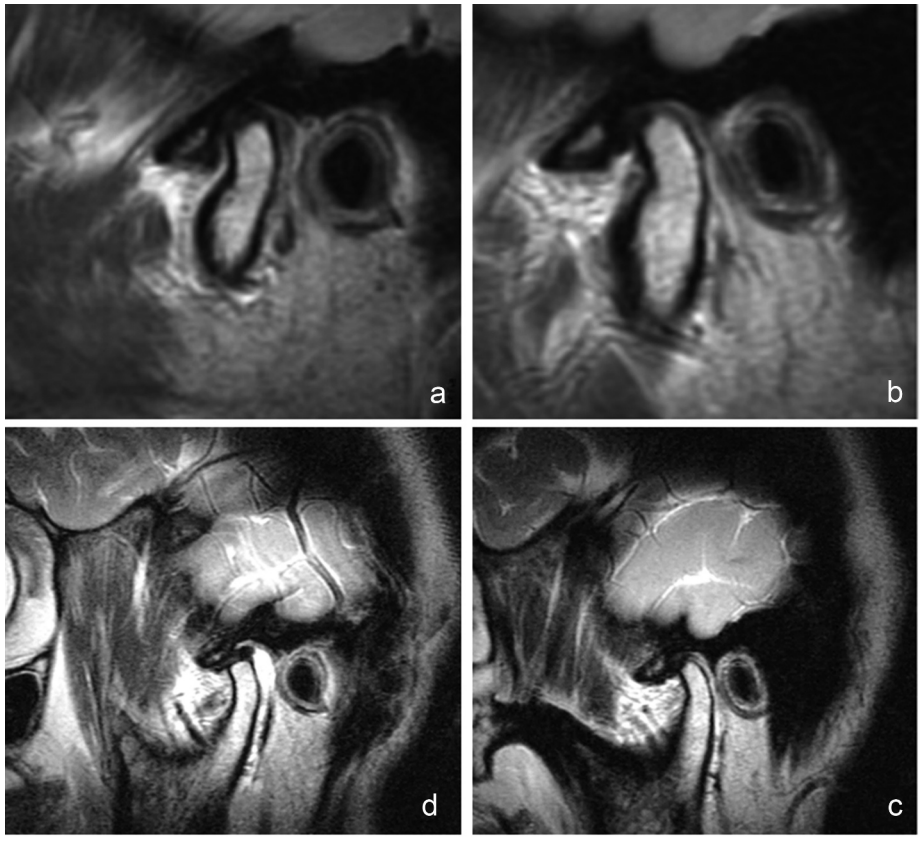

Figure 3. Sagittal view of proton density-weighted resonance imaging (closed mouth), showing partial displacement of the articular disc: (A) right side and (B) left side. T2-weighted images, taken from the dynamic sequence, show the insertion of the temporal muscle in the coronoid process on the right side (C) and the greater extension of the same on the left side (D).

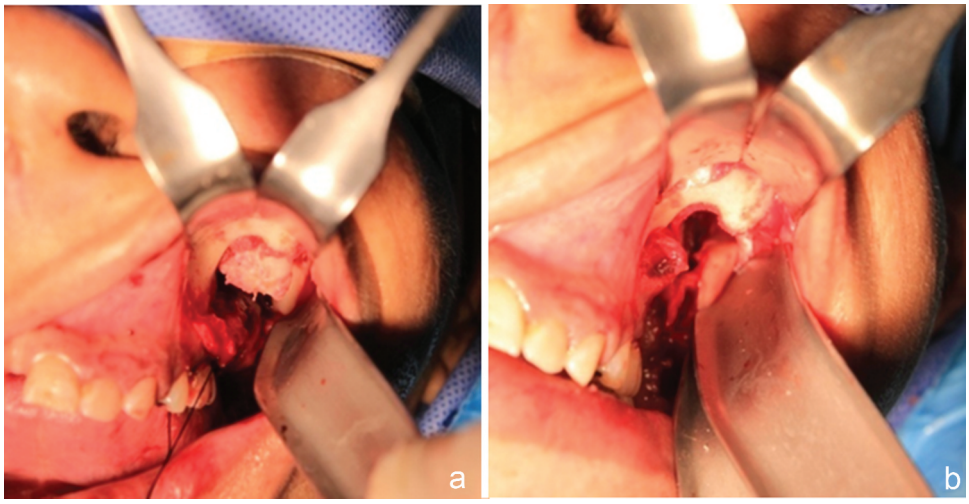

Figure 4. Intraoperative view shows the enlargement of the coronoid apophysis covered with fibrous tissue mimicking a capsule (A) and (B) the cavity where it was articulated.

a caudal traction was exerted, exposing the muscular insertions of the temporal and lateral pterygoid muscles. These were detached to allow extraction of the apophysis specimen followed by hemostasis of the arterioles coming from the muscles. Bilateral TMJ arthroscopy confirmed the absence of adherences potentially restricting condyle movements, the presence of a grade I synovitis, and grade II chondromalacia in the right TMJ; the left TMJ showed IV synovitis, without chondromalacia. The synovial membrane of both TMJs was infiltrated with 2 cc of triamcinolone acetonide. An immediate mouth opening of 32 mm was achieved.

Histopathological analysis of the decalcified specimen revealed a lesion formed by trabecular bone tissue; one end showed cartilaginous tissue surrounded by a layer of fibrous tissue. No neoplastic lesion was observed (Figure 5a,b). The diagnostic conclusion was the presence of histopathological characteristics compatible with AMC.

Postoperative edema, mouth opening in millimeters, surgical wound and rehabilitation protocol performed by the patient were recorded to evaluate the patient's progress. All data are summarized in Table 1. One year postoperatively, there were no CT signs of recurrent growth of the CA (Figure 6), and the maximal interincisal distance was 22 mm. The long-term result and follow-up of the radiolucent areas in the temporal bone adjacent to the former apophysis could not be obtained because the patient moved abroad one year post-surgically.

Discussion

To the authors' knowledge, there are no reported cases of AMC before 2011, when Peacock et al. [8] described it

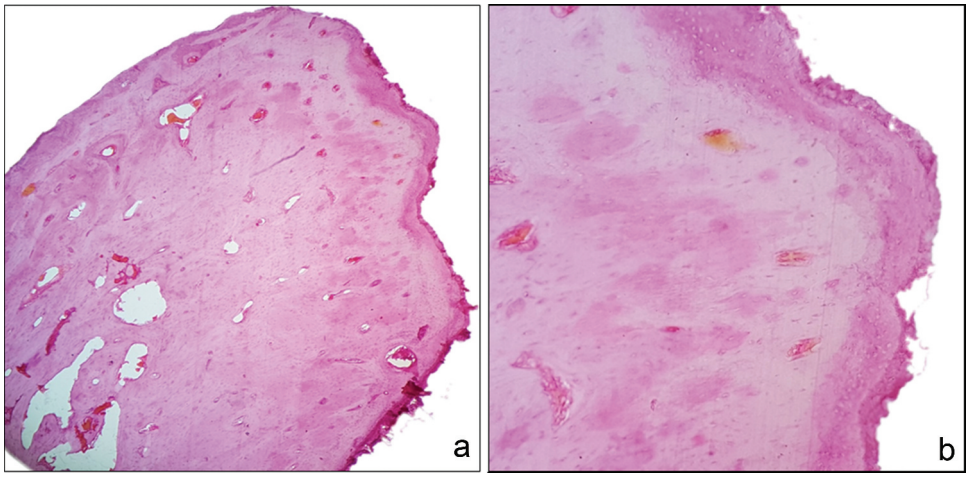

Figure 5. A. Decalcified sample using H&E staining 4X magnification, condylar normal layers are identified. B. H&E staining 10X of received sample; observe the absence of active neoplastic tissue.

Table 1. Description of the evolution of the patient in relation to post-operative edema, mouth opening, surgical wound, rehabilitation, and physiotherapy protocol.

Control time	Postoperative edema	Mouth opening (mm)	Evolution of the surgical wound	Rehabilitation and physiotherapy protocol
7 days	++++/+++++	13	Suture points in position, without dehiscence or exudates.	Start of protocol, mouth opening exercises, lymphatic drainage, infrared light, ultrasound and local heat.
15 days	+++/+++++	15	Suture points in position, in frank healing process without dehiscence or exudates.	Start of protocol, mouth opening exercises, lymphatic drainage, infrared light, ultrasound and local heat.
21 days	++/+++++	17	Surgical wound in frank healing process, some stitches present.	On alternate days a protocol with local heat and opening exercises was performed by a physiatrist.
30 days	+/+++++	18	Surgical wound in frank healing process, some stitches present.	On alternate days a protocol with local heat and opening exercises was performed by a physiatrist.
6 months	Not present	20	Eutrophic scar, slight local fibrosis.	On alternate days a protocol with local heat and opening exercises was performed by a physiatrist.
12 months	Not present	22	Pale pink mucosa, eutrophic scar without areas of fibrosis.	–

++++ severe edema; +++ moderate edema; ++ medium edema; + mild edema

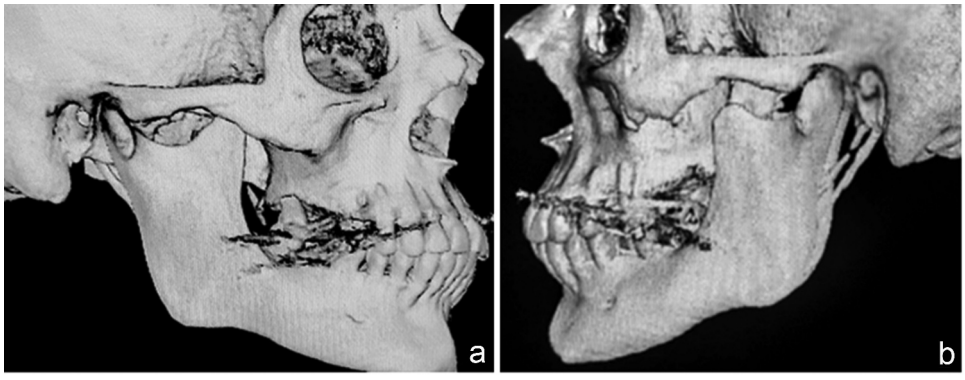

Figure 6. Computed tomography 3D volumetric reconstructions lateral views: A. Right side with normal coronoid apophysis and condyle; B. Left side showing no signs of recurrent growth of the apophysis.

for the first time in a 7-year-old male patient with a progressive and painless CMH, the current case being the second reported.

The etiology of an enlarged CA is controversial but seems to be associated with the type of abnormality. The following aspects have been reported: a relationship to trauma, failure in the cartilaginous center of jaw growth, some disturbance in the growth mechanism of CA, increased activity of the temporal muscle, hyperplasia of a neoplastic nature, and the presence of a lesion type OC [10,11,13–15]. In the reactive enlargement of CA, it is thought that temporal muscle hyperactivity plays an important role. The muscle is released by the arthrokinetic reflex of the displacement of the TMJ disc, which probably impresses a constant tension in the CA. The continuous "pull" of the muscle probably decreases the local blood circulation at the tendon insertion of the CA, favoring degenerative changes with calcium deposits of the tendon and thus, creating a reactive bone hyperplasia [1].

The initial symptoms of CA enlargement are a sensation of tightness in the TMJ region accompanied by a progressive reduction in the mouth opening without malocclusion. In late stages, the patient shows trismus, and the facial asymmetry is regularly associated with unilateral cases [2,5,6,15,16]. This condition can cause respiratory distress, delayed mandibular growth, muscular atrophy, as well as difficulty eating and performing adequate oral hygiene [3]. In the current case, the patient manifested a chronic and progressive limitation of the mouth opening that adversely affected eating solid food, speech, and oral hygiene for the last 30 years.

CT has become the gold standard in presurgical assessment and surgical planning, as 3D images are particularly useful in the evaluation of CA length and changes in the internal surface of the zygomatic arch [6,7,14,15]. In this patient's CT, an image of heterogeneous density was observed with evidence of erosion in its cortex and convexity in a lateral-medial direction. It was localized in the cephalic end of the left CA, which articulated with the posterior surface of the zygomatic bone.

The presumptive clinical diagnosis of the case was hyperplasia of the CA, while the imaging characteristics

were suggestive of OC. However, histological examination revealed trabecular bone tissue with no neoplastic features, surrounded by fibrocartilage, which made it possible to rule out OC or OM. Concerning CA hyperplasia, morphology on this process is usually preserved and, in the histopathological study, reports a bone tissue of regular appearance without the presence of fibrocartilage. On the other hand, Jacob [17] in 1899, described the findings in a 62-year-old corpse: limitation of mouth opening of 2 cm, produced by a hyperostosis of compact tissue originating from the temporal aspect of the left zygomatic bone and which considerably occupied the space of the zygomatic fossa, preventing the normal excursion of the CA of normal appearance and dimensions. Considering the original article, the name of Jacob's disease should be reserved for that condition in which a normal CA articulates with a zygomatic bone of abnormal contours. For this reason, the presented case is not thought to be Jacob's disease.

The treatment of AMC is the same for any of the different forms of AP alterations that may cause CMH, the primary objective being the restoration of the range of the mouth opening through surgical excision, which can be addressed under two approaches: a coronoidotomy, with or without a gap, and a coronoidectomy. In the first, there is less stress and trauma to the adjacent tissues, thus decreasing the incidence of fibrosis in the treated area; the latter alternative is most commonly used to avoid a possible recurrence due to the meeting of the segments [5,8].

Regarding surgical access, two approaches have been reported: a direct or indirect extraoral approach, an intraoral approach, or a combination of both [5,6,8,15]. In the extraoral approach, Al-Kayat-Bramley, bicoronal, hemicoronal, submandibular, or transzygomatic incisions have been used [7]. The intraoral approach provides similar results without cutaneous scars and decreases the risk of injury to the facial nerve. The choice of the surgical approach will depend on the lesion`s size as well as its conformation; a "bulbous" mass hinders its removal from the space between the temporal bone and the zygomatic arch [5,14]. The surgical approach used in this case was intraoral, which allowed the adequate resection of the AMC. The arthroscopy showed synovitis of different degrees in both joints and chondromalacia in the right TMJ, both findings commonly associated with the presence of osteoarthrosis [13].

Although postoperative CT imaging showed a satisfactory result in relation to CA, the mouth opening of 32 mm immediately after surgery was not maintained and at one-year follow-up was only 22 mm. This can be attributed to several factors, among them, the bilateral disc displacement, the initial multiple steroid injections into the left TMJ that could have caused fibrous or muscular changes, and a possible inconsistency in physiotherapy, which is of vital importance for the maintenance of the obtained results and the limitation of fibrosis at the surgical site [5,15]. It is unknown whether the patient received treatment for joint dysfunction, which could also be another factor to consider.

Conclusion

In conclusion, histological characterization is essential to differentiate an AMC from other entities, such as OM, OC, and CA hyperplasia. The identification of fibrocartilage, typical of physiological areas of the bone structures that make up the TMJ, specifically the mandibular condyle, supports the definitive diagnosis. The intraoral approach used in this case provided an adequate resection of the AMC without cutaneous scars or the risk of injury to the facial nerve.

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Disclosure statement

No potential conflict of interest was reported by the authors.

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