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Bilateral coronoid hyperplasia causing painless limitation of mandibular movement

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ABSTRACT

The coronoid process is a beaklike process in the ramus of the mandible. Coronoid process hyperplasia (CPH) is a rare possible cause of reduced mouth opening. An overgrown process interferes with mandibular rotation and lateral excursion and hence leads to restricted mouth opening (RMO). Although some factors are suggested, etiology of CPH is not completely known. Prescription of suitable radiography is necessary for an accurate diagnosis. This article reports a 30-year-old man with bilateral CPH and progressive RMO since childhood. This disorder affected his oral hygiene and quality of life. With the help of different types of radiography, CPH was diagnosed and coronoidectomy was the only treatment option. The patient showed normal jaw movements after the surgery and postoperative physiotherapy. General dentists have an important role in noticing RMO and referring the patients to maxillofacial radiologists. Although it is a rare phenomenon, general dentists need to keep CPH in mind as a possible cause. Panoramic imaging accompanied by computed tomography or cone beam computed tomography is the best imaging option in such cases.

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Introduction

Opening the mouth is a result of coordinated function of muscles and bones especially in the temporomandibular joint (TMJ). Therefore, anything that interferes with the TMJ's correct function can cause restricted mouth opening (RMO) and even complete lock. One of the possible causes of progressive RMO

is hyperplasia of the coronoid process, known as coronoid process hyperplasia (CPH). In Greek, korone means like a crown. The coronoid process is a beaklike process in the superioanterior part of the ramus of the mandible [1].

Bilateral CPH is a rare developmental condition characterized by abnormal overgrowth of the histopathologically normal coronoid processes. Movements of a larger coronoid process interfere with the medial or temporal surface of the zygomatic

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Fig. 1 – The patient's photographs illustrate the maximum mouth opening and lateral excursion.

bone. In addition, as this process grows gradually, the infratemporal space needed for rotation and translation of mandible is reduced, which results in reduction of the ranges of mouth opening and lateral excursion [2,3].

The etiology of CPH is not completely known. However, several factors have been suggested as possible etiology, such as temporalis muscle hyperactivity, trauma, hormonal factors, genetics, and familial factors [4,5].

The purpose of this article is reporting clinical and radiographic characteristics of CPH and imaging modalities that may be helpful in accurate diagnosis of this disorder.

Case report

A 30-year-old man who visited his dentist, complaining about pain in the left mandibular premolar, was referred to the Department of Radiology, School of Dentistry of Isfahan because of insufficient mouth opening and difficult dental operation.

Restriction in mouth opening was found to be present since the patient's childhood; which had progressed gradually in years. There was no record of childhood disease or trauma, or familial history of trismus. The patient also complained about episodes of migraine headache.

In clinical examination, interincisal space was 21 mm. In addition, lateral excursions to the left and right were possible, but they were very limited (Fig. 1).

The normal range for interincisal space in maximum mouth opening is 35-50 mm and normal lateral excursion is 8-12 mm toward mandibular incisors [6].

Click and crepitation in the right TMJ were observed and palpated. Muscles of the area were not tender.

Based on clinical examinations, primary diagnosis of the TMJ dysfunction was indicated and radiographic examinations were prescribed.

In the panoramic image, the coronoid processes were larger in length in both sides and were observed to be higher than the zygomatic arch, although the bone trabeculae were normal in the processes. Most of the posterior teeth were extracted, root canalled, or affected with remarkable dental caries, whereas all the anterior teeth were sound and healthy, which was probably because of insufficient oral hygiene due to the reduction in mouth opening and lack of access to the posterior areas when brushing (Fig. 2).

In the next step, computed tomography (CT) scan was obtained and the coronoid processes were surveyed in different views including 3-dimensional (3D), bone window, and soft tissue window.

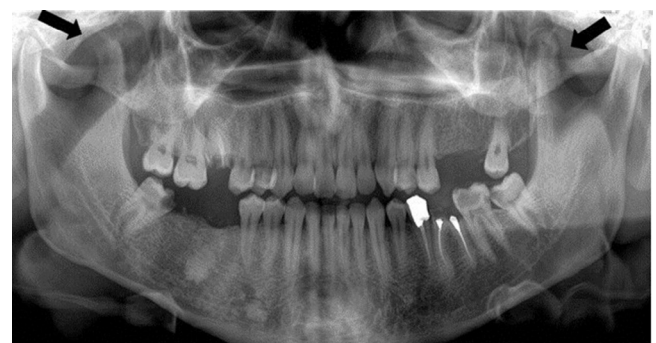


Fig. 2 – Panoramic radiography: the arrow indicates bilateral coronoid process hyperplasia.

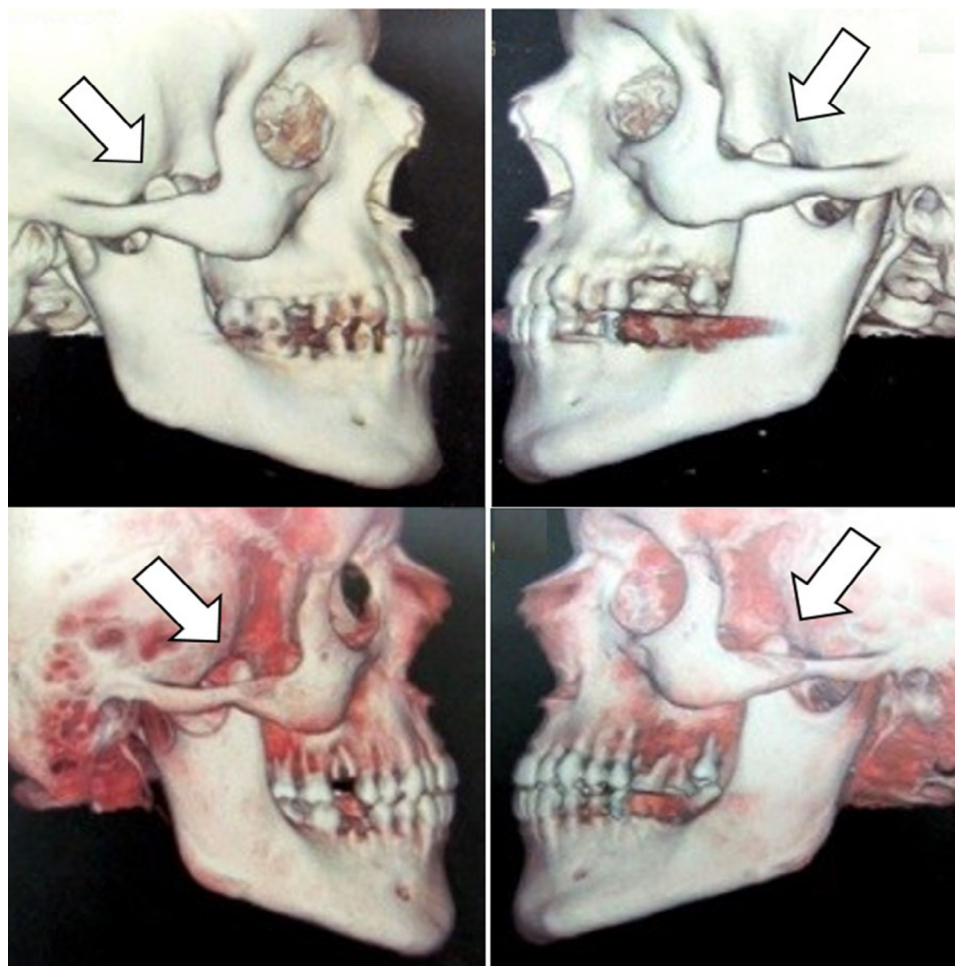


Fig. 3 – A 3D reconstructed CT scan shows the relation of hyperplastic coronoid process and the zygomatic bone. CT, computed tomography.

In 3D view, apices of the coronoid processes were significantly higher than the zygomatic arch and the condylar head (Fig. 3).

In axial images that were obtained using bone window, bone changes and the contact of medial parts of the zygomatic arch and the coronoid processes at bone sides were studied. In soft tissue window images, relation of the bones and the muscles were investigated (Fig. 4).

Based on the radiographic finding, CPH was diagnosed. The patient was referred to a maxillofacial surgeon for coronoidectomy. Bilateral coronoidectomy was carried out intraorally. After the operation, the patient underwent physiotherapy for 3 months to rehabilitate normal jaw movements.

Discussion

The purpose of this article is to report clinical and radiographic characteristics of CPH and imaging modalities that may be helpful in accurate diagnosis of this disorder.

The coronoid process is a triangular bone, which projects upward and slightly forward from the ramus of the mandible. Its posterior border is bounded by the mandibular incisure,

and its anterior border aligns with the ramus [6]. Normal development of the coronoid process includes intramembranous ossification of the accessory nucleus of the cartilage during the fourth week of intrauterine life. This cartilage is covered by a thick fibrocellular capsule that is not evident before birth. Skeletal growth continues until adulthood along with transformation and proliferation of the fibrocellular layer's cells [3].

With evolution of the mammals and a shift of diet to more vegetarianism and changes of the bite forces, the coronoid process has gradually grown larger and has turned to its present form [7]. Intraoral removal of this process causes no functional defects or facial malformations. Thus, this bone can be used for reconstruction of orbital floor defects, alveolar defects, nonunion mandibular fractures, sinus augmentations, reconstruction of bone abnormalities, and other craniomaxillofacial surgical procedures [1].

This abnormality is manifested as malocclusion and reduction in mouth opening. Patients usually complain about limitation in mouth opening. It might be mistaken for more prevalent causes for RMO such as TMJ internal derangement, masticatory muscle contraction disorders, and ankylosis [4]. CPH and chronic disk displacement without reduction have similar signs and symptoms, except that pain is not a common

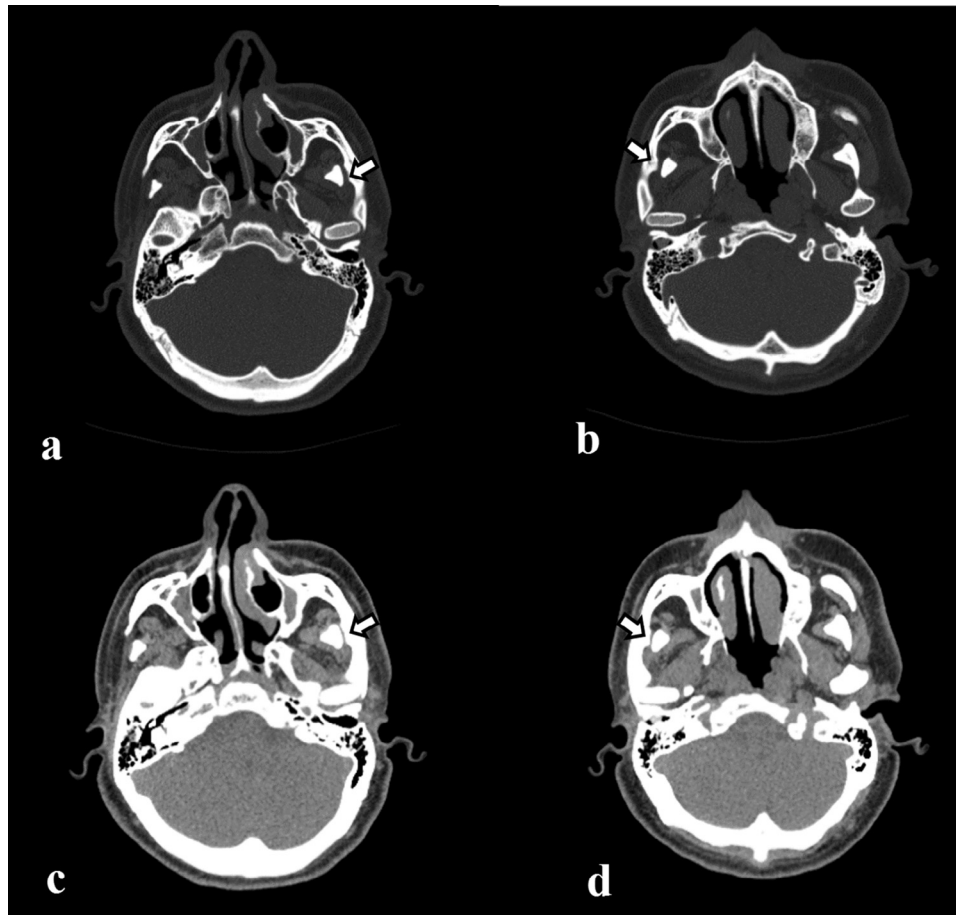


Fig. 4 – Axial CT in 2 views: soft tissue window (A and B) and bone window (C and D). The arrows indicate the relation between the coronoid process and the zygomatic bone. CT, computed tomography.

symptom in CPH. Thus, correct diagnosis of CPH does not usually happen in the first appointment, and this disorder is mostly misdiagnosed. The first case of mandibular hypomobility due to CPH was reported in the second half of the nineteenth century and few cases have been reported since then. Disorders involving the coronoid processes have been diagnosed in only 5% of mandible hypomobility cases, in both men and women, at a ratio of 5:1, and bilateral CPH is more prevalent in men [8–11].

In spite of low prevalence, this disorder needs to be considered as a possible diagnosis in patients with painless, progressive, and chronic RMO. Correct diagnosis of this condition has an important effect on the treatment plans and patients' quality of life. As a result, prescription of suitable images is necessary and mandatory for an accurate diagnosis. Coronoidectomy with physiotherapy following the surgery is the only treatment option for this disorder.

The etiology of CPH is almost unknown, but several causes have been proposed to explain the abnormal growth of this bone and the formation of CPH. However, there is not enough evidence to support any of proposed causes [12].

The most probable proposed cause for CPH is hyperactivity of temporalis muscle [7]. Temporalis muscle originates from temporal fossa upper than inferior temporal line and deep surface of temporalis fascia. Muscle fibers then converge

downward and are connected to the coronoid process and the upper part of the zygomatic arch. This muscle elevates the mandible and closes the mouth [6].

Studies carried out on postnatal surgeries on laboratory animals showed that changes in the muscular connection result in changes in size and form of the coronoid process. In addition, deficits in myogenic factors (MYF5/MYOD) cause coronoid and angular processes to be small or maintain complete defects [7]. Genetics and hormonal factors can also affect craniofacial size and morphologic characteristics [13,14]. Makek and Obwegeser indicated that different individual factors related to growth, which control generalized hypertrophy and longitudinal growth, may cause deformities [15]. Growth hormone (GH) is a peptide that is secreted from the anterior part of hypophysis and has an important role in growth and development of maxillofacial complex. GH bonds to GH receptors (GHR_s) which exist on the surface of cells and activate signals inside cells. Variations of GHR gene and mutations may cause disorders in craniofacial growth [13]. Some studies show that varieties in size and form of the coronoid process are related to expression of Paired box gene 9 (pax9) and SRY-box gene 9 (sox9). Sox9 is a transcription factor which is normally related to skeletal chondrogenesis. In the coronoid processes, its expression depends on the temporalis muscles' tension [7].

Diagnosis of CPH is mostly through radiography, scintigraphy of bone, and histopathologic studies. Thus, clinical examinations alone cannot diagnose and differentiate this disease from other disorders related to mouth opening. Progressive and painless mouth opening reduction through several years, especially in men, is a common clinical sign of CPH. Progressive RMO is also observed in syndromes of pain and dysfunction of the TMJ (especially chronic disk displacement without reduction) and also uncorrected fracture of the jaw or the zygomatic bone, radiotherapy-induced fibrotic changes of the masticatory muscles, rheumatoid arthritis, primary and secondary neoplastic diseases (including osteoma and osteochondroma), progressive fibrotic change of the oral mucous membrane, ankylosing spondylitis, myositis ossificans, and tetanus [3,16].

Among these causes, CPH is mostly mistaken for chronic disk displacement without reduction, which is the most common cause of mouth opening reduction. Chronic disk displacement without reduction is recognized with its typical clinical signs such as history of click in the TMJ which is vanished suddenly, pain, an experience of previous locked jaw, and unilateral deviation during opening of the mouth. However, there are no prevalent signs for CPH [4]. Thus, reaching a correct diagnosis is a very important and critical issue.

Patients with CPH usually receive treatments before the diagnosis. Because pain is not a common symptom in this disorder, patients with CPH do not usually notice its progress, and they do not pursue its medical treatment until there is too much restriction in mouth opening (such as what was observed in this case). Hence, the quantity of cases may be more than the reported ones.

Severe RMO causes considerable problems such as problems in oral health care. Thus, due to reduced oral health, caries rate is elevated in these patients, as was observed in this case. On the other hand, because of insufficient access to the posterior teeth, dental operations for such cases may be very difficult and even impossible for the dentist. In addition, surgical and prosthetic problems and even problems in intubation for general anesthesia are present in these patients; therefore, direct fiber-optic bronchoscopic nasal intubation is advocated [17,18].

Radiographic examination and observation of bony relations have an important role in correct diagnosis of this abnormality. Plain radiography is the first step in radiographic examination of patients with CPH. Panoramic radiography needs to be prescribed routinely for the cases in which clinical signs of jaw dysfunction are observed. Then, the TMJ areas should be surveyed for finding the cause of the restriction. The next step is prescription of CT, cone beam computed tomography (CBCT), or magnetic resonance imaging (MRI). The relation between the coronoid process and zygoma and every change in their bony surface is reviewable in CT and CBCT especially in 3D view. Not only are these techniques suitable for precise diagnosis, but they are also adequate for surgical protocols. Although MRI may help differentiate CPH from chronic disk displacement without reduction, this imaging is costly and not always accessible. On the other hand, with observing the coronoid process in CT and CBCT, MRI is no longer necessary.

Surgery improves the quality of life of patients with CPH. The purpose of surgery is to remove the coronoid process and

eliminate the mechanical obstacle of mouth opening. Access area for the surgery may be intraoral or extraoral [3]. Removing a severely hypertrophic coronoid process is not possible intraorally. The access will be gained through submandibular, (bi) temporal, preauricular, or coronal flap [19]. In addition, endoscopic methods have been introduced in the recent years as methods leaving little scars [20,21]. Because early comprehensive postoperative physiotherapy has an important role in correct mouth opening and preventing relapse of the treatment, it is advised after the surgery. Active and passive stretching exercises with or without the use of bite blocks, dynamic devices, wedges, mouth screws, spatulas, or TheraBites were reported to be helpful [22].

Relapse of the treatment is reported in some cases [19]. Possible causes of relapse may be persistence of underlying causes of hypertrophy, hematoma or postoperative fibrosis, inadequate physiotherapy program, or a poor diet. Relapse may lead to a second coronoidectomy; hence, regular follow-up and routine panoramic radiography especially with open mouth are advised for a better observation of margins of the restriction, the degree of mouth opening, and the condylar excursion. For postoperative follow-up in young patients and in cases with abnormal mouth opening range after surgery or a possibility of treatment relapse, MRI is a suitable option because ionizing rays are not used in this method and also hematoma, fibrosis, or muscular atrophy in the surgical site are well detectable [23].

Conclusion

Many etiologies may cause reduction in mouth opening, but the important issue is to diagnose and to eliminate the causes at the right time. As mentioned previously, this problem may lead to deterioration of quality of life and missing several teeth of a young patient.

General dentists have an important role. Despite CPH being a rare condition, it has to be considered as one of the etiologies of progressive mouth opening reduction. The patients had better be referred to maxillofacial radiologists who can perform further examination and confirm the final diagnosis. Panoramic imaging accompanied by CT or CBCT is the most suitable option in these patients because it helps the surgeon to plan the surgical protocol and to find the right diagnosis.

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