

Treatment of hyperplasia of the coronoid process of the mandible in adults: analysis of 42 literature reports and illustrative case

Tratamento da hiperplasia do processo coronoide de mandibular em adultos: análise de 42 relatos da literatura e caso ilustrativo

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ABSTRACT

Objective: The objective of this research was to review the literature, compare different methods of surgical treatment for coronoid hyperplasia and report a clinical case of unilateral coronoid hyperplasia treated by coronoidectomy with intraoral access. **Methods:** A critical review of the literature was performed by selection of papers published in the last 20 years on the treatment of coronoid in adults in the PubMed, Medline, Scielo and Lilacs databases, with the terms coronoid hyperplasia OR coronoid elongation AND treatment OR management. The data was extracted for analysis. **Results:** twenty-four articles were selected. It included 42 patients, among them 69% were male patients and 81% were bilaterally affected. The age group with the highest prevalence was the people in the 20's and 30's. Sixty-nine percent of the patients were treated with coronoidectomy and 26.2% with coronoidotomy. The majority (83.3%) with intraoral access. No cases had surgical complications reported, and 71.4% underwent physiotherapy after surgery. Regarding the results, 83.3% were considered satisfactory, 11.9% were unsatisfactory. **Conclusion:** The great majority of the cases that brought significant results in the improvement of the pre and postoperative mouth opening were treated by coronoidectomy, proving to be an efficient and safe surgical treatment for the coronoid hyperplasia.

Keywords: Adults. Hyperplasia. Mandible.

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RESUMO

Objetivo: O objetivo deste estudo foi revisar a literatura e comparar diferentes métodos de tratamento cirúrgico para hiperplasia do coronoide, além de relatar um caso clínico de hiperplasia coronoide unilateral tratada por coronoidectomia pelo acesso intraoral.

Métodos: A revisão eletrônica crítica da literatura foi feita selecionando artigos publicados nos últimos 20 anos sobre o tratamento da hiperplasia do coronoide em adultos nas bases de dados PubMed, Medline, Scielo e Lilacs com os termos coronoid hyperplasia OR coronoid elongation AND treatment OR management. Os dados foram extraídos para análise. **Resultados:** Foram selecionados 24 artigos. Incluindo um total de 42 pacientes, dentre eles 69% eram pacientes do sexo masculino e 81% foram acometidos bilateralmente. O grupo etário com a maior prevalência compreendeu de 20 a 30 anos. Sessenta e nove por cento dos pacientes foram tratados com coronoidectomia e 26,2% com coronoidotomia, a maioria (83,3%) com acesso intra-oral. Nenhum caso teve complicações cirúrgicas relatadas e 71,4% foram submetidas à fisioterapia após a cirurgia. Em relação aos resultados, 83,3% foram considerados satisfatórios, e 11,9% insatisfatórios. **Conclusão:** A grande maioria dos casos que trouxeram resultados significativos na melhora da abertura bucal pré e pós-operatória foram tratados pela coronoidectomia, demonstrando ser um tratamento cirúrgico eficiente e seguro para a hiperplasia do coronoide.

Termos de indexação: Adulto. Hiperplasia. Mandíbula.

INTRODUCTION

Coronary process hyperplasia (CPH) of the mandible is a rare developmental disorder characterized by abnormal growth of the mandible coronoid, such that the process collides with the medial surface of the zygomatic arch causing progressive limitation of the buccal opening [1,2]. It has unknown etiology, but several factors have been suggested as possible causes, among them, temporal muscle hyperactivity, trauma, hormonal factors, genetics and family factors [3].

CPH may be unilateral, which is more frequent in females, or bilateral. The latter form is the most frequent, affecting mainly men between 10 and 20 years. It is manifested by malocclusion and reduction of the oral opening. Trauma, ankylosis, masticatory contraction disorders, and dislocation of the articular disc without reduction should be included as differential diagnosis. However, in CPH, pain is not a frequent feature and imaging tests are essential in its definitive diagnosis [1,4].

The treatment for CPH must be surgical through intraoral or extraoral access, when complete bone removal of the coronoid process (coronoidectomy), or partial removal (coronoidotomy) [5]. should be performed.

Thus, the objective of the present study is to compare and discuss CPH of the mandible through a critical review of the literature, as well as to report a clinical case, seeking a better understanding of the characteristics and methods of surgical treatment of this developmental disorder.

METHODS

Study Design

This study was carried out in 2 stages. In the first stage a critical review of the literature on CPH of the mandible in adults and their treatment options was performed. In the second stage, the authors present a case of hyperplasia process.

Stage 1 – Critical review of the literature

Search strategy

Scientific papers regarding treatment of CPH were searched in the PubMed, Medline, Scielo and Lilacs databases published until May 2018. The following terms were used in the strategic search: (Coronoid hyperplasia OR coronoid elongation) AND (treatment OR management). The bibliographic reference of the papers included was also analyzed in an attempt to find suitable studies to the inclusion criteria, which were not found by the electronic search strategy.

Papers selection

Scientific papers on case reports, case series, clinical trials and retrospective and prospective studies on treatment of mandibular CPH in adult patients (above 18 years of age) published in English, Portuguese or Spanish in the last 20 years were included.

The defined exclusion criteria were: papers that were not available in their full form, literature review papers, papers with insufficient data, papers in which CPH was related to syndromes.

The papers were evaluated in two phases: first by reading their titles and abstracts, followed by reading the paper in full if, selected after the first stage of the process.

Data extraction and analysis

Data was analyzed, such as year of publication, age, gender, duration of symptoms, maximum oral opening (pre, trans and postoperative), diagnosis, affected side, treatment used, surgical access, physiotherapy done or other type of complementary treatment, postoperative period, complications and outcome.

The results were analyzed, as well as compared to the described and discussed case.

Stage 2 – Case report

The authors describe a case of a female patient, 28 years old, complaining about limitation of oral opening with 8 years of evolution, without painful symptomatology. The left unilateral CPH hypothesis was observed after tomographic examination, and the treatment was performed surgically by unilateral coronoidectomy by intraoral access, followed by complementary physiotherapy.

RESULTS

The research conducted in the databases resulted in 110 papers, of which 30 were in accordance with the inclusion criteria after the first reading step and were analyzed. Among those, 15 were excluded after reading. In addition, another 9 papers found in the bibliographical references of the read papers were included in the sample because they were adequate to the established criteria. At the end of the research, a total of 24 scientific papers on the treatment of CPH were obtained (figure 1). Among those, 21 papers of the clinical case report type, 2 prospective studies and 1 retrospective study. The data was then extracted for study (table 1).

The sample consisted of forty-two patients. Twenty-nine males (69%) and thirteen females (31%). Thirty-four cases had bilateral CPH (81%), of which twelve were female (35.3%) and twenty-two male (64.7%). Seven cases were unilateral CPH (16.6%), six males (87.7%) and one female (14.3%). One patient (2.4%) did not have the type of CPH identified in the study.

The average age of the patients was 19.1 years, ranging from 18 to 53 years. The evolution time had an average of 6.04 years, ranging from 2 to 28 years. Seventeen (40.5%) did not report this data.

The maximum preoperative buccal opening showed an average of 14.2mm, ranging from 9.5 to 30mm. The maximum transoperative buccal opening showed an average of 23mm, ranging from 26 to 50mm. Eighteen cases (42.9%) did not report this data. The maximum postoperative buccal opening showed an average of 24.9mm, ranging from 22 to 65mm. Four cases (9.5%) did not report this data.

In thirty-five patients the access was made intraorally (83.3%), three extraoral pre-auricular accesses (7.1%), two had the combination of intraoral and extraoral pre-auricular (4.8%), and two cases (4.8%) did not have the type of access identified in the study. Twenty-nine patients were submitted to coronoidectomy (69%), eleven coronoidotomy (26.2%), one patient had both surgical techniques (2.4%), and one had no technique identified in the study (2.4%).

The post-surgery follow-up period ranged from 1 to 30 months. Among forty-two, sixteen reported no surgical complications (38.1%) and for the other 26 patients (61.9%), this information was not obtained. Thirty patients underwent physiotherapy after surgery (71.4%), and for 12 patients this data was not obtained (28.6%). Thirty-five patients had satisfactory results (83.3%), five were unsatisfactory (11.9%) and two could not be evaluated (4.8%).

A 28-year-old female patient went to the maxillofacial surgery clinic complaining about limited oral opening with approximately 8 years of evolution. During the anamnesis she did not have painful symptomatology and reported a history of facial trauma 2 years before the clinical signs. The physical examination showed that the patient presented limited oral opening with an interincisal distance of 10mm, with a slight deviation of the mandibular

Table 1. Data of cases reported in the literature presenting coronoid process hyperplasia in adults.

Authors	Age		History (yrs)	Type	Sex	Preoperative		During		Postoperative		Approach	Type surgical	Follow-up (months)	Complications	Physiotherapy	Results
	(yrs)	(yrs)				MMO (mm)	MMO (mm)	MMO (mm)	MMO (mm)								
Yamaguchi et al. [17]	25	8	Uni	M	24	NS	43	IO	ectomy	8	NS	Yes	Satisfactory				
Gibbons, et al. [2]	28	12	Bi	M	15	30	40	IO + EO (preauricular)	ectomy	NS	NS	Yes	Satisfactory				
Colquhoun et al. [13]	32	3	Uni	M	21	NS	NS	IO	ectomy	18	NS	Yes	Satisfactory				
Agurto et al. [6]	26	3	Bi	M	22	35	22	IO	ectomy	30	NS	Yes	Unsatisfactory				
Bertacci et al. [31]	36	NS	Bi	M	24	NS	41	IO	ectomy	NS	NS	Yes	Satisfactory				
Gibbons et al. [14]	25	10	Bi	M	27	NS	42	IO	ectomy	6	NS	Yes	Satisfactory				
Kursoğlu et al. [10]	34	18	Bi	M	20	30	20	IO	ectomy	1	NS	Yes	Unsatisfactory				
Leovic et al. [29]	24	NS	NS	M	27	NS	NS	NS	NS	NS	NS	NS	NS				
Ferro et al. [8]	35	18	Bi	M	15	NS	35	NS	tomy	NS	NS	Yes	Satisfactory				
Yoshida et al. [28]	28	NS	Bi	M	13	NS	40	IO	ectomy	12	NS	Yes	Satisfactory				
Yura et al. [5]	34	NS	Bi	F	18	25	38	IO	tomy	6	None	NS	Satisfactory				
Ferreira et al. [23]	28	13	Uni	M	30	50	43	IO	tomy	3	NS	Yes	Satisfactory				
Bayar et al. [24]	26	3	Bi	M	18.51	29	31.12	IO	ectomy	1	None	Yes	Unsatisfactory				
Costa et al. [27]	21	2	Uni	M	23	NS	38	IO + EO (preauricular)	ectomy	NS	None	Yes	Satisfactory				
Min Kim et al. [18]	18	NS	Bi	M	20	NS	40	IO	ectomy	5	None	Yes	Satisfactory				
Behrends et al. [11]	21	NS	Uni	M	28	43	63	IO	ectomy	15	NS	Yes	Satisfactory				
Jenkins et al. [4]	19	4	Bi	M	32	35	65	IO	ectomy	18	NS	Yes	Satisfactory				
Surianti et al. [12]	18	6	Uni	M	12	NS	54	IO	ectomy	15	NS	Yes	Satisfactory				
Murakami et al. [30]	38	10	Bi	M	16	40	30	IO	ectomy	6	NS	Yes	Unsatisfactory				
Vaidhyanath et al. [32]	28	NS	Bi	M	9.5	30	33	IO	ectomy	10	NS	Yes	Satisfactory				
Ghazizadeh et al. [1]	25	10	Bi	M	28	48	40	IO	ectomy	NS	None	Yes	Satisfactory				
Jensen et al. [7]	29	NS	Bi	F	22	45	30	IO	tomy+ectomy	2	None	Yes	Unsatisfactory				
Silva et al. [22]	43 (max)	20 (max)	Bi (9)														
Romano et al. [25]	22 (min)	21 (max)	Uni (1)	F	25.6	41	36.6	IO (10)	tomy (8) ectomy (2)	1	None	NS	Satisfactory				
The authors' patient	29,2 (avg)	3 (avg)															
	53	NS	Bi	F	10	28	NS	IO	ectomy	NS	NS	Yes	NS				
	30	NS	Bi	M	21	NS	NS	IO	ectomy	NS	NS	Yes	Satisfactory				
	18	NS	Bi	M	22	42	32	IO	ectomy	3	NS	Yes	Satisfactory				
	43	28	Bi	M	11	NS	46	IO	ectomy	12	NS	Yes	Satisfactory				
	30 (avg)	NS	Bi (6)	M	16,3	NS	41	EO (3) (preauricular)	ectomy (6)	6	NS	Yes	Satisfactory				
	28	8	Uni	F	10	26	22	IO (3)	ectomy	NS	None	Yes	Satisfactory				

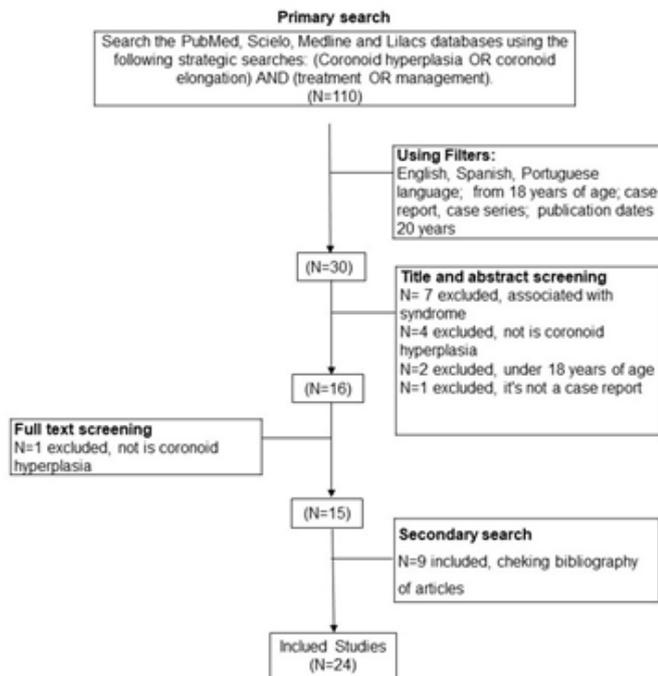


Figure 1. Flowchart for the search.

midline to the left, as well as, on palpation, no movement of the condylar translation (figure 2A). The tomographic examination showed an enlarged left coronoid process and in close contact with the inner face of the zygomatic body (figure 2B). Thus, the diagnostic hypothesis was unilateral CPH.

The patient underwent general anesthesia under nasotracheal intubation and intraoral access. After intraoral incision in the posterior region of the mandible, sub periosteal detachment was performed until the coronoid process was localized and the musculature of the temporal muscle bundles was removed. This was followed by the pinching of the process using Allis clamp, performance of mandibular incision osteotomy towards the anterior border of the mandibular ramus with surgical drill number 702, chisel and hammer (figure 2C) until complete removal of the hyperplastic coronoid process (figure 2D). In the transoperative period, a 26mm mouth opening could be observed. Histological examination showed healthy bone tissue, confirming the initial hypothesis.

The patient returned after 10 days of surgery with good evolution, without complaints, with a 22mm mouth opening and was then directed for complementary physiotherapy (figures 2E e 2F).

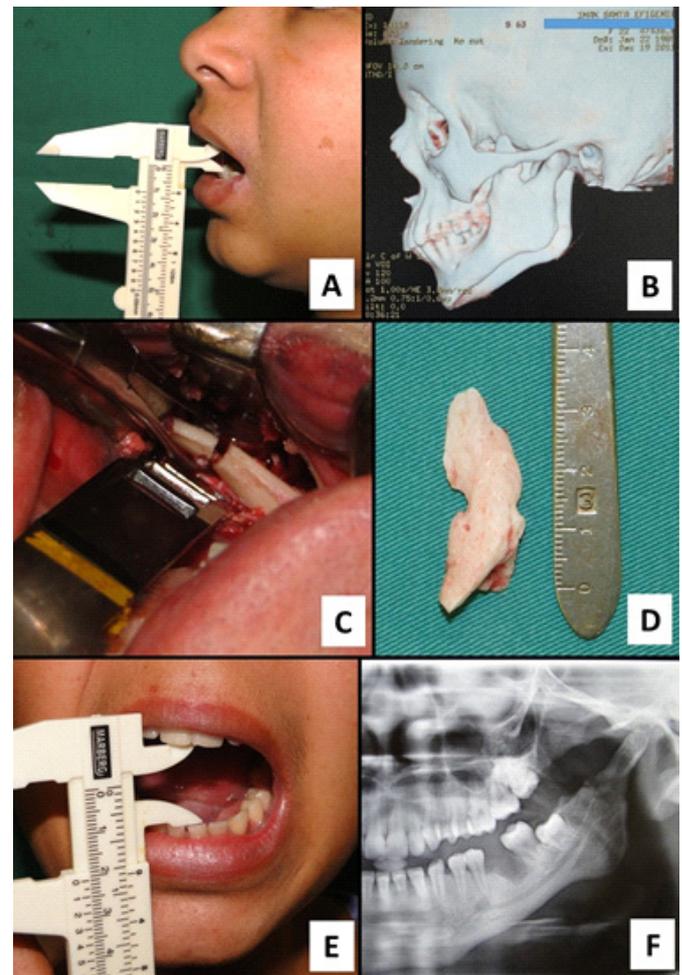


Figure 2. A) Preoperative mouth opening. B) Computed tomography in 3D reconstruction demonstrating enlargement of coronoid process of the mandible. C) Intraoral access and coronoid process osteotomy. D) Removed surgical part. E) Mouth opening on the 10th postoperative day. F) Postoperative panoramic radiography.

DISCUSSION

Coronary process growth was first described in 1853 by Von Langenbeck and the first case of mandibular hypomobility due to enlargement of the coronoid process was reported in 1899 [6,7]. In 1899, the formation of a synovial joint associated with the elongation of the coronoid process and the homolateral zygomatic bone was described [8]. Since then, several cases have been reported, creating a relative confusion between CPH and pathologies such as Jacob's disease, osteoma, osteochondroma and exostoses [9,10]. However, the pattern of bone growth in CPH is histologically normal [11], which is not the case in other diseases, in addition to the formation of pseudoarticulation between the coronoid and the zygomatic arch [12].

CPH is a rare and usually asymptomatic condition, with no occlusion abnormality and progressive oral limitation due to the contact of this structure in the temporal face of the zygomatic bone or the medial surface of the zygomatic arch [11,13-15]. As this process gradually increases, the infratemporal space necessary for the rotation and translation of the mandible is reduced, which results in the reduction of the mouth opening intervals and lateral excursion [1].

In addition, this developmental disorder can also lead to respiratory difficulties, delayed mandibular growth, muscular atrophy, difficulty to eat, or difficulties for adequate oral hygiene, which can lead to dental caries and make it difficult for the buccal surgeon to treat it because of the reduced operative space [11].

Its etiology and pathogenesis are still controversial, but the reviewed authors suggest that temporal muscle hyperactivity, temporomandibular dysfunction, endocrine stimulation, basal cell nevus carcinoma, traumas, genetic factors and heredity are factors that influence and can lead to muscle and skeletal alterations, whose occurrence favors the presence of morphological abnormalities of the coronoid process and the zygomatic bone [6,8,11,12,16,17]. Some cases presented a family history of CPH and trauma [12,21]. In the present study, the patient did not have painful symptomatology and had a history of facial trauma 2 years before clinical signs.

When considering a diagnosis of CPH, the clinician should keep in mind other more common conditions that have similar presentations. In fact, some patients with CPH often receive unnecessary treatment for other conditions before a diagnosis is reached. Limitation of buccal opening is often present in temporomandibular joint (TMJ) dysfunction, rheumatoid arthritis, TMJ ankylosis, uncorrected zygomatic bone fracture, tetanus, primary or secondary neoplastic disease [4].

Thus, imaging analysis in the diagnosis of CPH is essential through panoramic radiographs and computerized tomography, the latter is the gold standard in detecting the elongation of the coronoid process. In addition to revealing more precise information on the location of the contact of this structure with the adjacent bone, when the mouth is open, the examination will help in the differential diagnosis between CPH and other abnormalities. Magnetic resonance imaging is not recommended for the diagnosis

of bone abnormalities, but it may be used in cases of association with temporomandibular disorder [12,15].

Among 42 cases reviewed in this study, the majority were male and the age of diagnosis with the highest prevalence was between 10 and 30 years old confirming several publications [7,15,18]. In this case report, the patient is female, therefore, she doesn't correspond to the gender of higher prevalence and was diagnosed when she was 28 years, a very common age group in patients diagnosed with CPH.

This condition can occur as unilateral or bilateral hyperplasia. Most of the cases reviewed in this study were bilateral hyperplasia [6]. In cases of unilateral hyperplasia, the male gender is the most affected. In the present case, hyperplasia is unilateral in a female patient, gender of lower prevalence in unilateral hyperplasia.

Early diagnosis and proper treatment plan are important to reduce patient discomfort and avoid wasting time and money on conservative treatments, which would be ineffective. Surgical treatments followed by physiotherapy are successful in treating this disorder [18].

The surgical treatment of CPH has the purpose of removing the coronoid process and eliminating the mechanical obstacle that prevents adequate oral opening of the patient [1]. Two surgical techniques were described: coronoidectomy, more often used [11], and coronoidotomy, both of which can be performed through intraoral or extraoral access [19,20,21]. In this study, more than two thirds (69%) of the cases were treated by coronoidectomy, 26.2% by coronoidotomy, and only one case (2.4%) was treated with both surgical techniques.

Due to the ease of access and lower risk of complications, the vast majority (83.3%) of the cases had intraoral access. In two cases, the two surgical approaches were combined [14,22].

In one of the cases, the extraoral approach was performed initially, since the patient presented hypertrophy of the coronoid process, there was also great exostosis of the zygomatic arch at the point of contact with the coronoid process. The zygomatic arch was sectioned, connected from the masseter and rebuilt with mini plates after coronoidectomy, which was possible after the mouth opening evolved from 15mm to 30mm [14]. In another case, the patient's coronoid process was much hypertrophied, so the extraoral approach was initially chosen, due to the need for a greater extension for the resection of the

temporal muscle and its attachments. Subsequently, the intraoral surgical resection and removal of the coronoid process by coronoidectomy [22] were scheduled.

In coronoidectomy, the ascending ramus of the mandible is exposed the same as the upper part of the coronoid process, and then the temporal muscle is separated from the coronoid process. The entire coronoid process is removed. Alteration in muscle activity with temporal muscle detachment and postoperative fibrosis may lead to mandibular displacement [7]. In contrast, other studies affirm that coronoidectomy is an accepted treatment to obtain stable results in the correction of coronary malar interference [22] and prevention of postoperative fibrosis, besides guaranteeing the maintenance of the surgical part to obtain histopathological examination [12].

Coronoidotomy is based on the surgical detachment of the coronoid process of the mandibular ramus, and notably reduces the need for bone exposure and consequent surgical trauma in comparison with the coronoidectomy. In addition, this technique without removal of the coronoid process also prevents the formation of a postoperative hematoma, with consequent fibrosis at the operated site [5]. However, the stability of the result is considered questionable due to the risk of reconsolidation of the coronoid process [6]. In addition, there is no possibility to perform analysis [20].

In cases of intense trismus and minimal buccal opening, the temporal / pre-auricular approach is the best option to access the coronoid process of the mandible [14]. The submandibular approach can also be used in the case of zygomatic-coronary ankylosis and the coronal approach in cases of very elongated coronoid processes or associated lesions such as osteochondroma [20].

Intraoral access is the most appropriate [22], because it is aesthetically more convenient, since there is no apparent scar, in addition, it eliminates the risk of facial nerve injury [12,23-25] and offers sufficient space for the resection of the coronoid process and for the cut of the temporal tendon [22], despite the risk of postoperative hematoma formation [14]. Thus, despite the restricted preoperative oral opening of 10mm, aiming at the benefits of the intraoral approach for the accomplishment of the coronoidectomy, this was the treatment chosen for the reported case.

The decision on the type of surgical technique and approach should depend on the visibility, risk of complications and aesthetic requirements of the patient [8].

The great majority of the cases that showed significant results in the improvement of the oral opening were treated by coronoidectomy with a difference between pre and postoperative buccal opening varying around 10-42mm, whereas in patients treated by coronoidotomy this difference varied about 13-20 mm, only.

Only 5 cases presented unsatisfactory results and all these results came from treatment by coronoidectomy. One of the patients described in the present study had recurrence of symptoms 8 months after treatment, in addition to a new growth of the coronoid process after 2 and a half years [13]. In other publications [11,12,13,14,23], although there was no new growth in the coronoid process and the postoperative buccal opening was improved in comparison to the preoperative period, the result was considered unsatisfactory since the final buccal opening was less than 35mm, which has been proposed as a significantly restricted opening [26].

None of the cases presented operative complications, which makes the procedure not only efficient but also safe. In the case reported in this study, the patient had no surgical complication and the procedure provided an improvement in the buccal opening after the physiotherapy treatment.

In the postoperative period, the recommendation of physiotherapy to assist in improving the oral opening, restoring mandibular movements [27] and improving the patient's quality of life through daily exercise sessions is very important [23]. Almost all of the cases reviewed had physiotherapy as part of the treatment, confirming its importance for the treatment of CPH. It is recommended to start physiotherapy between 3 days and 1 week after surgery, with exercises performed for 10 minutes, 3 times a day, for 3 to 6 months [8].

The recurrence of CPH after a surgical procedure occurs due to the bone regeneration at the top of this structure, which may eventually join the mandibular ramus, causing, consequently, limitation of the buccal opening. Thus, monitoring the patient for a long time after surgery is essential in order to avoid recurrence [12].

CONCLUSION

CPH is a change in bone development that provides functional discomfort and gradual limitation of the oral opening. The treatment must be surgical, performed preferably by intraoral access and using the

technique of coronoidectomy, whose success is observed in the immediate postoperative period and also associated with long-term physiotherapy. Thus, it was verified that this type of treatment is effective, easy to perform, safe and with satisfactory results.

Collaborators

EFC NOGUEIRA, surgeon responsible for the patient, development of the manuscript. CMCT MARANHÃO, development of the manuscript. PL AGUIAR, development of the manuscript. RAC ALMEIDA, development of the manuscript. BCA TORRES, surgeon responsible for the patient, development of the manuscript. RJH VASCONCELLOS, manuscript supervisor.

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