

Case Report

Mandibular Coronoid Process Hypertrophy: Diagnosis and 20-Year Follow-Up with CBCT, MRI and EMG Evaluations

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Abstract: Coronoid process hypertrophy (CPH) consists of an abnormal volumetric increment of the mandibular coronoid process; as this process grows gradually, the infratemporal space needed for the rotation and translation of the mandible is reduced, which results in a reduction of the range of mouth opening and lateral excursion, limiting mouth opening. The purpose of this case report was to describe a rare case of hypertrophy of coronoid processes with associated temporomandibular ankylosis, monitored for over 20 years. The patient was first visited when he had a facial trauma at the age of 4. Then he was followed through clinical, functional, instrumental, bi-dimensional and three-dimensional radiological evaluations up to the age of 24. Physical therapy was initiated at the age of 10 to improve the condition of the masticatory muscles, while at the age of 14, Transcutaneous Electrical Nerve Stimulations were performed to reduce muscle tension and, a bite plane was delivered to control the parafunctional activity of the jaw in the night and self-control instruction was provided for daytime habits. The adult patient has not accepted surgical intervention; thus, the future objective is to continue monitoring over the years to avoid a detrimental progression of the medical condition through physical and functional therapies while waiting for patient consent to surgery if needed.

Keywords: temporomandibular joint; temporomandibular disorders; hypertrophy of the coronoid process; joint ankylosis; early diagnosis; mandibular movements; physical therapy; magnetic resonance imaging; cone-beam computed tomography; electromyography



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1. Introduction

Coronoid process hypertrophy (CPH), also defined as giant coronoid syndrome [1], was firstly described by Lagenbeck in 1853 [2] and consists of an abnormal volumetric increment of the mandibular coronoid process [1]; as this process grows gradually, the infratemporal space needed for the rotation and translation of the mandible is reduced, which results in a reduction of the range of mouth-opening and lateral excursion, limiting mouth opening.

CPH affects both genders with a male to female ratio of 5:1 and a peak of prevalence at around 25 years of age [1]. It can be mono or bilateral, although the bilateral form seems to be more frequent. The etiology of CPH is still not conclusive and in literature, different causes are reported [1]. In the clinical diagnosis of CPH, mouth-opening reduction (<20 mm) is quite always present [1]; however, the diagnosis should be confirmed by imaging of the head and neck region [3]. Cone-beam computed tomography (CBCT) can be considered the gold standard for radiologic diagnosis, providing detailed information about the length and width of the coronoid process and its relation to the zygomatic bone and arch [3–6]. Additionally, panoramic radiographs can be used as a routine examination for all patients with limited mouth opening to first assessed enlarged coronoid processes through Levandoski

panorgraphic analysis [7]. The limited mouth opening can be categorized as intra-articular or extra-articular [3,8,9], and the muscular component is usually involved in CPH due to its role as a stabilizer of the craniomandibular system during movements [7,8].

An early diagnosis is advisable to monitor and limit the progression of hypertrophy and consequent limited movements in oral multifunction [1].

However, this is not always feasible, due to delayed visits of patients stemming from the painless nature of CPH, and to general dentists being unaware of this rare TMJ pathology.

Regarding the type of treatment, it mainly depends on pathological severity, although surgery should be considered—usually an intraoral coronoidectomy together with postsurgical physiotherapy at the end of growth to achieve a normal range of mandibular opening maintained in the long-term period.

Thus, most authors agree that patients should be treated at the end of craniofacial skeletal growth to prevent any relapse with growth asymmetries [5,10–12].

However, a non-surgical treatment approach, with monitoring during growth until the major age, may be achievable with good results when an early diagnosis is performed.

The available literature showed a lack of studies presenting various therapies and long-term follow-up to evaluate treatment stability.

Thus, the purpose of this study was to describe a rare case of hypertrophy of coronoid processes with associated temporomandibular ankylosis, monitored for over 20 years. For this patient, strict monitoring was performed together with non-surgical treatments at different stages of growth through several clinical and imaging evaluations.

2. Clinical Case

After falling, a 4-year-old child had facial trauma and was referred to the First Aid Center of the Maresca Hospital Presidium in Torre del Greco, near Naples, in January 2001. He presented a chin cut that was sutured with three stitches; during this visit, no damage of the temporomandibular joint was diagnosed.

About 1 year after the incident, a schoolteacher noticed the difficulty of the child while eating. Thus, the parents were suggested to go to the Orthodontic Program of the Multidisciplinary Department of Medical-Surgical and Dental Specialties of the University of Campania Luigi Vanvitelli in Naples. A full visit with occlusal and functional examinations was performed by an orthodontist. The intraoral evaluation revealed mouth opening reduced (13 mm), lateral mandibular movements of 3 mm and a protrusion of 2 mm without pain or clicking sounds in the temporomandibular joints (TMJ) (Figure 1).



Figure 1. Reduced mouth opening (13 mm) at the first visit (age 5).

After excluding a muscle contracture through functional exercises to decontract TMJ and masticatory muscles, a possible diagnosis of coronoid process hypertrophy was provided through bi-dimensional panoramic X-ray evaluation that showed larger and higher coronoid processes over the zygomatic arch with normal bone trabeculae in the processes.

A clinical reevaluation and further radiological diagnostic examinations were performed at the age of 7. The patient was in mixed dentition and the elongation of the coronoid processes was confirmed by panoramic X-ray. Levandoski panographic analysis, described in Kubota et al. [7], was performed on the panoramic X-ray to measure the elongation or hypertrophy of the coronoid process through the evaluation of three reference points: condylion (Cd), gonion (Go) and koronion (Kr) [13]. The ratio of Kr-Go/Cd-Go on the right side was 1.10 and 1.09 on the left side (Figure 2).

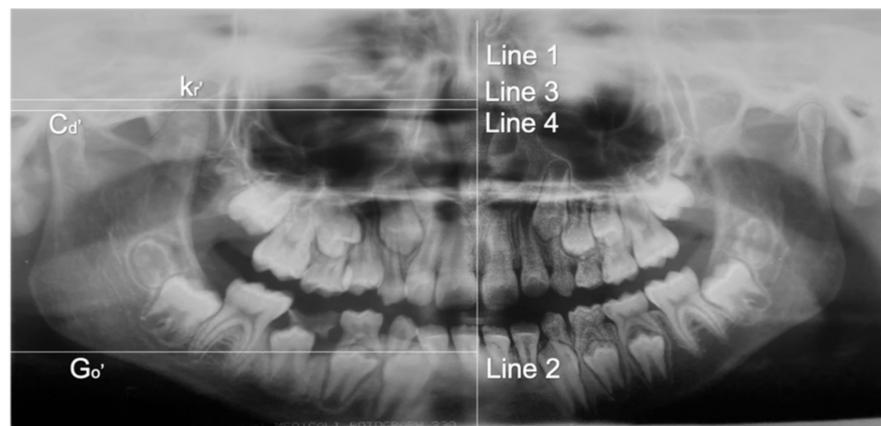


Figure 2. Diagram of the Levandoski panographic analysis on the panoramic X-ray. Line 1 is the maxillary vertical midline. Lines 2, 3 and 4 are perpendicular to line 1 as they cross the lower border of the symphysis of the mandible (Go'), the tip of the condyle (Cd') and the tip of the coronoid process (Kr'), respectively (age 7).

At the age of 10, the previous diagnosis was confirmed after clinical monitoring supported by a three-dimensional evaluation through CBCT and Magnetic Resonance Imaging (MRI). More in detail, CBCT scans showed bilateral enlargement of the coronoid processes and their close relationships with the anterior part of the zygomatic arches (Figure 3a–d). The MRI images confirmed a correct bilateral condyle–disk relationship. Bilateral TMJ fibrosis anchyloses seemed to be revealed, differentiating CPH from chronic disk displacement without reduction (Figure 4a,b).

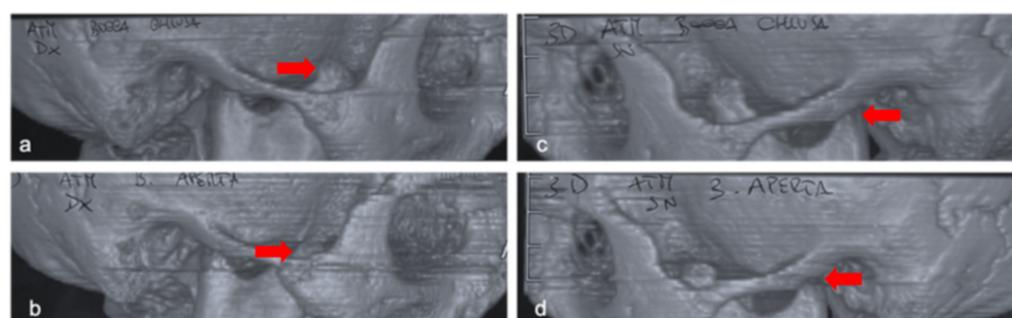


Figure 3. (a–d) CBCT scans: (a) right side—closed mouth; (b) right side—open mouth; (c) left side—closed mouth; (d) left side—open mouth (age: 10 years).

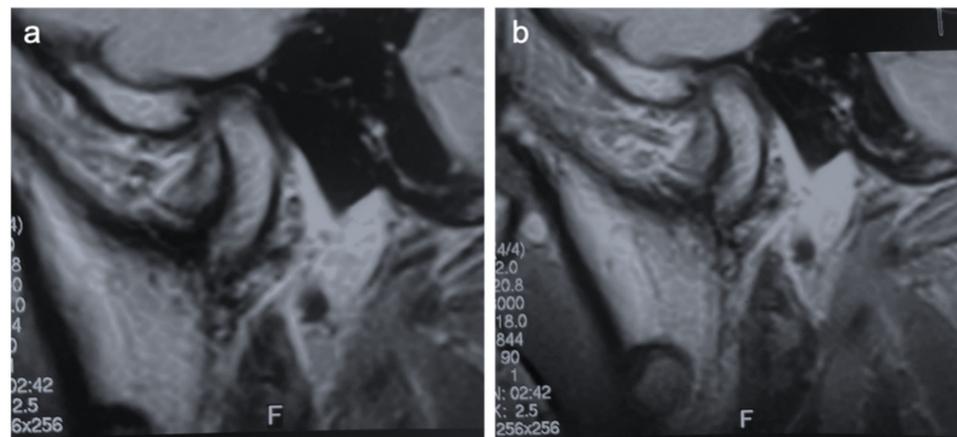


Figure 4. (a,b) MRI images at the right side: (a) closed mouth; (b) open mouth (age 10).

The patient's parents were given explanations and counseling regarding the diagnosis and treatment options. Due to the young age of the subject, only physical therapy was initiated to improve the condition of the masticatory muscles and avoid further mandibular movement reduction, mainly in mouth-opening [14–16] exercises involving active hinge opening and manual finger stretching in front of a mirror, combined with heat and massage through stretching and against-resistance exercises within painless limits. Over the years, periodical clinical checks were performed and the therapy resulted in an increased mouth opening from 13 mm to 21 mm after 1 year. This slight increase in mouth opening made daily activities easier, such as feeding and oral hygiene. After that achievement, the patient did not come to follow-ups, maybe being satisfied enough with the results obtained.

Only after 8 years (at the age of 14 in 2010), he came back to the Orthodontic Program, referring to recurrent headaches and muscle fatigues in the orofacial district (at the masseters and temporal muscles), mainly when keeping his teeth in contact for many hours a day.

He was subjected to clinical, photographic and radiological reevaluation, and an electromyographic examination of the masseter and temporal muscles was performed.

About the functional examination, the maximum opening of the mouth was limited but without any deflections, showing a maximum mouth opening of 20.05 mm (Figure 5).



Figure 5. Mouth opening increased to 20.05 mm (age 14).

CBCT (Figure 6a,b) and MRI (Figure 7a–d) images confirmed the coronoid process elongation on both sides which were locking above the zygomatic arch, confirming the absence of growth asymmetries and suspected TMJ intracapsular fibrosis ankylosis. The pain was mainly referred bilaterally by palpation of the masseter and anterior temporalis

muscles. The K7/EMG (Myotronics-Noromed, Inc., Tukwila, WA, USA) was also used, and it showed dystonia of all masticatory muscles and hypoactivity of the anterior right temporalis at rest.

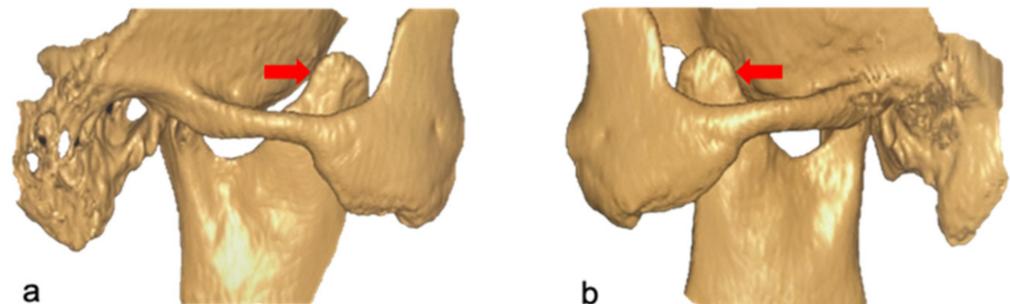


Figure 6. (a,b) CBCT examinations at closed mouth: (a) right side; (b) left side (age 14).

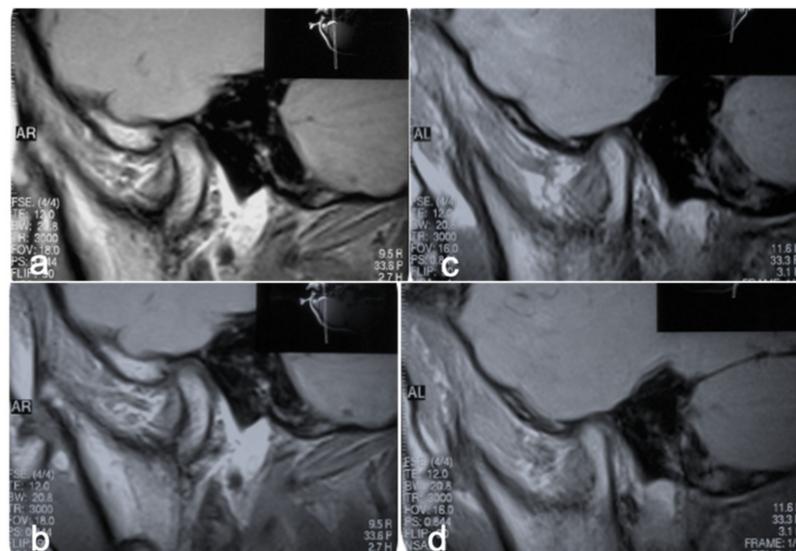


Figure 7. (a–d) MRI images: (a) right side—closed mouth; (b) right side—open mouth; (c) left side—closed mouth; (d) left side—open mouth (age 14).

The patient accepted treatment with Transcutaneous Electrical Nerve Stimulation (TENS) to reduce muscle tension, a bite plane to control the parafunctional activity of the jaw at night and self-control instruction for daytime habits. The patient was treated 30 min daily, two times weekly for 3 months, obtaining a significant reduction in algic symptomatology.

However, it was reiterated that no conservative therapy could solve the patient's condition except bilateral coronoidectomy and removal of the ankylotic mass with ostectomy at the end of growth, then allowing correct mandibular movements together with better mouth opening.

In 2020, in a long-term follow-up of 19 years, the patient was recalled for a monitoring check-up at the age of 23. A complete check-up was carried out. At the functional examination, the range of movement (ROM) was unchanged compared to the previous evaluation of 9 years before. Intraoral examination showed severe tooth decays at second and third lower molars on the left side that were extracted despite the limited opening of the mouth (Figure 8). On the panoramic X-ray, the Levandoski panoramic analysis was used to compare measurements with the previous analysis performed at the age of 7. The ratio of Kr-Go/Cd-Go on both sides was increased with a value of 1.11 on the right and 1.10 on the left side.



Figure 8. Diagram of the Levandoski panoramic analysis on the panoramic X-ray (age 23).

CBCT imaging was obtained to evaluate the coronoid process changes in comparison to scans of 13 years before (Figure 9). The mouth opening remained constant whereas the joint disks and condyle at the MRI examination showed an ankylotic aspect despite the acceptable position (Figure 10).

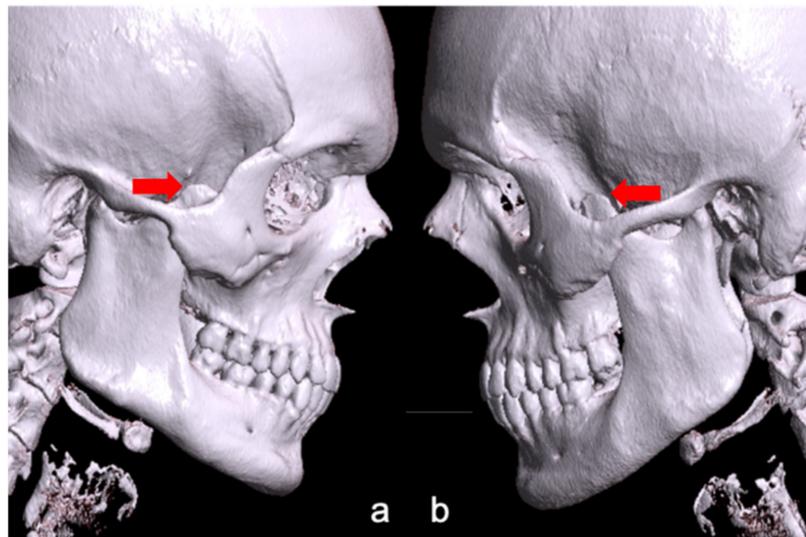


Figure 9. (a,b) CBCT examinations at closed mouth: (a) right side; (b) left side (age 23).

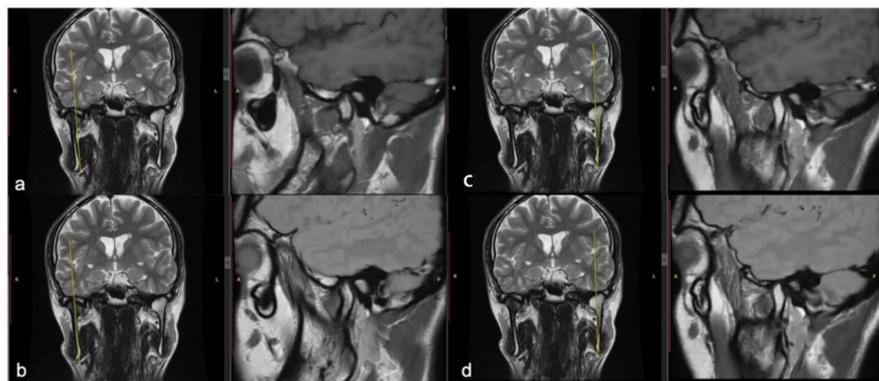


Figure 10. (a–d). MRI images: (a) right side—closed mouth; (b) right side—open mouth; (c) left side—closed mouth; (d) left side—open mouth (age 23).

The K7/EMG confirmed a mild hypotony of the masseter and anterior right temporal muscles (Figure 11).

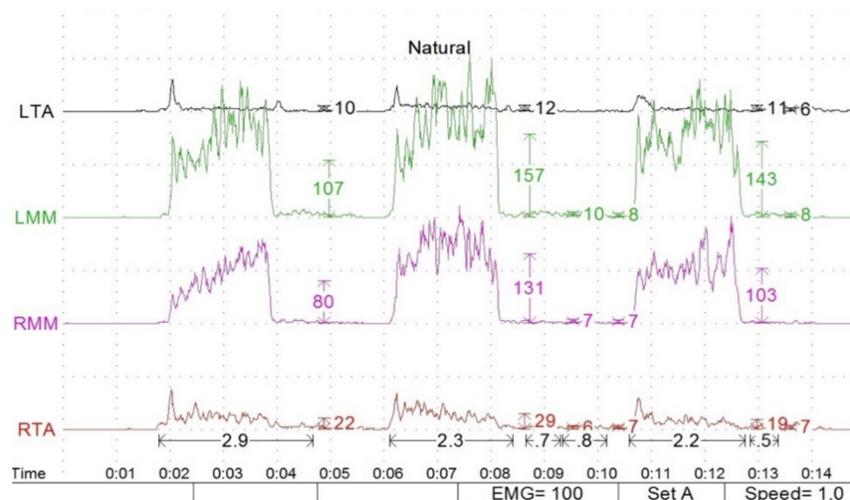


Figure 11. EMG/K7 analysis at rest (age 23). The following muscles were recorded: left temporalis anterior (LTA), left masseter (LMM), right masseter (RMM), right temporalis anterior (RTA).

Considering the adult age of the patient and his diagnostic records over time, surgical intervention of coronoidectomy was suggested as the gold standard of therapy to resolve the limited mouth opening. In this regard, new, minimally invasive surgical techniques associated with early postsurgical physiotherapy are available to reduce the main possible complications [1]. This was referred to the patient, along with the provision of all detailed information on treatment modalities and the consequent benefits to his oral functions. Nevertheless, the patient denied surgery for correction of the hypertrophy of the coronoid process, arguing that, at the time, the condition did not cause any reduction of his quality of daily life. He preferred to delay the surgical intervention to the future even though he understood the benefits of this approach.

3. Discussion

CPH is often misdiagnosed due to its rarity and the absence of pain [12], even if restrictions in mouth opening and mandibular movements are present since childhood [17]. Thus, a deep knowledge of any clinical and instrumental analyses available to monitor this condition during growth is necessary to make an early diagnosis and correct treatment plan. The normal range of mouth opening is 53–58 mm, measured between the incisal edges of the maxillary and mandibular teeth.

Diagnosis of CPH is effective mostly through imaging [17]. First, a panoramic X-ray is recommended [2,17], and if there are any suspicions, the subsequent step is the prescription of three-dimensional imaging such as CBCT and MRI to detect any intraarticular derangements in hard or soft tissues [5,18]. In particular, MRI may help differentiate CPH from chronic disk displacement without reduction [17,18]. The etiology of coronoid hypertrophy is not well-described, but it has been found in association with many conditions [4,7,19]. In this case, it may be supposed that the trauma was the main cause for the onset of the pathological process [1]. Studies carried out on postnatal surgeries on laboratory animals showed that changes in muscular connection result in changes in size and form of the coronoid process and deficits in some myogenic factors or in the expression of specific genes that can provoke muscular and articular disorders [17,20]. Patients with restricted mouth opening tend to eat softer foods, which may lead to reduced temporalis muscle activity. This results in fatigue and chronic hyperemia, further stimulating an inflammatory response. The chronic persistence of inflammatory cells in the muscles induces an excess of cytokines and growth factors, which contributes to the formation of permanent

fibrosis [6,21]. So, increased duration of TMJ ankylosis leads to an increased incidence of coronoid elongation.

The surgical management of CPH patients is very complex, being that the treatment of associated TMJ ankylosis is highly challenging for surgeons [9]. The major problems faced during such surgeries are the risk of excessive blood loss with all of the complications of blood transfusions [7], joint reankylosis [3], function reduction, growth defect postoperatively [8] and the mandibular inability to intubate in an emergency [22]. Obtaining a satisfactory outcome depends largely on proper post-operative rehabilitation [2] to avoid relapse, and to emphasize results, it is suggested to start physiotherapy 5–7 days after surgery [2,9,23].

It can be difficult to define the time to administer treatment in infants, although most authors agree that, except in patients with very severe limitations in mouth opening, it is better to perform the operation once the growth process has finished, in order to avoid recurrence, deformity or even restricted movements [2,12,24].

Kumar et al. demonstrated that patients >16 years old showed greater improvement in mouth opening at >1-year follow-up compared to patients aged <16 years [6]. The rate of joint reankylosis in a child group was 19.1%, which was significantly greater than that in the adult group (7.3%; $p < 0.05$) [9]. Yang et al. demonstrated that the rate of facial growth and function after the reconstruction of the mandibular condyle in children with ankylosis seemed to be less than on the undisturbed side, even after surgical treatment of the ankylosis [25].

A review of the literature found that vertical amplitude of normal mandibular movements for the production of speech is equal to one-third of the amplitude of the maximum opening of the jaw, around 7–18 mm with a slight anteroposterior component of 2–3 mm [26]. Moreover, a typical chewing pattern consists of a few cycles on one side, after which the food bolus is moved to the other side by tongue and cheeks for a similar number of cycles. Each cycle commences with an opening phase in which the teeth move away from the intercuspal position along a path close to the median line but gradually approaches the food bolus side. Rarely does the total movement exceed the minimum necessary to grasp the food bolus; on average, the opening phase involves about 18 mm incisal movement [27]. Effectively, our patient did not refer to any problems during chewing function.

Thus, physical and manual therapy (including joint mobilization and the manipulation of soft tissues) is amongst the 10 most common treatments used for temporomandibular disorders to improve range of motion and promote exercise to maintain healthy function [5,28]. Mazzetto et al. showed that after physical therapy, patients demonstrated an improvement in symptoms and expressed satisfaction with the treatment, which had improved both esthetics and mastication [29].

Chen et al. instead supported the high recurrence rate in children and found that ankylosis should be treated as soon as possible to improve patient cooperation with physical exercise post-operation. Untreated TMJ ankylosis in children leads to several adverse effects, including facial asymmetry, muscle dysfunction and psychological disorders [9].

Farronato et al., in a review of the literature, reported an increased mouth opening from 5 to 25 mm postoperatively, and intra-oral and extra-oral approaches were described. The intra-oral approach usually provides enough exposure to allow the removal of the hyperplastic process without visible scarring. The major risk is postoperative hematoma and fibrosis. Several extra-oral approaches have been proposed, such as sub-mandibular, pre-auricular, (bi)temporal or endoscopically assisted. The advantages of the extra-oral surgical approach are less fibrosis and/or hematoma formation and better exposure to reset the coronoid process and release the temporalis muscle. The damage of the facial nerve and a visible scar during the healing process are the main risks of the extra-oral technique.

The future goals are monitoring and ROM evaluation: as reported by Kumar et al., the unique resolute therapy for these cases consists of coronoidectomy and the removal of the ankylotic mass [30]; surgery will be performed when the patient is motivated, probably only when the mandibular function has reduced his quality of life. A minimally

invasive approach with postsurgical physiotherapy is proposed to reduce any possible complications reported in previous cases [1].

The maximum mouth opening is about 40 mm since the age of 6 and only 1.2% of young adults open less than that value. So, a restriction of mouth opening of less than 40 mm may be considered a pathologic condition [31].

However, the patient's willingness and perceptions have to be considered and the resolution of the restriction of mandibular movements may not always be feasible through adequate surgical interventions. In our case, the future objective is to continue monitoring over the years to avoid a detrimental progression of the medical condition through bi- and three-dimensional evaluations [32,33] and physical and functional therapies, while waiting for patient consent to surgical intervention.

4. Conclusions

Mandibular restriction is found in different diseases; thus, the formulation of a precise diagnosis is more difficult. Complementary exams for clarifying a clinical condition when in doubt should be considered significant. Even when the diagnosis is conclusive, it is not always viable to make a recognized therapeutic procedure and a more conservative approach is required, provided that this is duly explained to the patient.

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