Therapeutic Protocol for Orthosurgical Management of Class III Malocclusion in Patients With Cleidocranial Dysostosis

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Abstract: Cleidocranial dysostosis (CCD) is a congenital skeletal disorder with significant manifestations in facial and dental development. Patients are affected with CCD present maxillary deficiency, late dental eruption, and supernumerary teeth. Early and multidisciplinary approach is necessary to treat CCD patients, especially to manage dental eruption and Class III malocclusion with maxillary deficiency. Several orthodontic and surgical interventions are performed to enable traction and extraction of teeth. Yet the maxillary deficiency may be protracted followed by orthodontic dental compensation. On the other hand, it is important to note that CCD patients' treatment is closely related to the severity of transversal and sagittal deformities, as well as the discrepancies in the lower third of the face. In this context, patients with facial impairment highly affected by CCD may need ortho-surgical decompensation to reach more aesthetic outcomes. The present study reports a case of a 14-year-old young patient affected by CCD. Clinically, the patient presented Class III malocclusion, maxillary deficiency, short lower facial third, posterior crossbite, and anterior open bite leading to facial disharmony. The patient underwent treatment in 2 stages: the interceptive approach aimed to transversally expand the maxilla and promote its protraction; and the corrective phase combined with the orthognathic surgery treated the patients' main complains; the anterior open bite, unerupted teeth, and chin prominence. The treatment approach applied in the clinical report allowed the correction of the malocclusion and facial profile satisfying completely the patient's expectations.

Key Words: Class III malocclusion, cleidocranial dysostosis, corrective orthodontics, oral surgical procedures

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Cleidocranial dysostosis (CCD) is a rare autosomal-dominant inheritance that manifests clinically with shoulder hypermobility—due to clavicle hypoplasia or aplasia; delayed closure of cranial fontanelle; brachycephalic skull; short stature; late exfoliation of the primary dentition; late eruption of permanent teeth; supernumerary teeth; and morphological dental alterations. This disorder was first reported in 1765 and results from a mutation in the gene *CBFA1*, specifically in the short arm of the chromosome 6p21. The current prevalence rate of CCD reaches 1 in a million, without preference for sex and ethnicity.[1–7]

Genetic and molecular studies in humans point toward dental phenotypic alterations in the gene *Runx2* (CBFA1), which is a critical transcriptional regulator of bone and dental development. Heterogenic mutations in *Runx2* lead to CCD. This data support the hypothesis that *Runx2* is one of the main mesenchimatic factors influencing dental morphogenesis, especially the differentiation of ameloblasts and odontoblasts.[1,5,8–11]

Supernumerary teeth impact considerably occlusion. Predicting the development of these teeth has an important role to treat CCD patients in the dental practice. The quantity and morphology of supernumerary teeth (found in identical genetic mutations in RunX2) may vary from patient to patient. The environmental influence, as well as the complex combination of Epigenetics and the variation in the amount of teeth, may be involved in the expression of supernumerary teeth in CCD.[6,9,12,13]

Dental treatment for CCD patients must be implemented in a multidisciplinarily approach between deciduous and permanent dentition. Therapeutics include the extraction of supernumerary teeth and the correction of transversal and sagittal discrepancies with maxillary protraction.[1,2] According to the literature, 4 treatment approaches for CCD patients have been proposed: Toronto-Melbourne, Belfast-Hamburgh, Jerusalem, and Bronx.[6,14]

The Toronto-Melbourne protocol comprises a serial extraction of the deciduous teeth based on permanent root development favoring their spontaneous eruption. The supernumerary teeth are extracted during the deciduous teeth extraction under general anesthesia. The Jerusalem protocol requires 2 surgical interventions, initially the deciduous teeth are removed at the same time as the successor permanent teeth are exposed for tractionning. The second approach is accomplished when patients are approximately 13-year old and the remaining deciduous teeth are extracted, and canines and premolars are exposed to begin the tractionning phase. In the Bronx protocol all deciduous and supernumerary teeth are removed initially and in the second intervention all impacted teeth are exposed for tractionning. During this second approach the corrective appliances are bonded to prepare for the orthognathic surgery. The Belfast-Hamburgh protocol is characterized by only 1 surgical intervention to remove all deciduous teeth and to expose all supernumerary and permanent to begin their traction.[6,15]

After the interceptive approach, a new treatment phase will take place considering the need for ortho-surgical interventions based on the severity of the malocclusion and the facial deformities. In adults, the dentomaxillofacial alterations are consolidated and may have deleterious effects on facial pattern and occlusion. In these cases,

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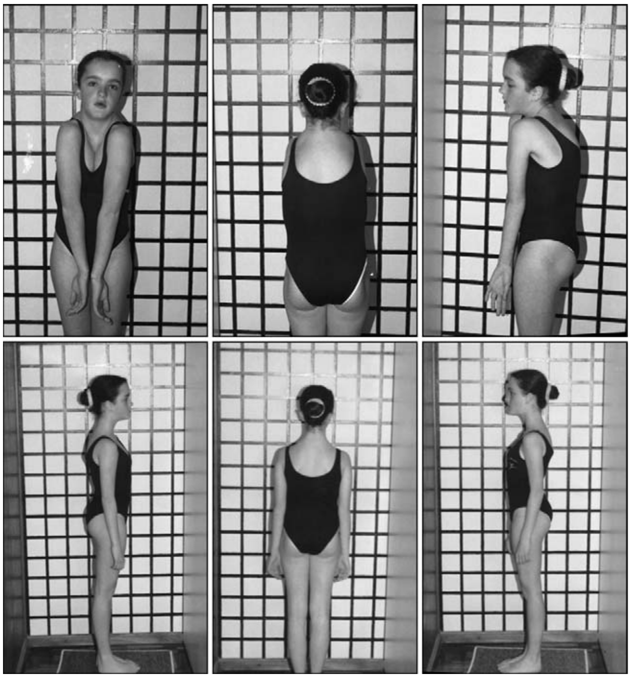

FIGURE 1. Diagnostic photographs of cleidocranial dysostosis. (A) Frontal dosiflexion. (B) Posterior dorsiflexion. (C) Right profile. (D) Left profile. (E) Posterior. (F) Right profile.

the corrective approach with orthognathic surgery is often necessary.[4,16]

Advancement of the maxilla or upper and lower repositioning via Le Fort I Osteotomy orthognathic surgery has become an established procedure for the restoration of facial function and esthetics, correction of skeletal and occlusal discrepancies, and treatment of obstructive sleep apnea in patients with facial deformities. Definitive correction of these anomalies leads to long-term stability of the skeletal changes.[1,3]

Based on the exposed, the present study aims to discuss the CCD ortho-surgical approach by presenting a successfully treated patient.

DIAGNOSIS

A female patient (G. M. S.) aging 28 years and 4 months was referred for orthodontic treatment. She presented history of previous orthodontic treatment with CCD diagnosis confirmed by shoulder hypermobility due to clavicle hypoplasia evidenced (Fig. 1A–F). The first orthodontic approach based on the Bronx protocol was accomplished when she was 14 years old. The deciduous and supernumerary teeth were extracted and rapid maxillary expansion combined with maxillary protraction was performed at the same time to correct the transversal and sagittal problems.

After that a follow-up period is indicated to wait for the adequate root development of the permanent teeth to start their traction but the patient abandoned the treatment.

When she returned and sought for treatment she presented Class I malocclusion based on maxillary deficiency associated with anterior open bite. The facial analysis revealed a slightly concave profile with a discrete asymmetric frontal view. Passive lip competence was also observed, as well as a defined horizontal line between the chin and the neck. The lower third of the face was deficient with lacking expression of the nasolabial sulcus (Fig. 2A–H).

In specific, the lateral radiograph (Table 1) revealed a sagittal discrepancy (ANB: -2.5^o) with potential participation of the mandible (SNB: 81.8^o) and relative involvement of the maxilla

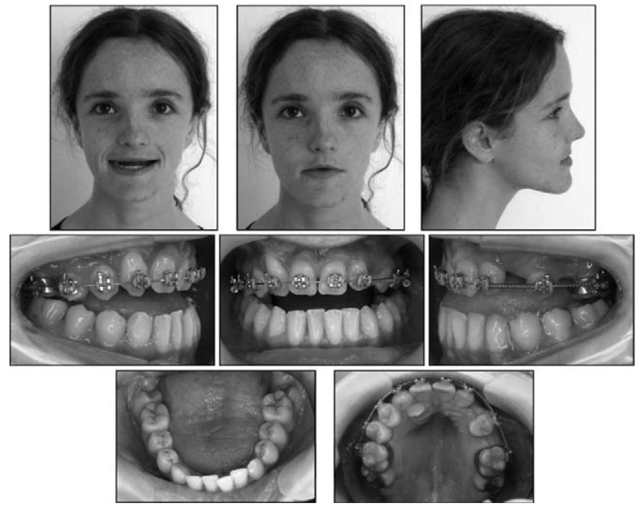

FIGURE 2. Initial photographs of the corrective phase (A—frontal smiling, B—frontal, C—lateral in 45°); intraoral photographs (D—right lateral, E—frontal, F—left lateral, G—occlusal lower, H—upper occlusal).

(SNA: 79.3^o). The mandible body was not proportional in relation to the ramus, and the gonial angle was excessively opened. The lower permanent incisors presented lingual inclination (1.NB: 18.0^o) as a compensation for the sagittal discrepancy.

TREATMENT PLAN

The therapeutic strategy was elaborated after analysis of the orthodontics records. It was decided to continue the treatment plan based on the Bronx protocol comprising the surgical approach to correct the sagittal and vertical discrepancies. The main factors contributing to the treatment approach consisted of the severity level of the maxillary deficiency, as well as the dentoalveolar discrepancy and their impact on facial aesthetics. The interceptive treatment had been performed previously as long as the supernumerary and deciduous teeth extractions followed by the fixed corrective appliance to provide space for permanent teeth traction. The leveling phase to decompensate the teeth was performed to enable the future surgical treatment. Combined, the corrective and surgical treatments contributed to overcome the skeletal, occlusal, and aesthetic deficiencies presented by the patient.

TREATMENT PROGRESS

The corrective approach was performed with metallic brackets 0.022″ × 0.030″ (Kirium Capelozza I, 3 M) in the upper arch from right second premolar to left first premolar, except for the upper right lateral incisor (palatal displacement), the upper left canine,

TABLE 1. Cephalometric Analysis Before and After Surgery

Measurement	Norm	Initial	Final
SNA	82	79.3	80.5
SNB	80	81.8	80.3
ANB	2	−2.5	−0.3
1.NA	22	23	29.3
1.NB	25	18.0	11.5
1-NA	4	1.2	4.4
1-NB	4	2.2	1.3
IMPA	87	86.2	80.8

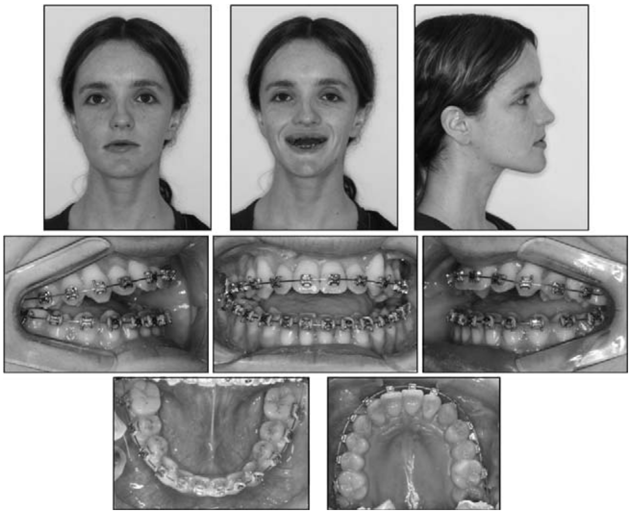

FIGURE 3. Upper right lateral incisor, upper left canine, and second premolar included in the dental arch (A—frontal smiling, B—frontal, C—lateral in 45°); intraoral photographs (D—right lateral, E—frontal, F—left lateral, G—lower occlusal, H—upper occlusal).

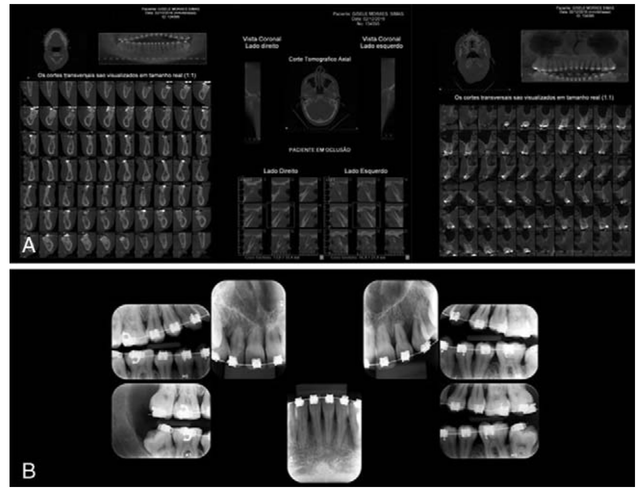

FIGURE 4. Orthodontic records to support the surgery. (A) Preoperative cone-beam computed tomography. (B) Preoperative periapical radiographs.

and the upper left second premolar (both unerupted). Orthodontic bands were installed in the first molars. By that time, new panoramic and lateral radiographs, and cone-beam computed tomography (CBCT) scans were requested for planning the traction of the unreputed teeth. Space was obtained to align the upper right lateral incisor, while upper left canine and second premolar were tractioned. The conventional orthodontic brackets were replaced for self-ligating metallic brackets $0.022'' \times 0.030''$ (Portia Capelozza I, 3 M) in the upper arch, which increased in perimeter and conventional brackets were bonded in the lower arch (Fig. 3A–H).

Before surgery, the same system of self-ligating metallic brackets was replaced in the lower arch from right to left second premolars. Special attention was given to the second lower left molar (impacted), which was treated with elastics and spacers for further inclusion and leveling in the arch. After this phase, the patient underwent the extraction of third molars as requested by the surgeon. The alignment of the lower arch, including the traction of the lower left second molar, was conducted progressively up to the stainless steel wire $0.019'' \times 0.025''$. Surgical hooks were used for the postoperative intermaxillary fixation with elastics. Elastics were also used after the surgery for discrete corrections toward conclusion of the case. For the same reason, slight occlusal corrections were performed. In accordance with the maxillofacial surgeon, the final position of the incisors was set only after the surgical traction of the maxilla (forward and downward). The final alignment was achieved with a stainless steel wire of $0.019'' \times 0.025''$.

The orthodontic and preoperative phases were concluded. Cone-beam computed tomography scans of the maxilla and mandible (Fig. 4A) and complete orthodontic records were requested to support the surgery, including periapical radiographs (Fig. 4B).

In this presurgical phase, when analyzed in frontal view, the patient had an unfavorable exposition of the anterior teeth because the occlusal plan was inverted. Even when smiling, the anterior teeth were exposed discretely. The patient ranked this problem among his main complaints. Based on the severe sagittal discrepancy with facial impairment and considering the patient's complaint on the lack of dental exposure while smiling, the treatment plan involved the maxilla exclusively.

Presurgical documentation, including full face CBCT (in centric relation) with study models scanned in final occlusion, was imported into the Dolphin Imaging Version software (11.9) for image manipulation and consequent surgical planning. Due to the great inversion of the maxillary plane and the patient's complaints, maxillary surgery was necessary. After discussion between surgical and orthodontic team, the treatment plan was presented to the patient. The treatment proposed consisted of maxillary advancement, with anterior bone extrusion, clockwise rotation, and quadrangular Le Fort I osteotomy with a bone step in the zygomatic buttress. This management reduces the bimaxilar surgery morbidity and mandibular surgery adverse effects, such as lower alveolar nerve paresthesia.

Six millimeters of anterior bone extrusion was planned for this case. This amount of extrusion allowed the correction of aesthetics without modifying the posterior region. The exposure of incisors was improved for both smiling and resting. Despite invasive, the surgical approach adopted in this case had low morbidity and enabled better postoperative healing.

Surgery occurred under general anesthesia with nasotracheal intubation with hypotension throughout the transoperative period. Linear maxillary incision was performed 5 mm above the inserted gingiva, by means of foursquare osteotomy with surgical saw for maxillary downfrature with Rowe forceps. An interocclusal splint, through 3-dimensional printing from virtual planning was used to reposition the maxilla in the new occlusion. The external measurements idealized the ideal measure of maxilla extrusion. Internal bone fixation was performed using four rigid plates of 1.5 mm.

As expected, a large bone defect became apparent in the anterior region of the maxilla due to the extrusion previously performed. To minimize the risk of pseudoarthrosis in the maxilla, bone grafting from the iliac crest was considered. However, the patient rejected this alternative due to the potentially prolonged healing and the local scars. A second alternative was proposed and accepted by the patient: xenogenous bone grafting. Bio Oss Collagen and BioGide collagen membrane were used to fill the gap of 5 to 7 mm in the anterior region of the maxilla. The Bio Oss colagen has this advantage of being a 3-dimensional scaffold, leading to a good stability of the bonegraft, fulfilling the gap. Surgery underwent with no intercurrence (Fig. 5A–C), which was confirmed by a postoperative tomography (Fig. 6A–D), showing a good bone formation, mainly close to the anterior plates.

In the immediate postoperative time patients were maintained with no elastics, so she could have a good recuperation from

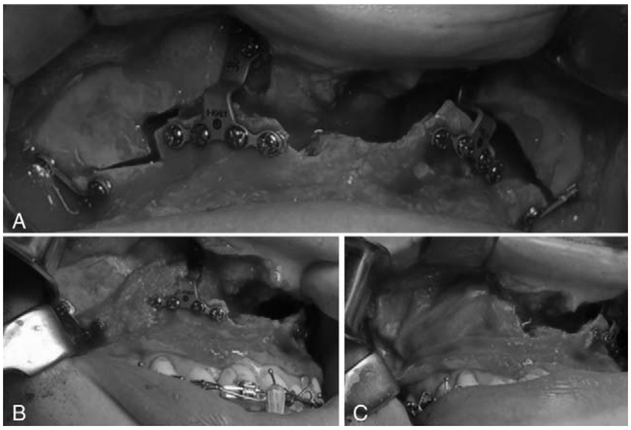

FIGURE 5. Xenogenous bone grafting. (A) Bone gap after maxillary repositioning. (B) Bio Oss collagen placed in the gap. (C) Placement of BioGide collagen membrane.

anesthesia. After 2 days and for the next 14 days the patient was blocked with intermaxillary elastics. One month after surgery, the patient was sent back to the orthodontist to do the final adjustments.

The techniques used to close the posterior occlusion in the postoperative period were folds in the orthodontic wires and mainly the use of intermaxillary elastics. After 2 years and 8 months of orthosurgical therapy, final orthodontic records were requested and the orthodontic retainers were provided to the patient (Hawley retainer in the upper arch and 3×3 in the lower arch). In a clinical evaluation immediately after the closure of the gap on the posterior and anterior region, a stable occlusion was observed, and there was no relapse in the postoperative movement of the maxilla.

Significant improvement of the periodontal condition including the reduction of the gingival retraction (specially in the upper left canine) was observed within 3 months after treatment. Functional corrections were necessary on the occlusal surface of the lower right and left second molars due to the premature contact commonly observed in CCD patients.

The orthodontic appliances were removed (Fig. 7A–H). Hawley retainer was used in the upper arch, while in the lower arch a 3×3 retainer was bonded. In total, the treatment was performed in 18 months. At the end of the treatment, the patient was aged 31 years and 3 months. At this stage, panoramic, lateral cephalometric, and periapical radiographs of the upper and lower incisors were requested.

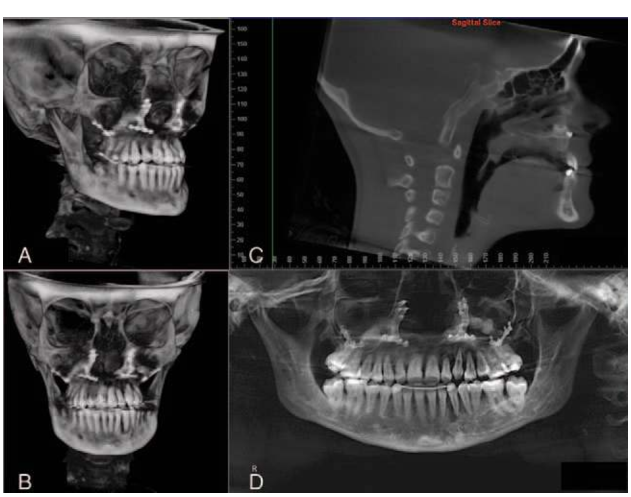

FIGURE 6. Postoperative tomography showing a good bone formation (A) lateral view in 45°, (B) frontal view, (C) sagittal slice, (D) panoramic slice.

FIGURE 7. Photographs on the removal of orthodontic appliances. (A) Frontal smiling, (B) frontal, and (C) lateral in 45°. Intraoral photographs (D) right lateral, (E) frontal, (F) left lateral, (G) lower occlusal, (H) upper occlusal.

TREATMENT OUTCOMES

At the end of treatment, the patient presented Class I molar relationship bilaterally without anterior open bite. The skeletal discrepancy was corrected (SNA: 80.5; SNB: 80.3; ANB: −0.3) (Table 1) with orthognathic surgery. It was possible to observe the skeletal profile changes with the superposition of the initial and final tracings (Fig. 8), with forward and downward displacement of maxilla and anticlockwise mandibular rotation. Moreover, a harmonious aspect was achieved in the facial profile. The reduction of the AFAI resulted from the decompensation of the maxilla forward and downward and the corrected angulation of the upper incisors. The nasolabial sulcus became more expressive in the middle third of the face with the decrease in facial concavity. Within 1-year follow-up, relative stability was observed indicating success of the treatment approach for a CCD patient with maxillary deficiency. A slight posterior bite opening has occurred, which is perfectly acceptable in patients presenting anterior open bite. Accordingly, patient's satisfaction confirmed the treatment effectiveness. On the other hand, it is important to note the gingival retraction in the upper incisors and the upper left canine justified on the lack of bone

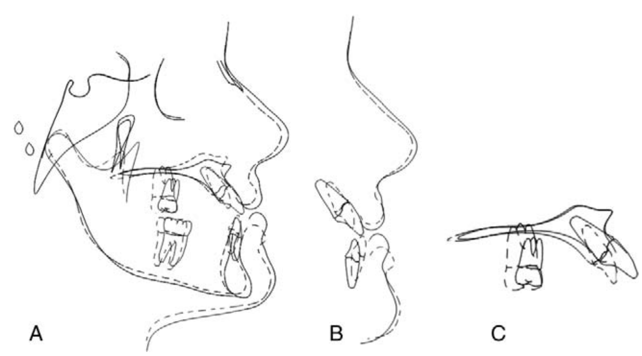

FIGURE 8. Superpositions. The dashed line is the initial tracing and the continuous line the final tracing. (A) Initial and final cephalometric tracings. (B) Initial and final maxillary position. (C) Initial and final incisor position.

support in CCD patients and the traction sequelae. Even with these limitations, the orthosurgical protocol adopted in this study fulfilled optimally the clinical needs and patient's expectations.

DISCUSSION

The treatment of the orthodontic sequelae caused by CCD is long and requires a multidisciplinary approach lasting up to growth cessation. There are many treatment modalities to correct the dentoskeletal discrepancies of CCD patients and the choice for one protocol may depend on individual health, facial aspects, and patients expectations. Nowadays, the orthopedic and orthodontic treatments combined with orthognathic surgery are the most common approaches.[7,16]

According to the literature, 4 treatment approaches for CCD patients have been proposed: Toronto-Melbourne, Belfast-Hamburgh, Jerusalem, and Bronx.[6,14] In this clinical report it was adopted the Bronx protocol and initially all deciduous and supernumerary teeth were removed and in the second intervention all impacted teeth were exposed for their traction.[6,15]

The Bronx Protocol, as well as the Jerusalem Protocol, adopt 2 surgical times. However, in Jerusalem Protocol the second intervention must happen at the age of 13. In this case it was no longer possible. Furthermore, it has fewer surgical times than Toronto-Melbourne Protocol as the deciduous teeth are all extracted at the same time, instead of the different interventions for serial extraction.

On the other hand, the Bronx protocol is disadvantageous in comparison with Belfast-Hamburgh because it has an extra surgical time. Nevertheless, due to the patient's advanced age at the time of the initial consultation, the Bronx protocol was chosen due to patient's age at the initial consultation. It was expected that after deciduous and supernumerary teeth removal, impacted permanent teeth could erupt spontaneously. Unfortunately, this did not happen and a second surgical intervention was required.

The only modification in this case consisted of the attempt to push forward the maxilla. This procedure has been widely reported in the literature as the ideal approach to correct the negative sagittal skeletal discrepancy. In this patient, the anterior crossbite and the cephalometric measurements defining the Class III malocclusion enabled this treatment adoption (SNA: 79.3^{o}; SNB: 81.8^{o}; ANB: -2.5^{o}).[6,17,19–21]

Baccetti et al,[18] in a randomized clinical trial of patients treated later (initial mean age of 10 years and 3 months), concluded that treatment performed in older patients did not significantly change the individual facial profile. Despite the age of the patient, the orthopedic approach was justified by the fact that CCD patients may present late cranial suture ossification.[1] But, unfortunately this approach was not well succeeded and significant improvement in the sagittal maxillomandibular relationship was not observed 1 year after therapy.

Concomitant to the interceptive procedure, the management of dental eruption and extraction of deciduous and supernumerary teeth took part. Several authors indicate this therapeutic procedure as the best way to conduct CCD patients' treatment to achieve the best functional and esthetic postadolescence outcomes.[1,6,10,11,13,22]

The moment for the orthognathic surgery may depend on functional problems, psychosocial impairment, and growth pattern. Wolford et al[24] reported that when performed early, the final result of the skeletal correction in young patients with maxillary hypoplasia might become unpredictable due to facial growth reminiscent.[15,24] Based on that, the surgery approach was post-pone until the end of growth in this patient.

After the follow-up period and when the craniofacial growth was finished, the corrective phase was initiated. This phase aimed at providing space for the permanent teeth and to decompensate teeth for the surgical intervention. Park et al[15] reported a similar approach regarding the treatment of a CCD individual. In that case a modified Bronx technique was applied and the orthognathic surgery was also necessary after the corrective orthodontic phase to achieve a satisfactory occlusion and the best facial profile for the patient.

Based on the severe sagittal discrepancy with facial impairment and considering the patient's complaint on the lack of dental exposure while smiling, the treatment plan involved the maxilla exclusively. The treatment proposed consisted of maxillary advancement, with anterior bone extrusion, clockwise rotation, and quadrangular Le Fort I osteotomy with a bone step in the zygomatic buttress. Dann et al[23] showed that the Le Fort I osteotomy can provide predictable and stable good results regarding facial middle third. Besides the better contour of the middle third, the forward maxillary advancement still results in better support for the upper lip. Daskalogiannakis et al[7] reported occlusal and facial good results applying the same technique in the treatment of a CCD patient.

Bone grafting is necessary in around 25% of patients and both autogenous bone and bone substitutes have been reported as options in the literature. The granular nature of the material facilitated its application between the bone segments, and we subsequently observed stabilization of the biomaterial and newly formed bone, preventing pseudoarthrosis and occlusal instability. In addition, the properties exhibited make Bio-oss a valid alternative to autogenous grafting, preventing the added morbidity of a donor surgical site.[25]

The bone substitute used in this study (Bio Oss Collagen, Geistlich Pharma AG) offers several advantages. Its composition consists of mineralized granules incorporated into collagen, which means that BOC can be molded and modeled into the shape desired. It adheres to bone structures when wetted and can easily be fitted to the surgical site of interest. The granules are kept in place by the collagen structure. As demonstrated by the clinical and tomographic results, the bone substitute chosen promoted and induced effective bone formation in the defect.[26,27]

CONCLUSION

The sagittal and vertical discrepancies correction performed in this report shows that complex cases involving genetic factors must be treated with well-defined protocols. Yet these protocols may range from an interceptive to a corrective phase, and usually may be combined with orthognathic surgeries in CCD patients. This multidisciplinary approach results in balanced occlusal and facial outcomes. Additionally, it provides to the patient a socially acceptable appearance that overcomes the main complaints reported in the beginning of treatment. The use of bone substitutes was a safe protocol to enable stability of the maxilla.

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