Two-phase orthodontic treatment of a complex malocclusion: Giving up efficiency in favor of effectiveness, quality of life, and functional rehabilitation?

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The interceptive orthodontic treatment of patients with complex dentofacial abnormalities is frequently inefficient and produces less than ideal outcomes. Therefore, postponing therapy to a single-phase surgical-orthodontic approach might be considered a reasonable option. However, other relevant aspects of the patient's quality of life, such as possible psychosocial problems and functional impairments, should also be considered before deciding whether to intercept a severe dentofacial malocclusion while the patient is still growing, or wait and treat later. This case report describes the nonsurgical treatment of a young patient with a severe Class III open-bite malocclusion associated with a cervical cystic lymphangioma. Despite the poor interceptive therapy prognosis, a 2-phase approach was effective. A reflection about giving up efficiency in favor of effectiveness, functional rehabilitation, and the patient's quality of life is included. (Am J Orthod Dentofacial Orthop 2013;143:547-58)

The treatment of patients with complex dentofacial abnormalities is always a great challenge to orthodontists.[1] Intercepting these problems early frequently lowers the efficiency of treatment because of an excessive treatment time, often without improving the efficacy.[2] When orthodontists examine children with malocclusions severely affecting at least 2 planes of space, postponing therapy to a single-phase surgical-orthodontic approach might be considered a reasonable treatment option.[3,4] However, as an essential member of any craniofacial team, an orthodontist must consider other relevant aspects of the patient's quality of life, such as possible psychosocial problems and functional impairments.[5] These variables should also be considered before deciding whether to prescribe early treatment for a severe dentofacial malocclusion.

Several environmental factors can be associated with the development of a complex skeletal malocclusion. The macroglossia and the anterior tongue posture caused by a cervical cystic lymphangioma (cystic hygroma) exemplify some of these etiologic determinants.[6] Children with neoplastic lesions of the tongue usually develop major myofunctional imbalances, resulting in significantly abnormal craniofacial growth patterns. The aim of this article was to report the treatment of a patient with a severe malocclusion caused by a cervical cystic lymphangioma in which the interceptive phase was effective but not efficient. A reflection about giving up efficiency in favor of effectiveness, functional rehabilitation, and patient's quality of life is included.

DIAGNOSIS AND ETIOLOGY

A 3-year-old girl was referred to an orthodontic office, and her mother's main concern was "the need to do something to hold the tongue." The child was born with a cervical cystic lymphangioma, and her tongue was visibly enlarged already in her first year of life. Posterior cervical masses were fingerlike processes extending bilaterally into the tongue. The child had neck resection of the lesions when she was 2 months

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old. Complete surgical excision of the cystic lesions was not possible because of the marked lymphangiomatous infiltration in the muscle fibers of the tongue. When she was 2 years old, a wedge glossectomy was performed to decrease the tongue's size.

Clinically, she had a straight profile, severe lip incompetence, and a striking tongue protrusion (Fig 1). The intraoral examination showed a full-step mesiocclusion with 10 mm of negative overjet, a posterior cross-bite tendency, and a severe anterior open bite (8 mm) (Fig 2). The only occlusal contact was between the left second deciduous molars, and the erythematous lesions on the tongue were still quite evident (Fig 1). The cephalometric evaluation (Fig 3) with the Sassouni[7] archeal analysis showed a skeletal Class III relationship despite the borderline ANB angle (−0.6°). There was also a severe vertical discrepancy confirmed by the steep mandibular plane angle (FMA, 30.6°; SN-GoGn, 42.7°) (Table). The panoramic radiograph showed, at the age of 3, incomplete root formation of the maxillary second deciduous molars; this was completed by the age of 5 years (Fig 3). The child's father had a Class III malocclusion, and her mother also showed signs of a Class III profile.

TREATMENT OBJECTIVES

After the orthodontic team carefully reviewed the patient's initial records, her parents received a detailed explanation about the poor prognosis of any interceptive orthodontic approach. Heredity, severity of the malocclusion, and the difficulty in controlling the tongue habit since the tumor could not be fully removed were mentioned as significant complicating factors. Because the efficacy and efficiency of an early treatment phase had an unfavorable prognosis, a surgical-orthodontic intervention at the end of growth was the first treatment option presented. However, the patient's mother was a dentist, and she adamantly refused postponing any interceptive intervention. Therefore, an alternative treatment plan was formulated, and its objectives were to try to (1) control the sagittal, vertical, and transverse dentofacial abnormal growth tendencies; (2) correct both molar and canine relationships to Class I relationships; (3) obtain normal incisor relationships; (4) develop a more adequate masticatory function; (5) improve lip closure and facial esthetics; and (6) develop a better muscular balance to increase the chances of obtaining stable results.

TREATMENT ALTERNATIVES

Two treatment plans were presented to the patient's parents. The first option was to wait until craniofacial growth was completed and perform surgical-orthodontic treatment. However, the patient's mother expressed her deep concerns about the psychosocial problems her daughter had been facing. Consequently, she refused the first treatment plan proposed and insisted that some interceptive therapy be performed to try to improve her daughter's oral function and facial harmony. Therefore, an alternative 2-phase treatment plan was developed. It consisted of a first phase of dentofacial orthopedics to try to induce a more harmonious growth pattern and improve function and facial esthetics. The second phase of treatment would consist of fixed orthodontic appliances to correct the remaining dentoalveolar discrepancies. The parents were carefully informed that the first phase would not necessarily eliminate the need for orthognathic surgery during the second phase of treatment. They were also aware that the stability of the results would be a challenge. After thorough deliberation of the advantages and disadvantages of both treatment options, the parents chose the 2-phase therapy and signed an informed consent authorizing the treatment of their daughter.

TREATMENT PROGRESS

Phase 1 treatment started when the panoramic radiograph showed complete root formation of the maxillary second deciduous molars. The patient was 5 years old, and her behavior in the dental chair was acceptable for performing orthodontic procedures. Rapid palatal expansion was carried out with a hyrax expander; immediately after the last screw activation, maxillary protraction with a Delaire facemask began. Five hundred centinewtons of force per side were applied, and 12 to 16 hours of daily wear were requested. However, the patient's compliance was poor; after 6 months of treatment, the facemask therapy was discontinued.

Our failure to obtain adequate cooperation with facemask wear led us to raise questions about the success of the interceptive treatment. However, the patient's mother was satisfied with the improvement of both the posterior and anterior open bites, as well as the better canine relationship. Therefore, she once again insisted that another attempt to control the poor growth tendencies should be made. The patient then received a chincup with vertical-pull traction for nighttime wear to try to control the excessive vertical growth pattern. In addition, a fixed acrylic tongue crib was inserted in the maxillary arch (Fig 4). The acrylic crib was chosen because of the risk of injuring the child's tongue lesions if a conventional wire-only crib was used. After the crib was

Fig 1. Pretreatment extraoral and intraoral photographs.

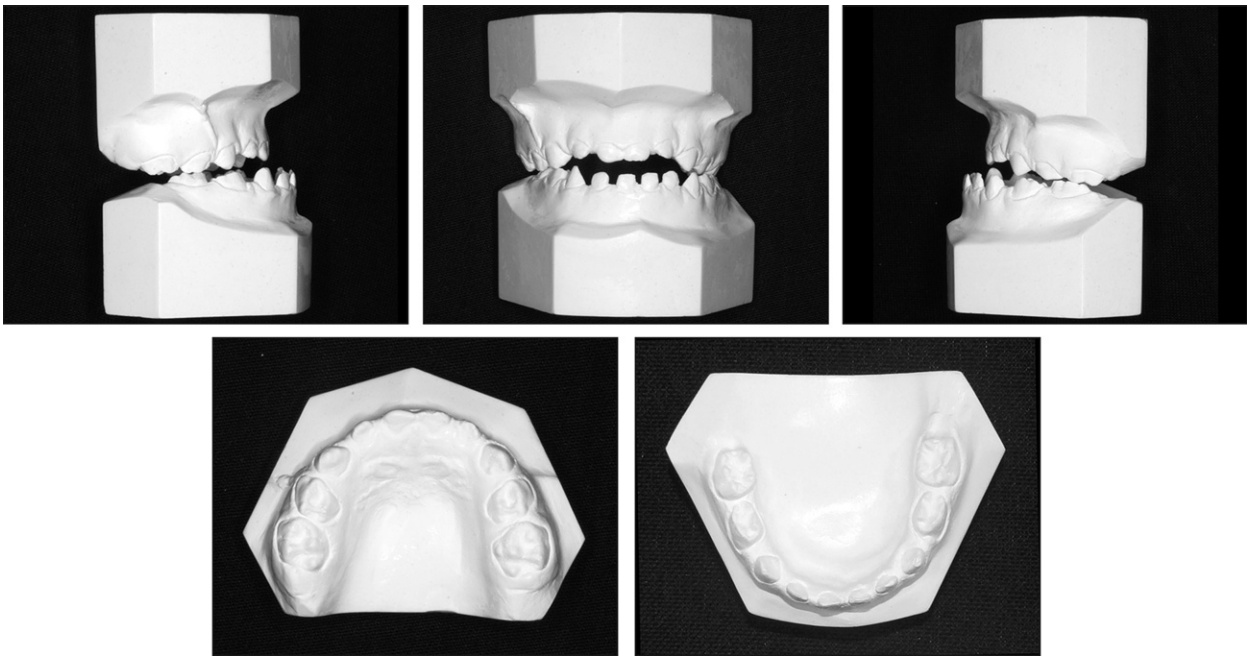

Fig 2. Pretreatment study models.

inserted, the patient could not thrust her tongue outside her mouth (Fig 4). Therefore, she developed a better tongue posture, improved her speech pattern, and her social behavior. She also became a more cooperative patient, increasing her adherence to the interceptive orthodontic treatment.

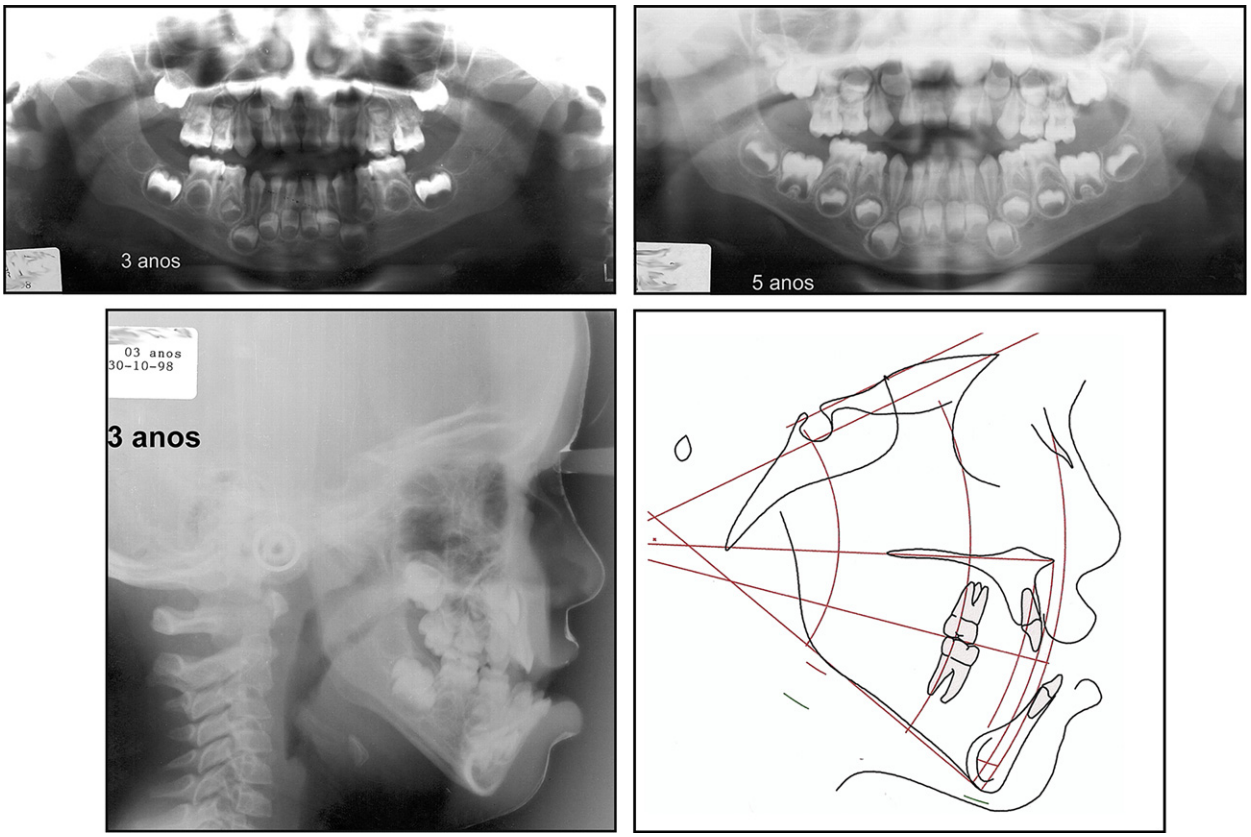

Fig 3. Pretreatment radiographs and tracing.

Table. Cephalometric measurements at pretreatment, after phase 1 of interceptive treatment, and after phase 2 of fixed appliance treatment

Measurement	*Pretreatment*	*After phase 1*	*After phase 2*
SNA (°)	81.7	79.7	79.8
SNB (°)	82.3	77.0	77.8
ANB (°)	0.7	2.7	2.8
SNGoGn (°)	42.7	45.2	44.9
ArGoGn (°)	145.3	138.9	134.2
NSGn (°)	68.8	74.9	74.6
FMA (°)	30.6	37.2	35.1
U1.NA (°)	10.9	24.9	24.0
U1-NA (mm)	1.1	8.3	5.9
L1.NB (°)	34.1	10.1	27.3
L1-NB (mm)	9.1	4.1	8.7
IMPA (°)	88.1	64.7	82.2
U1.L1 (°)	134.3	135.1	127.5

Progress records were taken 3 and 6 years after tongue crib and chincup insertion (Fig 5). They showed favorable growth between the maxilla and the mandible. After cautiously studying the progress records, we concluded that the remaining skeletal malocclusion could be camouflaged with fixed orthodontic appliances after the completion of her craniofacial growth. Follow-up consultations were scheduled bimonthly to check the status and adjust (if necessary) both the tongue crib and the chincup.

However, when the patient reached adolescence at 12 years of age (Figs 6, 7, 8, and 9), she refused to keep wearing the chincup and asked us to start the next phase of treatment with fixed appliances. She was at stage 3 of cervical vertebral maturation, and her hand-wrist radiograph showed that she was at the peak of her pubertal growth (Fig 8).[8] Maxillary and mandibular fixed appliances (0.022-in slot brackets) were bonded, leveling and alignment took place satisfactorily, and the mandibular arch-space discrepancy caused by the lingual inclination of the mandibular anterior segment was corrected (Figs 10 and 11). Vertical, triangular, quarter-inch, 4.5 oz elastics were used in the canine region bilaterally to improve intercuspation. Class III elastics (3/8 in, 6.5 oz) were used to maintain some growth-restraining forces when the 0.019 × 0.025-in archwires were in place. The overall second phase of treatment lasted 22 months, and debond was performed

Fig 4. Progress intraoral photographs immediately after maxillary protraction.

Fig 5. Progress intraoral photographs after 3 and 6 years of tongue crib and chincup therapy.

when the patient achieved stage 6 of cervical vertebral maturation (Fig 12). Fixed retention was bonded on both the mandibular (canine to canine) and the maxillary (first premolar to canine on each side) arches to minimize the chances of open-bite relapse. The patient was also instructed to wear a maxillary Essix retainer (type C+; DENTSPLY Raintree Essix, Sarasota, Fla) at night.

TREATMENT RESULTS

After 7 years of phase 1 treatment, the lip seal and tongue posture were significantly better, and the overall soft-tissue profile improvement was remarkable (Fig 6). The posterior open bite decreased, both maxillary and mandibular molars were in occlusion, overjet was overcorrected, and the anterior open bite was minimized (Figs 6 and 7). The patient was satisfied with both facial and dental treatment outcomes, and her mother reported that she became more confident and outgoing. Cephalometric superimpositions (Fig 9) showed that, after phase 1, Points A and B moved forward 2.5 and 3 mm, respectively, despite the ANB increase from 0.7° to 2.7° (Table). The mandibular plane angle rotated by 2.5° clockwise, and the palatal plane tipped 0.5° counterclockwise (Fig 9). The severity of the malocclusion was reduced after interceptive orthodontics, and the second phase of treatment was implemented to camouflage the remaining skeletal discrepancies and finish the correction of the malocclusion.

The phase 2 results showed adequate molar and canine relationships, normal overjet and overbite, and a substantial overall improvement of the occlusion (Figs 10 and 11). The posttreatment cephalometric analysis (Table) showed modest skeletal changes since the end of phase 1 in both sagittal (ANB, 2.7°-2.8°) and vertical (SN-GoGn, 45.2°-44.9°) dimensions. Conversely, significant dental modifications were registered. The interincisal angle decreased from 135.1° to 127.5°, and this change was primarily due to the proclination of the mandibular incisors (IMPA, 64.7°-82.2°; L1.NB, 10.1°-27.3°). The maxillary incisor inclination remained basically unchanged (U1.NA, 24.9°-24°). However, they were bodily retracted by 2.4 mm (U1-Na, 8.3-5.9 mm). The mandibular arch-space discrepancy was corrected with the forward bodily movement of the mandibular incisors (L1.NB, 4.1-8.7 mm). The panoramic radiograph confirmed adequate root parallelism, good overall alveolar bone height, and 3 unerupted third molars (Fig 12). Cephalometric superimpositions comparing the results of phases 1 and 2 showed insignificant facial growth (Fig 13) and good vertical control. They also showed that overbite and overjet corrections were achieved with tooth movement.

The patient was pleased with her smile, and the treatment highlights in her opinion were successful bite closure, good alignment of the maxillary anterior teeth, and the acquired capacity to hold her tongue inside her mouth. The results of this difficult and long treatment remained stable 18 months after treatment was completed (Fig 14).

Fig 6. Progress extraoral and intraoral photographs at the end of phase 1.

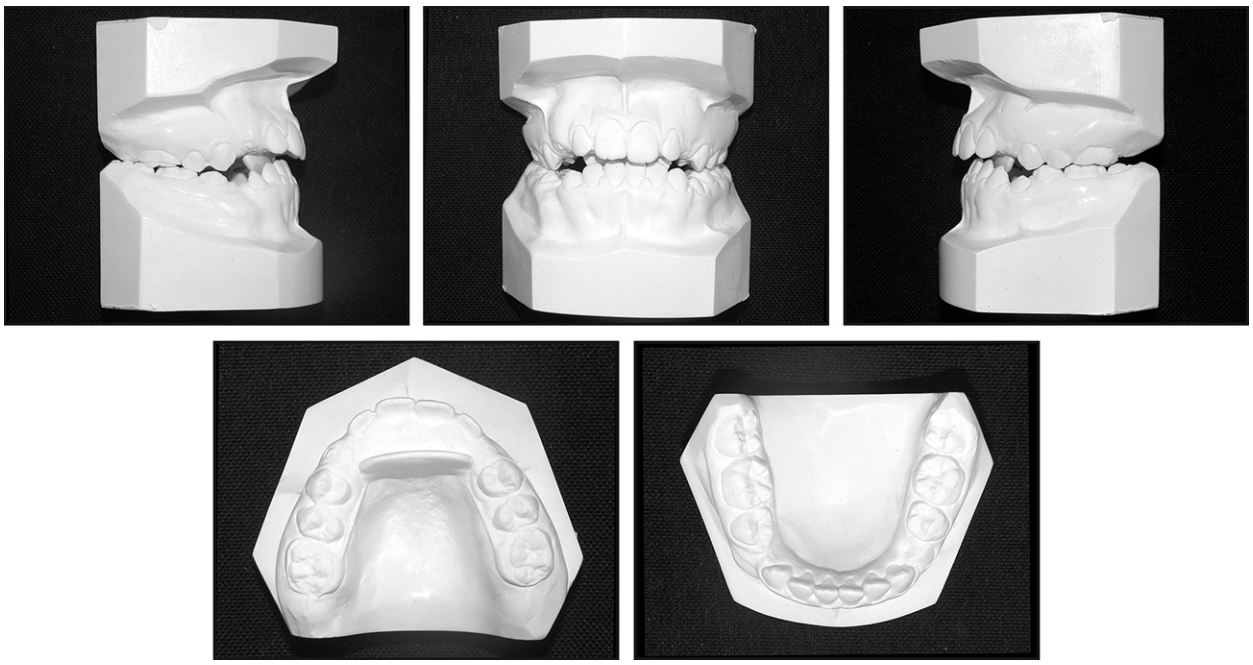

Fig 7. Progress study model at the end of phase 1.

DISCUSSION

The case presented here is only the second report of orthodontic treatment of a malocclusion caused by cystic hygroma. The other case was reported 4 decades ago.[6] The patients in both reports had similar skeletal open-bite features that might indicate similar growth patterns because of their significant macroglossia. Congenital lymphatic malformations are observed in

Fig 8. Progress radiographs.

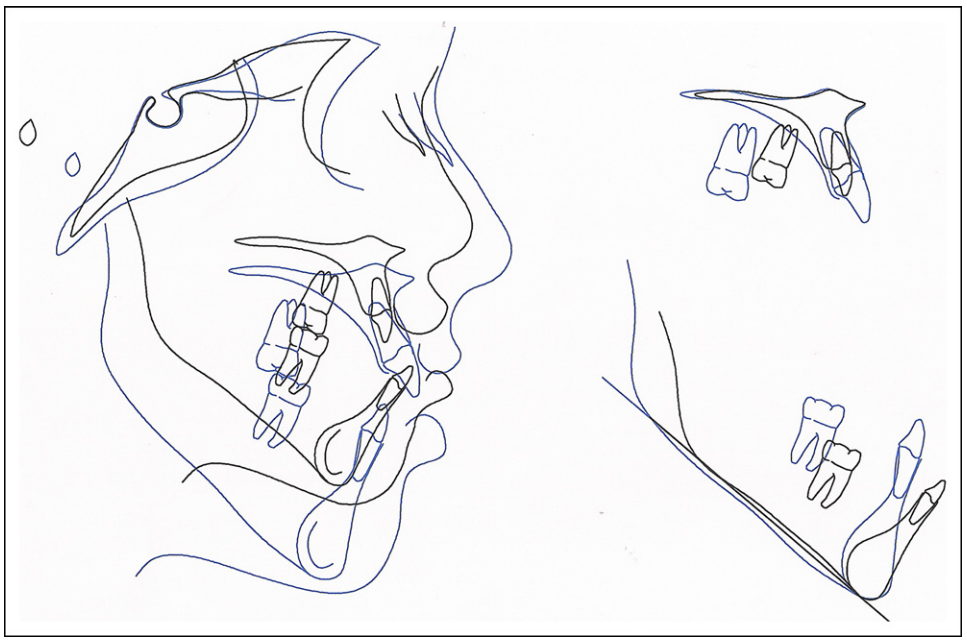

Fig 9. Superimposition of pretreatment (*black*) and end of phase 1 (*blue*) cephalometric tracings.

Fig 10. Posttreatment extraoral and intraoral photographs.

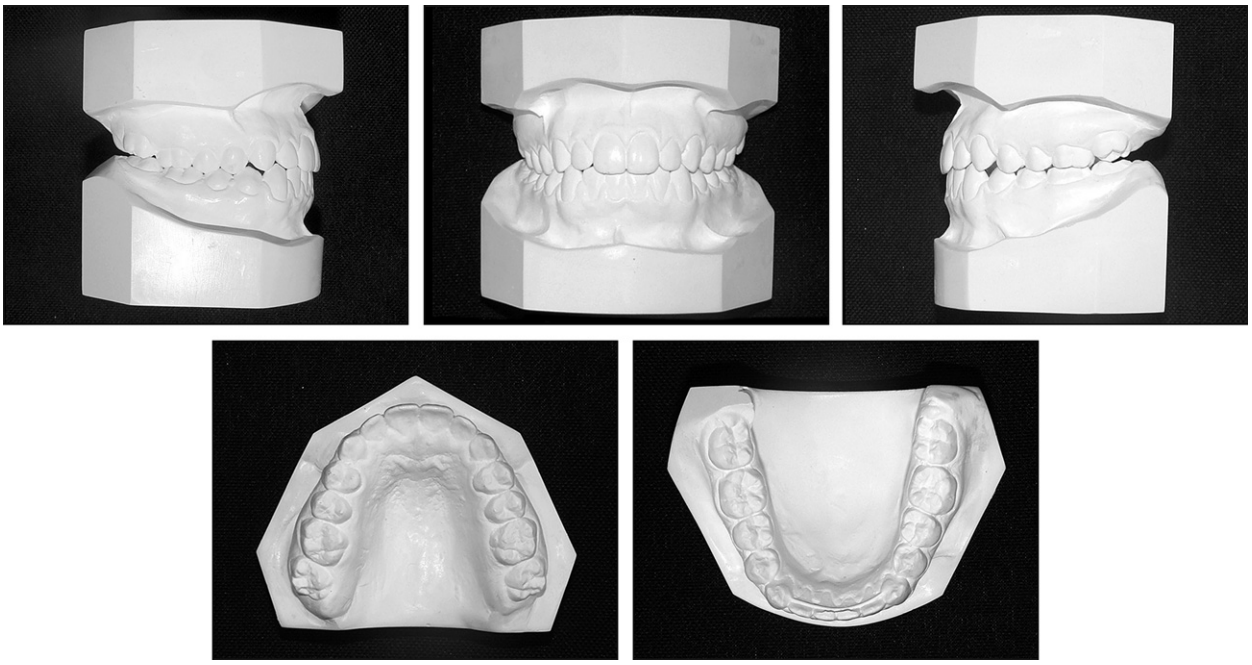

Fig 11. Posttreatment study models.

1 of every 6000 life births[9]; although this is not as frequent as other craniofacial deformities, it is reasonable to assume that some orthodontists might need to treat a patient with a complex malocclusion associated with tongue enlargement and malfunction caused by cystic hygroma.[10]

When these pathologies develop after the thirtieth week of gestation or after birth, they usually are not

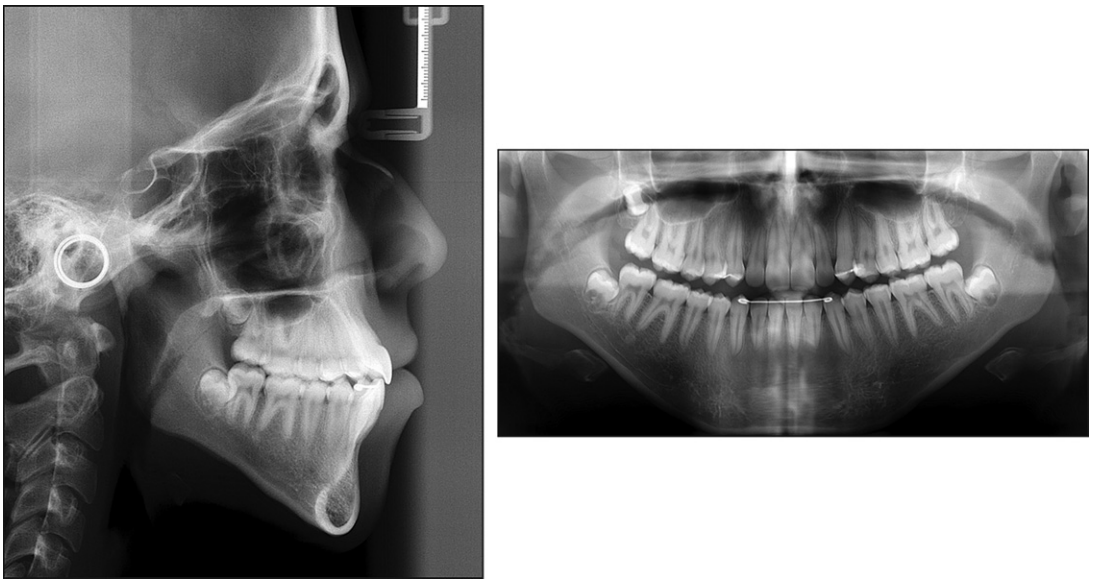

Fig 12. Posttreatment radiographs.

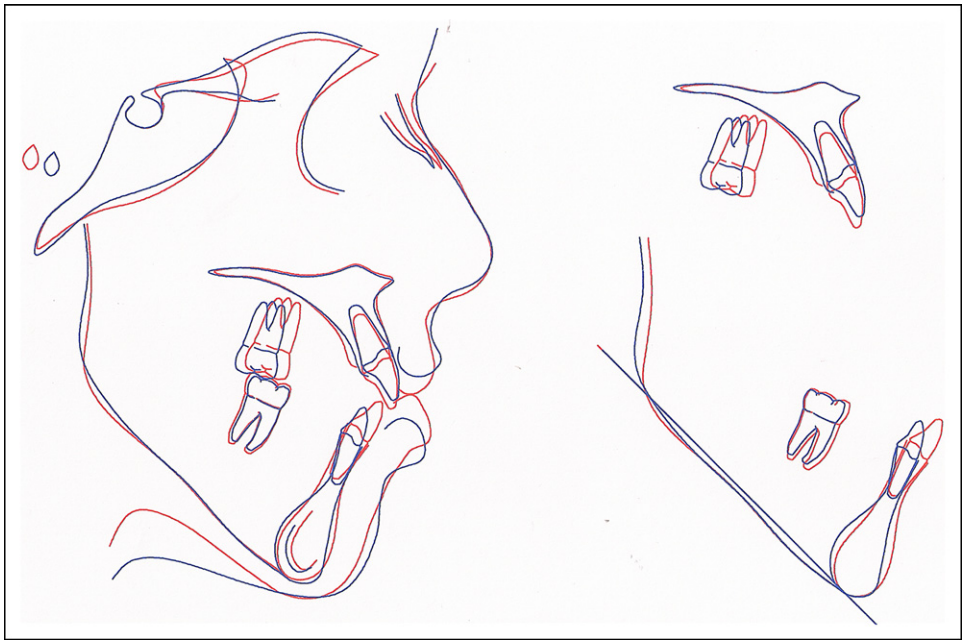

Fig 13. Superimposition of end of phase 1 (*blue*) and end of phase 2 (*red*) cephalometric tracings.

associated with chromosome abnormalities.[11] However, genetic counseling should be considered when evaluating the risks for future pregnancies. The surgical management of these pathologies might be limited by the invasiveness of the procedure or the complex anatomy of this region. Recently, new sclerotherapy methods have been proposed, when complete surgical removal of the lesions is not possible.[12] This approach was not used in our patient because, at that time, the sclerotherapy protocols were deemed too risky by her physicians. Another concern when treating patients with this pathology is its high recurrence rate.[12] However, our patient has shown only minor signs of relapse of the tumor lesions, which have been controlled with corticosteroid injections.

Fig 14. Photographs after 18 months of retention.

This case report had a rare outcome. The parental Class III heredity and the severity of the malocclusion, especially the macroglossia that could not be anatomically or functionally controlled, contributed to a poor prognosis for any interceptive treatment considered.[13,14] A classic surgical-orthodontic approach after growth completion would probably be the first treatment plan for most orthodontists. It was also the first option we presented to the patient's parents because it would certainly be a more efficient way to treat such a severe malocclusion.

However, the patient's mother was a dentist and refused this approach. She actually insisted that immediate interception of her daughter's inadequate tongue posture should be tried because of psychological concerns. There is considerable evidence to support the benefits of early orthodontic treatment to correct malocclusions and parafunctional habits as well as to improve oral function in a growing child, thus contributing to better skeletal and occlusal development.[15-18] Conversely, other authors have suggested that 2-phase therapy should not be implemented when the interceptive phase is inefficient or its outcomes are uncertain.[14,19] When deciding on the optimal timing to start treatment in a young patient, an orthodontist must consider the effectiveness and efficiency of all treatment options.[20] However, focusing this decision only on treatment efficiency might underestimate the positive effects of early orthodontic treatment on the patient's oral function and quality of life when the malocclusion is severe.

Facial appearance is a fundamental factor in determining interpersonal relationships.[21] Thus, the early treatment of complex malocclusions that compromise dental and facial esthetics can have important psychosocial implications for some patients. Children with significant dentofacial deformities are often perceived as less attractive by their peers, and differences of behavior toward attractive and unattractive people have been well documented.[22] Choosing to postpone orthodontic treatment of severe skeletal malocclusions until growth is completed might compromise some patients' self-esteem.[5] The phase 1 orthodontic treatment described here allowed the patient to pass through childhood and adolescence with good self-esteem, and attend a socially demanding high school with no major psychological problems related to the severity of her malocclusion or the neoplastic lesions in her tongue. Delaying her treatment until adulthood could have exacerbated problems related to speech pathology, temporomandibular joint disorders, masticatory function impairment, and inadequate tongue-resting posture.[23] We recognize that efficiency was compromised with a much longer than average treatment, but we gave up efficiency in favor of effectiveness, improving the patient's quality of life, and achieving functional rehabilitation.

Another topic that must be addressed is the unexpected efficacy of the alternative mechanics used during the first phase of treatment of our patient. The stability of early intervention on a skeletal Class III

open-bite malocclusion is a concern for every orthodontist. The application of cephalometric predictive parameters to estimate the potential effectiveness of interceptive orthodontics in Class III patients indicated that the chances of failure with facemask maxillary protraction were high in our patient.[24,25] The severe maxillomandibular discrepancy observed in her pretreatment facial morphology, the increased vertical dimension, and the prognathic mandible were unfavorable factors for long-term treatment stability.[26] Also, we could not achieve overcorrection of the Class III skeletal malocclusion as recommended in the literature to improve stability.[27-29] So, how do we explain the facial and occlusal results obtained as well as the good stability at 18 months posttreatment?

Since the patient did not cooperate with the facemask maxillary protraction during the first months of treatment, the use of reverse-pull headgear was postponed and ultimately was not even necessary. Therefore, the good maxillary advancement observed cannot be credited to conventional facemask therapy. We hypothesize that the combination of the fixed maxillary acrylic crib and the chincup therapy used for 7 continuous years had a major role in the final treatment outcome. The improvement in both the ANB and SNA angles might be due to the forward force that her powerful tongue exerted on the crib, forcing the maxillary dentoalveolar complex anteriorly. This continuous and intense force might have stimulated its growth in that direction, acting as an intraoral stimulus for maxillary protraction during all those years. The significant mandibular incisor lingual inclination registered at the end of phase 1 might indicate that the crib functioned as an "inverted lip bumper," inhibiting the tongue to rest on the lingual surface of the anterior mandibular teeth. Similarly, the tongue did not touch the anterior portion of the mandible and therefore might have performed as a mandibular "deactivator." Both assumptions could help to explain the 4° decrease in the SNB angle from pretreatment to the end of phase 1.

Several studies have shown that a Class III growth pattern does not change into a Class I pattern after facemask maxillary protraction.[30] This might be due to the relatively short duration of facemask therapy[14] and the fact that the suggested ideal timing for this orthopedic approach is not coincident with the pubertal growth spurt.[30] Therefore, stability is a concern when treating children with a skeletal Class III relationship. The long-term combined use of tongue crib and chincup for this patient might have been the decisive factors that contributed to the good sagittal stability 18 months after treatment, corroborating a previous report.[31] However, after the removal of both crib and chincup, the child still had a great growth potential (cervical vertebral stage 3). The use of fixed appliances with Class III elastics might have added an orthopedic growth control up to the end of phase 2, when she was already at the end of her mandibular growth (cervical vertebral stage 6), and this should not be overlooked.

The treatment and long-term stability of anterior open bites have frustrated numerous orthodontists for many years.[32] Previous reports have shown that several open bites corrected with either dentofacial orthopedics, orthodontic tooth movement, or orthognathic surgery can relapse after treatment.[29-35] According to Denison et al,[33] although dental and skeletal malocclusions can be adequately corrected, the role of orofacial musculature as an etiologic factor must also be addressed during the orthodontic therapy. The stability of open-bite corrections might increase if the functional etiology is eliminated during treatment.[33] Clinical examinations of our patient throughout treatment showed a progressively better adaptation of her tongue in the improving intraoral environment. Such functional improvement probably contributed to the stability of the overall treatment results, especially the open bite. There is previous evidence to support that the use of tongue cribs can alter tongue posture and increase open-bite treatment stability.[32] The use of a sharp metal tongue spur was contraindicated for this child because of the risk of injuring her tumorous tongue lesions. Therefore, a polished acrylic crib was used and might have contributed to improving her tongue function. Despite the good treatment stability at 18 months posttreatment, long-term follow-ups are recommended, since there is still a risk of vertical relapse.[32-35]

Another procedure that might have contributed to the stability of the open-bite correction was the glossectomy performed when the patient was 2 years old. One centimeter of her tongue's anterior portion was removed; this was approximately a quarter of the tongue. During the initial consultation, her parents reported a significant improvement in her ability to taste foods after the glossectomy. Throughout the orthodontic treatment, the patient reported normal tasting abilities. A cautious and precise indication of glossectomy should be considered when treating this type of anomaly.

CONCLUSIONS

This case report illustrated the treatment of a patient with a complex dentoskeletal malocclusion and a poor interceptive therapy prognosis. Surgical-orthodontic correction after growth completion was the most obvious therapeutic choice. However, a long-term phase 1 treatment was implemented to try to improve the patient's masticatory function, speech abilities, and

quality of life. The treatment was effective but not efficient. An orthodontist should always try to implement efficient clinical approaches. However, when facing severe dentoskeletal malocclusions in children, the decision about whether to treat such problems at an early age should be made along with the family, considering not only efficiency, but also the potential benefits to the patient's quality of life.

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