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Surgically Assisted Maxillary Expansion and Orthognathic Surgery in the Treatment of Hypoplastic Maxilla: An Ortho-Surgical Case Report

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Abstract: Skeletal Class III malocclusion, when associated with a moderate to severe discrepancy, compromises not only occlusal and masticatory function but also facial aesthetics, often resulting in a concave profile. In such cases, orthognathic surgery is the treatment of choice. In addition to the maxillary deficiency in the anteroposterior direction, transverse impairment is also common, leading to unilateral or bilateral posterior crossbite. In adult patients, due to skeletal maturation and ossification of the midpalatal suture, Surgically Assisted Rapid Maxillary Expansion (SARME) becomes an effective and indicated alternative. This paper aims to report the clinical case of a young female patient with Class III dentoskeletal deformity associated with maxillary atresia, treated through an orthodontic-surgical approach involving SARME followed by orthognathic surgery. Despite the complexity of the treatment, which included two surgical procedures, it was completed in 20 months—shorter than the average time reported in the literature. This outcome is attributed to the patient's high adherence to treatment and the coordinated effort of a multidisciplinary team.

Keywords: Orthognathic surgery; Maxillary expansion; Angle Class III malocclusion.

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1. Introduction

Primary neuroendocrine carcinomas of the mediastinum are exceedingly rare, typically associated with the thymus and paraganglionic structures adjacent to major mediastinal vessels. In less common cases, these tumors may also affect retroperitoneal structures, the inferior vena cava, the presacral region, the parathyroid gland, the ovary, as well as the gastrointestinal and biliary systems [1–3]. The etiology of small cell neuroendocrine carcinoma of the mediastinum (SCNCM) remains uncertain, with two main hypotheses: one suggests that the tumor arises from ectopic tissue due to aberrant embryonic migration, while the other proposes an origin from a teratomatous component [2].

The estimated incidence of SCNCM is approximately 1 case per 50 million individuals annually, with no sex predilection, and a mean patient age of around 58 years [4]. Clinically, symptoms include cough, dyspnea, dysphagia, weight loss, fatigue, and fever [1, 3]. For differential diagnosis, it is essential to consider Ewing sarcoma, primitive neuroectodermal tumor (PNET), lymphoma, neuroblastoma, and rhabdomyosarcoma, as well as primary or metastatic basaloid carcinoma [2].

The mortality rate of SCNCM reaches 50% within two years, and only half of the patients show any response to chemotherapy. The estimated median survival is approximately 14 months. Given the severity and rarity of this neoplasm, early diagnosis is crucial, as neoadjuvant therapies may improve survival rates compared to surgical resection alone [3, 5, 6]. Historically, the management of SCNCM has been challenging; therapeutic interventions that initially demonstrate efficacy are frequently followed by tumor recurrence and metastases [1, 3, 5, 7].

Given the relevance of the early diagnosis of this malignant neoplasm, this case report shows histopathological findings from the biopsy at the time of diagnosis and necroscopic findings that confirm its primary presentation. Moreover, we discuss macroscopic and microscopic criteria and immunohistochemistry staining even as differential diagnoses.

2. Case Report

A 17-year-old melanoderm female patient was referred by an oral and maxillofacial surgeon to begin orthodontic preparation for surgical purposes. The patient presented complete orthodontic documentation, including panoramic radiograph, lateral cephalometric radiograph with cephalometric analysis, intraoral and extraoral photographs, and plaster study models. Her main complaints included: "narrow smile, large chin, open bite, and crooked teeth," in addition to episodes of muscle pain associated with the masticatory muscles. She had no prior history of orthodontic treatment or systemic comorbidities and was in good general health.

Clinical examination revealed a concave facial profile, skeletal maxillary atresia with palatal/lingual inclination of the posterior teeth, dolichofacial pattern, facial asymmetry with mandibular deviation to the left, moderate dental crowding in the upper arch and mild in the lower arch, bilateral Class III molar and canine relationships, anterior and posterior crossbite, and anterior open bite (Figures 1A to 1F). The panoramic radiograph showed normal aspects except for a radiolucent image suggestive of a root remnant from the extracted lower left first molar (tooth 36), and the presence of a semi-impacted lower left third molar (Figure 2).

Cephalometric analyses were performed according to USP, McNamara, and Ricketts parameters. Among the findings were slightly proclined upper incisors relative to the cranial base (1.NA = 27.25°) and lingually inclined lower incisors (1.NB = 22.08°). A marked divergence between the skeletal bases was also observed (FMA = 32.98°; S-N^Gn = 73.23°), as well as a maxillomandibular discrepancy compatible with mandibular protrusion (ANB = -2.09°) and maxillary retrusion (N-Perp A = -1.94 mm). The sagittal and vertical relationship of the incisors was altered, with a negative overjet of -4.87 mm and an overbite of -3.50 mm (Figures 3A and 3B).

During the manipulation of the plaster models mounted in Class I occlusion—simulating the surgical movements required to correct the skeletal Class III pattern—occlusal interferences and persistence of posterior crossbite were identified. Given this condition, Surgically Assisted Rapid Maxillary Expansion (SARME) was indicated, along with the extraction of the lower left third molar (tooth 38), prior to orthognathic surgery. A Hyrax-type expander was placed on the upper arch, and the patient was referred to the surgeon for the surgical procedure.

Seven days after SARME, activation of the appliance was initiated with 1/4 turn twice a day, resulting in an expansion rate of 0.5 mm per day. After full activation of the expansion screw (11 mm), the device was locked and maintained in position as a retainer for a period of six months.

Concurrently, alignment and leveling of the lower arch were initiated using a fixed metallic orthodontic appliance, Roth prescription with 0.022" slot, following this archwire sequence: 0.014", 0.016", and 0.018" Nickel-Titanium wires, followed by 0.018", 0.020", 0.017x0.025", and 0.019x0.025" stainless steel wires. During this phase, space opening for

the missing tooth 36 was achieved using an open coil spring, aiming for future rehabilitation with an osseointegrated dental implant.

Figure 1. A to F. Pre-treatment intraoral and extraoral photographs.

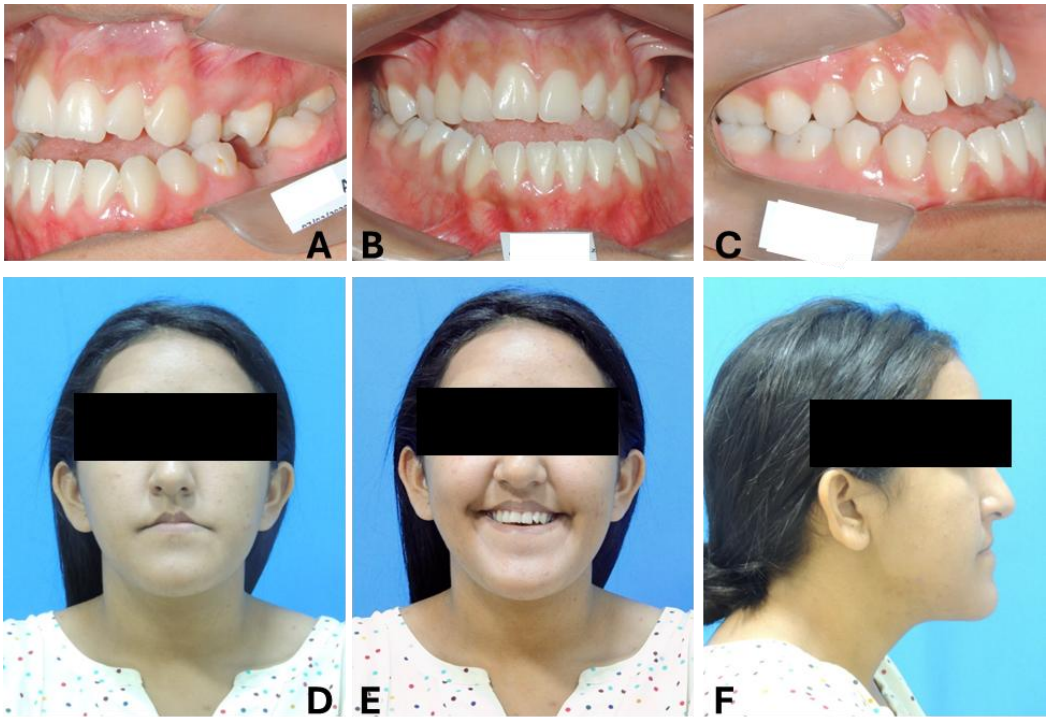
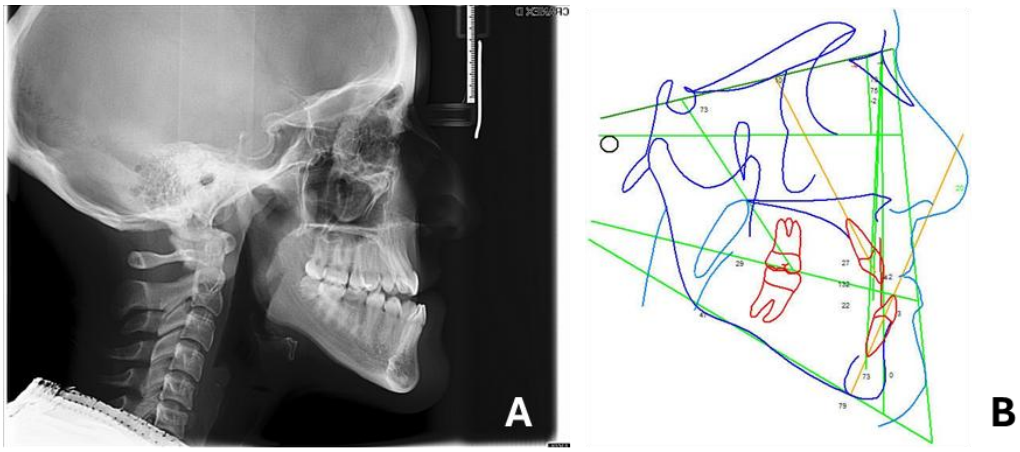


Figure 2. Pre-treatment panoramic radiograph.

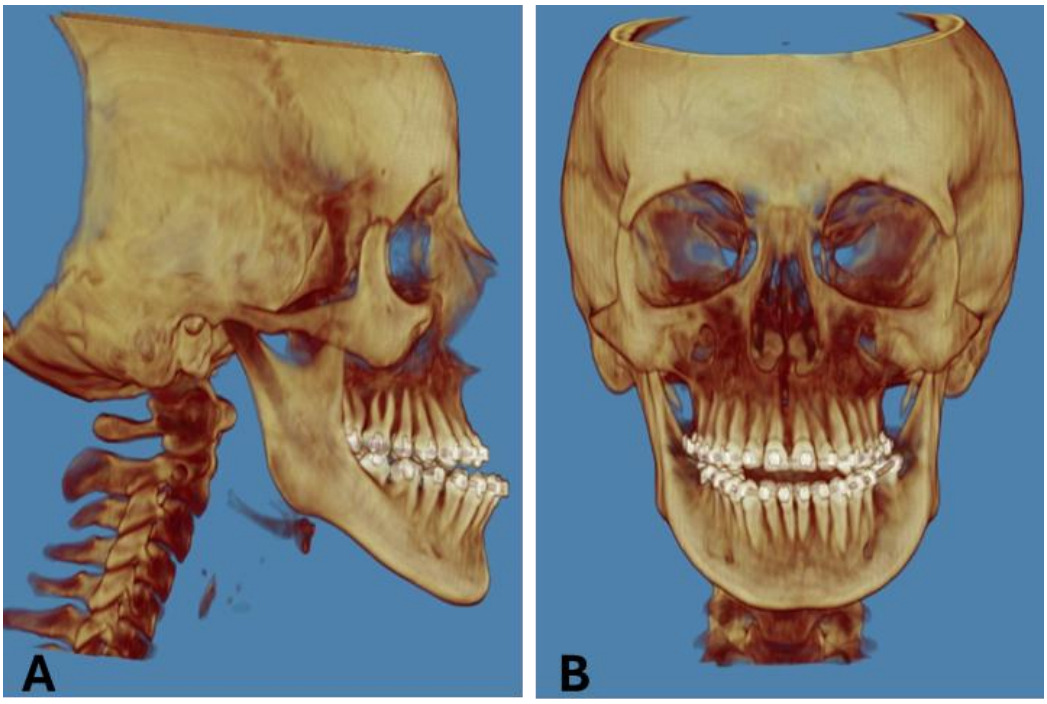
After the six-month retention period in the upper arch, the Hyrax appliance was removed, and alignment and leveling of the upper teeth were initiated, following the same wire progression used in the lower arch. After 12 months of orthodontic treatment, the patient was ready for surgical intervention, presenting with proper dental alignment and leveling, a leveled Spee curve, and a clearly defined maxillomandibular discrepancy. Surgical movement simulation on the plaster models demonstrated correct Class I occlusion, adequate overjet and overbite, coincidence of upper and lower dental midlines, a favorable transverse relationship, and absence of occlusal interferences. Interproximal hooks were installed on the 0.019x0.025" stainless steel wire, along with individual metallic ligatures for intraoperative intermaxillary fixation. The patient was then referred to the surgeon.

Figure 3. Pre-treatment lateral cephalometric radiograph and tracing.



The surgical planning was based on cranial computed tomography (Figures 4A and 4B) and three-dimensional digital models processed using the Dolphin Imaging 11.95® software. The surgical plan included advancement of point A (maxilla) by +5.82 mm with clockwise rotation, retreatment of point B (mandible) by -2.08 mm with counterclockwise rotation, retreatment of the soft pogonion by -0.85 mm, and correction of the upper and lower midlines by 1.00 mm and 2.05 mm, respectively (Figures 5A and 5B). Based on the planned movements, an occlusal surgical guide was made using 3D printing in UV resin (Anycubic 405 nm®), with a FlashForge Hunter 3D® printer. Thirty days after the orthognathic surgery, the patient returned to the clinic to continue the orthodontic refinement phase (Figures 6A to 6C). Intermaxillary elastics were used, and finishing bends were made in the arches to promote final occlusal adjustments.

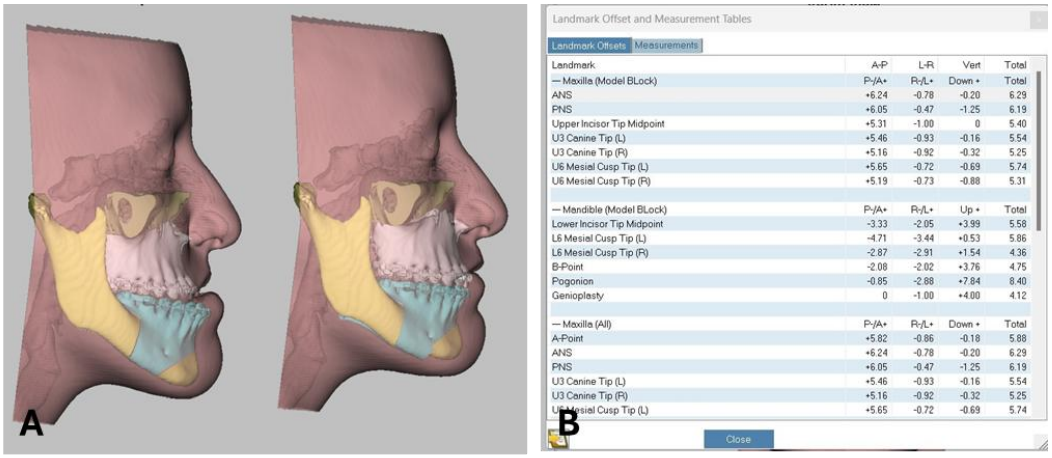
Figure 4. A and B. Preoperative Cranial CT Scan.



After achieving functional and aesthetically satisfactory occlusion, the fixed orthodontic appliance was removed. As retention, fixed 3x3 retainers were placed in the upper and lower arches, as well as a fixed retainer between teeth 35 and 37, acting as a space maintainer for the future installation of an implant in the region of tooth 36. Additionally,

a Hawley retainer with a palatal acrylic cover was made for nightly use, aiming for greater transverse stability during the post-maxillary expansion period. The prosthetic space for the rehabilitation of tooth 36 was properly prepared; however, the installation of the osseointegrated implant was postponed by the patient's personal decision. The orthodontic treatment was completed in a total of 20 months (Figures 7A to 7E).

Figure 5. A and B. Digital Planning Images in Dolphin Imaging 11.95® Software and Linear Movement Measurement Table.



Landmark	A-P	L-R	Vert	Total
— Maxilla (Model BLock)	P->A+	R->L+	Down +	Total
ANS	+6.24	-0.78	-0.20	6.29
PNS	+6.05	-0.47	-1.25	6.19
Upper Incisor Tip Midpoint	+5.31	-1.00	0	5.40
U3 Canine Tip (L)	+5.46	-0.93	-0.16	5.54
U3 Canine Tip (R)	+5.16	-0.92	-0.32	5.25
U6 Mesial Cusp Tip (L)	+5.65	-0.72	-0.69	5.74
U6 Mesial Cusp Tip (R)	+5.19	-0.73	-0.88	5.31
— Mandible (Model BLock)	P->A+	R->L+	Up +	Total
Lower Incisor Tip Midpoint	-3.33	-2.05	+3.99	5.58
L6 Mesial Cusp Tip (L)	-4.71	-3.44	+0.53	5.86
L6 Mesial Cusp Tip (R)	-2.87	-2.91	+1.54	4.36
B-Point	-2.08	-2.02	+3.76	4.75
Pogonion	-0.85	-2.88	+7.84	8.40
Genioplasty	0	-1.00	+4.00	4.12
— Maxilla (All)	P->A+	R->L+	Down +	Total
A-Point	+5.82	-0.86	-0.18	5.88
ANS	+6.24	-0.78	-0.20	6.29
PNS	+6.05	-0.47	-1.25	6.19
U3 Canine Tip (L)	+5.46	-0.93	-0.16	5.54
U3 Canine Tip (R)	+5.16	-0.92	-0.32	5.25
[illegible]sial Cusp Tip (L)	+5.65	-0.72	-0.69	5.74

Figure 6. A, B, and C. Post-operative intraoral photographs, showing the setup of the appliance with interproximal hooks and individual ligatures, and the new occlusion.

After the completion of the orthodontic-surgical treatment, new orthodontic documentation was requested, including photographic and radiographic records, to compare the pre- and post-operative bone and dental positions (Figures 8A and 8B). The cephalometric analysis confirmed the success of the dental decompensation, evidenced by the reduction of the vestibular inclination of the upper incisors from 27.25° to 16.671° and the correction of the lingual inclination of the lower incisors, which changed from 22.08° to 27.933° (Figures 9A and 9B).

The correction of the divergence between the bone bases was achieved through surgical movements, with a significant improvement in the FMA and S-N.Gn angles, demonstrating balance in the maxillomandibular relationships. To better visualize the skeletal repositioning, the pre- and post-operative cephalometric tracings were superimposed, highlighting the advancement of the maxilla and the retrusion associated with the counterclockwise rotation of the mandible. These modifications led to a significant improvement in the patient's facial profile, providing greater harmony and aesthetic balance. The superimposition of the pre- and post-operative cephalometric tracings allowed for a clear visualization of the changes promoted by the orthodontic and surgical interventions. The advancement of the maxilla, as well as the retrusion and counterclockwise rotation of the mandible, resulted in a significant improvement in the harmony and balance of the facial profile.

The changes in the bone bases were also quantified: the ANB angle, initially negative (-2.09°), reached values compatible with an adequate maxillomandibular relationship, while the FMA and S-N.Gn angles, which initially showed marked divergence, showed

significant improvement after surgery, compatible with a more balanced skeletal pattern (Figure 10 and Table 1).

Figure 7. Intraoral photographs (A to C) and extraoral photographs (D and E) taken at the final stage after the removal of the orthodontic appliance, showing an excellent facial profile, smile aesthetics, bilateral Class I Angle dental occlusion, coincident dental and facial midlines, and prepared space for rehabilitation of tooth 36.

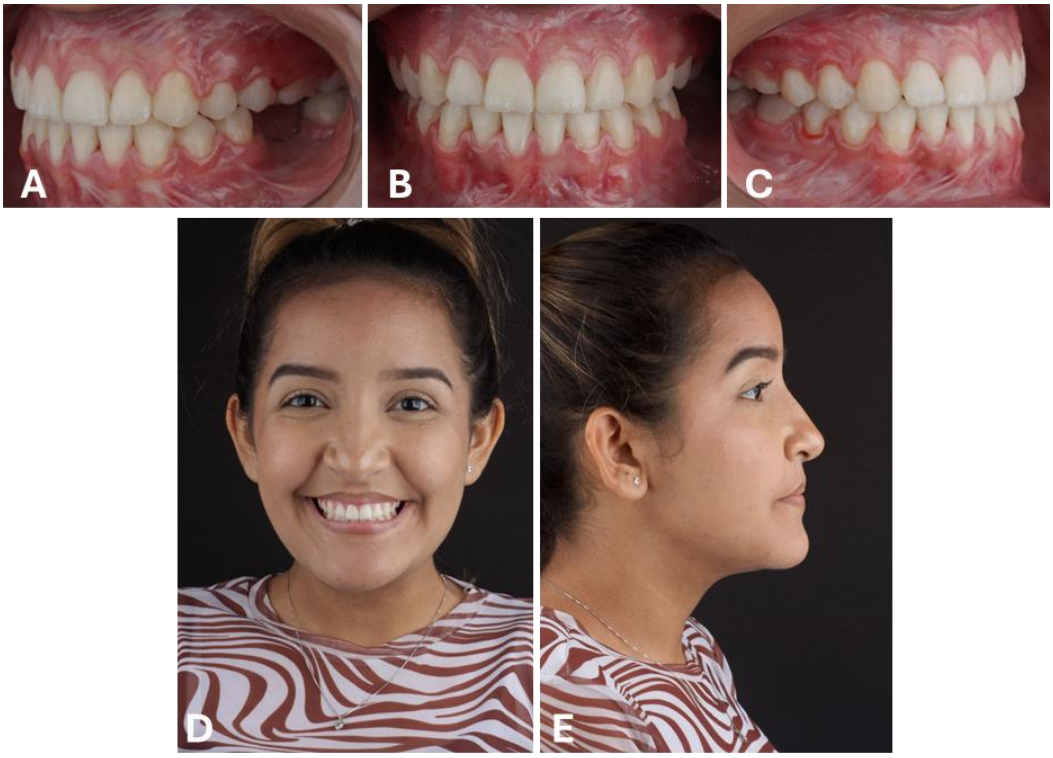
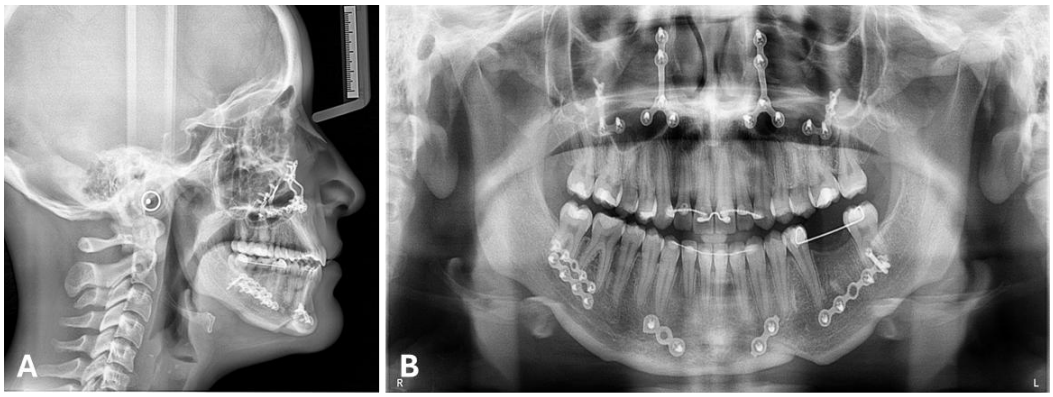


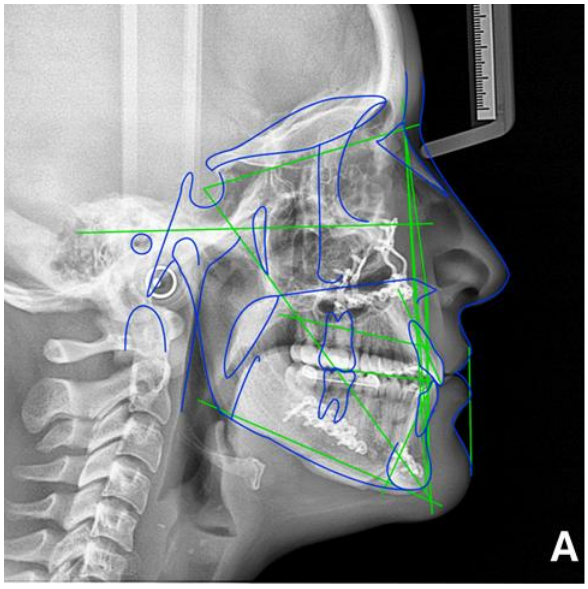
Figure 8. A and B. Post-operative lateral cephalometric radiograph and panoramic radiograph, showing fixation material and fixed retainers between 13 and 23, 33 and 43, and 35 and 37.



Photographs taken 12 months post-operatively demonstrate stable bilateral Angle Class I occlusion, coincident midlines, adequate alignment, absence of diastemas, and maintenance of space for rehabilitation of tooth 36 with adhesive fixed retainer (Figures 11A to 11C). The impact of the treatment on the patient's quality of life was assessed using the OHIP-14 (Oral Health Impact Profile-14) questionnaire. The OHIP-14 questionnaire consists of 14 items that address different dimensions of quality of life, such as pain, discomfort, functionality, self-image, and social well-being, related to oral health. Participants are asked about the frequency with which they experienced certain issues in the

past few weeks, with response options ranging from "never" to "always." The patient answered "never" to 12 questions and "almost never" to 2 questions, justifying the response due to the absence of tooth 36, which had not yet been rehabilitated for personal reasons.

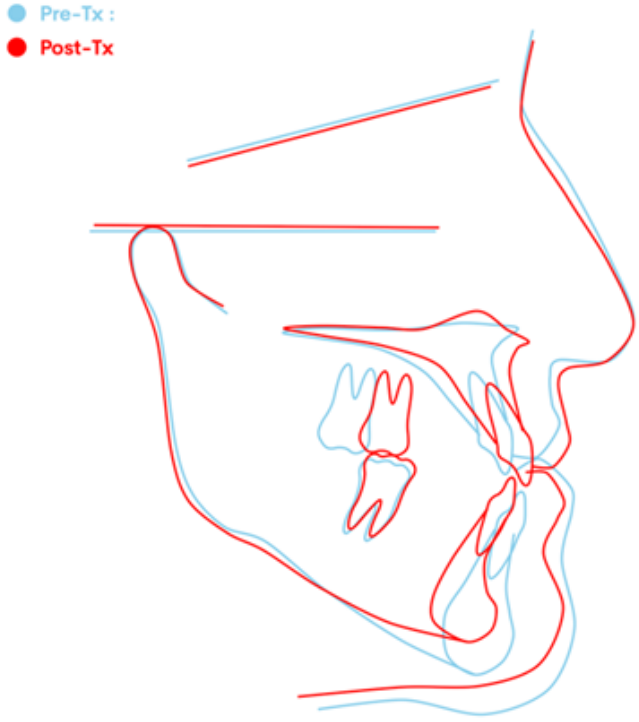
Figure 9. A and B. Final cephalometric tracing and table containing cephalometric measurements.



#	Descrição	Valor	Padrão
	Campo I		
1	(N-Pog).(Po-Orb)	92.611 gr	88.0 ± 1.0
2	N-A.Pog	7.236 gr	0.0 ± 2.0
3	S-N.A	81.201 gr	82.0 ± 2.0
4	S-N.B	76.905 gr	80.0 ± 2.0
5	A-N.B	4.296 gr	2.0 ± 2.0
6	S-N.D	74.476 gr	76.0
	Campo II		
7	S-N.Gn	70.305 gr	67.0
8	S-N.Ocl	23.739 gr	14.0
9	(S-N).(Go-Me)	39.948 gr	32.0
10	(Go-Gn).Ocl	13.732 gr	18.0
	Campo III		
11	Ângulo Interincisivo	131.1 gr	131.0
12	1/.NS	97.872 gr	103.0
13	1/-Órbita	0.755 mm	5.0
14	1/.NA	16.671 gr	22.0 ± 5.0
15	1/-NA	2.259 mm	4.0
16	/1.NB	27.933 gr	25.0 ± 4.0
17	/1-NB	4.383 mm	4.0
18	/1-NPog	3.443 mm	0.0
	Campo IV		
19	H.(N-B)	2.526 gr	10.5 ± 1.5
20	H-Nariz	12.58 mm	10.0 ± 1.0
21	Pog-NB	1.261 mm	-
22	Eminência Mentoniana	7.81 mm	7.0 ± 1.0
	Campo V		
23	FMIA	64.025 gr	68.0
24	FMA	24.896 gr	25.
25	IMPA	91.08 gr	87.
26	TPi	-8.044 mm	-

B

Figure 10. Overlay of pre- and post-operative cephalometric tracings.



3. Discussion and Conclusion

The treatment of adult patients classified as class III, with no growth potential, can be carried out through compensatory approaches, with orthodontic camouflage, or cor-

rective approaches, through orthognathic surgery [10]. In patients with moderate to severe discrepancies and compromised facial aesthetics, the surgical approach is considered the most effective and stable [11]. In the case of the patient in question, the predominant complaint was facial aesthetics, associated with a severe intermaxillary discrepancy, which made compensatory treatment unfeasible and likely to result in an unsatisfactory and unstable outcome.

Figure 11. Follow-up photographs, 12 months post-operative.

Table 1. Comparative table of pre- and post-operative cephalometric measurements.

	Preoperative	Postoperative	Interpretation
1. NA	27.25°	16.671°	Overcorrected proclination of maxillary incisors
1. NB	22.08°	27.933°	Corrected retroclination of mandibular incisors
ANB	-2.09°	4.296°	Corrected mandibular protrusion
FMA	32.98°	24.896°	Decreased skeletal base divergence
S-N.Gn	73.23°	70.305°	Decreased skeletal base divergence

The main objectives of pre-surgical orthodontic treatment involve dental alignment and leveling, descompensation of the inclination of the incisors, coordination of the arches, correction of the Spee curve, and elimination of occlusal interferences [12-14]. Studies indicate that the average time for pre-surgical orthodontic preparation varies from 15.4 to 25 months [8]. In the case presented, a shorter pre-surgical preparation time was observed, probably due to the absence of previous compensatory treatments, mild to moderate dental crowding, significant maxillomandibular discrepancy, and early and effective management of maxillary atresia. Additionally, the initial condition of the patient, with upper incisors vestibularized and lower incisors lingualized, was benefited by the natural expansion of the lower arch, promoting initial alignment and leveling, and by the vestibularization of the lower teeth, as well as the space gain in the upper arch after surgical disjunction, which contributed to the reduction of the vestibular inclination of the upper incisors during the space closure.

The literature suggests that about half of patients with class III malocclusion and skeletal maxillary retrusion present with maxillary deficiency syndrome associated with transverse palatal atresia [15]. In young patients, the low interdigitation of the sutures facilitates the orthopedic disjunction of the maxilla [4]. However, in adults, the fusion of the maxillary, zygomatic, and pterygopalatine structures limits the results of orthopedic expansion, requiring this resistance to be broken through ERMAC [5].

The orthodontic appliances used for ERMAC can be dentally supported, dentomucosal supported, or bone-supported. For this case, a Hyrax dental-supported expander was chosen due to its convenience for both the dentist and the patient, ease of installation, simplified hygiene, and low cost. Some authors note that dental-supported devices tend to favor dental vestibularization, recommending the use of auxiliary skeletal anchorage [6]. However, controlled clinical trials have not found statistically significant differences in maxillary disjunction results with or without bone anchorage [7]. It is also recommended to perform a transverse overcorrection of the maxilla, as some degree of relapse is expected [8]. After 12 months of post-operative follow-up, no relapse was observed, and arch coordination remained satisfactory, as illustrated in Figures 11A to 11C.

The reported case demonstrated the success of the planning and its execution, with complete correction of the dentoskeletal deformity, providing excellent aesthetics and functionality to the patient. Post-operative follow-up was carried out without signs of relapse. Although the treatment involved two surgical moments, the completion time was satisfactory, thanks to the commitment of both the patient and the team. This report reinforces the importance of questioning limiting concepts that consider ortho-surgical cases as highly complex and with excessive treatment time.

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Research Ethics Committee Approval: We declare that the patient approved the study by signing an informed consent form, and that the study followed the ethical guidelines established by the Declaration of Helsinki.

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Conflicts of Interest: The authors declare no conflicts of interest.

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