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Interdisciplinary full digital restorative treatment of a young patient with severe open bite and amelogenesis imperfecta: a case report

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ABSTRACT

Background Amelogenesis Imperfecta (AI) is a syndromic entity comprising several clinical conditions, mostly genetic-based, affecting quality and quantity of enamel. The use of digital tools can be advantageous to enhance communication between the dental team and patients. Moreover, a full digital approach would allow to check each single clinical step according to the treatment plan with a backward approach. The aim of this case report is to describe a multidisciplinary treatment program of a young patient with severe open bite and amelogenesis imperfecta, whose restorative rehabilitation was planned in a full-digital workflow, from the initial communication phase up to the final prosthodontic rehabilitation.

Case report An 8-year old female patient was diagnosed with a Class II division 1 malocclusion, severe open bite and AI. The treatment plan included 3 phases: the elimination of the tongue interposition habit and the anterior open bite, the orthodontic correction of dental deviation and leveling of both dental arches and a prosthetic rehabilitation. The extraction of the four first molars corrected the canine and molar Angle Class II relationships, deviation of the midline and repositioning of the maxillary incisors on the sagittal plane. Feldspathic ceramics was used in anterior sites to enhance the esthetic outcome. CAD/CAM hybrid ceramo-polymeric restorative material (PICN) was chosen for posterior regions.

Conclusion The outcomes of the present case report proved that the planned objectives were satisfactorily obtained thanks to proper treatment planning, full digital workflow and the patient's optimal compliance with the extraoral device.

KEYWORDS Digital dentistry; Open bite; Amelogenesis imperfecta; CAD/CAM; Feldspathic ceramic.

INTRODUCTION

Anterior open bite is a condition found in 17% of patients seeking orthodontic treatment (1,2), with an overall prevalence ranging from 25% to 38%, depending on demographic factors (3). The treatment of this malocclusion is often challenging and in literature various mechanical alternatives are available: palatal crib, bonded spurs (4), orthodontic camouflage with premolars or first molars extraction (5), TAD's (6), mini-plates (7) and orthognathic surgery (8). However, appropriate diagnosis and treatment plan are needed before determining the most appropriate approach (9). The etiology of anterior open bite is multifactorial, often requiring comprehensive, extended and interdisciplinary treatments (10). Moreover, given the multifactorial etiology, accurate treatment timing and planning are particularly required. Authors in literature described cases involving non-nutritive sucking habits where orthodontic appliances promoted habit suspension and tongue repositioning (11,12). In these cases a two-phase treatment is suggested: stop the habit in the early mixed dentition as a first step, then, during permanent dentition, various treatment alternatives are possible as second stage approach, like fixed orthodontic camouflage (with or without extractions) or orthodontic-orthognatic approach (13-15).

Amelogenesis imperfecta (AI) is a syndromic entity comprising several clinical conditions, mostly of genetic origin, affecting the tooth enamel in either quality or quantity. This disease is characterized by crown malformation and abnormal enamel density and shows a prevalence variable from 1:700 to 1:14.000. The enamel appears as mulberry-shaped, easily undergoes fractures and is esthetically highly unpleasant, impairing both tooth color and shape. In the affected patients, negative psychological attitude subsequent to the compromised appearance and function, often reduce the attractiveness, self-esteem and social interaction.

AI usually occurs as a stand-alone disease, although it has been rarely reported in association with multiorgan syndromes such as cone-rod dystrophy, platyspondyly, nephro-

calcinosis, hypothalamo–hypophyseal insufficiency, Kohlschutter syndrome. AI has also been unfrequently reported in association with metabolic disorders such as hypocalciuria and Bartter-like syndrome (16,17).
Mutations or alterations in any of the genes encoding specific enamel proteins have been associated with AI: Enamelin gene (ENAM), Amelogenin gene (AMELX), Kallikrein 4 gene (KLK4), Matrix Metalloproteinase 20 gene (MMP-20), and Distal-less homeobox 3 gene (DLX3). Some studies showed that AI is a family pathology and can be inherited as autosomal dominant, autosomal recessive or x-linked dominant and x-linked recessive (18-20).
Although AI can affect both deciduous and permanent dentition, more frequently it involves permanent teeth (incisors and first molars both in upper and lower jaws). AI may be associated to several dental and skeletal developmental defects or abnormalities, affecting teeth number and eruption, root and periodontal morphology and maxillo-mandibular shape.
AI patients also complain tooth sensitivity, unsatisfactory esthetics and loss of occlusal vertical dimension due to the rapid wearing of teeth. Restorative treatment planning for AI patients aims to remove surface stains, reduce sensitivity, maintain vertical dimension of occlusion and restore appealing esthetics, preferably with fixed prostheses, according to the fundamentals of minimum invasiveness, adhesion and biomimetics.
The aim of this case report is to describe a multidisciplinary treatment program of a young patient with severe open bite and AI, whose restorative rehabilitation was planned in a full-digital workflow, from the initial communication phase up to the final prosthodontic rehabilitation.

CASE REPORT

Diagnosis and treatment plan

An 8-year old female patient came to the Division of Orthodontics of the Department of Neurosciences, Reproductive and Odontostomatological Sciences of the University "Federico II" of Naples (Italy) accompanied by her parents, complaining of a severe open bite, an unappealing aesthetics of the dental arches and a troublesome dental sensitivity (Fig. 1).
The clinical examination showed the presence of mixed dentition, atypical swallowing, long face, convex facial profile and absence of lip closure. All teeth, both in the lower and upper arch, were affected by enamel hypoplasia, with evident areas of exposed dentin (Fig. 2). From a periodontal point of view, severe gingivitis with marginal tissues swelling and bleeding on probing was detected. Probing depth was never beyond 2 mm. The young patient was diagnosed with a Class II division 1 malocclusion, severe open bite and AI.
Radiological examination with orthopanoramic radiography showed mixed dentition and all permanent

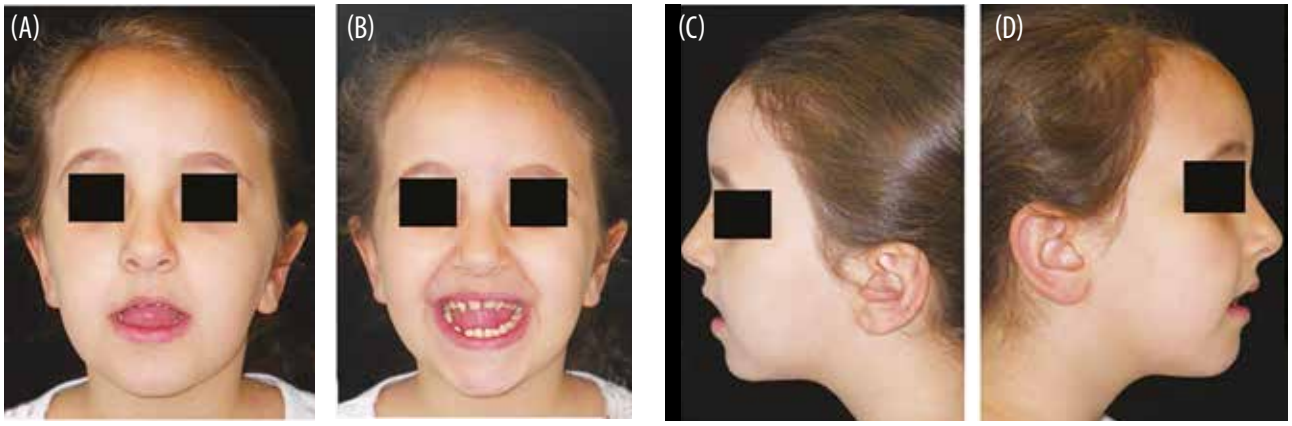

FIG. 1 Pre-operative front view. A: rest position; B: smile position; C: left silhouette profile; D: right silhouette profile.

FIG. 2 Pre-operative intraoral views. A: frontal view; B: maxillary occlusal view; C: mandibular occlusal view.

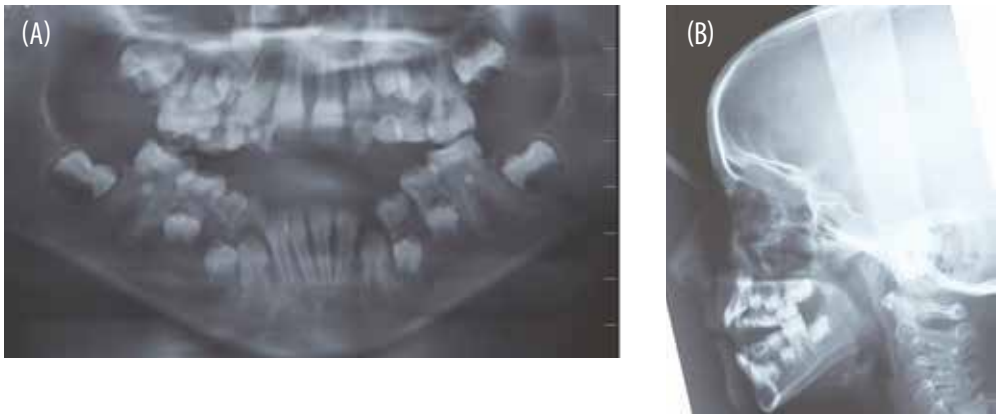

FIG. 3 Pre-operative orthopanoramic (A) and lateral cephalometric radiograph (B).

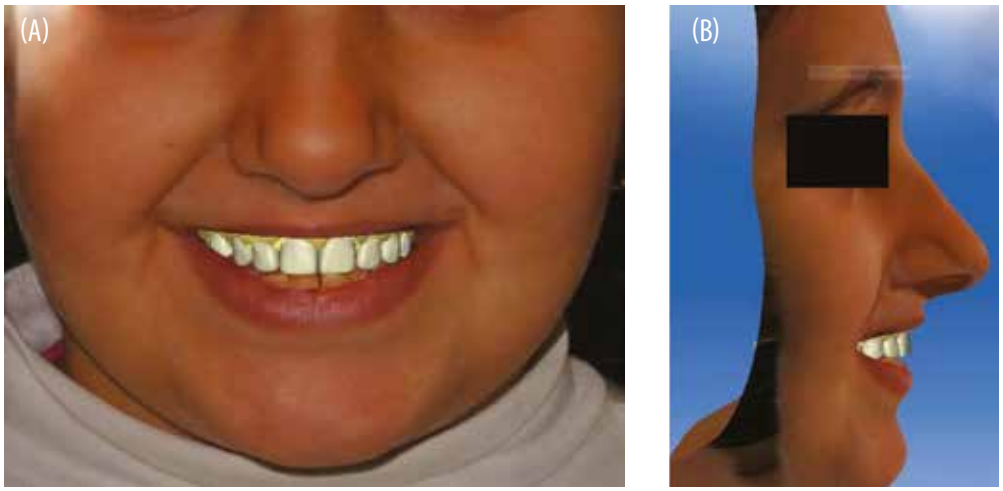

FIG. 4 Digital esthetic analysis and previsualization. A: front view; B: lateral view.

teeth were present, with the only exception of the third molars (Fig. 3A).
The cephalometric analysis indicated a Class II sagittal skeletal pattern with clockwise rotation of the mandible and a hyper-divergent growth tendency (Fig. 3B).
The objectives of the first phase of treatment were the elimination of the tongue interposition habit and the anterior open bite. The second phase of treatment planned the orthodontic correction of dental deviation and leveling of both dental arches by extractions of all the first molars. In the third phase the authors performed a final prosthetic rehabilitation in order to obtain a good aesthetic result and a preservation of dental health. The patient and the parents were briefed on the treatment plan and signed the informed consent

Treatment progress

The first treatment phase consisted of two steps. Firstly, a conventional palatal expander modified with a palatal grid was applied to correct the posterior cross-bite and anterior open bite: the screw was expanded with one quarter turn daily for 20 days. Then, following a passive retention period of about 6 months, the expander was removed and a fixed palatal grid was placed to prevent persistent tongue thrust and interposition. This interceptive phase lasted 20 months, during which speech therapy was performed.

At the end of the first phase, the patient was addressed to the Division of Prosthodontics and Digital Dentistry of our School. Digital impressions of both arches were taken with an intraoral scanner (TRIOS 3, 3Shape), subsequently imported into CAD (inLab Cad & inLab Model, Dentsply Sirona) and digitally combined with a picture of the patient's face. The program designed a previsualization of the final orthodontic alignment, useful both as an operative reference and as a communication tool with the patient's parents (Fig. 4).
Treatment phase II began 36 months after the end of phase I, with the application of an orthopedic device in combination with a pull headgear in order to control vertical mandibular growth. This extra-oral traction reduced the sagittal discrepancy enhancing a counterclockwise rotation of the mandible and restricting the horizontal growth of the maxilla. Later on, extractions of upper and lower first permanent molars on both sides were performed and a corrective orthodontic treatment was started with standard .022"× .028" edgewise appliances. Complete leveling and alignment was achieved over 16 months with a sequence of .014 "Ni-Ti" archwires; 0.14"x.025", later looped .018"× .025" and .019"× .025" stainless steel archwires in order to close the extraction spaces. To improve intercuspation in the final stages, the patient had been wearing Class II intermaxillary elastics (¼") and vertical elastics (3/16")

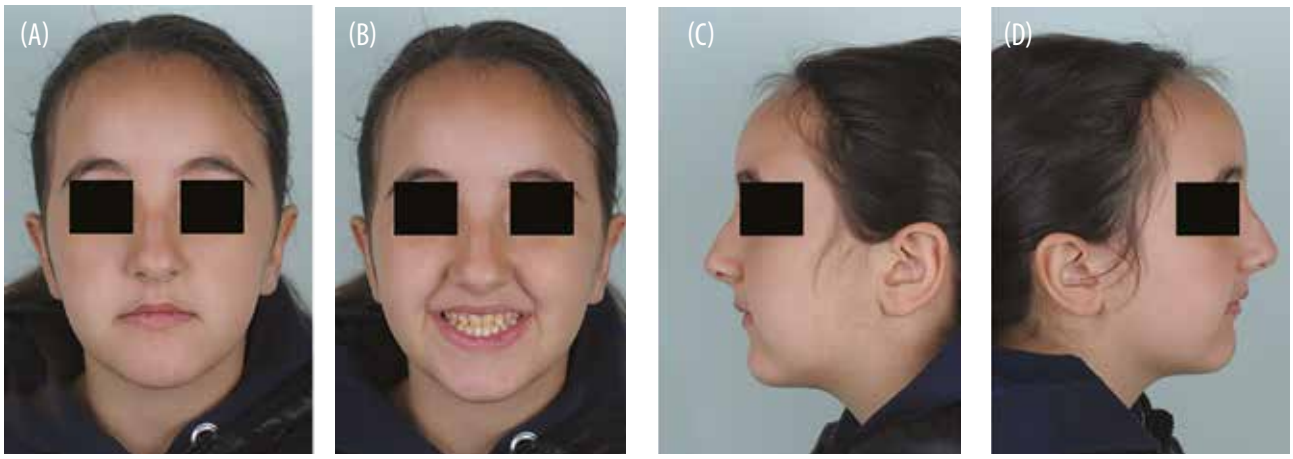

FIG. 5 Post-orthodontic front view. A: rest position; B: smile position. C: left silhouette profile; D: right silhouette profile.

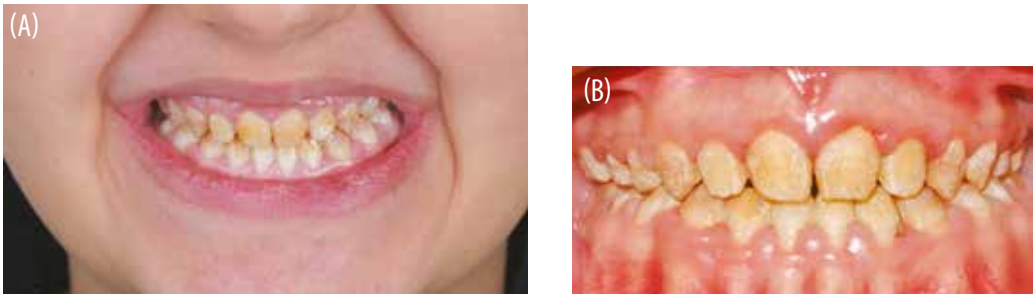

FIG. 6 Smile (A) and occlusion (B) view after orthodontic treatment.

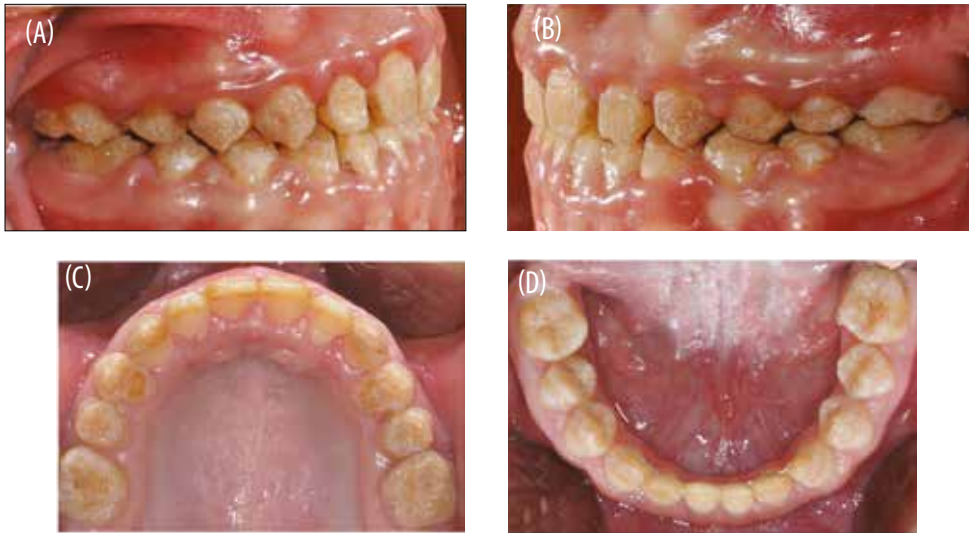

FIG. 7 Post-orthodontic intraoral views. A: left lateral view; B: right lateral view. C: maxillary occlusal view; D: mandibular occlusal view.

bilaterally, along with intermaxillary elastics (¼") in the anterior region. The total orthodontic correction lasted 39 months overall (Fig. 5-8).

The patient was then addressed to the Division of Prosthodontics and Digital Dentistry Department of our School, in order to plan the restorative phase of the treatment. This third and final phase aimed to give the patient a new, appealing smile along with functional stabilization of occlusion, protection of the exposed dentin and reduced teeth sensitivity. As a first step, digital impressions of the aligned arches were taken with an intraoral scanner and imported in CAD environment as previously described to develop a new CAD/CAM project of the restorations (Fig. 9).

By means of a 3D printer (Form2, Formlabs), two resin models were made (Grey resin, Formlabs), on which two

FIG. 8 Post-orthodontic orthopanoramic radiograph (A) and lateral cephalometric radiograph (B).

silicone templates of the new dental arches were created, aimed at getting an intra-oral mock-up (Fig. 9B, 9C).
This procedure was followed for three reasons: to provide a communication tool, preview the final result and show it to the young patient and her parents and as a guide for tooth preparation depth during the operative phase. A thin layer of petroleum jelly was applied onto the surfaces of teeth as a separating medium, then bisacrylate resin (Acrytemp, Zhermack) was poured inside the templates, finally the templates were positioned onto the dental arches. After waiting for the setting time, the templates were removed and the mock-up checked as to occlusion, esthetics and phonetics (Fig. 10).
Afterwards, all affected teeth were prepared for full-coverage, adhesively bonded restorations with iuxta-gingival margins (Fig. 11), using the calibrated bur/depth guide technique, carefully checking the minimum amount of dental tissues to be removed in accordance with the principles of minimum invasiveness.
The material choice fell upon a feldspathic ceramic for the anterior teeth (Vitablocks TriLuxe, VITA), in order to

FIG. 9 A: digital mock-up; B: 3D-printed model of the digital maxillary mock-up; C: 3D-printed model of the digital mandibular mock-up; D: digital impressions of maxillary and mandibular arches after orthodontic treatment.

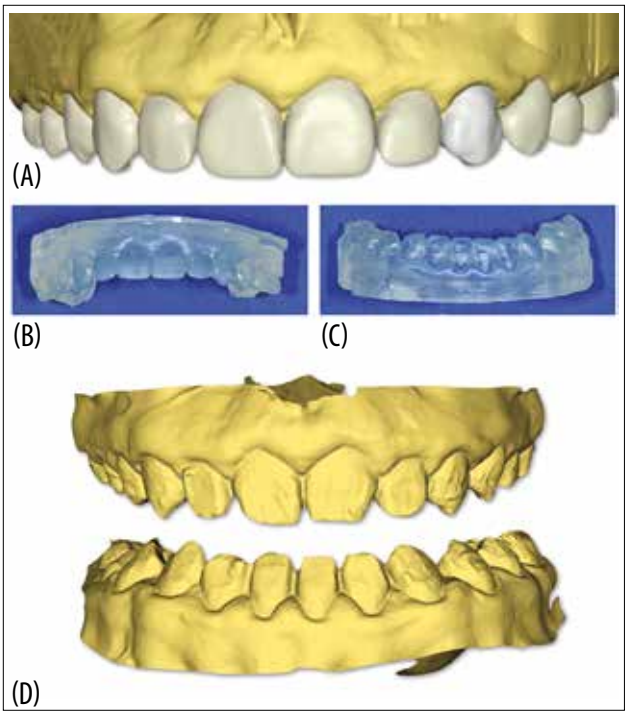

FIG. 10 A: Isolation of the interproximal spaces by means of PTFE tapes before the maxillary mockup; B: Silicone template filled with bisacrylate to mold the maxillary mock-up; C: Intraoral placement of the silicone template to mold the maxillary mock-up; D: Smile view of the maxillary mock-up.

FIG. 11 Tooth preparations at maxillary and mandibular arches.

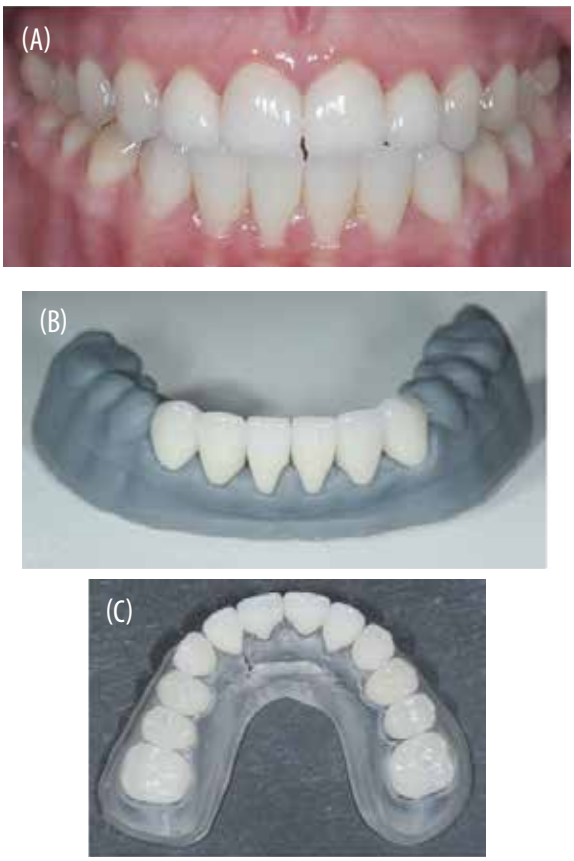

FIG. 12 A: Post-operative front view in occlusion. B: 3D printed model of the mandibular arch with feldspathic ceramic veneers. C: 3D printed model of the maxillary arch with final restorations: feldspathic ceramic veneers, in anterior areas and polymer-infiltrated ceramic network (PICN) onlays in posterior regions.

ensure a good esthetic result and natural appearance, translucency and color stability; for the posterior sites, a Polymer-infiltrated Ceramic-network material (PICN, Enamic, VITA) was chosen to produce single crowns, with a minimum occlusal thickness (0.7 mm) (Fig. 12). This hybrid material, made of a sintered ceramic matrix (86% in weight) infiltrated with a polymer matrix (14% in weight), is characterized by low abrasion toward opposing teeth surfaces, possibility of material repair and chemical compatibility with adhesive resin cements (21). Ultimately, the restorations were adhesively cemented using a dual cure cement (Panavia V5, Kuraray) and the final, minimal occlusal adjustments done by dedicated instruments (Enamic polishing clinical set, VITA), applying minimal hand pressure (Fig. 13, 14).

RESULTS

Post-treatment images and 3D scans shows a Class I molar and canine relationships, harmonious dental arches and dental midlines coinciding with the facial midline. The radiographic evaluation showed an ideal root parallelism. The cephalometric analysis indicated a slight counter-clockwise rotation of the mandible. The facial profile was improved, and the mentalis muscle was noticed to be less stressed at rest. Furthermore, the vertical and horizontal relationships of the incisors were successfully corrected. The radiographical follow-up exhibited periodontal and bone tissue health. Tracing superimpositions showed a decrease in lower facial height due to the extraction of posterior teeth and counter-clockwise rotation of the mandible.

DISCUSSION

Orthodontic treatment with first molar extractions is a controvert therapeutic option, since some authors consider them as fundamental keys to the occlusion, while other authors experienced better functional results and more stability in patients with skeletal open bite (22-24).

The success of this kind of treatment is dependent on the clinician's ability and experience to select the appropriate patient and have control over biomechanics. In the present case report, the extraction of the four first molars was essential to correct the Angle Class II molar and canine relationships, to change arch shape, correct the midline deviation and sagittal repositioning of the maxillary incisors. Since the patient had an unfitting tongue position, which influenced the malocclusion, speech therapy was implemented. The aim was to neurologically reprogram the patient's consciousness on having an adequate tongue rest position during both treatment and retention period.

The outcomes of the present case report demonstrated that the objectives proposed were satisfactorily achieved thanks to the association of the treatment procedures and the patient's excellent compliance with the extra-oral device. Regarding the aesthetic effects, the camouflage of a

FIG. 13 Post-operative front view of maxillary (A) and mandibular arch (B).

FIG. 14 : Post-operative smile view (A) and extraoral front view (B).

severe Class II open-bite malocclusion, with a hyperdivergent growth pattern, is a treatment with aesthetic limitations when compared to the surgical benefits on the facial profile and the patient's smile. However, the patient's aesthetics were not compromised by the restorative phase, since the treatment plan followed for AI aimed to remove dental surface irregularities, improve sensitivity and maintain regular occlusion. Perioral muscle balance was furthermore improved: relevant attenuation of the mentalis strain was shown post-operatively.

Feldspathic ceramics was used in anterior sites to enhance the esthetic outcome, whereas a CAD-CAM hybrid ceramo-polimeric restorative material (PICN) was chosen for posterior regions in order to allow possible endodontic interventions and occlusal adjustments over time.

The satisfactory outcomes were achieved thanks to the treatment procedures in association with the patient's excellent compliance with the extra-oral device.

The full digital workflow allowed verifying the correctness of the desired teeth positions at the end of the orthodontic treatment, so as to perform the prosthodontic procedures saving a significant amount of sound tooth tissues necessary for the adhesive cementation of the final restorations. The use of feldspathic ceramics and CAD-CAM PICN material proved to be effective in achieving optimal esthetics in anterior areas and successful biomechanics in posterior regions.

Author contributions

Conceptualization, R.V.; software, R.S.; writing—original draft preparation, R.V.; review and editing, S.P.; writing-project administration, F.Z.; All authors have read and agreed to the published version of the manuscript.

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Conflicts of interest

The authors declare no conflict of interest.

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