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A Patient with a Nonsyndromic Cloverleaf Skull Managed with Orthopedics and Orthodontics

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ABSTRACT

A patient with a nonsyndromic cloverleaf skull deformity with previous craniofacial surgery at 3, 7 and 9 years of age sought orthodontic treatment at 12 years of age; he presented with a class II div 1 malocclusion with an overjet of 13 mm with average upper and lower incisors inclined in a skeletal class II pattern and with increased lower face height and a retrognathic mandible. Furthermore, the upper and lower arches were levelled and aligned with the preadjusted MBT brackets after 0.019 × 0.025 stainless steel archwires were inserted; thereafter, a Scandi Tubular fixed functional appliance was placed. This appliance remained in his mouth for six months. The patient achieved anteroposterior correction with orthodontic treatment, which improved aesthetics and facial proportions and enhanced the patient's confidence in his social interactions.

Keywords

Class II div 1 malocclusion, Nonsyndromic cloverleaf skull, Retrognathia, Scandi tubular fixed functional appliance.

Introduction

Craniosynostosis is the premature fusion of one or more cranial sutures [1]. A cloverleaf skull shape results from the premature fusion of the coronal, lambdoid, and sometimes sagittal sutures [2]. Thereafter, the brain expands in areas of least resistance, producing a trilobed skull. This type of craniosynostosis may be syndromic or nonsyndromic. Thanatophoric dysplasia, a type II thanatophoric dysplasia, accounts for 40% of all cloverleaf skull syndromes and includes Crouzon syndrome, Pfeiffer syndrome, Carpenter syndrome, Apert syndrome, and Antley–Bixler syndrome [3]. Syndromic cloverleaf skulls often cause midface hypoplasia, leading to class III malocclusions with severe crowding and dental anomalies; nevertheless, in some cases nonsyndromic cloverleaf skulls present with mandibular retrognathia, which is a rare craniofacial condition characterized by a three-lobed skull and receding chin and which could be a congenital anomaly that may present as an isolated defect [4]. Hydrocephalus is also common; despite its multifactorial nature [5], the prevalence of hydrocephalus is unknown, and it may make up less than 10% of all cloverleaf skull presentations. In nonsyndromic cases, mandibular retrognathia might occur due to cranial base restriction. This restriction occurs because the skull base is abnormally shaped due to suture fusion, which alters the normal angle and growth trajectory of the midface and mandible. As a result, the cranial base can become shortened or overly flexed, reducing the space for mandibular projection. Mandibular retrognathism can be treated during the phase of mixed dentition or early in the phase of permanent dentition with functional appliances, including the Bionator, Twinblock, and Activator appliances, among others [6]; and fixed functional appliances, such as the Herbst appliance [7], and the Forzus appliance [8], among others [9]. Distraction osteogenesis of the mandible is used as a treatment in some cases [10], and mandibular advancement with orthognathic surgery is utilized when the growth of the maxillary and jaw has ceased [11]. Here, a case involving treatment with a functional fixed appliance during mixed dentition is presented; good results were obtained at

the end of treatment.

Case Presentation

A twelve-year-old male patient presented with a class II div 1 malocclusion on a skeletal class II pattern with increased lower face height and an overjet of 13 mm (Figures 1-3).

Figure 1: Pretreatment extraoral photographs.

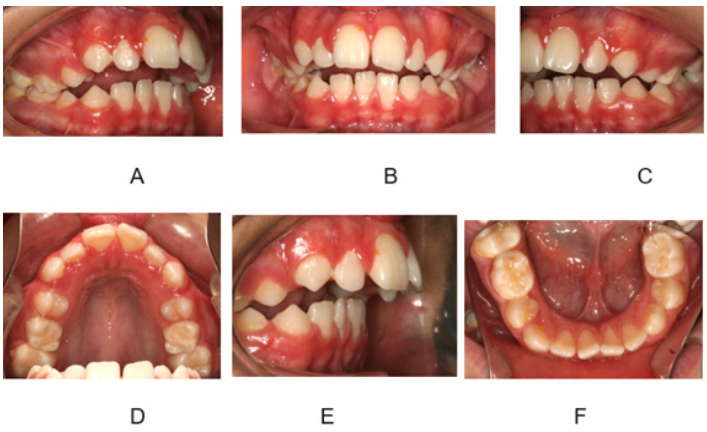

Figure 2: Intraoral pretreatment photographs (A-F).

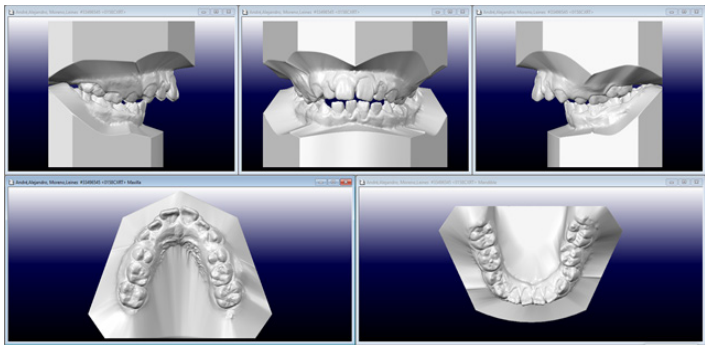

Figure 3: Pretreatment digital study models (iTero 2- Orthocad).

Medical History

The mother reported that her son had a nonsyndromic cloverleaf skull; before orthodontic treatment, the neurosurgeon performed three surgeries on the patient. The first surgery involved posterior fossa decompression for apneas and intracranial hypertension; the second surgery involved bilateral frontal–orbital advancement; and the third surgery involved a zygomatic orbital retouch. No general abnormalities were present according to the relevant medical history except for the nonsyndromic cloverleaf skull. The parents brought their child for examination because they were concerned about his mandibular retrognathia, and the patient's attitude was receptive.

Clinical Examination of the Soft Tissue Pattern

During the evaluation of the patient's soft tissue pattern, the lips were incompetent, and the tongue exhibited a lower incisor/lip swallowing pattern. Moreover, there was no relevant history of relevant past habits, and the temporomandibular joint showed no evidence of TMJ dysfunction. The patient had mixed dentition and facial asymmetry (Figure 1).

Occlusion

There was a class II Div 1 incisor relationship, and the centerline was not coincident; the lower angle shifted 2 mm to the right, with an overjet of 13 mm and an anterior open bite of 3 mm. The buccal occlusion and canine relationships on both sides were class II relationships; there was no crowding in the upper and lower labial segments, and the upper and lower buccal segments were well aligned (Figure 2).

Etiology

The patient was determined to have a nonsyndromic cloverleaf skull deformity, a skeletal class II pattern, an anterior open bite and a lower centerline that shifted to the left 2 mm (Figures 3 and 4).

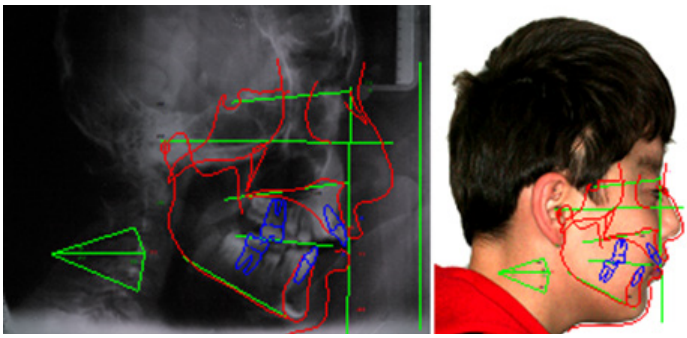

Figure 4: Pretreatment McLaughlin cephalometric tracing confirmed the class II skeletal pattern; the lower face height is increased, with an average positioning of the upper and lower incisors.

Cephalometric Pretreatment

McLaughlin cephalometric analysis confirmed the class II skeletal pattern (ANB 5.9°), with a tendency toward a retrognathic maxillary (SNA 78.8°) and retrognathic mandibula (SNB 72.9°), and the lower face height was slightly increased (FMA–MP-FH 29.0°), with the palatal–mandibular angle at 37.6°, the upper incisors at 108° (UI-palatal plane) and lower incisors at 90.5°. (IMPA) (Figure 4 and Table 1). Panoramic X-ray revealed mixed dentition without any pathological features. All teeth were present during this period of normal dentition development, and the third molars were not visible (Figure 5).

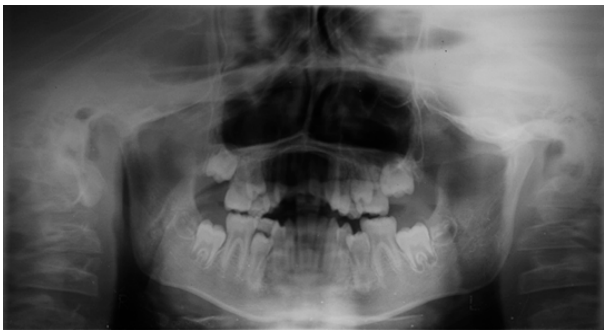

Figure 5: Normal development during mixed dentition, with no pathological features; the lower third molars are visible.

Table 1: Pretreatment Cephalometric Analysis.

Measurements	Norm	Value
Horizontal skeletal		
SNA (°)	82.0	78.8
SNB (°)	80.0	72.9
ANB (°)	2.0	5.9
Vertical Skeletal		
FMA (MP-PH) °	26	29.0
MP-SN °	33.0	34.8
Palatal-Mand Angle	28.0	37.6
Palatal-Occ Plane	10.0	14.2
Mand Plane to Occ	14.2	23.4
Mx occlusal Plane (Mx OP-Na-Perp °	95.0	95.6
Anterior Dental		
U-incisor protrusion UI-Apo (mm)	6.0	9.7
LI Protrusion (LI-Apo) (mm)	2.0	-4.6
UI-Palatal Plane (°)	110.0	108.1
UI- Occ Plane (°)	54.0	57.7
LI – Occ Plane (°)	72.0	66.1
IMPA (°)	95.0	90.5

Aims of Treatment

The therapeutic goals were to reduce the overjet of 13 mm, correct the centerline, establish a class I molar and canine relationship, and improve the anteroposterior discrepancy.

Treatment Plan

The treatment plan involved alignment of the upper and lower arch, correcting the centerline, and establishing a class I incisor, canine and molar relationship with a fixed functional appliance (Scandi Tubular appliance, Norwegian) for nonextraction treatment. The retention incorporated an upper Hawley retainer and lower lingual bonded retainer, and the prognosis was good.

Treatment Progress

Preadjusted 0.022 × 0.025 MBT brackets (3 M Unitek, Monrovia, USA) were placed for the patient. Levelling and aligning the upper and lower arches were accomplished with 0.012, 0.014 and 0.016 round NiTi arch wires (High Land Metals, IN 46131, USA) followed by working arches with 0.016, 0.018 and 0.020 Australian stainless steel wires to control the overbite (A. J. Wilcock wire; Hay Mills B25 8DW, Birmingham, England); afterward, 0.017 × 0.025 and 0.019 × 0.025 rectangular stainless steel preformed arch wires (High Land Metals, IN 46131, USA) were tied with a 0.010 s.s. ligature for torque and tip expression; afterward, the Scandi tubular appliance was placed, and continuous s.s. ligatures were tied from the lower 46 to 43, 43 to 33 and 33 to lower 36 to avoid proinclination of the lower incisors during the forward mandibular position with the fixed functional appliance. The patient wore the Scandi tubular appliance for 6 months in his mouth during this period, after which the appliance was removed and the patient underwent 6 months of consolidation with only the fixed appliance (Figure 5).

Treatment Results

The posttreatment assessment revealed the following: the patient achieved a Class I incisor, canine and molar relationship (Figures 6 and 8) and achieved facial balance (Figure 7). The overall superimposition before and after treatment revealed changes in the skeletal pattern from class II to class I (Figure 11). Facial growth occurred, and the height of the lower face was reduced (Figure 10). All permanent teeth were present (Figure 9). The prognosis was good.

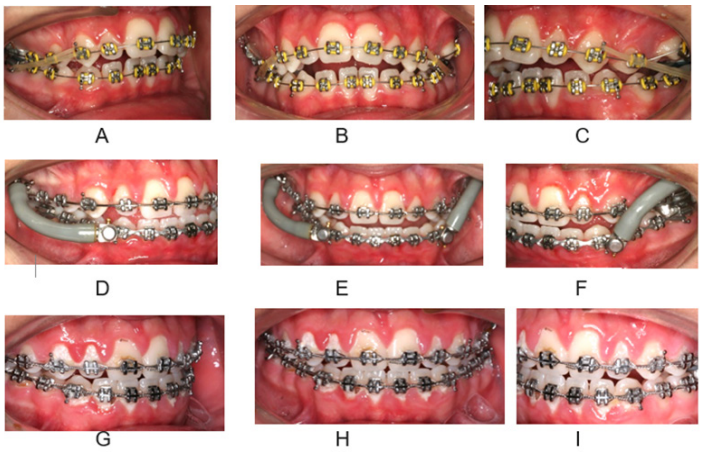

Figure 5: Levelling and aligning setup (A to C). The Scandi tubular appliance (Norwegian) was placed (D-F). In the Class I molar and canine relationship, the overjet was reduced from 13 to 1 mm (G-I).

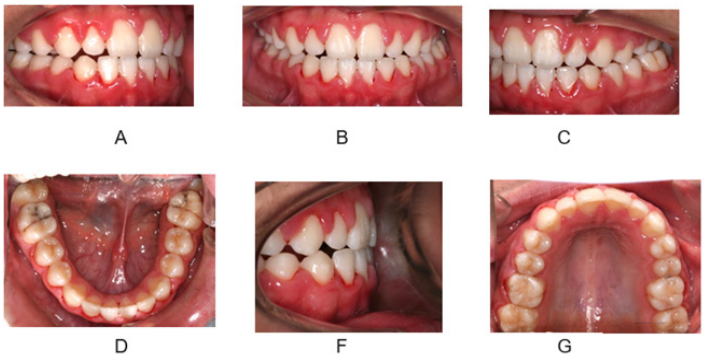

Figure 6: Intraoral post treatment photographs (A-F). A Class I molar and canine relationship was obtained. Oral hygiene was fair.

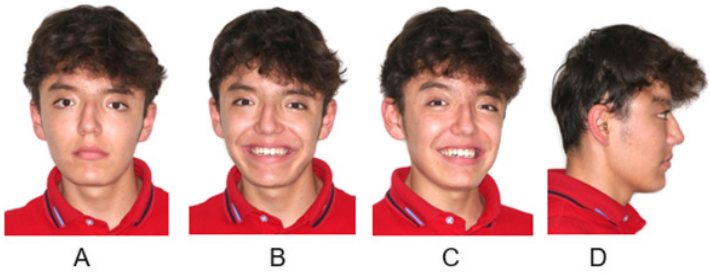

Figure 7: Extraoral post treatment photographs (A-D).

Figure 8: Digital study models at the end of treatment. The models show a Class I molar and canine relationship.

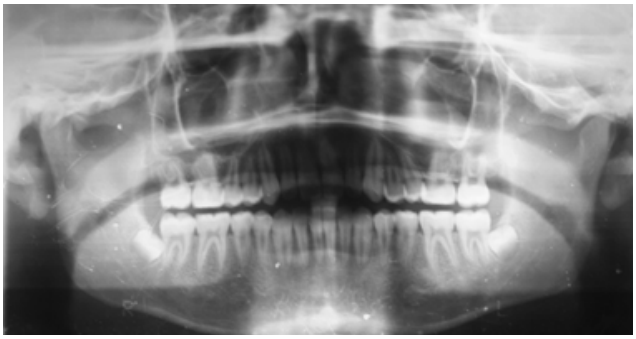

Figure 9: After orthodontic treatment, a panoramic view of the X-rays revealed permanent dentition with no pathological features. The lower third molars were present but not the upper third molars.

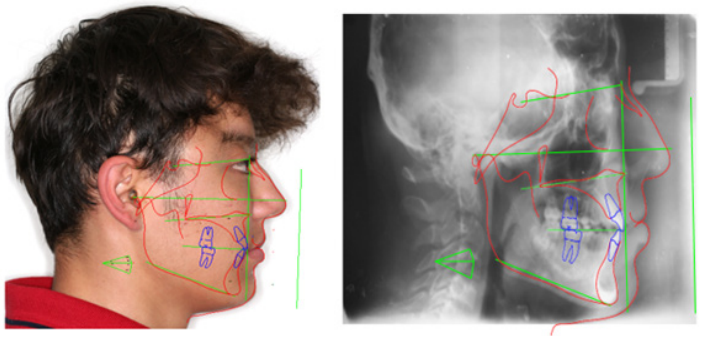

Figure 10: Post treatment McLaughlin cephalometric tracing. The ANB is at 1°. Confirming the class I skeletal pattern, the lower face height was reduced FMA (MP-FH) (°) at 21.5°, the upper incisors were normal (108.1°), and the lower incisors were slightly retroclined (88.8°).

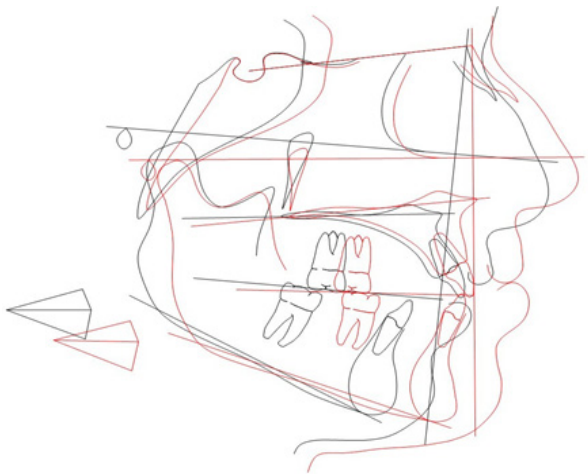

Figure 11: Overall superimposition, registered on the sella-nasion line at the sella: black line: before treatment; red line: at the end of treatment.

Cephalometric Posttreatment

McLaughlin cephalometric analysis confirmed the Class I skeletal pattern (ANB 1°), with an almost average maxillary position (SNA 79.2°), an average mandibular position (SNB 78.2°), a reduced lower facial height (FMA–MP-FH 21.5°) and a palatal-mandibular angle of 27.4°. The upper incisors were at 108.1° (UI-palatal plane), and the lower incisors were at 88.8°. (IMPA) (Figures 10 and 11 and Table 2). Panoramic X-ray revealed a permanent dentition of the patient without any pathological features. All teeth were present during this period, and the lower third molars were visible (Figure 9).

Table 2: Cephalometric Tracing: (A) Norms, (B) Before Treatment, (C) Posttreatment.

Measurements	Norm A	Value Pre-Treatment B	Value Post treatment C
Horizontal skeletal			
SNA (°)	82.0	78.8	79.2
SNB (°)	80.0	72.9	78.2
ANB (°)	2.0	5.9	1.0
Vertical Skeletal			
FMA (MP-PH) °	26	29.0	21.5
MP-SN °	33.0	34.8	28.6
Palatal-Mand Angle	28.0	37.6	27.4
Palatal-Occ Plane	10.0	14.2	7.9
Mand Plane to Occ	14.2	23.4	19.5
Mx Occlusal Plane (Mx OP-Na-Perp °	95.0	95.6	92.1
Anterior Dental			
U-Incisor protrusion UI-Apo (mm)	6.0	9.7	7.7
LI Protrusion (LI-Apo) (mm)	2.0	-4.6	2.9
UI-Palatal Plane (°)	110.0	108.1	108.1
UI- Occ Plane (°)	54.0	57.7	64.0
LI – Occ Plane (°)	72.0	66.1	71.7
IMPA (°)	95.0	90.5	88.8

Discussion

The patient had a retrognathic mandible with an overjet of 13 mm and increased lower facial height. Functional appliances, which are indicated for a retrognathic mandible [12], are among other types of appliances, such as the Klöen headgear [13] and fixed functional appliances [14], that are used during the facial growth period. However, functional appliances are contraindicated for use with high-angle mandibular and palatal plane cases [15,16], as these types of appliances increase the vertical dimension after treatment due to the eruption guidance to correct the molar class II to class I relationship. It would have been helpful to use a Twin Block with high pull Interlandi headgear to maintain the lower face height [17,18] and to reassess the case after functional appliance treatment had ended. Nevertheless, the patient did not want to wear any bulky appliances or headgear for treatment; therefore, it was decided to treat him with a comfortable fixed functional appliance, such as the Scandi Tubular appliance (Saga Dental, Norway). After aligning and levelling the occlusion with fixed, preadjusted appliances (MBT, Monrovia, California, USA), I decided to place the Scandi tubular appliance, positioning the mandible forward, edge to edge, of the upper and lower incisors over a period of 6 months. The forward positioning of the mandible for a period of time has been shown to scientifically alter the change in the position of the glenoid fossa [19,20], contributing to forward jaw location and condylar growth [21,22] via the expression of the Sox9 gene, which accelerates chondrocyte differentiation and regulates the expression of collagen type II, increasing bone formation in the glenoid fossa [23]. Remodeling of the glenoid fossa may occur at the inferior part of the anterior surface by decreasing the glenoid fossa angle and forward shifting of the mandibular condyle in height and length [24] during the growth period, which is driven by the biomechanical loading transcription of posterior fibrous tissues of the articular disc. This process can induce bone resorption and bone formation when the lateral pterygoid, masseter and anterior belly of the digastric muscles are stimulated by a functional appliance through viscoelastic tissues via a Mecano-transduction process [25]. *In vitro* studies on bone remodeling stimulated by mechanical loading have contributed to an understanding of these processes; the studies have shown that the processes upregulate interleukin-1β (IL-1β), TNF-α, and IL-6 expression, thereby upregulating RANKL expression to induce osteoclast differentiation through the RANK receptor on circulating monocytes. Therefore, IL-4 and IL-13 are expressed by osteoblasts to stimulate osteoprotegerin (OPG), a decoy of RANKL, and to regulate the homeostasis of osteoclastic activity. Interleukin-12-mediated expression of matrix metalloproteinases leads to degradation of the bone matrix, and the anti-inflammatory cytokine IL-10 expressed by osteoblasts inhibits metalloproteinase via tissue inhibitors of metalloproteinases (TIMPs). Therefore, IL-17 stimulates cathepsin K in osteoclasts to induce bone resorption, thereby increasing bone matrix degradation, which stimulates osteoblastic bone formation and accelerates bone mineralization [26-30]. Therefore, there is strong scientific evidence that functional appliances cause remodeling of the glenoid fossa as part of the biological response to forward mandibular positioning. Nonetheless, orthopedic fixed functional appliances induce growth modification and adaptation of the mandible to a new position through 30–40% orthopedic movement and 60–70% dentoalveolar changes [31], not only in skeletal class II patients but also in skeletal class III patients [32].

Patients with high-angle class II div 1 patterns are typically contraindicated for treatment with functional appliances or fixed functional appliances [16]. They are usually treated with extractions of the upper first premolars and lower first premolars [33] to allow some anterior mandibular rotation. Nevertheless, this patient had a high-angle class II skeletal pattern with mandibular retrognathia. Surprisingly, treatment with the fixed Scandi tubular appliance limited the vertical eruption of the posterior teeth, thereby preventing mandibular posterior rotation; the mandible was positioned forward. Therefore, the masseter muscle generated more horizontal forces, which rotated the mandible anteriorly, stimulating condylar remodeling and bone deposition in a more favorable horizontal rotation direction; this repositioning contributed to chin projection, reduced the lower facial height and enhanced the sagittal correction of the class II skeletal pattern in this patient [34-36].

Finally, the patient finished orthodontic treatment with upper and lower s.s. rectangular archwires of 0.019 × 0.025. The retention consisted of a bonded lingual retainer and a removable upper Hawley with a tongue trap, as the patient habitually projected his tongue toward the upper incisors.

Conclusion

The nonsyndromic cloverleaf patient achieved anteroposterior correction with a fixed functional Scandi tubular appliance, which improved aesthetics and facial proportions and enhanced the patient's confidence in his social interactions.

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