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TURKISH JOURNAL of
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Turkish Journal of Orthodontics (Turk J Orthod) is an international, scientific, open access periodical published in accordance with independent, unbiased, and double-blinded peer-review principles. The journal is the official publication of Turkish Orthodontic Society and it is published quarterly on March, June, September and December.

Turkish Journal of Orthodontics publishes clinical and experimental studies on on all aspects of orthodontics including craniofacial development and growth, reviews on current topics, case reports, editorial comments and letters to the editor that are prepared in accordance with the ethical guidelines. The journal's publication language is English and the Editorial Board encourages submissions from international authors.

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Abstract: An abstract should be submitted with all submissions except for Letters to the Editor. The abstract of Original Articles should be structured with subheadings (Objective, Methods, Results, and Conclusion). Please check Table 1 below for word count specifications.
Keywords: Each submission must be accompanied by a minimum of three to a maximum of six keywords for subject indexing at the end of the abstract. The keywords should be listed in full without

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ORIGINAL ARTICLE	4500	250 (Structured)	30	6	7 or total of 15 images
REVIEW ARTICLE	5000	250	50	6	10 or total of 20 images
CASE REPORT	1000	200	15	No tables	10 or total of 20 images
LETTER TO THE EDITOR	500	No abstract	5	No tables	No media

Tables

Tables should be included in the main document, presented after the reference list, and they should be numbered consecutively in the order they are referred to within the main text. A descriptive title must be placed above the tables. Abbreviations used in the tables should be defined below the tables by footnotes (even if they are defined within the main text). Tables should be created using the "insert table" command of the word processing software and they should be arranged clearly to provide easy reading. Data presented in the tables should not be a repetition of the data presented within the main text but should be supporting the main text.

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Book Section: Suh KN, Keystone JS. Malaria and babesiosis. Gorbach SL, Barlett JG, Blacklow NR, editors. Infectious Diseases. Philadelphia: Lippincott Williams; 2004.p.2290-308.

Books with a Single Author: Sweetman SC. Martindale the Complete Drug Reference. 34th ed. London: Pharmaceutical Press; 2005.

Editor(s) as Author: Huizing EH, de Groot JAM, editors. Functional reconstructive nasal surgery. Stuttgart-New York: Thieme; 2003.

Conference Proceedings: Bengisson S. Sothemin BG. Enforcement of data protection, privacy and security in medical informatics. In: Lun KC, Degoulet P, Piemme TE, Rienhoff O, editors. MEDINFO 92. Proceedings of the 7th World Congress on Medical Informatics; 1992 Sept 6-10; Geneva, Switzerland. Amsterdam: North-Holland; 1992. pp.1561-5.

Scientific or Technical Report: Cusick M, Chew EY, Hoogwerf B, Agrón E, Wu L, Lindley A, et al. Early Treatment Diabetic Retinopathy Study Research Group. Risk factors for renal replacement therapy in the Early Treatment Diabetic Retinopathy Study (ETDRS), Early Treatment Diabetic Retinopathy Study Kidney Int: 2004. Report No: 26.

Thesis: Yılmaz B. Ankara Üniversitesindeki Öğrencilerin Beslenme Durumları, Fiziksel Aktiviteleri ve Beden Kitle İndeksleri Kan Lipidleri Arasındaki Ilişkiler. H.Ü. Sağlık Bilimleri Enstitüsü, Doktora Tezi. 2007.

Manuscripts Accepted for Publication, Not Published Yet: Slots J. The microflora of black stain on human primary teeth. Scand J Dent Res. 1974.

Epub Ahead of Print Articles: Cai L, Yeh BM, Westphalen AC, Roberts JP, Wang ZJ. Adult living donor liver imaging. Diagn Interv Radiol. 2016 Feb 24. doi: 10.5152/dir.2016.15323. [Epub ahead of print].

Manuscripts Published in Electronic Format: Morse SS. Factors in the emergence of infectious diseases. Emerg Infect Dis (serial online) 1995 Jan-Mar (cited 1996 June 5): 1(1): (24 screens). Available from: URL: http:/ www.cdc.gov/ncidodlEID/cid.htm.

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Accepted manuscripts are copy-edited for grammar, punctuation, and format. Once the publication process of a manuscript is completed, it is published online on the journal's webpage as an ahead-of-print publication before it is included in its scheduled issue. A PDF proof of the accepted manuscript is sent to the corresponding author and their publication approval is requested within 2 days of their receipt of the proof.

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Original Article

Does Maxillary Protraction with Alt-RAMEC Protocol Affect Inferior Sclera Exposure? A Controlled 3dMD Study

Yasemin Bahar Acar, Hanife Nuray Yılmaz, Elvan Önem Özbilen

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Main Points

- Sella-nasion-A point angle (SNA) increased in the study group. However, this increase was limited compared to many studies on alternate rapid maxillary expansions and constrictions in the literature.
- The ratio of S : E in percentage was calculated to standardize the sclera exposure relative to the overall eye height.
- The ratio of S : E in percentage decreased significantly in both study and control groups on right and left sides.
- Intergroup comparisons revealed that the increase in SNA was highly significant whereas the decrease in S : E ratio was not significant.

ABSTRACT

Objective: The purpose of this controlled retrospective study was to measure and evaluate the inferior sclera exposure changes using 3dMD stereophotogrammetric images in a prepubertal Class III patient sample that underwent maxillary protraction with alternate rapid maxillary expansions and constrictions protocol followed by facemask.

Methods: The study group included 15 prepubertal patients (mean age: 9.85 $\pm$ 1.44 years) with Class III malocclusion due to maxillary retrognathism. Nine weeks of alternate rapid maxillary expansions and constrictions protocol was followed prior to 7 months of face mask treatment and 3 months of retention with Bionator. Pretreatment (T0) and post-retention (T1) lateral cephalometric radiographs and 3dMD images were retrieved from clinical archive. The same records were used for a control group of 15 well-matched, untreated patients (mean age: 9.4 $\pm$ 0.79 years). The distance between the upper eyelid margin and the lower eyelid margin was recorded as the overall eye height (E), and the distance between inferior limbus and the lower eyelid margin was recorded as inferior sclera exposure (S). The S : E ratio in percentage was calculated. Sella-nasion-A point angle (SNA) was used as the skeletal variable.

Results: SNA angle, right S : E, and left S : E changed significantly in both groups at T1-T0. The intergroup comparison was highly significant for SNA angle but was not significant for right and left S : E variables.

Conclusion: The S : E ratio decreased significantly in both alternate rapid maxillary expansions and constrictions/facemask and the control groups. However, the change in S : E ratio between groups was not significant.

Keywords: Alt-RAMEC, Angle Class III, maxillary retrusion, sclera, stereophotogrammetry

INTRODUCTION

Maxillary retrognathism has an important contribution in the etiology of skeletal Class III problems. Many researchers reported that 60% of Class III malocclusions are related with maxillary deficiency.[1,2] In growing patients with Class III malocclusion of maxillary origin, orthopedic treatment with facemask (FM) therapy has become a conventional technique with well-documented effects.[3,4] The rapid maxillary expansion (RME) prior to FM application facilitates the circummaxillary sutural response.[5,6]

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In 2005, Liou and Tsai[7] introduced an alternative method called "alternate rapid maxillary expansions and constrictions" (Alt-RAMEC) with a double-hinged expansion screw. This protocol proposed sequential expansion and constriction sets for 7-9 consecutive weeks prior to FM to increase the disarticulation of circummaxillary sutures.[7,8] The reported amounts of maxillary protraction with the Alt-RAMEC protocol were significantly more (4-6 mm in 5 months) in a shorter treatment duration[7,9] than the conventional method with RME (1.5-3 mm in 10-12 months).[10,11] Wang et al[12] concluded that the sutural opening was quantitatively more in the Alt-RAMEC group than the conventional RME.[12]

Anterior maxillary deficiency has several facial characteristics such as flat or sunken cheekbones, flat and long midface, flattened paranasal area, and prominent chin, resulting in a concave profile. Another common facial feature of these patients is increased scleral exposure of the eyes due to the decreased anterior maxillary projection of the underlying skeletal structures. Increased scleral exposure is associated with an undesirable elderly appearance and therefore is clinically undesirable.[13,14] Orthopedic and surgical (i.e., orthognathic) interferences can have an indirect impact on the periorbital area. The effect of orthognathic surgery on the scleral exposure has been documented in a limited number of studies, and they have agreed on the positive effects of maxillary advancement on inferior sclera exposure.[15-18] To the best of our knowledge, only one study has investigated the changes in inferior sclera exposure after maxillary protraction on facial photographs.[19]

The purpose of this controlled study is to measure and evaluate the inferior sclera exposure changes using 3dMD stereophotogrammetric images in a growing Class III patient sample that underwent maxillary protraction treatment with Alt-RAMEC protocol followed by FM.

METHODS

This controlled cohort retrospective study was approved by the ethical committee of Marmara University, Dental School (approval date: 01.06.2020, İstanbul; protocol number: 2020-401) and was conducted in accordance with the Declaration of Helsinki of 1975 as revised in 2013.

G*Power (version 3.1.9.4) software was used for the power analysis. The sample size was calculated based on a previous study[20] with a significance level of .05 and a power of 95% to detect a clinically meaningful difference of 1.43˚ (±1.15˚) for Sella-nasion-A point angle (SNA) between the groups. The power analysis showed that 15 patients in each group were required. The study sample was derived from the population of patients who presented to the Department of Orthodontics Marmara University, Dental School for evaluation and management of skeletal Class III malocclusion from January 1, 2009 through January 1, 2018. Inclusion criteria were anterior crossbite ≥1 mm; skeletal Class III malocclusion due to maxillary hypoplasia (diagnosed by decreased distance from N perpendicular to A point (<−1 mm), SNA (<80°), and maxillary depth (<90°)); normal to low-angle vertical growth pattern (Sella-Nasion to Gonion-Menton angle (SN-GoMe) ≤ 32 ± 6˚); Wits appraisal <−1 mm; age between 8 and 11 years; prepubertal growth stage according to the cervical vertebral maturation method; no previous orthodontic treatment; maxillary protraction treatment with FM; complete records. Exclusion criteria were patients with a large mandible (corpus length ≥ anterior cranial base+7 mm); pseudo Class III malocclusion; high-angle vertical growth pattern; presence of systemic diseases; craniofacial anomalies or temporomandibular joint disorders; history of facial trauma and orbital surgery; patients who failed to follow the treatment protocol. The informed consent was obtained from all the patients included in the study and the control group.

The study group (Alt-RAMEC/FM) comprised 15 patients (8 males, 7 females; mean age: 9.85 ± 1.44 years). Alternate rapid maxillary expansions and constrictions protocol was followed with the double-hinged screw (US patent number: 6334771B1) attached to posterior acrylic bite blocks with bilateral hooks for the attachment of elastics. The screw was activated 1 mm/day (twice in the morning and twice in the evening) for the first week and closed at the same rate for the second week. The expansion and constriction sets were continued for 9 weeks in total. A Petit-type FM (Adjustable Dynamic Protraction Facemask™, Ormco, Orange, Calif, USA) was then prescribed for a minimum of 16 h/day and 500 g/side until a full Class II molar and canine relationship was achieved. The duration of face mask treatment was 7 months on average. Patients were then given Bionator for retention for 3 months. At Pretreatment (T0) and after FM+Bionator (T1), lateral cephalometric radiographs and 3dMD stereophotogrammetric images were derived from the clinical archive of the department of orthodontics.

The control group comprised 15 subjects (9 males, 6 females; mean age: 9.4 ± 0.79 years). Lateral cephalometric radiographs and 3dMD stereophotogrammetric images were acquired initially (T0) and at the end of 9-months of observation period (T1). No orthodontic treatment was performed during that observation period.

T0 and T1 lateral cephalometric radiographs were traced to analyze the skeletal changes on NemoStudio NX Pro 10.4.2 cephalometric tracing software (Nemotech, Madrid, Spain). SNA angle was taken into consideration as a study variable for maxillary changes. 3dMDface system (3dMD Inc., Atlanta, Ga, USA) was established in a separate room using 2 modular units of 6 medical-grade, machine-vision cameras. For standardization, the 3dMD cameras were calibrated prior to every use with its calibration tray. The lighting was standardized with the powerful industrial-grade flash systems of the 3dMD module. Patients were positioned on a height-adjustable chair, looking into a mirror to establish natural head position. The position of the mirror was fixed on the wall between the cameras and opposite to the patient. The operator could manipulate the patient's head for orientation, adjusted the chair height for the patients to face the mirror, and instructed the patients to look directly into their eyes on their reflection in the mirror. Every image was quality-checked immediately on a 20-inch desktop computer screen for artifacts, and shooting was repeated when necessary. The acquired images were saved automatically in a .tbs (tricorder surface binary) format. A template was customized for landmark

Figure 1. The landmarks used in 3dMD stereophotogrammetric images: upper eyelid margin (A), the inferior limbus (B), and lower eyelid margin (C). The proportional relation between the inferior sclera exposure (S) and eye height (E).

measurements using 3dMDvultus software (3dMD Inc.). One investigator, blinded to the cephalometric analyses, measured inferior sclera exposure on both sides. The landmarks were upper eyelid margin (A), the inferior limbus (B), and lower eyelid margin (C). The distance between the upper eyelid margin and the lower eyelid margin was recorded as the overall eye height (E), and the distance between inferior limbus and the lower eyelid margin was recorded as inferior sclera exposure (S) (Figure 1). The S : E ratio in percentage was calculated to standardize the sclera exposure relative to the overall eye height.[17]

Statistical Analysis

During the assessment of the data obtained in the study, the IBM Statistical Package for the Social Sciences software (IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY, USA) was used for statistical analysis. Data were analyzed using descriptive statistical methods (mean, standard deviation). T0 and T1 values were compared statistically by the Wilcoxon signed-rank test while Mann–Whitney *U* test was used for intergroup comparisons. Significance was evaluated at a level of $P < .05$. To assess the intraexaminer reliability, 2 weeks after the first measurements, 20% of all the records were randomly selected and analyzed by the same examiner.

RESULTS

The intraclass correlation coefficient of all parameters showed a high rate of consonance between the measurements and ranged from 0.820 to 0.958. The groups presented comparable values in all of the cephalometric analyses except Incisor-Mandibular plane angle (IMPA) which was higher in the control group ($P < .05$) (Table 1).

Table 1. Evaluation of the homogeneity between the initial (T0) cephalometric values of the groups

Variables	Alt-RAMEC/FM Mean ± SD	Control Mean ± SD	*P*
SNA	77.84 ± 2.33	78.07 ± 3.15	.823
SNB	77.96 ± 2.27	79.13 ± 3.44	.280
ANB	−0.12 ± 1.18	−1.06 ± 2.12	.142
FMA	26.45 ± 3.7	29.2 ± 5.12	.102
U1-SN	100.11 ± 5.76	102.27 ± 9.3	.452
IMPA	84.57 ± 4.1	89.4 ± 5.71	.013*

Mann–Whitney *U* test $^{*}P < .05$.
Alt-RAMEC, alternate rapid maxillary expansions and constrictions; FM, facemask; SNA, Sella-nasion-A point angle; SNB, Sella-Nasion-B point angle; ANB, A point-Nasion-B point angle; U1-SN, Upper incisor to Sella-Nasion angle.

Table 2 summarizes the changes in study variables in both groups. SNA angle increased 2.12 ± 1.2° ($P < .001$) in Alt-RAMEC/FM group and 0.87 ± 0.92° ($P < .01$) in control group. The decreases in S : E ratios in percentage were 1.82 ± 2.75 ($P < .05$) and 1.68 ± 2 ($P < .01$) for the right and left eye, respectively, in Alt-RAMEC/FM group. In the control group, the decreases in S : E ratios in percentage were 1 ± 1.59 ($P < .05$) and 1.37 ± 2.02 ($P < .05$) for the right and left eye, respectively. All changes were found statistically significant.

Table 3 shows the comparison of the changes in S : E ratios (T1-T0) between the groups. The change in SNA between groups was statistically significant ($P < .01$), whereas the changes in S : E ratios in percentage were not significant.

DISCUSSION

In the literature, there are studies that evaluated the alteration of inferior sclera exposure after orthognathic surgery, and they all agree on the improving effects of maxillary advancement. In this study, we hypothesized that a similar improvement in the scleral

Table 2. Analyses of the changes in study variables in both groups

Groups	Variables	T0 Mean ± SD	T1 Mean ± SD	Difference Mean ± SD	*P*
Alt-RAMEC/FM	SNA	77.84 ± 2.33	79.96 ± 2.58	2.12 ± 1.2	.001**
	FMA	26.45 ± 3.7	26.68 ± 2.9	0.23 ± 1.39	.427
	Right S : E	7.08 ± 3.51	5.26 ± 3.69	−1.82 ± 2.75	.017*
	Left S : E	6.97 ± 3.61	5.29 ± 3.37	−1.68 ± 2	.008**
Control	SNA	78.07 ± 3.15	78.93 ± 2.81	0.87 ± 0.92	.008**
	FMA	29.2 ± 5.12	29.33 ± 5.6	0.13 ± 2.39	.812
	Right S : E	4.29 ± 4.33	3.29 ± 3.5	−1 ± 1.59	.01*
	Left S : E	4.31 ± 4.81	2.94 ± 3.74	−1.37 ± 2.02	.031*

Wilcoxon signed-rank test $^{*}P < .05$; $^{**}P < .01$.
Alt-RAMEC, alternate rapid maxillary expansions and constrictions; FM, facemask; SNA, sella-nasion-A point angle; E, overall eye height; S, inferior sclera exposure; S : E, ratio of sclera exposure relative to the overall eye height in percentage.

Table 3. Comparison of the changes in S : E ratios (T1-T0) between the groups

Variables	Alt-RAMEC/FM Mean ± SD	Control Mean ± SD	*P*
SNA	2.12 ± 1.2	0.87 ± 0.92	.003**
Right S : E	−1.82 ± 2.75	−1 ± 1.59	.325
Left S : E	−1.68 ± 2	−1.37 ± 2.02	.679

Mann-Whitney *U* test; ***P* < .01.
Alt-RAMEC, alternate rapid maxillary expansions and constrictions; FM, facemask; SNA, sella-nasion-A point angle; E, overall eye height; S, inferior sclera exposure; S : E, ratio of sclera exposure relative to the overall eye height in percentage.

exposure to that achieved by maxillary advancement surgery could be detected in the maxillary protraction treatment as well. Our primary aim was to measure the inferior sclera exposure on 3dMD images before and after maxillary protraction with Alt-RAMEC/FM protocol in prepubertal Class III subjects and compare the results with a well-matched untreated control group.

Studies that evaluated the periorbital changes after orthognathic surgery were performed on standardized facial photographs. In the present study, 3dMDface system, which provides stereophotogrammetric images, was used. Stereophotogrammetry is a non-invasive method that offers clinicians a comprehensive 3-dimensional representation of the craniofacial complex. In 2-dimensional photography, there are several factors that must be controlled for standardization. The distance between the object and camera, camera angulation, amount of magnification, lightning, and head orientation are some of these variables. In a clinical setting, stereophotogrammetry is advantageous over conventional photography by the reduced number of these variables and by its ability to reproduce 1 : 1 surface imaging. Lightning conditions, camera angulations are more standardized because of the fixed nature of the 3dMDface system apparatuses. Furthermore, the system is calibrated prior to every use, and the resultant image can immediately be evaluated on a computer screen for artifacts. The precision and reliability of 3D anthropometric data collected with the 3dMDface system have been found to be high and useful for phenotypic measures by many studies.[21-24]

Natural head posture is the upright position of the head, while it is balanced by the post-cervical and masticatory–suprahyoid–infrahyoid muscle groups, with the eyes directed forward so that the visual axis is parallel to the floor.[25] Three-dimensional images captured in natural head position are advantageous for several reasons: they are shown to be highly reproducible,[26] allow standardized patient orientation, and the visual axis is parallel to the floor during capturing also allowing more standardized eyelid and eye globe positions. Krause et al[27] has used the same method of stereophotogrammetry in natural head position for 3-dimensional analysis of changes in scleral show after surgical treatment of endocrine orbitopathy. They concluded that this method proved to be effective as it allowed exact analysis of lid contour and proptosis measurement and comparison even in cases where the bony orbit was changed.

In the Alt-RAMEC protocol, Liou[8] introduced a double-hinged screw and explained that the center of rotation of the maxilla would be located near the maxillary tuberosity. Moreover, the resistance structures are weakened, and more mobilization in the sutures is expected due to the repeated expansion and constriction sets, resulting in the forward movement of tuber maxilla without any resorption and enhanced anterior displacement of the A-point.[8] The amount of protraction with the Alt-RAMEC protocol in many studies was found greater than the conventional RME + FM protocol. Liou and Tsai[7] reported 5.8 ± 2.3 mm; Canturk and Celikoglu[28] reported 3.84 mm; and Isci et al[9] reported 4.13 mm forward movement of A-point. However, Vieira et al[29] reported less advancement (1.92 mm) with the Alt-RAMEC in comparison to the RME (2.74 mm) protocol. The promising results of the Alt-RAMEC protocol encouraged the authors to use this method in the present study where the SNA changed 2.12 ± 1.2°. Masucci et al[30] reported 2.7° and Do-de Latour et al[31] reported 1.4° improvement. These variations might result from several factors such as the severity of Class III malocclusion, age, timing of the record collection, type of expansion device, patient cooperation, and treatment duration.

To the best of our knowledge, only one study by Kale et al[19] evaluated the change in inferior sclera exposure after maxillary protraction with and without skeletal anchorage. They used facial photographs to evaluate scleral changes and concluded that in both methods the visibility of sclera reduced significantly with more improvement in the skeletal anchorage group. However, they lacked a control group, and the question remained unanswered as to whether this result was a pure effect of the treatment intervention or an effect to be expected with normal growth. In the present study, the S : E ratio decreased significantly in both Alt-RAMEC/FM and the control groups. However, when the change in S : E ratio between 2 groups was compared, there was no significant difference. Therefore, we interpret the improvement in the scleral exposure as an effect of active growth rather than an effect of the maxillary protraction. The hypothesis was rejected. Moreover, unlike the study of Kale et al.[19] the present study was conducted on stereophotogrammetry which has advantages over conventional photography by more standardized conditions such as lightning, camera angulations, etc., as was already mentioned above. Therefore, the measurements performed on stereophotogrammetric images can be considered more reliable and precise.

In the literature, age-related soft tissue changes of the palpebral fissure were evaluated by Hreczko et al.[32] They evaluated the surface measurements and age-related changes of the palpebral fissure in 1552 healthy Caucasian subjects between 2 and 18 years of age. They concluded that (1) at age 2, the height of the palpebral fissure was the most developed feature by 93.3%; (2) the measurements reached adult size between ages 8 and 16; and (3) the periods with minimal growth were at ages 5-7 and 9-10.[32] The age group of the present sample corresponds with these minimal growth periods. As a further precaution to maximize standardization of the sclera shown by the size of the eye, the S : E ratio was calculated as in the study by Norouzi et al.[17] Regarding the age-related surface remodeling of the maxillary base and orbital floor, Björk and Skieller[33] found that apposition occurs on the orbital floor and that the sutural lowering of the maxillary body is compensated by the lowering of the orbital floor. Iseri et al[34] conducted an implant study including 8- to 25-year-old female subjects. They reported a posterosuperior relocation of the orbital floor during the growth period and that not only the sutural lowering but also the forward displacement of the maxilla was compensated by the appositional growth on the orbital floor. However, they also added that the amounts of true rotation and the surface remodeling of maxilla are much smaller than those of the mandible; therefore, the errors incurred by superimposition will probably be relatively small in the analysis of relatively short periods of growth or treatment such as 1 year.[34] In the present study, this interval was 9-10 months.

In the present study, sclera exposure might not have been affected from the vertical changes since the Frankfort horizontal line to mandibular plane Angle (FMA) angle did not show any significant change after the treatment. In the literature, there are studies[28,30] reporting an increase in the vertical height while some others report no changes[20,35] as it was in the present study. The unchanged vertical height might be the result of the continued growth in the posterior face height and possible control of the vertical dimension with the posterior acrylic bite block. The studies in the literature evaluated the effects of maxillary advancement with or without impaction by orthognathic surgery or maxillary protraction with FM on sclera exposure. To the best of our knowledge, there is no study evaluating the effects of down-fracture or the increase in vertical dimension on sclera exposure. However, Soydan et al[15] reported that the impact of sagittal movement was being superior to the impact of vertical movement on the reduction of inferior sclera exposure. Moreover, they asserted that a severe amount of isolated maxillary impaction such as more than 3 mm might have some effects on exposition of inferior sclera. In the light of this information, we might infer that the increase in the vertical dimension should be in severe amounts in order to affect the sclera exposure.

A limitation of this study can be considered as the small number of subjects, which was due to the retrospective study design. The change in SNA was limited also (i.e., smaller than previous reports on Alt-RAMEC/FM). However, the T1 measurements in this study were collected after a 3-month retention period with Bionator. Most studies reflected immediate effects of Alt-RAMEC/FM protocol. For future studies, different treatment protocols (like corticotomy-assisted FM treatment where greater maxillary movement is anticipated) with larger group of subjects can be considered. Using pretreatment and posttreatment cone-beam computed tomography (CBCT) images could be beneficial to document more precise skeletal effects of the treatment. However, in that case, taking CBCT from an untreated control group can be questionable in terms of ethical concerns.

CONCLUSION

Within the limitations of this study, we conclude that the observed reduction in the inferior sclera exposure after the 9-week Alt-RAMEC protocol and 7 months of FM treatment followed by 3 months of retention was an effect of continuing active growth rather than the treatment intervention.

Ethics Committee Approval: Ethics committee approval was received from the Ethical Commitee of Marmara University, Dental School, İstanbul, Turkey (approval date and number: 01.06.2020, 2020/35; protocol number: 2020/401).

Informed Consent: Written informed consent was obtained from the parents of all patients.

Peer-review: Externally peer-reviewed.

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Original Article

Does the Alternate Rapid Maxillary Expansion-Constriction/Reverse Headgear Therapy Enhance Pharyngeal Airway Dimensions?

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Main Points

- The Alternate Rapid Maxillary Expansion-Constriction/Reverse Headgear (AltRAMEC/RH) protocol produced more significant improvement in the nasopharyngeal airway dimensions as compared to the Rapid Maxillary Expansion/Reverse Headgear (RME/RH) protocol.
- Changes in the oropharyngeal airway were insignificant with both the protocols.
- Lateral cephalograms can serve as important tools in evaluating the airway, avoiding unnecessary additional radiation exposure.

ABSTRACT

Objective: The enhanced effect of maxillary protraction following the Alternate Rapid Maxillary Expansion-Constriction/Reverse Headgear (AltRAMEC/RH) protocol over the Rapid Maxillary Expansion/Reverse Headgear (RME/RH) protocol has been well documented. However, it is not known if the airway dimensions also follow a similar enhancement. This retrospective cohort study therefore aims to compare dimensional changes in the pharyngeal airway after maxillary protraction following RME/RH, versus AltRAMEC/RH.

Methods: Pre- and post-treatment lateral cephalograms of 46 skeletal Class III patients with maxillary retrusion, who had undergone maxillary protraction using the AltRAMEC/RH or RME/RH protocol were compared for 20 dentoskeletal and airway variables. The waiting period of 6-8 months before initiating treatment served as the control period. The results were statistically evaluated using the paired *t*-test, the independent *t*-test, and the intraclass correlation coefficient.

Results: The nasopharyngeal airway indicators in the AltRAMEC/RH group (PNS-ad1, PNS-ad2, UPD) showed a statistically significant mean increase of 2.09 mm, 2.74 mm, and 1.30 mm respectively. This was significantly more pronounced than the RME/RH group ($P < .001$). The control period did not show any significant change, thus showing the negligible effect of growth on the airway dimension. No significant changes were observed in the oropharyngeal airway indicators for both groups ($P > .001$).

Conclusions: The AltRAMEC/RH protocol produced more significant improvement in the nasopharyngeal airway dimensions as compared to the RME/RH protocol. The changes in the oropharyngeal airway were insignificant with both the protocols.

Keywords: Skeletal Class III, maxillary protraction, pharyngeal airway, nasopharynx, rapid maxillary expansion

INTRODUCTION

Rapid maxillary expansion (RME) along with reverse headgear (RH) therapy is accepted as the cornerstone of early orthopedic interception in developing skeletal Class III malocclusions.[1] Maxillary expansion is an important part of protraction with facemask (FM) as it disarticulates the circummaxillary suture, which is postulated to prime the sutures for more pronounced orthopedic effects.[2] The average protraction by using rapid maxillary expansion/reverse headgear (RME/RH) is reported to be 1.5-3 mm in 10-12 months.[3]

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To open the circummaxillary sutures more extensively and to improve the effectiveness of maxillary protraction, another novel breakthrough protocol was presented by Eric Jein-Wein Liou, which consisted of repetitive maxillary expansion and constriction (Alternate Rapid Maxillary Expansion-Constriction/ Reverse Headgear (AltRAMEC) protocol) for a period of 7 weeks followed by maxillary protraction. The amount of maxillary protraction achieved thus was 5-6 mm.[3]

The benefits of these 2 protocols on dentoskeletal and soft tissue structures is now well documented.[4-6] The changes in the nasopharyngeal and oropharyngeal airway following the RME/RH protocol have also been studied, but with conflicting results.[6] Many of them have reported short-term improvement in airway dimensions—the majority of which have reported nasopharyngeal improvement following protraction using FM therapy.[7-11] However, there are reports which do not support the above findings, reporting no significant changes in sagittal nasopharyngeal and oropharyngeal dimensions, with or without RME.[12,13]

Two recent investigations on the effect of the AltRAMEC protocol have reported both dimensional and volumetric increase in the upper pharyngeal airway. One assessed volumetric changes after AltRAMEC without maxillary protraction, while the other used cephalograms to compare dimensional changes in the pharyngeal airway between 2 groups, after undergoing 5 and 9 weeks of the AltRAMEC protocol, followed by maxillary protraction.[6,14] Increase in dimension of the upper airway was reported in both groups, without any significant difference.

The enhanced effect of the AltRAMEC/RH protocol over the RME/RH protocol in improving maxillary protraction has been well documented in various investigations.[4-6] Do the airway dimensions also follow the same improvement? This has not been investigated so far. This study intends to address this lacuna.

The null hypothesis generated was that the dimensional changes in airway occurring as a result of RME/RH or the AltRAMEC/RH protocols may not be different. Thus, the aim of this study was to compare sagittal dimensional changes in the upper and lower airway following treatment with AltRAMEC /RH therapy, versus RME/RH therapy.

METHODS

This was a retrospective cohort study based on data collected from departmental treatment records, including lateral cephalograms of skeletal Class III patients with retrusive maxilla treated with protraction FM, at the Department of Orthodontics, Government Dental College, after obtaining approval from the Institutional Ethics Committee (IEC number: 65/15/DC 28/12/15). A consent waiver was obtained from the IEC Board, as patients who had completed treatment were unavailable and the identity of the patients in the records was not disclosed to the researcher analyzing the cephalograms.

Data from a previous study formed the basis for calculating the sample size.[10] The standard deviation for the change in the nasopharyngeal airway parameter before and after treatment by maxillary protraction was 3.35.

Sample size calculation was done by the formula

$$n = \frac{(Z_\alpha + Z_\beta)^2 s}{d^2}$$

Where, n = sample size

σ = Standard deviation

$Z_\alpha = 1.96$ for $\alpha = 0.05$ (α = type I error)

$Z_\beta = 0.84$ for (1- β) power = 0.20 (β = type II error)

d = difference in mean

To detect a difference in mean of 2 mm (d), a sample size of 23 was deemed necessary to be able to reject the null hypothesis that the means of the 2 groups are equal.

Accordingly, the records of patients with developing skeletal Class III malocclusions who had received dentofacial orthopedic therapy were examined by 2 clinicians (V.P. and P.S.). The pre- and post-treatment cephalograms of patients with skeletal Class III malocclusion due to retrognathic maxilla (CVMI $\leq$ CS3), who had undergone either RME/RH or AltRAMEC/RH therapy, were selected. The records of patients with craniofacial deformities and endocrine problems were excluded.

The study involved 2 groups. Group 1 comprised 23 patients (10 females and 13 males; mean age 10.27 $\pm$ 1.26 years) treated using the AltRAMEC protocol for 5 weeks, using a bonded RME appliance followed by protraction of maxilla. Group 2 consisted of 23 patients (12 females and 11 males; mean age 9.53 $\pm$ 1.50 years) who had undergone maxillary protraction after RME. Hyrax RME screws (A2620 rapid expander, Leone Orthodontic Products, Sesto Fiorentino, Firenze, Italy) and Petit-type FM (Leone Facemask-Dynamic Vertical Adjustable, Leone Orthodontic Products, Sesto Fiorentino, Firenze, Italy) had been used in this study.

A modified acrylic-bonded RME was constructed for each patient. In group 1, the screw of the RME appliance was activated/deactivated twice daily (0.20 mm per turn) during alternate weeks for constriction/expansion respectively over a course of 5 weeks. In group 2, the screw was activated twice daily (0.20 mm per turn) for 2 weeks and then sealed with acrylic after maximum activation.

In both groups, a Petit-type FM was used simultaneously with a maxillary protraction force of 500 g per side with an antero-inferior force vector of approximately 30° to the occlusal plane, applied from the hooks placed in the canine region on the buccal sides of the expanders. The patients were instructed to wear the appliances for at least 15-16 hours per day until a 2-mm positive overjet was achieved as per the institutional protocol. The parents were asked to replace the elastics at least once a day and to record the daily use of the appliances.

At the time of first visit, all patients were routinely registered and records including lateral cephalograms were taken as per institutional protocol (T0). This being a government institution, it caters to the oral health needs of 4 thickly populated districts. Despite early scheduling of appointments for patients needing interceptive treatment, this leads to a delay of 6-8 months for initiation of treatment, at which time the second cephalogram was taken (T1). The third cephalogram was taken at the end of therapy (T2). Accordingly, T1-T0 represented the effect of normal growth and T2-T1 represented the effect produced as a result of appliance therapy. Thus the period from T1-T0 served as a control period and T2-T1, the active treatment period. All lateral cephalometric radiographs of patients had been taken by the same institutional technician using standard operating protocols (PlanMeca2002 CC Proline Cephalostat) and the treatment results were evaluated cephalometrically.

Cephalometric tracings were done on 0.003-inch matte acetate paper by 1 investigator (N.B.) under optimal illumination, and identification of landmarks by another (S.S.) who was unaware of the group to which the radiograph belonged. A digital Vernier caliper was used for recording linear measurements (6-inch, 0.01 mm accuracy, 506-196-20 Absolute Digimatic Digital Caliper, Mitutoyo) and the angular measurement was obtained with a semi-circular protractor to the nearest 0.5°. Structures were retraced in the event of any disparity. A total of 20 parameters were evaluated of which 9 were airway parameters. (Tables 1 and 2, Figure 1) The tracings were done randomly, and later reassembled in the specific groups. After 2 weeks, 15 radiographs were randomly selected and all procedures repeated by the same researcher.

Statistical Analysis

All the statistical analyses were performed by investigator N.B., using the SPSS statistical package (version 18 SPSS Inc., Chicago, IL, USA) for Windows. The analyses performed were the paired *t*-test (to quantify the treatment changes within each group), and the independent *t*-test (to compare the treatment changes between groups 1 and 2). Intra-examiner reliability was tested using the intraclass correlation coefficient (ICC) (Table 3).

RESULTS

Table 4 shows the measurements taken at 2 time periods T0 and T1. The airway variables were not seen to be significantly different. The maxillary skeletal parameters A-Np and ANB had become more negative (−3.46 to −4.20 and −2.17 to −2.93 respectively). The mandibular parameters SNB and Co-Gn showed statistically significant increase ($P < .001$).

Presented in Table 5 is the baseline comparison of cephalometric variables for the skeletal and airway parameters between group 1 and group 2 at the start of treatment. No significant differences were observed.

Table 1. Cephalometric landmarks

S	Sella: The point representing the midpoint of the pituitary fossa (Sella Turcica).
N	Nasion: The most anterior point on the fronto-nasal suture in the mid-sagittal plane.
A	A point: The deepest point of the curve of the maxilla between the anterior nasal spine and the dental alveolus.
B	B point: The deepest point on the bony curvature between the crest of the alveolus and pogonion.
Po	Porion: The most superior point of the outline of the external auditory meatus ("anatomic porion").
Or	Orbitale: The lowest point on the inferior orbital margin.
ANS	Anterior nasal spine: The tip of the anterior nasal spine.
PNS	Posterior nasal spine: The tip of the posterior nasal spine.
Co	Condylion: The most superior point on the head of the condylar head.
Go	Gonion: The midpoint at the angle of the mandible.
Pog	Pogonion: The most anterior point on the mandibular symphisis.
Gn	Gnathion: The most posterior-inferior point on the outline of the mandible, lies on the contour of the chin at the point of intersection of the facial axis.
Me	Menton: The most inferior point on the symphyseal outline.
Ba	Basion: The most posteroinferior point on the anterior margin of the Foramen magnum.
ad1	Lower adenoid tissue: The point where posterior nasal spine (PNS) – basion (Ba) line intersects the posterior pharyngeal wall.
ad2	Upper adenoid tissue: The point where a line perpendicular to sella (S) – Ba plane passing through PNS intersects the posterior pharyngeal wall.
H	Hormion: The point located at the intersection between the perpendicular line to S–Ba from PNS and the cranial base.
C4	The most infero-posterior point on the corpus of the fourth cervical vertebra.

Table 2. Cephalometric parameters

Head Posture	1	SN-CVT (°)	Downward angle between SN (sella-nasion) and CVT (cervical vertebral tangent).
Maxillary Skeletal	2	Point A to Np (mm)	Point A to a line drawn perpendicular to Frankfort horizontal from nasion.
	3	Co-A (mm)	Effective midfacial length: Distance between condylion and subspinale.
	4	SNA (°)	The angle between the anterior cranial base (SN) and NA line.
Mandibular Skeletal	5	Pog to Np (mm)	Point Pog to a line drawn perpendicular to Frankfort horizontal from nasion.
	6	Co-Gn	Effective mandibular length: Distance between condylion and gnathion.
	7	SNB (°)	The angle between the anterior cranial base (SN) and NB line.
Anteroposterior Relationship	8	ANB (°)	The angle between the NA line and NB line
	9	Wits (mm)	Distance between the two points of intersection of the two perpendicular lines from points A and B to the functional occlusal plane.
Vertical Relationship	10	SN to Go-Gn (°)	Mandibular plane angle: Angle between SN plane and the mandibular plane (between Go-Gn).
	11	Jarabak ratio (%)	Percentage of the anterior and posterior facial proportions: This ratio is obtained by the formula posterior facial height/anterior facial height x 100. Anterior facial height is measured from nasion to menton and the posterior facial height is measured from sella to gonion.
Nasopharynx	12	PNS-ad1 (mm)	Upper airway thickness: The distance from PNS to the pharyngeal wall along the line from basion Ba to PNS.
	13	ad1-Ba (mm)	Upper adenoid thickness: The soft tissue thickness at the posterior nasopharynx wall through the PNS-Ba line.
	14	PNS-ad2 (mm)	Lower airway thickness: The distance from PNS to the adenoid tissue along the line from PNS to the midpoint of the line intersecting Ba to sellaturcica.
	15	Ad2-H (mm)	Lower adenoid thickness: Soft tissue thickness at the posterior nasopharynx wall through the PNS-H line.
	16	UPD (mm)	McNamara's upper pharynx dimension: The minimum distance between the upper soft palate and the nearest point on the posterior pharynx wall.
Oropharynx	17	SPAS (mm)	Superior posterior airway space: The anteroposterior width of the pharynx measured between the posterior pharyngeal wall and the dorsum of the soft palate on a line parallel to the FH plane (the line through Po and Or).
	18	MAS (mm)	Middle airway space: The anteroposterior width of the pharynx measured between the posterior pharyngeal wall and the dorsum of the tongue on a line parallel to the FH plane.
	19	IAS (mm)	Inferior airway space: The anteroposterior width of the pharynx measured between the posterior pharyngeal wall and the dorsum of the tongue on a line parallel to the FH plane that runs through C2i. (inferior-most point on posterior border of C2 vertebra).
	20	LPD (mm)	McNamara's lower pharynx dimension: The minimum distance between the point where the posterior tongue contour crosses the mandible and the nearest point on the posterior pharynx wall.

Table 6 shows the results of the analysis of changes within each group and between the 2 groups. Significant maxillary advancement ($P < .001$) and mandibular restriction ($P < .001$) were seen in both groups, leading to improvement in the sagittal discrepancy ($P < .001$).

A comparison of the 2 groups showed the changes in group 1 to be more pronounced, as evidenced by the A-Np, Co-A, SNA, SNB, Pog-Np, ANB, and Wits values ($P < .001$, $=P < .01$). The nasopharyngeal airway also followed the same improvement but only in group 1, as evidenced by the statistically significant increase in PNS-ad1, PNS-ad2, and UPD. There were no significant changes in nasopharyngeal airway in group 2. The oropharyngeal airway dimensions did not show significant changes in either of the groups.

DISCUSSION

A major drawback in most of the investigations on airway changes has been the lack of a control group.[8-11] A few studies have used age- and sex-matched Class III controls[7,12,13] and some have even used Class I as controls.[14,15] However, dentoalveolar and skeletal growth trends differ from person to person, making valid comparisons questionable.[16] To overcome this individual variability, this research used the same patients as the "delayed controls" group, on whom active treatment was administered after a waiting period of 6-8 months only. This made it possible to evaluate and make the comparisons of treatment results more valid. The results of this study showed no significant changes during the period when active treatment had not been started. Hence, the changes observed during treatment period may be attributed to therapy rather than growth.

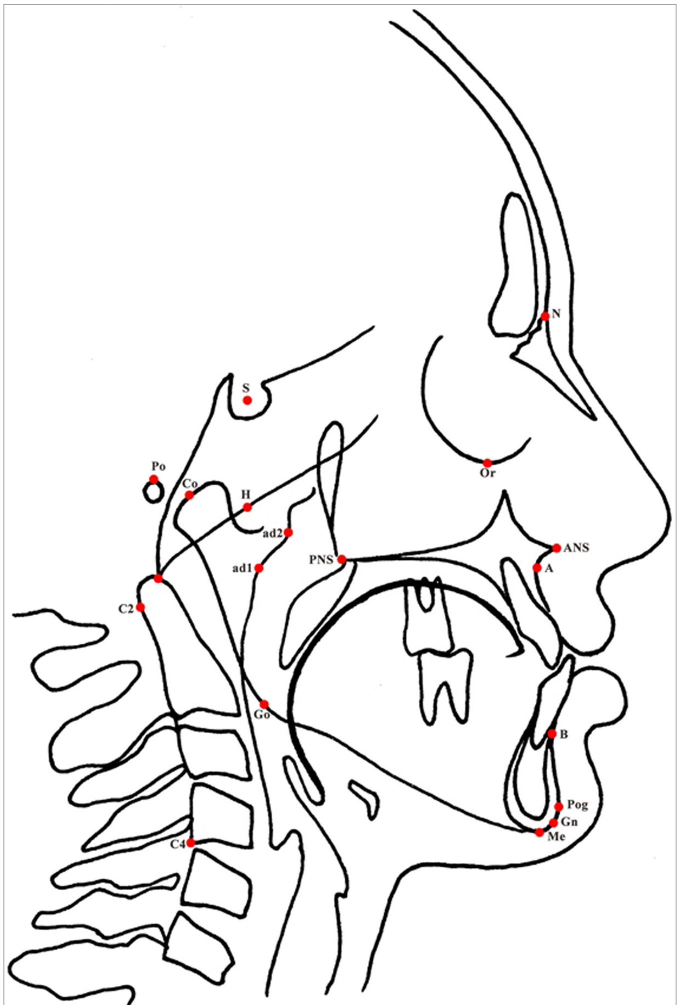

Figure 1. Cephalometric landmarks used (Table 1)

The increase in the anteroposterior dimensions of the nasopharyngeal airway was more significant in group 1 as compared to group 2. This is an important observation of this study. This may be attributed to the increased maxillary protraction obtained in the AltRAMEC protocol being reflected in the pharyngeal structures. Though there are studies proving the efficacy of AltRAMEC/RH over RME/RH in improving skeletal parameters,[4,5] the literature is scant in regard to comparison of airway dimensional changes between these 2 protocols. Hence, a comparison with previous studies was not possible.

The significant increase in sagittal dimensions of upper airway in group 1 are comparable to the cephalometric study by Celikoglu which showed similar significant increase in the linear variables of the upper airway in both treated groups following a 5-week and 9- week AltRAMEC protocol.[17] They attributed the results to the anterior movement of the maxilla.

Table 3. Intraclass correlation coefficients showing the level of agreement

Cephalometric Parameter	ICC
SNA	.988
Pog-Np	.997
PNS-ad1	1.000
LPD	.993

This result also complies with a CBCT study reporting significant increase in nasopharyngeal volume following the AltRAMEC protocol. However, their study did not use an FM for maxillary protraction. They concluded that for retrognathic maxillae, the AltRAMEC protocol alone, without the use of a maxillary protraction device, could not be considered a treatment option.[6]

When analyzing the therapeutic effects of airway modifications, an important consideration is the lymphoid tissue located on the posterior pharyngeal wall.[12] The comparison of the lymphoid parameters ad1-Ba and ad2-H in this study did not show a significant change during the control and treatment periods, attributing the obtained increase in nasopharyngeal airway dimensions to active interventional therapy. This is contradictory to the results obtained by Bacetti et al.[13] who reported a decrease of lymphoid tissue in both treated and control groups. No significant changes in any of the airway variables were reported by Bacetti et al. This could probably be attributed to the younger age of the group in the study by Bacetti et al., when compared with the mean age of the patients in our study group.

Group 2 also showed an increase in the nasopharyngeal airway measurements but this increase was not statistically significant. This is in agreement with previous studies which showed an increase only in upper airway but not in the lower airway.[9-11] A longitudinal follow-up study of 5 years has demonstrated that the changes in the nasopharyngeal airway remained more pronounced and stable during the follow-up period as compared to the treatment period.[11]

The observations made by Kilinç et al.[7] (2006) who studied the effect of RME/RH in 18 patients with Class III malocclusion reported short-term improvement in both nasopharyngeal and oropharyngeal airway dimensions. Later investigations have, however, contradicted these findings.[12,13] This increase in nasopharyngeal airway also complies with the reported improvement in nasopharyngeal airway due to maxillary advancement by other surgical methods of treating skeletal Class III malocclusion, such as conventional LeFort I advancement or anterior maxillary segmental distraction for cleft patients.[18]

The findings of this study indicate that facilitation of maxillary protraction in growing patients with an orthopedic appliance could contribute to enhancement in upper-airway dimensions, thus improving respiratory function in patients with maxillary hypoplasia. This increase is found to be more significant in protraction following AltRAMEC therapy over protraction following the RME protocol. Thus, the early airway improvement reported in our study coheres with the evidence, stressing the importance of stable skeletal changes obtained via early orthopedic intervention in skeletal Class III over later surgical intervention.[19] The advantage of early intervention in skeletal

Table 4. Comparison of changes at first visit (T0) and start of treatment (T1)

		T0		T1		
Variable		**Mean**	**SD**	**Mean**	**SD**	***P***
Head Posture	SN–CVT (°)	102.46	8.26	100.46	7.66	.146
Maxillary Skeletal	A-Np (mm)	−3.46	2.84	−4.20	2.82	.006†
	Co-A (mm)	84.28	4.10	84.41	4.07	.011‡
	SNA (°)	79.46	4.57	79.59	4.56	.030 ‡
Mandibular Skeletal	Pog-Np (mm)	−2.23	5.15	−2.43	5.71	.737
	Co-Gn (mm)	113.52	5.76	115.43	5.72	.000*
	SNB (°)	81.63	4.61	82.52	4.38	.000*
Antero Posterior	ANB (°)	−2.17	2.99	−2.93	2.59	.001‡
	Wits (mm)	−8.71	3.40	−9.07	3.50	.370
Vertical	Sn to Go-Gn (°)	34.28	5.24	33.83	5.49	.397
	Jarabak (%)	62.62	4.52	62.69	4.64	.794
Nasopharynx	PNS-ad1 (mm)	20.74	3.45	20.61	2.95	.695
	ad1-Ba (mm)	22.28	3.90	22.17	3.16	.846
	PNS-ad2 (mm)	17.33	2.23	16.87	2.40	.076
	ad2-H (mm)	17.00	5.05	17.39	4.53	.464
	UPD (mm)	11.65	1.82	12.00	1.73	.088
Oropharynx	SPAS (mm)	14.41	3.42	14.48	2.56	.919
	MAS (mm)	11.71	3.10	11.30	2.23	.464
	IAS (mm)	12.20	4.00	11.41	3.36	.266
	LPD (mm)	11.90	2.44	11.46	2.77	.343

*$P < .001$, †$P < .01$, ‡$P < .05$.

Table 5. Comparison of baseline parameters between the 2 groups: Group 1 (alternate rapid maxillary expansion-constriction/reverse headgear) and group 2 (rapid maxillary expansion/reverse headgear) before treatment

		Group 1 (T1)		Group 2 (T1)		
Variable		**Mean**	**SD**	**Mean**	**SD**	***P***
Head Posture	SN–CVT (°)	100.46	7.66	99.87	10.55	.830
Maxillary Skeletal	A-Np (mm)	−4.20	2.82	−3.11	2.34	.162
	Co-A (mm)	84.41	4.07	83.96	4.58	.723
	SNA (°)	79.59	4.56	80.02	3.54	.720
Mandibular Skeletal	Pog- Np (mm)	−2.44	5.72	−2.59	4.44	.920
	Co-Gn (mm)	115.43	5.73	113.09	5.02	.146
	SNB (°)	82.52	4.38	82.52	3.72	1.000
Antero Posterior	ANB (°)	−2.94	2.59	−2.50	1.64	.500
	Wits (mm)	−9.06	3.50	−6.65	6.03	.104
Vertical	Sn to Go-Gn (°)	33.83	5.94	33.39	5.74	.802
	Jarabak (%)	62.69	4.64	62.48	4.27	.874
Nasopharynx	PNS-ad1 (mm)	20.22	1.57	20.74	1.51	.257
	ad1-Ba (mm)	22.17	3.16	22.39	4.52	.851
	PNS-ad2 (mm)	16.87	2.40	15.35	3.16	.072
	ad2-H (mm)	17.39	4.53	17.61	4.22	.867
	UPD (mm)	12.00	1.73	12.67	3.05	.362
Oropharynx	SPAS (mm)	14.48	2.56	15.13	2.81	.415
	MAS (mm)	11.30	2.23	13.13	2.56	.013
	IAS (mm)	11.41	3.63	14.13	3.33	.011
	LPD (mm)	11.46	2.77	10.37	3.08	.214

*$P < .001$, †$P < .01$, ‡$P < .05$.

Table 6. Comparison of the mean changes between group 1 (alternate rapid maxillary expansion-constriction/reverse headgear) and group 2 (rapid maxillary expansion/reverse headgear) group

Variable		AltRAMEC/RH Difference (T2-T1)			RME/RH Difference (T2-T1)			Unpaired *t*-Test
		Mean	SD	*P*	Mean	SD	*P*	*P*
Head Posture	SN–CVT (º)	1.15	5.78	.35	−2.22	5.25	.055	.044‡
Maxillary Skeletal	A-Np (mm)	2.37	1.38	.000*	1.43	0.53	.000*	.004†
	Co-A (mm)	2.72	1.16	.000*	1.57	0.51	.000*	.000*
	SNA (º)	2.33	1.24	.000*	1.68	2.04	.001*	.197
Mandibular Skeletal	Pog- Np (mm)	−2.85	1.82	.000*	−0.48	1.92	.244	.000*
	Co-Gn (mm)	1.83	1.47	.000*	1.91	1.31	.000*	.833
	SNB (º)	−2.17	1.61	.000*	−0.74	1.91	.077	.009†
Antero Posterior	ANB (º)	3.84	1.82	.000*	3.20	2.15	.000*	.272
	Wits (mm)	4.85	2.57	.000*	2.91	2.92	.000*	.021‡
Vertical	Sn to Go-Gn (º)	1.78	1.68	.000*	0.65	1.27	.022‡	.013‡
	Jarabak (%)	−1.11	1.45	.001*	−0.38	1.65	.281	.119
Nasopharynx	PNS-ad1 (mm)	2.09	1.83	.000*	0.00	0.45	.133	.000*
	ad1-Ba (mm)	0.61	1.50	.064	0.28	3.83	.727	.706
	PNS-ad2 (mm)	2.74	1.18	.000*	0.00	0.45	1.000	.000*
	ad2-H (mm)	0.09	2.86	.885	0.00	2.89	1.000	.919
	UPD (mm)	1.30	1.11	.000*	0.02	1.93	.957	.008†
Oropharynx	SPAS (mm)	0.74	2.38	.150	0.54	3.12	.413	.812
	MAS (mm)	0.43	2.19	.352	−0.13	1.84	.737	.349
	IAS (mm)	0.93	3.17	.170	−0.15	3.15	.819	249
	LPD (mm)	0.59	1.84	.140	0.09	2.81	.884	.479

*$P < .001$, †$P < .01$, ‡$P < .05$.
AltRAMEC, alternate rapid maxillary expansion/constriction; RME, rapid maxillary expansion; RH, reverse headgear.

Class III was highlighted in this interesting study which compared the long-term treatment effects produced by protraction FM therapy (followed later by a second phase of comprehensive fixed-appliance therapy), with untreated Class III controls and with subjects surgically treated with LeFort I maxillary advancement. They have reported that early treatment with orthopedic forces to advance the maxilla might reduce altogether the need for surgical intervention later. If surgery becomes necessary, it might be restricted to only 1 jaw, thereby minimizing complications.[19]

The oropharyngeal airway parameters SPAS, MPS, IAS, OAW, and LPD did not show any significant increase. This is in agreement with previous investigations which have reported an increase only in the nasopharyngeal airway dimensions.[10,11]

The post-treatment posture of the head was seen to be more extended with respect to the cervical vertebrae, as indicated by the mean increase of 1.15° in the SN–CVT angle in the AltRAMEC group. This could be attributed to the counter-clockwise rotation of the maxillary complex, reported in previous studies with maxillary protraction.[16] The upper-airway dimension and head posture observed in our study seem to be in accordance with the findings from previous investigations.[20]

The findings of the present investigation have demonstrated that the AltRAMEC/RH protocol showed significant favorable effects over the RME/RH therapy with respect to the skeletal components of Class III malocclusion, as reported in previous studies.[21-23] The nasopharyngeal airway dimensions also followed a similar improvement. Thus, the null hypothesis stands rejected.

The present study is based on two-dimensional cephalometric measurements of airway structures, and thus, has limitations. However, as lateral cephalograms are used as routine diagnostic aids in orthodontics, they may also serve as important tools in evaluating the airway, avoiding unnecessary radiation exposure when subjected to computed tomographic imaging. Although there are problems of superimposition and magnification in the lateral cephalogram due to the complex anatomy of the pharyngeal airway, there have been studies stating that lateral cephalometric films stand significantly reliable and reproducible in assessing airway dimensions on successive radiographs when attention is given to the reproducibility of head position during image acquisition.[24] A positive correlation between nasopharyngeal airway size on cephalometric films and its true volumetric size as assessed from CBCT scans has also been reported.[25]

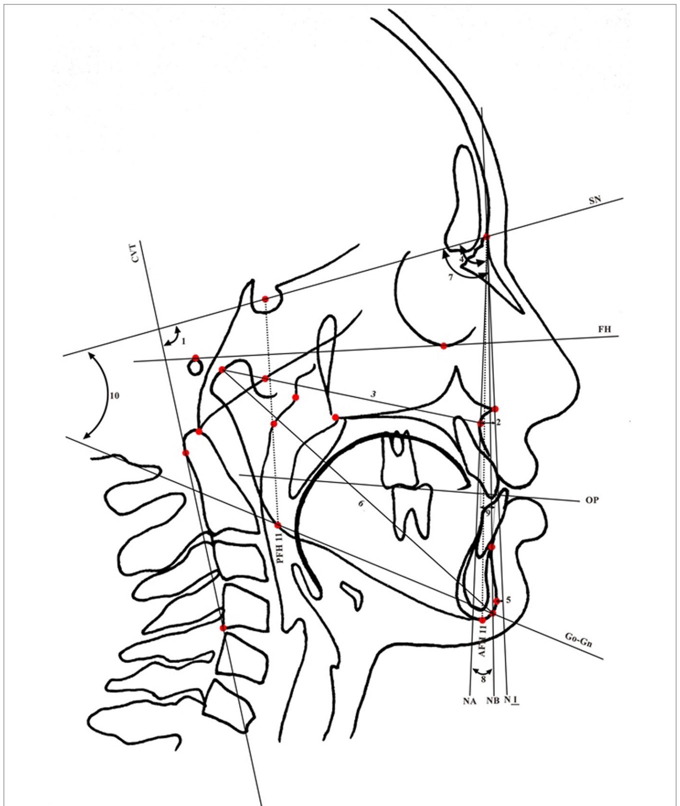

Figure 2. Skeletal parameters and planes used (Table 2)

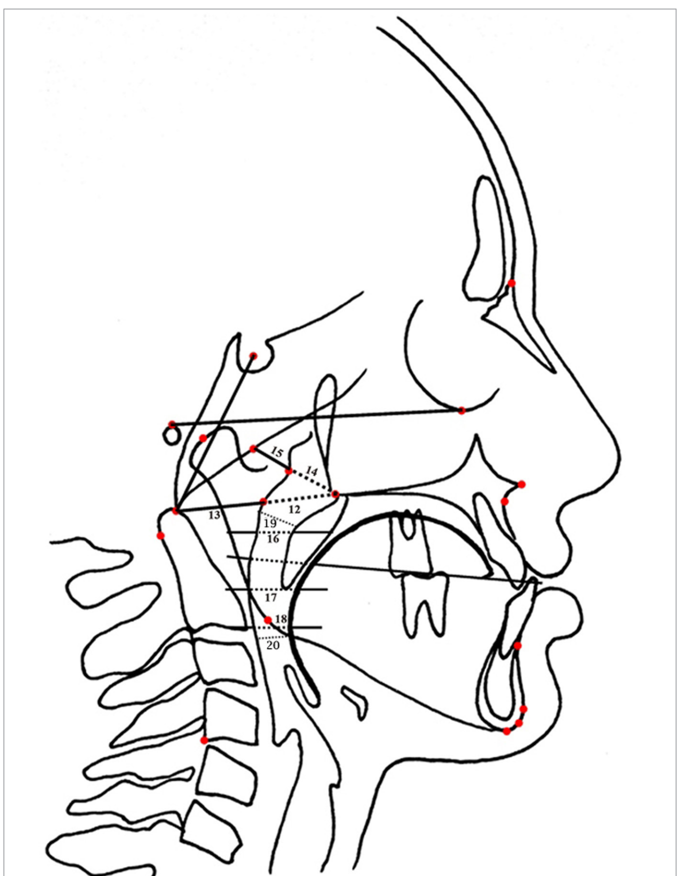

Figure 3. Airway parameters (Table 2)

CONCLUSION

The AltRAMEC/RH protocol produced more significant improvement in the nasopharyngeal airway dimensions as compared to the RME/RH protocol; whereas the changes in oropharyngeal airway were insignificant with both the protocols.

Ethics Committee Approval: Ethics committee approval was received for this study from the IEC number: 65/15/DC 28/12/15 of Government Dental College, Calicut, Kerala, India.

Informed Consent: Departmental Treatment Records used. Consent Waiver obtained from IEC Board.

Peer-review: Externally peer-reviewed.

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Conflict of Interest: The authors have no conflicts of interest to declare.

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Original Article

Obstetrician–Gynecologists' Knowledge and Awareness on Nasoalveolar Molding in Newborns with Cleft Lip and Palate

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Main Points

- Obstetrician–gynecologists (OB-GYNs) play an important role on the diagnosis of orofacial clefts with the help of prenatal ultrasound image scanning. However; despite their crucial contribution on diagnosis, their knowdge on nasoalveolar molding is insufficient.
- Nasoalveolar molding (NAM) provides aesthetic and functional anatomy, approximation of the maxillary segments and psychological support to the parents of the newborns with cleft lip and palate (CLP).
- Informative seminars and conferences should be organized to increase the awareness of Obstetrician–gynaecologists regarding NAM and multidisciplinary treatment approaches of newborn CLP patients.

ABSTRACT

Objective: This study aimed to determine the level of knowledge and awareness of obstetrician–gynecologists (OB-GYNs) about the presurgical orthopedic treatment of newborns with cleft lip and palate (CLP).

Methods: We conducted a 12-question survey by email to 532 OB-GYNs who were members of the Turkish Obstetricians and Gynaecologists Association. The participants were asked about their years in practice, region of practice, and knowledge of CLP and nasoalveolar molding (NAM).

Results: A total of 141 OB-GYNs agreed to participate and completed the survey. Fifty-nine (42%) of 141 OB-GYNs had never heard of NAM treatment in newborns with CLP. Twenty-seven percent had information about NAM, and 23% referred newborns with CLP for NAM. No statistically significant difference existed in the knowledge level about the preoperative treatment of newborns with CLP between experienced and inexperienced OB-GYNs ($P > .05$).

Conclusion: The knowledge levels of OB-GYNs about NAM were insufficient. We hope that this study will provide more effective results in OB-GYNs referring newborns with CLP for NAM.

Keywords: Cleft lip and palate, survey, nasoalveolar molding, obstetricians, gynecologists

INTRODUCTION

Cleft lip and palate (CLP) is the second most common congenital defect, according to the Centers for Disease Control and Prevention.[1] The frequency of various cleft lip types with or without cleft palate is 1 in 700-1000 live births worldwide.[2,3] A multidisciplinary team of experts evaluates newborns with CLP and provides a surgical treatment that is usually performed in the first year. However, the number of surgical operations these individuals undergo can vary from 2 to 20 until adulthood.[4]

Nasoalveolar molding (NAM) has emerged as a relatively new technique in cleft care over the past decade. The NAM technique uses acrylic nasal protrusions that are attached to the vestibular part of the acrylic

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Figure 1. Bilateral cleft lip and palate patient facial photograph

feeding plate to bring the nasal alar cartilage to the normal form and position in the neonatal period. Moreover, NAM contributes to the elongation of the columella before cleft lip surgery. The NAM technique facilitates the surgical reconstruction of the cleft and reduces the severity of the deformity (Figures 1-3).

Newborns with CLP start using the NAM appliance immediately after birth and before surgical treatment of the lip at around 5 months of age or when the cleft is reduced to less than 5 mm between segments. The parents perform daily adjustments to the bands on the infant's face and attend weekly or biweekly clinical appointments. Both short- and long-term studies have shown that NAM significantly improves nasal symmetry compared to surgical treatment alone. Additionally, NAM provides

Figure 2. Nasoalveolar molding appliance in situ

Figure 3. Extraoral view of repaired cleft lip and palate(CLP), after NAM therapy

aesthetic and functional anatomy and approximation of the maxillary segments.[5-8]

Obstetrician–gynecologists (OB-GYNs) are often the first to discover orofacial clefts and other craniofacial conditions in prenatal ultrasound image scanning. Prenatal ultrasound is also generally the first picture of the infant for parents, who do not expect the sonographer to detect a birth defect.[9] The sensitivity of the routine transabdominal ultrasound scan at 20 gestational weeks ranges from 16% to 93%.[10,11] However, when a fetus is diagnosed with CLP by ultrasonography, the treatment options should be planned in a multidisciplinary way. Obstetrician–gynaecologists play an important role in the diagnosis of CLP in terms of educating the parents about the treatment progress and interdisciplinary team approach, timely referral to an orthodontist, and early commencement of psychological support for the parents.

This study aimed to determine the level of knowledge and awareness of OB-GYNs about the presurgical orthopedic treatment of newborns with CLP.

METHODS

We conducted a questionnaire study with OB-GYNs who were randomly selected from all over Turkey. The study was approved by the Clinical Research Ethics Committee of Ordu University (No. 2020/220). The questionnaire was specifically designed by the researchers using a Google Forms platform. Responses from OB-GYNs that were received within 12 weeks were included. The purpose of the project was communicated to all the participants. The questionnaire was distributed by email to 532 OB-GYNs, of whom 141 agreed to participate. The questionnaire consisted of 12 questions in 2 parts (Table 1). First, demographic information was collected, including title, workplace, and years of practice. The participants were asked about their knowledge of CLP and

Table 1. Questionnaire

Questions	Answer
What is your title?	Specialist Doctor
	Assistant Professor
	Associate Professor
	Professor
What is your gender?	Male
	Female
How many years have you been an OB-GYN?	
What is your workplace?	Faculty of Medicine
	Public Hospital
	Private Practice/Hospital
Do you have any information about orthopedic applications performed before surgical treatment of CLP newborns?	Yes
	No
How many CLP newborn births have you ever seen?	None
	1-4
	5-10
	> 10
Have you heard about feeding plate/ NAM in CLP newborns?	Yes
	No
Do you know what NAM is?	Yes
	No
Do you know who performed the NAM?	Yes
	No
Do you know for what purpose NAM is performed?	Yes
	No
Do you refer patients for NAM application?	Yes
	No
When you diagnose CLP on ultrasound, which unit do you refer the patient to?	Plastic and Reconstructive Surgery
	Pediatrics
	Dentist
	Orthodontics
	I do not refer

CLP, cleft lip and palate; NAM, nasoalveolar molding.

NAM in the second part. Of the total participants, 78 (55%) were female and 63 (45%) were male. Those who had been OB-GYNs for 10 years or more were described as experienced, while those practicing for less than 10 years were described as inexperienced. Eighty-six (61%) of the participants were inexperienced, and 55 (39%) were experienced.

Statistical analysis used SPSS software (SPSS for Windows version 20.0; SPSS Inc, Chicago, IL, USA). Descriptive statistics were formed for the evaluated parameters. Percentages were calculated for categorical variables. The Pearson chi-square test was used to compare OB-GYNs according to their level of knowledge of preoperative treatment of newborns with CLP. Statistical significance was set at $P < .05$.

Table 2. Sociodemographic characteristics of OB-GYNs (N = 141)

	n	%
Gender		
Female	78	55
Male	63	45
Experience		
Less than 10 years	86	61
10 years or more	55	39
Workplace		
Faculty of Medicine	64	45
Public Hospital	43	31
Private Practice	34	24

%, Percentage; Sample (N) = 141.

RESULTS

A total of 141 OB-GYNs responded to the questionnaire. Sixty-four (45%) worked in a Faculty of Medicine, 43 (31%) in public hospitals, and 34 (24%) in private healthcare (Table 2). Sixty-four (45%) participants had knowledge of the orthopedic practices performed before the surgical treatment of newborns with CLP. Thirty-three (23%) OB-GYNs referred newborns with CLP for NAM, and 108 (77%) did not (Table 3).

While 11 (8%) OB-GYNs had never attended the birth of a newborn with CLP, 93 (66%) had attended 1-4, 22 (16%) had attended 5-10, and 15 (11%) had attended more than 10 (Table 4).

Fifty-nine of 141 (42%) OB-GYNs have never heard of NAM treatment for newborns with CLP. Thirty-eight (27%) had information about NAM, whereas 103 (73%) OB-GYNs did not know the purpose of NAM.

No statistically significant difference existed in the knowledge level of preoperative treatment of newborns with CLP between experienced and inexperienced OB-GYNs ($P > .05$; Table 4).

DISCUSSION

Orofacial clefts are among the most common congenital craniofacial abnormalities. The etiology of CLP is multifactorial, and the incidence may be affected by many factors, including

Table 3. Percentage distributions of the OB-GYN where to refer newborns with cleft lip and palate for nasoalveolar molding

Department to refer	%
Plastic and Reconstructive Surgery	41
Pediatrics	39
Dentist	4
Orthodontist	10
Do not refer	6

Table 4. Comparison of obstetrician–gynecologists according to their level of knowledge about preoperative treatment of newborns with cleft lip and palate

Queries	Answer	Inexperienced	Experienced	P*
Do you have any information about orthopedic applications performed before surgical treatment of CLP newborn?	Yes	44	20	.085
	No	42	35	
How many CLP newborn births have you ever seen?	None	8	3	.183
	0-4	60	33	
	5-10	9	13	
	> 10	9	6	
Have you heard about feeding plate/NAM in CLP newborns?	Yes	51	31	.730
	No	35	24	
Do you know what NAM is?	Yes	24	14	.749
	No	62	41	
Do you know who performed the NAM?	Yes	18	13	.705
	No	68	42	
Do you know for what purpose NAM is performed?	Yes	25	13	.478
	No	61	42	
Do you refer patients for NAM application?	Yes	20	13	.958
	No	66	42	

*Results of Pearson Chi-square test.
CLP, cleft lip and palate; NAM, nasoalveolar molding.

ethnicity, race, and geography. The combined prevalence of orofacial clefts is approximately 1 in 700 live births in Europe, with an ethnic and geographic variation.[12] According to Yılmaz et al.,[13] unilateral CLP was the most common cleft type, seen more on the left side, with patients mostly applying for treatment in university hospitals (64.9%).

Both genetic and environmental factors affect the risk of orofacial clefts. The development of facial structures occurs between the fourth and twelfth weeks of pregnancy, and the left and right sides of the facial structures fuse in the middle of these weeks. If these parts do not fuse properly, craniofacial clefts occur.[14] Cleft lip and palate can be diagnosed during pregnancy with ultrasonography and magnetic resonance imaging.[15]

An accurate prenatal diagnosis of CLP is critical for establishing long-term treatment planning, prognosis, and proper counseling with the parents.[9] The parents need to be informed and counseled about the severity of the cleft, the predicted outcome, and the options for repair by a trained cleft team. Although no intrauterine treatment exists for CLP, both parents and infants benefit from early diagnosis and counseling. The parents may take time to adjust to the reality of the condition and educate themselves about it. The initial shock of the diagnosis can usually be overcome with systematic and planned counseling.[16,17]

Maarse et al.[18] found a large discrepancy among studies, with prenatal detection rates with 2D ultrasound imaging ranging from 9 to 100% for cleft lip with or without cleft palate, 0-22% for cleft palate only, and 0-73% for all types of cleft. Using 3D imaging, the detection rate reached 100% for cleft lip, 86-90% for cleft lip with cleft palate, and 0-89% for cleft palate only. Additionally, Faure et al.[19] performed a study to define the prenatal ultrasound semiology of cleft palate without cleft lip using 3D visualization. They found that an axial transverse ultrasound view and visualization of the secondary fetal palate enables diagnosing a cleft palate without cleft lip. The prenatal diagnosis gave the parents time to manage their feelings, accept the child at birth, and prepare family and friends.[20] Early diagnosis also helped the parents to interact with similar parents and have a better understanding.[21] Clear and consistent information about CLP, possible treatments, and prognosis must be given during initial counseling at cleft centers to reduce anxiety, confusion, and uncertainty.[22,23] Most parents had concerns about the wellbeing of the child and especially the feeding techniques that can be adopted. Prenatal counseling helped to alleviate such concerns and led to more successful parenting.[24,25]

The general treatment protocol for CLP involves presurgical orthopedics, surgical repair of the lip and palate, and treatment of problems related to otology speech, and dental anomalies. Nasoalveolar molding is an important presurgical orthopedic technique for alignment and correction of the nasal cartilage, minimizing the formation of scar tissue and thus producing a more consistent postoperative result.[5] Nasoalveolar molding lengthens the columella, an important factor that can affect the aesthetic and functional results of lip surgery, especially in newborns with bilateral CLP. Eventually, a cleft is easier to repair by using NAM before surgery.[14]

Cleft lip and palate should be treated with a multidisciplinary approach involving many specialists including OB-GYNs, pediatricians, speech therapists, plastic and reconstructive surgeons, and orthodontists. There are cleft centers in some countries where the parents are guided by the cleft teams whereas there are no centers in some countries. The parents may be referred to an orthodontist specialized in cleft care. It is important for OB-GYNs at the birth of a newborn with CLP to refer the patient to an orthodontist for rapid orthopedic treatment before surgery. In addition to their specialty education, CLP centers and societies may organize courses, conferences, and seminars for postgraduates to increase awareness for OB-GYNs. The major importance of NAM is that when used in conjunction with surgical lip repair, it allows a single initial surgery to address the nose, lip, and alveolar complex, thereby reducing the need for secondary surgery.[26] Matsuo et al.[27] concluded that the cartilaginous tissues of newborns are softer and their plasticity is higher due to the level of estrogen transferred from the mother. This plasticity facilitates reshaping discrete fragments. The plasticity of cartilaginous tissues lasts until approximately 3-4 months of age, after which the level of estrogen decreases, and the cartilage regains elasticity, so performing NAM as soon as possible after birth is important.

The results of this study reveal that OB-GYNs in Turkey did not have a good knowledge of presurgical orthopedics and NAM therapy. Fifty-nine of 141 (42%) OB-GYNs had never heard of NAM for newborns with CLP. Thirty-eight (27%) had information about NAM. Thirty-three (23%) referred newborns with CLP for NAM, and 108 (77%) did not. This is an important ratio because the success of presurgical orthopedic treatments is closely related to its commencement as soon as possible after birth. Unfortunately, parents who are not informed after birth and not directed to a specialist for NAM will be deprived of the benefits of presurgical orthopedics.

No statistically significant difference existed in the knowledge level of preoperative treatment of newborns with CLP between experienced and inexperienced OB-GYNs ($P > .05$). Knowledge and awareness of NAM in newborns with CLP among OB-GYNs was insufficient in our sample. We could not compare our findings with the literature since no previous study has evaluated the awareness of OB-GYNs about NAM.

Our study has some limitations. It could be done with a larger sample of participants. Unfortunately, of 532 OB-GYNs, only 141 participated, which is a low rate. This study was also conducted with participants from a single country. Therefore, conducting a study with a greater participation rate, a larger sample size, and more global participants is important.

CONCLUSION

Newborns with CLP should be treated with a multidisciplinary approach that involves many specialists including OB-GYNs. The awareness and knowledge of NAM among OB-GYNs are limited. Commencing the presurgical orthopedic treatment process as soon as possible after the birth of newborns' CLP is crucial for treatment success. Therefore, it is important to increase the awareness of OB-GYNs of CLP treatment alternatives, NAM, and presurgical orthopedic treatment through various courses and seminars during their specialty education.

Ethics Committee Approval: Ethics committee approval was received for this study from the Clinical Research Ethics Committee of Ordu University (No. 2020/220).

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Original Article

Disinfection of Orthodontic Elastomers and Its Effects on Tensile Strength

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Main Points

- Different changes in mechanical properties of elastomeric ligatures (EL) were found between the disinfection methods.
- Seventy percent of alcohol showed negative changes in all mechanical properties of orthodontic elastics when immersed for 30 minutes.
- Two percent glutaraldehyde did not cause significant changes in the mechanical properties of orthodontic elastics when immersed for 30 minutes.

ABSTRACT

Objective: This study aimed to investigate the effect of different disinfection protocols on the mechanical properties of orthodontic elastomeric ligatures (EL), an important issue to biosafety improvement and infection control, and to avoid cross-contamination.

Methods: A total of 120 EL were randomly divided into 6 experimental groups (n = 20) according to the disinfection method employed: group 1, EL were not immersed in a disinfectant solution (control group); group 2, EL were immersed in 2% glutaraldehyde; group 3, EL were immersed in 70% alcohol solution; group 4, EL were cleaned in an ultrasound washing (UW) machine by immersion in 0.5% enzyme detergent solution; group 5, UW procedure was performed, followed by immersion in 2% glutaraldehyde; Group 6, UW procedure was performed, followed by immersion in 70% alcohol. After disinfection, EL were subjected to a tensile strength test where the maximum strength, maximum elongation, and work at failure were determined. Data were statistically evaluated using one-way ANOVA and Dunnett's *t*-test for multiple comparisons.

Results: Statistically significant different ($P < .05$) values were found between the disinfection methods, and 70% alcohol showed negative changes in all mechanical properties of orthodontic elastomers. By contrast, 2% glutaraldehyde did not show significant alteration in mechanical properties, whereas the UW procedure showed significant alteration in maximum strength and work at failure.

Conclusion: Of the tested substances for disinfection, 2% glutaraldehyde was the only substance that did not cause significant changes in the mechanical properties of orthodontic elastics and is considered as an alternative for elastic disinfection before its use.

Keywords: Disinfection methods, orthodontics elastomers, mechanical phenomena

INTRODUCTION

Given the increasing incidence of transmissible diseases, infection control and biosafety recently came into focus, as they have not been subjected to strict criteria as they are today.[1-4] As a result, sterilization and disinfection measures are routinely adopted to avoid cross-contamination.[1,3,4] The recent spread of coronavirus disease has gripped the entire international community and caused widespread public health concerns.[5] Global efforts have been taken to prevent COVID-19 and help control its epidemic.[6] Since then, disinfection control measures have been highlighted.[3,6]

Polyurethane elastomers are widely used in orthodontics as ligatures and elastic chains.[2,7] They are structurally organized by a long polymeric chain, presenting relatively weak forces of attraction between them.[2] Elastomeric

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ligatures (EL) are mainly used to tie the archwire, which transfers the forces needed for tooth movement, to the brackets.[8,9] Their elastic properties, easiness of application, and cost-efficiency make them a very important component in orthodontic treatment.[8,9] Nevertheless, EL quickly degrades in the oral cavity and may present substantial alterations in their physical and mechanical proprieties.[8,10] The main causes of the quick degradation are related to rapid breakage of polyurethane molecular crosslinks.[11]

Numerous studies have evaluated the strength of elastomers, in terms of force delivery and rate of force decay in various environments and different testing conditions.[2,7-9,11-13] Factors such as the action of salivary enzymes, humidity of the oral environment, pH, and temperature variation, contact with masticatory forces, use of mouthwashes, and bacterial biofilm buildup have all been associated with elastomer deformation and force degradation behavior.[9,11,14]

Elastomeric ligatures are considered semi-critical dental materials that need cold sterilization since they are not resistant to heat.[1] A high level of disinfection should proceed through the destruction or inactivation of potential microorganisms and their contaminants.[1,8] This process includes cleaning, disinfection, and storage.[15] Unfortunately, elastomers undergo degradation when subjected to repeated disinfection. The effects of disinfection on the mechanical proprieties of EL have been investigated in several studies.[2,7-10,12,13,16,17] However, little information exists on the effects of disinfection methods on tensile strength.[8,9] Thus, this study aimed to assess the effect of different disinfection protocols on the mechanical properties of orthodontic EL.

METHODS

This study was conducted in the Dental Materials Laboratory of the School of Health and Life Sciences of the Pontifical Catholic University of Rio Grande do Sul (PUCRS). The Scientific Commission of Dentistry School approved its implementation, under registration number 0023/11.

A total of 120 crystal-colored EL (Morelli, Sorocaba, SP), all within the expiry date, were used. They were stored according to the manufacturer's instructions up to the time of use. The EL were randomly divided into 6 experimental groups (N = 20), according to the disinfection methods employed (Table 1): group 1, EL were not immersed in a disinfectant solution (control group); group 2, EL were immersed in 2% glutaraldehyde for 30 minutes; group 3, EL were immersed in 70% alcohol solution for 30 minutes; group 4, EL were cleaned in an ultrasound washing (UW) machine (Sercon, São Paulo, Brazil) by immersion in 0.5% enzyme detergent solution (Riozime III, Rioquímica, Rio de Janeiro, Brazil) for 10 minutes; group 5, UW procedure was performed, followed by immersion in 2% glutaraldehyde for 30 minutes; group 6, UW procedure was performed, followed by immersion in 70% alcohol solution for 30 minutes. After the UW procedure, EL were washed with purified water for 5 minutes and dried with absorbent paper. The washing and drying procedure was repeated after 30 minutes in all ligatures submitted to disinfection for the complete removal of disinfectant residues. All samples were stored in closed test tubes at room temperature for 7 days before the tensile strength test.

Table 1. Method of disinfection employed

Group	Treatment
1	Control (not immersed in any solution)
2	2% glutaraldehyde immersion for 30 minutes.
3	70% Alcohol immersion for 30 minutes.
4	Ultrasound washing for 10 minutes with enzyme detergent.
5	Ultrasound washing and 2% glutaraldehyde immersion for 30 minutes.
6	Ultrasound washing and 70% Alcohol immersion for 30 minutes.

Tensile Strength Test

The tensile strength test was performed immediately after the samples were removed from the test tubes. The tensile strength test was carried out using a pair of stainless steel hooks (0.032 inches in diameter) attached to the fixed and movable crossheads of a universal testing machine (EMIC, DL 10000 Brazil). Elastomeric ligatures were attached to the pair of hooks and stretched until fracture occurred. Each ligature was loaded in tension at a crosshead speed of 5 mm/min and load cell of 50 N, according to the recommendation of a previous study.[18] Each side of the movable crosshead was previously adjusted to ensure no initial distention. The same pair of hooks was employed in all experiments. The tensile strength test measurements were not normalized by cross-section area. The tensile and manufacturing properties of orthodontics elastomers were the same in all specimens in the present study.

The load–extension curve was recorded graphically, and the maximum strength, maximum elongation, and work at failure were determined. Data were calculated and processed by EMIC DL software. Maximum strength was defined as the maximal tension registered, obtained by the peak of the load–extension curve. The work at failure was measured as the total area under the load–extension curve from 0 to the maximum strength, corresponding to the amount of energy required to fracture the material. Maximum elongation was defined by the amount of extension necessary to reach the maximum strength, corresponding to the longitudinal distance between the 0 point and the more distant point in the load–extension curve. Maximum strength was recorded in Newton (N). Work at failure was recorded in Newton per millimeter (N/mm), whereas maximum elongation was recorded in millimeters (mm).

Statistical Analysis

An exploratory analysis was performed to evaluate data related to the studied variables. The differences among the groups were determined statistically after confirmation of normality (Kolmogorov–Smirnov) and homoscedasticity (Kendall–Stuart test). One-way analysis of variance (ANOVA) and Dunnett's *t*-test were performed to find differences between the maximum strength, maximum elongation, and work at failure. The assessed factors were the disinfection treatment and UW procedures. Data were statistically analyzed using SPSS 17.0, based on a significance level of 0.05%.

Table 2. Comparison of maximum srength and elongation and work at failure between groups with ANOVA

	Maximum Strength* Mean ± SD	Maximum Elongation** Mean ± SD	Work at Failure*** Mean ± SD
Group 1	21.39 ± 3.35	17.27 ± 0.76	16.81 ± 4.85
Group 2	22.58 ± 2.88	17.11 ± 0.60	20.78 ± 4.63
Group 3	16.94 ± 3.50	15.59 ± 1.30	14.36 ± 5.70
Group 4	21.93 ± 3.05	14.64 ± 0.54	20.32 ± 4.24
Group 5	19.93 ± 1.86	15.13 ± 0.61	17.16 ± 4.93
Group 6	22.00 ± 3.71	14.22 ± 0.51	20.56 ± 5.31
ANOVA's *P*	<.01	<.01	<.01

*Maximum strength is expressed in N. **Maximum elongation is expressed in mm. ***Work at failure is expressed in N/mm. SD, standard deviation.

RESULTS

Tensile strength test results are descriptively presented by the mean and standard deviation for each group, as shown in Table 2.

Comparison of maximum strength, elongation, and work at failure between groups are presented in Tables 2 and 3, respectively by one-way ANOVA and Dunnett's *t*-test.

Maximum Strength

According to the obtained results, a statistically significant difference ($P < .05$) in maximum strength values was found among the disinfection methods. Dunnett's Multiple comparisons showed that Group 3 had a statistically significant decrease ($P < .05$) in the maximum strength (16.94 ± 3.50 SD), when compared with the control group (21.39 ± 3.35 SD). Group 5 presented a decrease in the mean values (19.93 ± 1.86 SD) when compared with the control group, but was not statistically significant ($P > .05$). Groups 2 (22.58 ± 2.88 SD), 4 (21.93 ± 3.05 SD), and 6 (22.00 ± 3.71 SD) had a slight increase when compared with the control group (21.39 ± 3.35 SD) but not statistically significant ($P > .05$) (Tables 2 and 3).

Maximum Elongation

Statistically significant difference was found between the disinfection procedures when analyzing the maximum elongation ($P < .05$). The groups showed a decrease of elasticity and the differences were statistically significant for all groups against control, except for group 2 (Tables 2 and 3).

Work at Failure

Statistically significant differences were found between the disinfection procedures ($P < .05$). However, Dunnett's test was not able to show statistically significant differences between the disinfection procedures against the control as can be seen in Tables 2 and 3.

DISCUSSION

The longer the exposure of an item to a disinfectant, all contaminating microorganisms will be more likely inactivated.[5,8,9,19] Unfortunately, with extended exposure to disinfectant solutions, some sensitive materials used in orthodontics may more likely cause degradation of mechanical and physical properties.[10,11,20-22]

Table 3. Comparison of disinfection groups with control group by Dunnett *t*-Test

Multiple Comparisons						95% CI	
Variable	Group (I)	Groups (J)	Mean Difference (I-J)	Std. Error	Sig.	Lower Bound	Upper Bound
Maximum Strength	Control	Glutaraldehyde 2%	−1.19	0.98	0.83	−4.05	1.66
		Alcohol 70%	4.46*	0.98	0	1.6	7.31
		UW	−0.54	0.98	0.99	−3.39	2.32
		UW+ Alcohol 70%	−0.61	0.98	0.98	−3.46	2.24
		UW + Glutaraldehyde 2%	1.44	0.98	0.69	−1.42	4.29
Maximum Elongation	Control	Glutaraldehyde 2%	0.16	0.24	0.98	−0.55	0.86
		Alcohol 70%	1.68*	0.24	0	0.97	2.39
		UW	2.58*	0.24	0	1.87	3.29
		UW+ Alcohol 70%	3.05*	0.24	0	2.34	3.76
		UW + Glutaraldehyde 2%	2.13*	0.24	0	1.42	2.84
Work at Failure	Control	Glutaraldehyde 2%	−3.97	1.59	0.13	−8.58	0.64
		Alcohol 70%	2.45	1.59	0.63	−2.16	7.05
		UW	−3.29	1.59	0.31	−7.90	1.32
		UW+ Alcohol 70%	−3.75	1.59	0.17	−8.36	0.85
		UW + Glutaraldehyde 2%	−0.35	1.59	1	−4.95	4.26

*The mean difference is significant at the .05 level.
UW, ultrasound washing; CI, confidence interval.

Many disinfectant agents have been employed in the dental office with regard to orthodontic ligatures.[1,10] Notwithstanding, few studies have investigated the effect of washing and disinfection on the mechanical properties of orthodontic ligatures.

This study demonstrated a change in the tensile behavior of EL according to the treatment received. Similar results were found by a study evaluating the effect of 2 disinfectant solutions on 3 orthodontic ligature brands, which were exposed from 1 hour until 28 days.[9] The authors further concluded that the brand and immersion time influenced the elastic resistance of the material.[9]

Singh et al.[8] evaluated the effect of extended exposure on the tensile load at failure of different orthodontic EL to 3 disinfectant solutions. The disinfectant solutions were 1.5% glutaraldehyde, 2% glutaraldehyde, and ortho-phthalaldehyde. Compared with unexposed specimens, the behavior of all EL in terms of tensile load at failure was different according to the disinfectant solutions used.[8]

Most of the orthodontic EL currently available had similar fabrication methods.[8] However, according to Evangelista et al.,[9] significant differences exist between manufactured ligatures mainly in terms of the glass transition temperature. A higher glass transition temperature, which indicates a more rigid polymer, is associated with higher tensile strength. Tensile strength is an important property of elastic ligatures because consistent force delivery is needed to sustain full engagement of the archwires in the bracket slot for tooth movement.[8] The finding of a decrease in tensile strength after exposure to the disinfectant solution in the present study has paralleled the results of Singh et al.[8]

The 70% alcohol significantly affected the maximum strength, maximum elongation, and work at failure. These results may be attributed to the fact that synthetic elastomers (polymers) are very sensitive to the effects of free-radical generating systems, notably ozone and oxygen.[11] The exposure to free radicals results in a decrease in the flexibility and tensile strength of the polymer.[8,11] Scission of macromolecule chains is responsible for the chemical degradation in the couple bond between carbon atoms.[11]

Polyurethane elastomers have the ability to act both as donor H through the HN group or simultaneously as receptor H through the C = O group.[23] This phenomenon may be associated with superficial changes because reactivity increased with the use of 70% alcohol. Kim and Lee[24] investigated the color change in EL, and their finding suggested that alcohol solutions cause superficial chemical degradation affecting its color in the early hours of immersion by plasticizer leaching. Similar color changes were found in aesthetic EL.[25,26]

Glutaraldehyde is recognized by the Food and Drug Administration (FDA) as a high-degree disinfection agent.[19] Evangelista et al.[9] observed time dependency and progressive degradation on elastomers immersed in glutaraldehyde solution. They assumed that the active compound product and water act by plasticizing the elastic polymer and cause a polymeric chain to slip past each other. Despite that, the present study demonstrated that 2% glutaraldehyde did not significantly influence maximum strength and maximum elongation. This can be explained by the short immersion period. While in the present study the elastics were exposed to the disinfectant solution for 30 minutes, in Evangelista et al.'s[9] study, they were exposed up to 28 days.

The UW procedure with an enzyme detergent showed to affect the maximum strength and maximum elongation of orthodontic ligatures. While the maximum strength increased, the maximum elongation decreased. Those results demonstrate that a reduction in elastic property is associated with an increment in the stiffness of elastomers. Similar results were found in a study evaluating the behavior of elastomers exposed to different disinfectant solutions.[20] In the study by Nattrass et al.,[20] the loss of the strength of the elastic chain, when kept tight in different media storage, was measured after 7 days. A maximum strength increment was attributed to an increased rigidity for the incorporation of the material solution. The same cannot be observed in samples that remained without immersion. When changes in the elasticity of elastomers stored in water were assessed, it was found that the leaching process that occurred with the material was time-dependent. The exposure to liquid allowed the weakening of non-covalent bonds and subsequently degraded the elastomer.[21,27] This may be true depending on the type of agent used. Thus, cross-infection control using liquid agents should have the shortest possible immersion time. In the present study, compared with the non-UW groups, the groups that underwent longer UW in aqueous solution with detergent showed reduced elasticity. This finding suggests that the aqueous component or the chemical substances in the disinfectant solution may plasticize or cause degradation of the elastomers.

There is a need for future studies comparing different EL and time of exposure to disinfectant solutions. Time dependency and manufacturing characteristics are, without any doubt, essential to fully understand the mechanical properties of EL. However, as previously mentioned, this study mainly focused on the effect of washing and disinfection procedures for 30 minutes on the mechanical properties of orthodontic ligatures, which are important factors in this time of coronavirus pandemic.

CONCLUSION

Based on the results of this study, 2% glutaraldehyde can be used for disinfection of orthodontic EL before its use, which showed no significant influence on maximum strength, maximum elongation, and work at failure. On the contrary, 70% alcohol and UW procedure should be avoided as they significantly influence the mechanical properties of EL.

Ethics Committee Approval: Ethics committee approval was received for this study from the Ethics Committee of the Scientific Commission of the School of Health and Life Sciences of the Pontifical Catholic University of Rio Grande do Sul (PUCRS) (Protocol number 0023/11).

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Conflict of Interest: The authors have no conflicts of interest to declare.

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Original Article

Effects of Tooth Brushing and MouthWashing on Leaching Bisphenol A Levels From an Orthodontic Adhesive: An In Vitro Study

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Main Points

- Bisphenol A (BPA) release from orthodontic adhesive samples after tooth brushing and mouth washing was investigated.
- Detected levels of BPA were found to be lower than referenced tolerable daily intake levels.
- Tooth brushing did not significantly affect the leaching BPA amount.
- Increased levels of leached BPA with alcohol-containing mouth washing solutions should be considered during fixed orthodontic treatment.

ABSTRACT

Objective: To assess the levels of bisphenol A (BPA) released from an orthodontic adhesive with respect to the effects of tooth brushing and mouth washing.

Methods: Three groups, each containing fifteen adhesive samples were prepared. In Group 1, samples were polymerized according to manufacturer instructions. In Group 2, after the same polymerization protocol, each sample was brushed with a fluoride-containing toothpaste. For Group 3, samples were immersed in a mouth washing solution after polymerization. Later, all samples were placed into glass tubes containing 5 mL distilled water. High-performance liquid chromatography (HPLC) measurements were performed to assess the leaching amount of BPA. Intergroup comparison was performed by one way ANOVA test.

Results: Mean amounts of BPA were found to be 0.2674 μg/L, 0.2692 μg/L, and 0.2705 μg/L, respectively. Only a significant difference was found between Group 1 and 3 ($P < .01$), revealing higher BPA levels with the mouth washing solution.

Conclusion: Measurable amounts of BPA release were observed in all groups of orthodontic adhesive samples, but the detected amounts were below the toxic levels. From a clinical point of view, alcohol-containing mouth washing solutions might increase the amount of leaching monomer, since alcohol is solvent of BPA.

Keywords: Bisphenol A, orthodontic adhesives, leaching, tooth brushing, mouth washing

INTRODUCTION

In recent years, peoples' concerns for the release of some chemicals from cosmetic and food products, kitchen tools, and/or various intraoral appliances have steadily been increasing. This is an important issue in terms of general public health. Endocrine-disrupting substances are chemicals that alter the development and functioning of the endocrine system.[1] They can be found in nature, synthetic and industrial products,[2] and affect production, release, binding, transport, activity, destruction, and excretion of hormones. One or more of these effects may be combined and clinical findings are based on the sum of all effects.[3] Some substances are known to cause impairments in the endocrine system those are polychlorinated biphenyls, plastic-associated and household products, pesticides, and heavy metals.[4]

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Among endocrine disrupters, bisphenol A (BPA) is a colorless and solid chemical that is well soluble in organic solvents but slightly soluble in water.[5] It is used in the manufacturing of polycarbonates for hard plastic products and epoxy resin linings for metal food and beverage cans.[6] Due to its wide application range, the demand and production for BPA are increasing year by year. It has been discovered that BPA is an artificial estrogen and can interact with other endocrine receptors such as the thyroid hormone receptor.[7] In the human body, it has been determined in blood, urine, breast milk, and some other tissues.[8]

Bisphenol A is not a component of dental products by purely itself but its derivates are used in dental materials. Therefore, BPA could be a clinical concern in dentistry, since exposure into the oral cavity can occur especially due to changes in intraoral temperature, salivary enzymatic structure, pH, or mechanical wear.[9,10] Commonly used BPA-derivatives in dentistry are bisphenol A diglycidylmethacrylate (Bis-GMA), ethoxylatedbisphenol A glycoldimethacrylate (Bis-EMA), and bisphenol A dimethacrylate (Bis-DMA).[11] It is stated that toxic results can occur as a result of the dissolution of non-polymerized BPA monomers from resin-based dental materials; some studies were conducted in the field of orthodontics as well.[12-15]

This should be particularly taken into consideration for orthodontics, because early treatment is usually performed in most cases, declaring that most of the orthodontic patients are children and adolescents.[16] Epidemiological and animal studies suggested that endocrine disruptors have adverse effects on birth weight, reproductive tract development and promote childhood obesity formation.[17]

Malkiewicz et al.[12] and Kloukos et al.[18] suggested that mouth rinsing can be useful to prevent exposure to the potential hazard of leaching monomers after orthodontic bracket placement. In a relevant systematic review, several recommendations have been made to reduce the amount of exposed BPA.[19] However, there is still no sufficient knowledge regarding a clinical way to prevent or decrease the possible release of BPA after orthodontic bonding. Therefore the aim of this in-vitro study was to assess the levels of BPA released from an orthodontic adhesive by evaluating the effects of tooth brushing and mouth washing.

METHODS

Ethical consent was not required since this was an experimental study. Three groups, each consisting fifteen samples of bonding adhesives were prepared at room temperature. All of the samples were prepared according to the experimental design by the same researcher at the same laboratory and under the same conditions. Samples were prepared in molds, made of Teflon matrices that were 5 mm in diameter and 2 mm in thickness, based on previous research.[12] Transbond XT Primer and Transbond XT Light Curing Adhesive (3M Unitek, Monrovia CA, USA) were chosen as primer and adhesive (Figure 1a). Primer was gently applied as a single layer with the help of an application brush (Figure 1b). Attention was paid to avoid exaggerated application. LED light source with 420-480 nm wavelength (COXO® DB-686 Cappu LED Curing Light; Foshan Coxo Medıcal Instrument CO. LTD, Guangdong Province, China) was used for photopolymerization. Light-curing was performed directly from the upper surface of the samples for 10 s.

Preparation of the Samples

The experiment procedures related to the preparation of the samples are shown in Figure 2.

In Group 1, adhesive samples were prepared, primer was applied gently with an application brush on each sample, and were polymerized. Samples were placed into glass tubes containing 5 mL distilled water (Figure 2a).

In Group 2, the polymerization process was performed in the same way as in Group 1. Then, each sample was manually brushed with a fluoride-containing toothpaste (Colgate® Triple Action Toothpaste; Colgate-Palmolive Company) and a toothbrush (Curaprox; Curaden International AG, Kriens, Switzerland). Standardization of the brushing force was targeted through manual brushing performed by the same researcher (BA), who was a right-handed 29-year-old woman, with gentle scrabbing

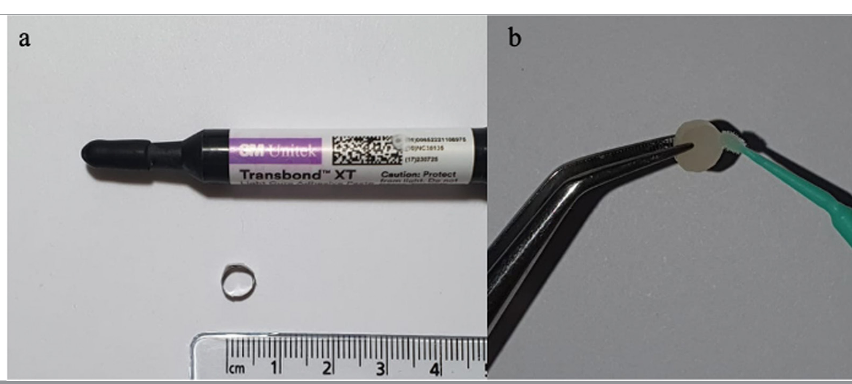

Figure 1. a, b. (a) Preparation of the adhesive samples (5 mm in diameter and 2 mm in thickness); (b) Application of primer

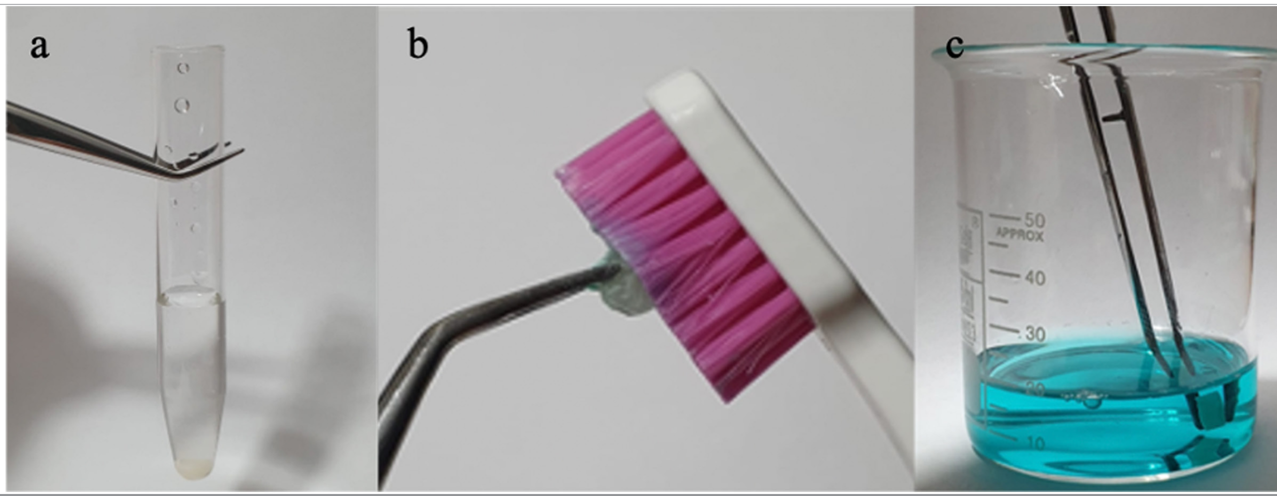

Figure 2. a-c. (a) Adhesive sample placed into glass tubes containing 5 mL distilled water in Group 1; (b) Adhesive for review the only sample manually brushed with a fluoride-containing toothpaste in Group 2; (c) Adhesive sample immersed and moved physically in 20 mL of mouth washing solution in Group 3

on the adhesive surfaces for 10 s. Later, samples were rinsed with water and placed in a glass tube containing 5 mL of distilled water (Figure 2b).

In Group 3, polymerization was performed with the same protocol. Then, each sample was immersed and moved physically in 20 mL of mouth washing solution (Cool Mint Listerine®; Johnson & Johnson Consumer Inc) in circular motions for 30 s and placed into a glass tube containing 5 mL of distilled water (Figure 2c).

In the second and third groups, a new glass of water and mouthwash was used with each sample. During all experimental procedures, samples were held with a dental tweezer from a single point, and all surfaces were included in application procedures. Care was taken not to use any plastic material to prevent any monomer interference. High-performance liquid chromotography (HPLC) measurements were performed 1 h after the placement of composite samples to glass tubes with distilled water. Ingredients of the orthodontic primer and adhesive, toothpaste, and mouth washing solution are given in Table 1.

Chemical Analysis

High-performance liquid chromotography was used for the determination of BPA in samples. The HPLC system consisted of Shimadzu LC-2010 (Shimadzu Corp., Kyoto, Japan) and Shimadzu HPLC solution software. Chromatographic separation was carried out on a C18 column (Phenomenex, 150 × 4.6 mm, 5 µm particle size), using a gradient solvent system comprising 80% acetonitrile acid (A) (for HPLC, gradient grade, ≥99.9%, Sigma-Aldrich) and 20% MeOH (B) (for HPLC, gradient grade, ≥99.9%, Sigma-Aldrich). Gradient profile: 0-20 min. The flow rate was adjusted to 1.0 mL/min, the detection wavelength was 228 nm and 2 µL of each sample was injected. All separations were performed at 25°C. Standard solutions were processed in a device according to this method and the time of peak for BPA was determined. The linear fittings of the calibration curves were used to calculate the concentration of BPA in the polymer solution based on the area of chromatographic peaks at the corresponding retention time. High-performance liquid chromotography assays were performed in triplicate for each time period, and the results were averaged. Under the conditions of the experiment, the detection threshold of BPA was estimated at 0.1 ppm (0.1 mg/L). Bisphenol concentration of the samples was determined by taking chromatograms on the HPLC instrument by autosampler.

Statistical Analysis

Data analysis was performed using the Statistical Package for Social Sciences software (SPSS, Version 21.0, USA). The Shapiro–Wilk test was used to determine the homogeneity of the data. Descriptive statistics were presented as mean and standard

Table 1. Ingredients of orthodontic primer and adhesive, toothpaste and antiseptic mouthwashing solution

	Ingredients
Transbond XT Primer	Bisphenol A Diglycidyl Ether Dimethacrylate (BISGMA), Triethylene Glycol Dimethacrylate (TEGDMA), 4-(Dimethylamino)-Benzeneethanol
Transbond XT Light Curing Adhesive	Silane Treated Quartz, Bisphenol A Diglycidyl Ether Dimethacrylate (BISGMA), Bisphenol A Dimethacrylate, Silane Treated Silica, Diphenyliodonium Hexafluorophosphate, Triphenylantimony
Colgate® Triple Action Toothpaste	Sodium Monofluorophosphate, Glycerin, Hydrated Silica, Water, Sodium Bicarbonate, PEG-12, Sodium Lauryl Sulfate, Flavor, Sodium Hydroxide, Cellulose Gum, Carrageenan, Sodium Saccharin, FD&C Blue No 1., D&C Yellow No.10
Cool Mint Listerine®	Eucalyptol, Menthol, Methyl Salicylate, Thymol, Water, Alcohol, Sorbitol, Poloxamer, Benzoid Acid, Sodium Saccharin, Sodium Benzoate, Flavor, Green 3

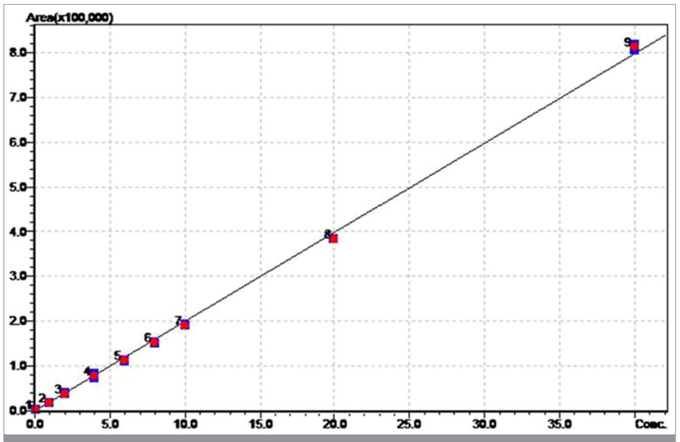

Figure 3. Standard calibration curve for high-performance liquid chromatography for detection of Bisphenol A in the polymer solution

deviation. Intergroup comparison was performed by one way ANOVA test. A *P* value of < .05 was considered to be statistically significant.

RESULTS

Bisphenol A amount in distilled water was analyzed with HPLC. The standard calibration curve for HPLC of detection of BPA was given in Figure 3. The calibration method was used for the determination of BPA in the prepared solution sample. The peak intensities that vary linearly with BPA concentration were given in the calibration curve, and it was proved appropriate that peak areas change linearly with concentration. In addition, the fact that the regression number was close to 1 ($R^2 > 0.99$) made it possible to analyze BPA with this method.

Descriptive statistics and comparison between groups are given in Table 2. The results declared that measurable amounts of BPA release were observed in all groups. Mean amounts of BPA was found to be 0.2674 , 0.2692, and 0.2705 µg/L, respectively in Group 1 to 3. The highest amount of BPA release was found in Group 3, whereas the lowest was detected in Group 1.

According to one way ANOVA test, no statistically significant differences were found between Group 1 and Group 2, as well as between Group 2 and Group 3. However, there was a significant difference between Group 1 and Group 3 ($P < .01$), declaring significantly higher BPA levels in mouth washing group.

DISCUSSION

Studies on the leaching of BPA monomer from dental materials were first performed on dental sealants in the 1990s.[20] In this in-vitro study we aimed to evaluate the amount of BPA released from a standard orthodontic adhesive under the influence of tooth brushing and mouth washing. Although tooth brushing did not reveal significant differences relative to the leaching BPA amounts, increased levels were noted with the mouth washing solution. Previous studies have shown that BPA was released into the oral environment after adhesive polymerization and it was noted that the highest BPA release occurred immediately after polymerization.[12,21,22] Inline, measurements were made immediately after polymerization in our study.

The tolerable daily intake level of BPA detected by the European Food Safety Authority is limited to 50 mcg/kg bw/day.[23] The current results declared that the detected BPA levels were considerably lower than the mentioned tolerable daily intake levels. Furthermore, all the adhesive surfaces were in contact with distilled water in this study, but in reality, only the adhesive parts at the bracket margins are in contact with the oral cavity. Previous studies mentioned the low dose effects of BPA.[24,25] Despite the small amounts of this chemical, long treatment durations of fixed orthodontic appliances might reveal clinical importance in this sense. Therefore, even small amounts of monomer may be considered to be important. It was mentioned that rinsing the mouth with water after orthodontic bonding could be useful to take the excess monomer out.[12,18] It is obvious that this process can be beneficial since the results of our study also showed measurable amounts of BPA. In addition, it can be explained that pure water was preferred in our study to avoid the interference effects of different substances in tap water.

Current results showed that brushing teeth with a fluorized toothpaste did not seem to affect the amount of leaching BPA monomer when compared to Group 1, in which samples had no additional process after polymerization. However, it was noted that antiseptic mouthwash can increase the amount significantly. This finding might be attributed to the fact that most of the mouth washing solutions—including the one used in this study—contain alcohol, which is an organic solvent for BPA.[25] Eliades et al.[13] investigated the effect of alcohol-containing rinsing solution and water on the release of BPA after orthodontic bonding in an in vivo study and demonstrated higher levels of BPA in patients who only rinsed their mouth with water, which was against the result they expected. They

Table 2. Descriptive values of data and comparison between groups with one way ANOVA test

	Group 1	Group 2	Group 3	*P* between Groups		
	Mean ± SD	Mean ± SD	Mean ± SD	Group 1-Group 2	Group 1-Group 3	Group 2-Group 3
BPA levels (mg/L)	0.2674 ± 0.000632	0.2692 ± 0.002549	0.2705 ± 0.003796	.146	.007*	.402

Group 1, samples were placed into pure water after polymerization.
Group 2, samples were brushed with a fluoride-containing tooth-paste, rinsed, and placed in pure water after polymerization.
Group 3, samples were immersed in mouth washing solution and placed in pure water after polymerization.
*$P < .05$.
SD, standard deviation; BPA, bisphenol A.

related their results to the reduced contact of the adhesive with the solution, which was limited with bracket–enamel margin, and also to the insufficient rinsing of patients due to the taste of ethanolic solutions.

Since alcoholic compounds contain a hydrocarbon group instead of hydrogen atom in water, alcohol is chemically less polar solvent than water. While the hydrocarbon part of alcohol gets van der Waals interaction with the hydrocarbon part of BPA, hydroxyl group of alcohol makes a hydrogen bond with the hydroxyl group of BPA which enhances the solubility of BPA in alcoholic compounds. Hence, using an alcohol-containing mouthwash after orthodontic bonding may increase the amount of BPA in the oral cavity. Many studies have been performed with mouth washing solutions and the results declared that these products provide antimicrobial and antigingivitis activities.[26-28] On the other hand, it should be considered that systematic toxicological studies have to be conducted to sustain the total safety of these products. Since in vivo reactions could provide different results, future studies with alternative strategies should be investigated to provide better knowledge on this topic.

The study had some limitations. First, the measurements were performed with HPLC at only 1 time interval, but the recurring brushing and rinsing procedures were not simulated, therefore further researches could be planned in this regard. Second, the clinical practice of bonding was not fully imitated in this experiment. Again, BPA release with different adhesives and mouth washing solutions could be addressed in the future.

CONCLUSION

The present in vitro study provided a basis for the measurable amounts of BPA release with orthodontic adhesive samples. However, the detected levels of BPA were below the referenced tolerable daily intake levels. Results declared no significant difference relative to the leaching BPA amounts. However, orthodontists should consider the increased levels of leached BPA with alcohol-containing mouth washing solutions during fixed orthodontic treatment.

Ethics Committee Approval: Ethical consent was not required since this was an experimental study.

Informed Consent: N/A.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - B.B.T., C.T., M.Ç.U.; Design - B.B.T., C.T., M.Ç.U.; Supervision - B.B.T., C.T., M.Ç.U.; Fundings - B.A.; Materials - B.A., E.Y., O.C.B., M.Ç.U.; Data Collection and/or Processing - B.A., E.Y., O.C.B., M.Ç.U.; Analysis and/or Interpretation - B.B.T., C.T., E.Y., O.C.B., M.Ç.U.; Literature Review - B.A., B.B.T., C.T., M.Ç.U.; Writing - B.A., B.B.T., C.T., M.Ç.U.; Critical Review - B.B.T., C.T., M.Ç.U.

Conflict of Interest: The authors have no conflicts of interest to declare.

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Original Article

The Effects of Increased Maxillary Canine Bracket Angulation on Tooth Movement and Alignment Efficiency: A Prospective Clinical Study

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Main Points

- Orthodontic bracket prescription was significant for the direction and amount of tooth movement.
- The distal movement of the canine was significantly higher in the control group than in the study group in treatment involving premolar extraction.
- Different canine bracket angulations had similar effects on crowding correction.

ABSTRACT

Objective: This study aimed to evaluate the effects of a 10° angulation of a maxillary canine (Mx3) bracket on Mx3 and maxillary central incisor (Mx1) tooth movements and alignment efficiency in treatments involving maxillary premolar extraction.

Methods: This split-mouth study included 29 individuals in a +10° angulation study group and a 0° angulation control group. The initial (T0) and 12th week (T1) orthodontic models were prepared and digitized with a three-dimensional scanner and superimposed using the OrthoAnalyzer analysis program. The movements of Mx3 and Mx1 were measured, and the alignment efficiency was assessed using Little's Irregularity Index. The Shapiro–Wilk test was used to test the data for a normal distribution. T1 and T0 measurements within the group and T1 and T0 differences between groups were compared using a paired samples *t*-test. The significance level was set to $P < .05$.

Results: The linear movements of Mx3 in the distal direction significantly increased, and Little's Irregularity Index values statistically significantly decreased in both groups. Distal movements of Mx3 were significantly higher in the control group than in the study group ($P < 0.01$ and $P < 0.05$). The movements of Mx1 and Little's Irregularity Index measurements did not show statistically significant differences between the groups ($P > 0.05$).

Conclusion: A +10° Mx3 bracket angulation increased the inclination of Mx3 to the mesial, but it decreased the Mx3 distal linear movement and the retraction of Mx1, with no difference in terms of alignment efficiency between the + 10° and 0° Mx3 bracket angulations.

Keywords: Maxillary canine, bracket angulation, alignment, tooth movement

INTRODUCTION

Over the years, clinicians have worked on bracket design to achieve the optimum aesthetics and functionality goals of orthodontic treatment.[1] In 1972, Andrews introduced the straight-wire technique with pre-adjusted brackets that included angulation, inclination, and in–out values in the bracket design.[2] Slot angulation is an important component for obtaining dental positions suitable for Andrews' 6 keys to normal occlusion.[3] Since then, different straight-wire bracket prescriptions have been introduced by Roth,[4] Alexander,[5] McLaughlin, Bennett and Trevisi (MBT),[6] and Capelozza.[7] These prescriptions have different angulation values for the maxillary

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canine (Mx3) brackets. The Mx3 angulation was 11° in Andrews' prescription[2] and 10° in Alexander's prescription.[5] In Roth's system, the Mx3 angulation was increased to 13° to facilitate canine guidance.[4] The MBT[6] and Capelozza[7] procedures used an Mx3 bracket angulation of 8° to achieve a more favorable relationship between Mx3 and the first premolar roots.[7,8]

Protrusive incisors and anterior crowding due to lack of space are the main indications for orthodontic treatment with first premolar extraction.[5] In previous studies, different Mx3 bracket angulations were evaluated for anchorage loss,[9] anterior tooth positions,[1,10] and dental arch perimeter.[11] Based on these studies, changes in the amount of tooth movement and position caused by different bracket angulations were considered to increase the alignment efficiency, which is related to the features of the preadjusted appliances.[3] Therefore, the present study evaluated the effects of Mx3 bracket angulation of 10° on Mx3 and maxillary central incisor (Mx1) tooth movements and alignment efficiency, in treatments involving maxillary first premolar extraction.

METHODS

The study protocol was reviewed and approved by the ethics review board of the Ondokuz Mayıs University Clinical Research Ethics Committee, resolution number OMÜ KAEK—2016/336. This was a single-center prospective clinical study, with a single operator (MT) participating in the orthodontic treatment of the patients. A signed informed patient consent form was obtained from all patients. The individuals included in the study had the following conditions:

- Patients with maxillary anterior crowding due to lack of space
- Indication of fixed orthodontic treatment with moderate anchorage in the upper dental arch and extraction of the maxillary right–left 1st premolars
- Both with and without an indication for extraction of mandibular right–left 1st premolars
- In permanent dentition
- No missing teeth
- No systemic diseases
- Not on any medication
- Good oral hygiene

The average age of the 29 participants (20 females and 9 males) was 15 years and 3 months (13 years and 8 months to 17 years and 10 months) (Table 1). The study groups were formed using the split-mouth method used in many studies,[12,13,14] as it produces more reliable data than those generated by comparing the variables on different patients. This design eliminated the differences due to gender, age, and other individual characteristics in the study participants. All individuals were bonded with Mx3 brackets with a +10° angulation in one-half of the upper dental arch (study group) and a 0° angulation in the other half of the upper dental arch (control group). The right–left direction distribution of the groups was conducted through simple randomization, including the use of a shuffled deck of bracket prescription cards. Prescription 1, with 0º in the left Mx3 bracket angulation and +10° in the right Mx3 bracket angulation; and prescription 2, with +10° in the left Mx3 bracket angulation and 0º in the right Mx3 bracket angulation were prepared. Fifteen patients were bonded with 0º in the left half of the dental arch and +10° in the right half of the dental arch, and equalized with 14 patients with +10° in the left-half of the dental arch and 0º in the right-half of the dental arch. Figure 1 presents the flowchart of this study.

The brackets used in this study were the 0.018-inch slot Level Arch Modern prescription Mini Diamond Twin® (Ormco, Glendora, CA, USA) metal brackets and Accent™ (Ormco, Glendora, CA, USA) maxillary first molar tubes. The bracket prescriptions used for the study and control groups are shown in Table 2. The bracket selection criteria were the same angulation and torque values for all symmetrical teeth, with only the Mx3 teeth having a different angulation alternative. The 0.018-inch slot Level Arch Modern prescription Mini Diamond

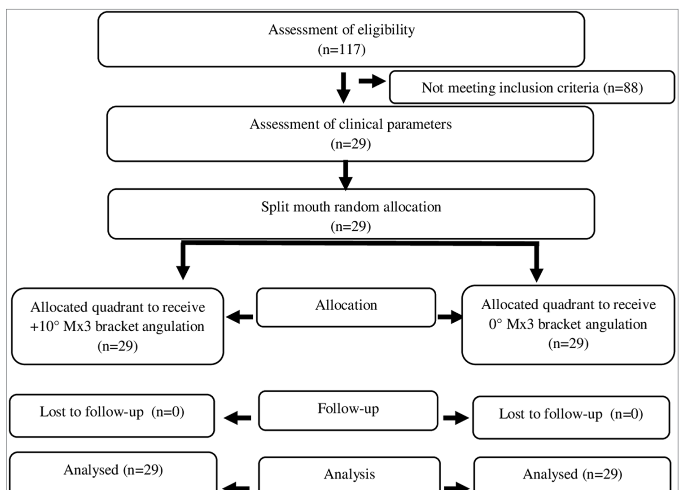

Figure 1. Flowchart of this study

Table 1. Mean values for age, arch/tooth size discrepancy and skeletal relationship of individuals included in this study

Measurements	Mean ± SD
Age	15.3 ± 1.65
Arch/tooth size discrepancy (mm)	5.96 ± 1.84
Palatal Plane—Mx1Angle (°)	113.92 ± 6.50
ANB Angle (°)	3.90 ± 2.03
SnGoGn Angle (°)	35.71 ± 5.53

Table 2. The angular values of the brackets used for the study and control groups

Brackets and tube (Maxillary)	Groups			
	Study		Control	
	Angulation	Torque	Angulation	Torque
Central	+5°	+14°	+5°	+14°
Canine	+10°	0°	0°	0°
First molar	+15° (distal offset)	−10°	+15° (distal offset)	−10°

Twin® (Ormco, Glendora, CA, USA) metal bracket was preferred because they met the criteria.

The initial arch wire was a 0.017 inch × 0.025 inch Turbo Wire (Ormco Corp., Orange, CA, USA). This wire is a nine-strand, rectangular, braided NiTi with low stiffness and great flexibility.[15] It can also be used as an initial arch wire in severe malocclusions.[16]

The brackets were bonded to Mx1, Mx3 and maxillary first molar teeth as measurement references. The upper orthodontic model (T0) was then prepared. Bonding in the upper arch was completed without the inclusion of the maxillary right–left first premolars and maxillary second molars (Mx7). The anchorage was prepared moderately. The maxillary right–left second premolars and maxillary right–left first molars were the anchorage segment, and the maxillary anterior teeth were the active segment. After the extraction of the premolars, lacebacks were applied to the Mx3 teeth during the same session using a 0.010-inch-long ligature wire and a Turbo Wire® arch, with the arch wire inserted into the brackets. The frequency of the control sessions was 4 weeks. In the fourth and eighth weeks of the control sessions, the arch wire was removed, and the laceback application was repeated with a new ligature wire. On the 12th week of the control sessions, a second upper orthodontic model (T1) was prepared. The T0 and T1 models were digitized with a three-dimensional scanner (3Shape R-700 Desktop Orthodontic Scanner, Copenhagen, Denmark) and superimposed using the OrthoAnalyzer (3Shape, Copenhagen, Denmark) analysis program. The medial and lateral points of the third palatal rugae and the medial point of the first palatal rugae were used as references for superimposition[17,18] (Figure 2). Sagittal and horizontal planes were formed in this model. The sagittal plane was created using the medial points of the first, second, and third palatal rugae on the medial palatal suture. The horizontal plane was formed perpendicular to the sagittal plane by passing through the medial point of the third palatal rugae on the right side and the lateral points of the right and left third palatal rugae.

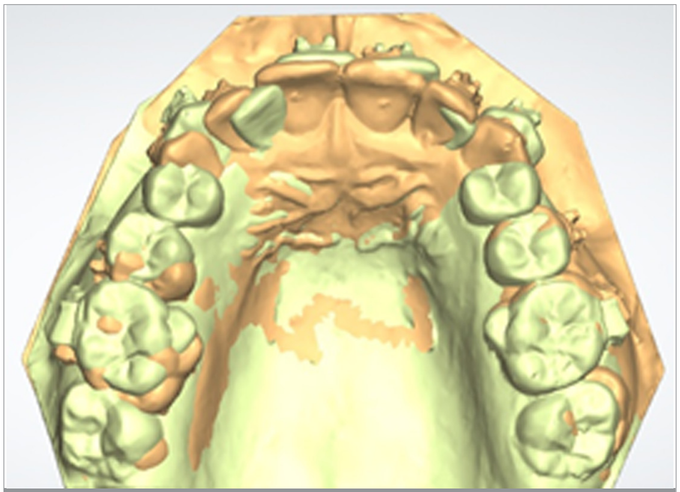

Figure 2. The superimposition model

Measurements

Mx3 millimeter (Mx3 mm): The distance between the tip of Mx3 and the tip of the Mx7 mesiobuccal cusp on the x-axis of the sagittal plane.

Mx1 millimeter (Mx1 mm): The distance between the tip of Mx1 and the tip of the Mx7 mesiobuccal cusp on the x-axis of the sagittal plane.

Mx3 degree (Mx3, °): The disto-occlusal angle between the line passing the disto-occlusal corner of the Mx3 bracket and the tip of the tooth and the line passing parallel to the bracket base.

Mx1 degree (Mx1, °): The disto-incisal angle between the line passing the disto-incisal corner of the Mx1 bracket and the tip of the tooth and the line passing parallel to the bracket base.

Linear and angular measurements are shown in Figure 3 and Figure 4, respectively.

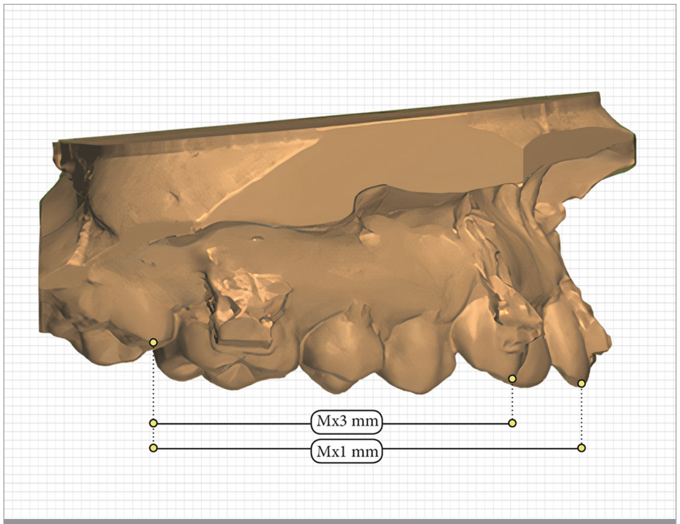

Figure 3. Mx3 and Mx1 linear measurements

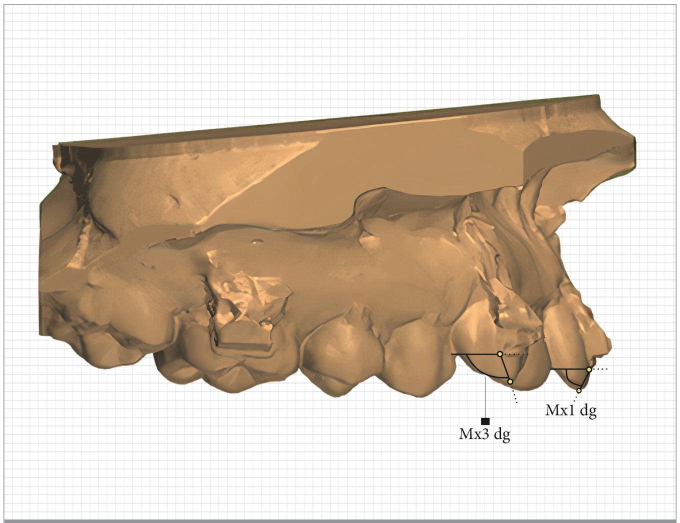

Figure 4. Mx3 and Mx1 angular measurements

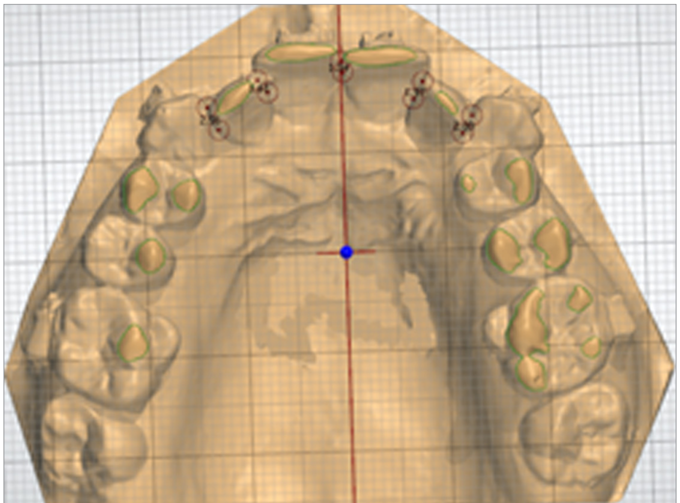

Figure 5. Little's Irregularity Index measurement

Little's Irregularity Index was calculated as the sum of the linear measurements of the distance between the tooth's respective anatomic contact points and the adjacent anatomic contact points in the upper jaw anterior region (Figure 5). Since this study had a split-mouth design, the current degree of crowding in the midline was evenly divided into 2 sides when calculating the Irregularity Index.

Statistical Analysis

The dataset consisted of measurements performed on 58 upper jaw digital orthodontic models of 29 patients. The margin of error in the measurements was calculated from 14 measurements repeated by the same researcher on 20 orthodontic models after 6 weeks. According to the Dahlberg formula,[19] the margin of error was calculated to not exceed 1.2° for the angular measurements and 0.4 mm for the linear measurements.

The data were analyzed with IBM SPSS V23, and the Shapiro–Wilk test was used to test the data for a normal distribution. Intragroup comparisons between the T0 and T1 stages and intergroup comparisons for T1–T0 difference values were made using a paired samples *t*-test. The results were presented as mean ± standard deviation, and the significance level was set to $P < .05$.

RESULTS

Intragroup Comparisons

The amount of linear movements of Mx3 in the distal direction significantly increased in both groups ($P < .001$). However, the angular movements of the Mx3 teeth did not show a statistically significant difference in either the study or the control groups ($P > .05$).

The Mx1s showed statistically significant retrusion in the control group ($P < .01$), but no statistically significant retrusion in the study group ($P > .05$).

Little's Index values showed statistically significant decreases in both the study ($P < .001$) and control ($P < .001$) groups.

Intragroup comparisons are shown in Table 3.

Intergroup Comparisons

The linear movement of the Mx3 in the distal direction was significantly higher in the control group than in the study group ($P < .01$). The Mx3 angulation showed a statistically significant difference ($P < .05$) between the study and control groups. The Mx3 inclined mesially in the study group and distally in the control group.

The linear movements of the Mx1s and the amount of change in the Little's Index measurements did not show any statistically significant differences between the groups ($P > .05$).

Intergroup comparisons are shown in Table 4.

Table 3. Paired samples *t*-test results for intragroup comparisons

Measurement	Group	Measurement Value: Bonding (T0) Mean ± SD	Measurement Value: 12th week (T1) Mean ± SD	*P*-value
Mx3 (°) (Angular)	Study	105.6 ± 24.2	110.1 ± 12.7	.225
	Control	113.5 ± 14.1	106 ± 21.1	.076
Mx3 (mm) (Linear)	Study	31.7 ± 1.7	29.4 ± 2.2	<.001***
	Control	31.7 ± 1.8	28.8 ± 1.7	<.001***
Mx1 (°) (Angular)	Study	88.4 ± 19.5	85.5 ± 11.4	.148
	Control	87.5 ± 13.8	83.2 ± 11.0	.009**
Mx1 (mm) (Linear)	Study	39.7 ± 2.9	39.2 ± 2.4	.105
	Control	39.5 ± 3.2	38.7 ± 2.4	.007**
Little index value	Study	4.46 ± 2.33	–0.49 ± 0.61	<.001***
	Control	4.78 ± 1.687	–0.59 ± 0.742	<.001***

**Statistically significant difference ($P < .01$).
***Statistically significant difference ($P < .001$).
Mx3, Maxillary canine tooth; Mx1, Maxillary central incisor tooth; SD, standard deviation.

Table 4. Paired samples *t*-test results for intergroup comparisons

Measurement	Group	Measurement (T1-T0) Mean ± SD	*P*
Mx3 (°) (Angular)	Study	4.46 ± 19.38	.015*
	Control	−7.52 ± 21.97	
Mx3 (mm) (Linear)	Study	−2.26 ± 1.12	.007**
	Control	−2.9 ± 1.09	
Mx1 (°) (Angular)	Study	−2.87 ± 10.39	.553
	Control	−4.35 ± 8.33	
Mx1 (mm) (Linear)	Study	−0.54 ± 1.73	.556
	Control	−0.74 ± 1.36	
Little index value	Study	−4.95 ± 2.13	.417
	Control	−5.37 ± 1.63	

*Statistically significant difference ($P < .05$)
**Statistically significant difference ($P < .01$)
Mx3, Maxillary canine tooth; Mx1, Maxillary central incisor tooth; SD, standard deviation.

DISCUSSION

In this study, treatment involving the maxillary first premolar extraction during the first 12 weeks was evaluated. During this period, a single arch wire was used to counteract the effects of arch wire change. Therefore, a 0.017 × 0.025 Turbo Wire was chosen as arch wire for increased breakage resistance. This arch wire was introduced as an initial arch wire by the manufacturer. The possible torque effect of the rectangular arch wire on Mx3 was eliminated by choosing 0° as the torque value of the Mx3 brackets in both groups.

The data set of this study was obtained by measuring the amount of tooth movement and crowding in digital models. Previous studies have also used digital jaw models for the three-dimensional analysis of orthodontic tooth movement[20] and for the calculation of Little's Irregularity Index.[21] The results of this study showed that Mx3 of the study group exhibited angular changes in the mesial direction while moving linearly in the distal direction. By contrast, the control group showed both angular and linear movements of Mx3 in the distal direction.

A comparison of the 2 groups revealed that the amount of linear and angular movements in the distal direction was significantly higher in the control group than in the study group. This result is consistent with the angulations of the Mx3 brackets, as increases in the angulation of the bracket were accompanied by increases in the angulation of the tooth in the mesial direction and with decreases in the amount of linear motion in the distal direction. In orthodontic treatment involving premolar extraction, excessive distal tipping of the canines can lead to posterior bite opening and prolong the total treatment time.[6] For this reason, the Mx3 movement observed in the study group was considered more desirable in the initial phase of the treatment with extraction.

In this study, a statistically significant retrusion was measured in the Mx1s in the control group, but no statistically significant retrusion occurred in the study group. Therefore, increases in the Mx3 bracket angulation were accompanied by a decrease in the amount of movement of Mx1 in the palatinal direction. According to Pontes et al.,[11] the upper dental arch length increased with increasing angulation in the 6 anterior maxillary teeth when using a straight-wire bracket. Although these previous researchers obtained this result from cases with no tooth loss in the upper dental arch, their finding is consistent with that which we obtained in our cases with premolar extraction.

In the present study, the amount of reduction in maxillary anterior crowding at the end of 12 weeks was defined as alignment efficiency. Here, Little's Irregularity Index was used to evaluate the effect of Mx3 bracket angulation on alignment efficiency, as this index has been used in many studies to evaluate the efficiency of crowding treatments.[22,23,24] This index has also been used to assess the performance of the arch and orthodontic bracket systems, orthodontic stability, retention of the upper jaw, and measurements of the lower jaw over time.[25,26]

However, in the present split-mouth study, the amount of crowding present in the midline was equally distributed on both sides according to Little's Irregularity Index calculations for each group. Specifically, the index value decreased from +4.78 mm to −0.59 mm in the control group and from +4.46 mm to −0.49 mm in the study group. Therefore, the decrease in anterior crowding did not exhibit a statistically significant difference between the 2 groups. Although the amount and angle of movement of Mx3 in the distal direction showed significant differences between the 2 groups, their alignment efficiencies were similar. This result is related to the effects of the Mx3 bracket angulations on the retrusion of Mx1. Increases in the Mx3 bracket angulation led to decreases in the distal canine tooth movement and in the retrusion of Mx1. The opposite was true for the control group. Therefore, the alignment activity, which is the total result of these tooth movements, did not differ between the groups. However, the differences measured for the individual movements of Mx3 and Mx1 were statistically significant.

The limitation of this study is that tooth movement was evaluated only at the crown level. The movement of the tooth root should also be measured by radiological assessment. Nevertheless, this prospective clinical study can serve as a reference for clinicians when choosing a bracket prescription appropriate for the Mx1 and Mx3 positions in a treatment involving premolar extraction. Future studies could examine the effects of different Mx3 bracket angulations and bracket torque combinations on tooth movement and alignment efficiency.

CONCLUSION

In this study, the following results were obtained:

- The +10° Mx3 bracket angulation caused an increase in the inclination of the canine tooth in the mesial direction and a decrease in the distal linear movement of Mx3 compared with the 0° Mx3 bracket angulation.
- The +10° Mx3 bracket angulation caused a decrease in the Mx1 retraction compared with the 0° Mx3 bracket angulation.
- No significant difference was found between the +10° Mx3 bracket angulation and the 0° Mx3 bracket angulation in terms of alignment efficiency in a treatment involving maxillary first premolar extraction.

Ethics Committee Approval: The study protocol was reviewed and approved by the ethics review board of the Ondokuz Mayıs University Clinical Research Ethics Committee, resolution number OMÜ KAEK —2016/336.

Informed Consent: A signed informed patient consent form was obtained from all patients.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.Y.; Design - S.Y.; Supervision - S.Y.; Fundings – M.T.; Materials - M.T.; Data Collection and/or Processing - M.T.; Analysis and/or Interpretation - M.T., S.Y.; Literature Review - M.T.; Writing - M.T., S.Y.; Critical Review - S.Y.

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Original Article

Knowledge and Attitude Toward Temporomandibular Disorders: A Survey in İstanbul

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Main Points

- General dental practitioners in Istanbul consider occlusal splint therapy to be the most important treatment method in the rehabilitation of TMD patients.
- Contrary to the point of view in other countries, only one GDP (0.9%) referred the TMD patients to orthodontists.
- There is a consistency between the clinical approaches of GDPs in Istanbul to TMD patients and the consensus papers.

ABSTRACT

Objective: The aim of this study was to identify the strategies used for the diagnosis and treatment of patients with temporomandibular disorders (TMDs) by general dental practitioners (GDPs) in the city of Istanbul.

Methods: A total of 154 GDPs were assessed by a single examiner in this questionnaire-based study. Descriptive statistics were calculated for all variables, and the results were analyzed at a 95% CI and statistical power of 80% with the significance level set at $P < .05$.

Results: The most frequently employed strategies for obtaining a diagnosis were patients' medical history (33.1%), physical examination (37.7%), and a combination of diagnostic methods (29.2%). The most commonly referred specialties were prosthodontics (62.2%) and maxillofacial surgery (36.9%). All GDPs treated their TMD patients with occlusal splints, and the majority of the occlusal splints were hard occlusal splints (62.8%). Half of the GDPs considered the etiology of TMD to involve stress, whereas 49.4% believed that TMD is of a multifactorial etiology.

Conclusion: The GDPs clarified that TMD patients were mostly treated with occlusal splints and this datum is consistent with the suggestions of previous consensus papers.

Keywords: Temporomandibular joint disorders, dentist practice patterns, health-care surveys

INTRODUCTION

The American Association for Dental Research (AADR) approved a statement about temporomandibular disorders (TMDs) in 2010 and explained that "TMDs encompass a group of musculoskeletal and neuromuscular conditions that involve the temporomandibular joints (TMJs), the masticatory muscles, and all associated tissues."[1] General dental practitioners (GDPs) are responsible for the diagnosis and management of TMD and associated structures.[2,3] To achieve better treatment results, GDPs should consider and understand the diagnostic strategies, treatment plans, and possible outcomes of TMDs. Numerous scientific papers have been published on evidence-based diagnosis and treatment strategies, and consensus papers have been published on TMD.[1,4-7]

Two consensus statements have been published by the European Academy of Craniomandibular Disorders and the AADR that provide guidelines and recommendations on the examination, diagnosis, and management of patients with TMD for GDPs.[1,4] Both statements recommend that the primary management of TMD should be

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based on conservative therapeutic modalities. The AADR statement also states that "many of the conservative modalities have proven to be at least as effective in providing symptomatic relief as most forms of invasive treatment."[1]

One of the most controversial topics in the field concerns the temporomandibular joint, its related structures, and the methods used to diagnose and manage TMD. The present study aimed to identify the strategies used for the diagnosis and treatment of TMD patients by GDPs in the city of Istanbul using a questionnaire-based survey.

METHODS

The total population of Turkey was estimated at 77.7 million people in 2014, and the most populated province was Istanbul, with 18.5% (14 377 018) of the total population. In Turkey, 15 412 private dentists are registered in the Turkish Dental Association; however, the number of specialists in TMD and orofacial pain (OFP) is unclear. Istanbul includes almost 37% (5743) of the private dentists in Turkey, indicating that there are an estimated 2504 individuals per practitioner in Istanbul. In this study, the contact information of 400 GDPs who were not affiliated to any academic institution or public corporation in Istanbul was requested from the Istanbul Chamber of Dentists. Venancio and Camparis[8] reported that this sample size was sufficient for this category of study. All contact numbers were dialed and 163 dentists answered the call. Detailed information about the study was explained before starting the survey. The researcher indicated to the participants that there was no obligation to participate and they were permitted to stop responding to the questionnaire at any time if they did not want to continue the survey. Explicit informed consent was obtained via cellular phone conversation. One hundred fifty-seven GDPs agreed to take part in the present study and 6 of the dentists contacted refused to join at the beginning of the telephone interview. All interviews were conducted individually by one researcher to avoid interrater variation. The telephone interviews lasted 15-20 minutes. Three participants chose not to complete the survey during the telephone interview and these questionnaires that were incompletely filled were discarded (n = 154). Inclusion in the sample was independent of the school of origin, age, gender, professional experience, or year of graduation. The study was approved by the Research Ethics Committee (2017/21).

The questionnaire used in the present study was similar to a survey conducted by Aldrigue et al.[3] in Brazil that focused on data from the AADR, the European Academy of Craniomandibular Disorders, and the American Academy of Orofacial Pain. They reported that a systematic review of agreements based on the international recommendations for TMD and OFP management was conducted to confirm the questionnaire used in their study. A revision was made by adding only 1 question (Q9) to the questionnaire used in the sample survey study. There were no differences in questions or their meanings, between the Turkish questionnaire used for this study and the original English questionnaire.

In the present study, while the first 5 questions (Q1-Q5) determined the behavior of GDPs when they first encounter TMD patients, Q6-Q12 determined the treatment approach of GDPs (n = 78) who answered "occlusal splinting" to Q5. Q13-Q14 were asked to determine the perspective regarding cause–effect relationship in TMD disorder. Participants could choose more than 1 answer in the questionnaire, except for the yes/no option questions.

Statistical Analysis

All interviews were conducted individually by a single researcher. For all statistical tests, the NCSS 2007 and PASS 2008 Statistical & Power Analysis Software (NCSS, Kaysville, Utah, USA) were used. Descriptive statistics (mean, standard deviation, median, correlation, minimum, maximum, and frequency) were calculated for all variables, and the results were analyzed at a 95% confidence interval and a statistical power of 80% with the significance level set at $P < .05$. Dichotomic answers were compared using a binomial test, and multiple answers were evaluated using a Clopper–Pearson test. Pearson's chi-square test and the Fisher–Freeman–Halton test were used for comparison of qualitative data.

RESULTS

Of the GDPs surveyed, 50.6% reported that they received potential TMD patients in their office ($P > .05$). The diagnostic procedure employed, the approach toward each patient, and the place of referral were significantly different among GDPs ($P < .05$). The medical history (33.1%), physical examination (37.7%), and a combination of diagnostic methods (29.2%) were the most frequently employed strategies for obtaining a diagnosis. Of the practitioners who responded, 44.2% reported that they referred their patients to an academic institution. Prosthodontics (62.2%) and maxillofacial surgery (36.9%) were common specialties for patient referral. All practitioners who received potential TMD patients in their office specified that they offered occlusal splinting as the treatment for TMD (Table 1).

Table 2 shows the responses to the occlusal splint-related questions. Hard stabilization appliances (62.8%) and soft stabilization appliances (35.9%) were the most frequently used types of splints ($P < .05$). During splint fabrication, 61.5% of GDPs did not use semi-adjustable articulators, and 43.6% performed occlusal adjustments at the time of application. Occlusal splints were fabricated in maximum habitual intercuspation or in a centric relationship by 50% of the GDPs, with the treatment based on the features of the individual patients ($P < .05$). A total of 64.1% of GDPs believed that an increase in the vertical dimension should be patient-dependent ($P < .05$), and although 46.2% of GDPs instructed their patients on nocturnal splint use, 46.2% believed that the duration of splint use should be patient-dependent ($P < .05$). Most GDPs (75.6%) followed up with their patients monthly ($P < .05$).

Table 3 indicates the responses to the cause–effect relationship questions for TMD. Half of the GDPs considered the etiology of TMD to involve stress-related factors, whereas 49.4% believed

Table 1. Distribution of Behavior-Related Questions

Questions	Frequency	Percentage	*P*
Q1. Have potential TMD patients sought treatment at your office?			
Yes	78	50.6	.935
No	76	49.4	
Q2. What procedures do you use to diagnose these patients?			
Medical history	51	33.1	.001**
Physical examination	58	37.7	
Radiological visualization	0	0	
Study model	0	0	
Combination of these methods	45	29.2	
Q3. What is your approach toward these patients?			
Offer treatment	42	27.3	.018*
Refer to another dentist	44	28.6	
Refer to an academic institution	68	44.2	
Q4. If you do not treat these patients, to what specialty do you refer them?			
Prosthodontics	69	62.2	.001**
Orthodontics	1	0.9	
Otorhinolaryngology	0	0	
Physiotherapy	0	0	
Neurology	0	0	
Maxillofacial surgery	41	36.9	
Q5. If you do treat these patients, what treatments do you offer them?			
Counseling	0	0	-
Diet plans	0	0	
Thermotherapy	0	0	
Physiotherapy	0	0	
Pharmacotherapy	0	0	
Occlusal splinting	78	100	
Occlusal adjustment	0	0	
Orthodontics	0	0	
Oral rehabilitation and prosthetic treatment	0	0	
Other	0	0	

Binomial test and Clopper–Pearson test, $^{*}P < .05$; $^{**}P < .01$.

that TMD is of multifactorial etiology ($P < .05$). All GDPs considered multidisciplinary medical and dental treatment to be necessary ($P < .05$).

Table 4 shows the answers of the participants, some of whom received potential TMD patients in their office (group 1), and some who did not (group 2), to the determinated questions (Q2 and Q13), and the significant differences between groups indicated with a sign. Answers of "physical examination" and "combination of diagnostic methods" were significantly higher in the group 1, and an answer of "medical history" was significantly higher in the group 2 ($P < .05$). Furthermore, answers to Q13 did not show significant differences between the groups ($P = .171$).

DISCUSSION

Most GDPs diagnose TMD based on the patient's medical history and a physical examination,[3] although some different diagnostic methods have been discussed in previous studies.[1,5,9-14] In the present study, all GDPs diagnosed TMD based on the medical history of the patient and a physical examination, and none applied a radiological visualization or achieved a study model.

Two consensus papers suggested that patients with TMD should be treated with conservative and reversible treatment modalities.[1,4] In the present study, GDPs who referred their TMD patients to a specialist preferred to refer them to prosthodontic (62.2%),

Table 2. Frequencies and Percentages of the Answers to Each Splint-Related Question, Considering That Splints Are the Most Common Choice of Treatment for TMD

Questions	Frequency	Percentage	*P*
Q6. What type of splint do you employ?			
Anterior bite appliances	0	0	.001**
Posterior bite appliances	1	1.3	
Hard stabilization appliances	49	62.8	
Anterior positioning appliances	0	0	
Soft stabilization appliances	28	35.9	
Q7. Do you use semi-adjustable articulators?			
Yes	30	38.5	.054
No	48	61.5	
Q8. In what occlusal relationship do you fabricate the splint?			
Maximum habitual intercuspation	8	10.3	.001**
Centric relationship	31	39.7	
Depends on the individual patient	39	50.0	
Q9. By how much do you increase the occlusal vertical dimension with the splint?			
1 mm	0	0	.001**
2 mm	8	10.3	
3 mm	19	24.4	
≥4 mm	1	1.3	
Depends on the individual patient	50	64.1	
Q10. Do you adjust the occlusal surface of the splint at the time of fitting?			
Yes	34	43.6	.308
No	44	56.4	
Q11. What are your instructions regarding the duration of splint use?			
Nocturnal	36	46.2	.001**
Daytime	2	2.6	
All the time	4	5.1	
Depends on the individual patient	36	46.2	
Q12. How often do the patients return to the office for follow-up?			
Weekly	0	0	.001**
Monthly	59	75.6	
Depends on the individual patient	19	24.4	

Binomial test and Clopper–Pearson test, $^{**}P < .01$.

maxillofacial surgery (36.9%), and orthodontic specialists (0.9%). The choice of TMD treatment modality by the prosthodontists and orthodontists is mostly conservative and reversible in accordance with the suggestions in the consensus statements. Thus, maxillofacial surgeons, who were the second common specialties for TMD patient referral, should know when and how to treat and/or when and to whom to refer the TMD patients. Only 1 GDP referred the TMD patients to orthodontics (0.9%), whereas the study by Aldrigue et al.[3] indicated that most GDPs in Brazil referred their TMD patients to orthodontists.

Velly et al.[15] examined the treatment modalities of GDPs for TMD patients, and occlusal splinting was the preferred treatment (96.6%). The same study revealed that hard acrylic custom stabilization splints were the common treatment appliance (60.1%), which was similar with the present study. A meta-analysis performed by Fricton et al.[16] found that the hard occlusal splints showed reasonable efficacy in the treatment of TMD pain when compared with non-occluding splints or no treatment. However, studies have revealed that both active and placebo splints equally improved the patient outcomes.[17] Alencar and Becker[18] randomly selected 42 patients with myofascial pain and treated 3 groups of patients with hard splints, soft splints, or non-occlusal splints. The results of their study showed that all 3 appliances reduced the symptoms, and no significant differences were observed among the 3 groups after 90 days. Thus, in the present study, the use of splints appears to be an appropriate choice for the initial treatment of TMD patients as a conservative treatment, regardless of the contact surfaces of the splints or the material from which they are produced. Otherwise,

Table 3. Frequency and Percentages of the Answers to Each Cause–Effect Question

Questions	Frequency	Percentage	*P*
Q13. To what do you attribute the etiology of TMD?			
Stress	77	50.0	.001**
Parafunction	1	0.6	
Trauma	0	0	
Occlusion-related factors	0	0	
Medical muscle-skeletal disorders	0	0	
Multifactorial	76	49.4	
Q14. Do you believe in multidisciplinary medical and dental treatment?			
Yes	154	100	.001**
No	0	0	

Binomial test and Clopper–Pearson test, $^{**}P < .01$.

Terebesi et al.[19] investigated the relationship between the motor unit recruitment and vertical changes in the jaws and reported that the amount of vertical separation of the splints had a positive correlation with the therapeutic outcome of patients with myofascial pain syndrome. In the present study, half of the GDPs (50%) adjusted the thickness of the occlusal splint depending on the needs of individual patients and 24.4% adjusted the thickness of the occlusal splint by 3 mm.

The pharmacotherapy of TMD patients has been defined by several publications.[2,4,20] Heir et al.[2] suggested that medication can include analgesics, antidepressants, antianxiety agents, muscle relaxants, corticosteroids, antihistamines, local anesthetics, antihypertensives, antiepileptic drugs, adjunctive neuropathic pain medications, tryptans, and ergot derivatives. The dentists' routine use of various categories of these advised drugs requires superior skills and knowledge. In addition, the side effects and the addiction risk of these drugs must be considered by practitioners.[21] In the present study, none of the GDPs preferred pharmacotherapy as the treatment for TMD.

Table 4. Comparison of Answers and Statistical Contrasts in Variables Between Groups

		Q1. Have potential TMD patients sought treatment at your office?		
Questions	Answer	Group 1, Yes (n=78), Mean (%)	Group 2, No (n=76), Mean (%)	*P*
Q2	Medical history	3 (3.8)	48 (63.2)	.001a,**
	Physical examination	35 (44.9)	23 (30.3)	
	Combination of these methods	40 (51.3)	5 (6.6)	
Q13	Stress	35 (44.9)	42 (55.3)	.171b
	Parafunction	0	1 (1.3)	
	Multifactorial	43 (55.1)	33 (43.4)	

[a]Pearson's chi-square test; [b]Fisher–Freeman–Halton test, $^{**}P < .01$.

The etiology of TMD remains unclear. Among the various hypotheses proposed to explain the onset and maintenance of symptoms, those advocating occlusal factors and psychological disturbances are among the most common, and the etiology of TMD is usually described as multifactorial in the literature.[22-24] In the present study, the greater part of GDPs considered the etiology of TMD to be multifactorial, and most of the GDPs believed that stress was the main issue, whereas only one GDP thought that parafunction was key to the etiology. Similarly, Lei et al.[25] reported that stress may contribute to the incidence of TMD.

There is no specific consensus on the treatment options for patients with TMD in the field of dentistry. The probable reason for this situation is that the etiologies of TMDs are multifactorial. Therefore, the subject of TMD disorders is covered in different specialties during the dental education and PhD programs.

Occlusal stability is a very important issue for dysfunctional patients. These patients have a lower capability to adapt to occlusal changes and are easily disturbed by occlusal instability. Thus, orthodontic treatment has to be performed according to the rules that allow an "ideal and stable" result to be achieved.[26] There are different opinions in the literature regarding the relationship between orthodontic treatment and TMD. In 1988, Greene and Laskin[27] explained that there were statistically positive correlations between orthodontics and TMD treatment. On the contrary, in 1995, McNamara et al.[28] explained that there was no raised risk for TMD associated with any type of orthodontic mechanics and concluded that while a stable occlusion is a reasonable orthodontic treatment goal, failure to achieve a specific gnathologically ideal occlusion does not result in TMD signs and symptoms. Even if most of the research do not support the correlation between orthodontic treatment and TMD, it should be highlighted that absolute conclusions cannot be drawn because the etiologies of TMDs are multifactorial.[26]

Before determining orthodontics–TMD relationship and the associated treatment approach, it is useful to determine whether this relationship occurs before or during orthodontic treatment.[26] If the patient has signs or symptoms of TMD before starting orthodontic treatment, diagnosis is crucial. When the main complaint is pain, a differential diagnosis is important to determine whether the pain is because of TMD.[26] After diagnosis of TMD-related pain, a conservative treatment protocol including pharmacotherapy, counseling, behavioral therapy, home exercises, physical therapy, and/or occlusal appliances should be assessed.[29] As a rule, orthodontic treatment should not be started as long as a patient suffers from facial pain.[26] Orthodontic treatment may be considered after a certain period of time after the facial pain subsides. Patients with generalized musculoskeletal pain or with a systemic inflammatory disease should be rehabilitated with an interdisciplinary perspective.[26]

If TMD signs and symptoms show up during active orthodontic treatment, the first step is always to make the diagnosis.[26] The second step is to stop active orthodontic treatment temporarily to avoid aggravating factors.[26] Activating orthodontic appliances subjects the teeth to forces that may cause temporary discomfort or pain.[26] Orthodontic pain induced by means of separators may result in a temporary reduction in the pressure pain thresholds of the muscles of mastication.[30] The third step is to manage the pain by following the same conservative treatment protocol as suggested above.[26] An occlusal splint can also be used to evaluate the interference-free position of the mandible. Finally, when the patient is pain-free, orthodontic treatment can be continued as previously planned, or, if necessary, modified according to the patient's condition.[26]

DC/TMD examination forms are frequently used in the diagnosis and treatment of TMD patients.[5] However, these forms cannot be used typically and it might be due to fact that they contain very detailed information and applications in TMDs. For this reason, a simplified examination form is needed for both GDPs and PhDs to make the initial diagnosis of patients with TMD. The use of this form should be aimed at providing accurate diagnosis and referral to the right specialty area before the treatment of TMDs. It is recommended that this examination form is created with the efforts of the practitioners in the specialty areas, and that it should be integrated into the curricula of dentistry and PhD education. Thus, both the early diagnosis of TMDs and the participation of the specialties in the treatment protocol can be provided.

There are some methodological limitations in this study. First, the study group was from a specific region and further studies can be focused on transnational comparisons, which can highlight the knowledge of GDPs and the education modality of each country regarding TMD and related topics. Second, a study population with greater sample size could provide clearer findings on strategies used by GDPs to diagnose and treat patients with TMD.

CONCLUSION

In the present study, GDPs clarified that TMD patients were mostly treated with occlusal splints and this datum is consistent with the suggestions of previous consensus papers. However, it cannot be concluded that the treatment approach has been successfully applied because there are no data on the clinical outcomes of the patients.

Ethics Committee Approval: Ethics committee approval was received for this study from the Ethics Committee of İstanbul University Faculty of Dentistry (2017/21).

Informed Consent: Explicit informed consent was obtained via cellular phone conversation.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - T.C.; Design - T.C., A.S.; Supervision - T.C.; Materials - T.C.; Data Collection and/or Processing - T.C.; Analysis and/or Interpretation - T.C.; Literature Review - T.C., A.S.; Writing - T.C.; Critical Review - T.C., A.S.

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Case Report

Minimum Surgico-Orthopedic Treatment using Computer-Assisted Single-Tooth Osteotomy in an Adolescent Skeletal Class III Patient with Anterior Ankylosed Tooth: A Case Report

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Main Points

- Children having skeletal Class III malocclusion and traumatic anterior teeth may show a difference in alveolar bone height and open bite due to bony ankylosis of traumatized teeth.
- In this case, the alveolar bone height cannot be improved by fixed orthodontic appliance, and esthetic results may not be obtained even if the implant or prosthetic treatments are attempted after the completion of growth.
- Orthodontic treatment accompanied with CAD/CAM-based minimal surgery is helpful especially when the alignment of the teeth is impossible and the overjet and overbite of the ankylosed tooth area are not improved. Therefore, it would be very esthetically disadvantageous by previous methods of orthodontic treatment.
- Minimal surgery such as single-tooth osteotomy during orthodontic treatment immediately improves the patient's quality of life in terms of socio-psychology or esthetics and the patient would not need alveolar bone augmentation on the anterior maxilla for dental implant at the time of growth completion.

ABSTRACT

Traumatic tooth avulsion can lead to ankylosis, which may interfere with growth of the alveolar bone in a growing patient. The resulting difference in alveolar bone height and position can lead to esthetic problems such as open bite.

A growing 13-year-old female patient presented skeletal Class III malocclusion with bone ankylosis of a maxillary anterior tooth. Even after 2 years of orthopedic and orthodontic treatment, little improvement was achieved regarding the positions of the anterior maxillary teeth, or the vertical position of the maxillary right central incisor. Therefore, surgical treatment by single-tooth osteotomy (STO) and corticotomy for the anterior ankylosed tooth were considered and performed using a CAD/CAM surgical guide, based on presurgical computer-based simulation surgery. Orthodontic and orthopedic treatments were completed at 10 months after surgery. The patient showed a favorable course of healing, with no mobility issues or gingival recession 3 years after single-tooth osteotomy and corticotomy surgeries. A favorable outcome was finally achieved by applying orthopedic treatment combined with STO and corticotomy for the anterior ankylosed tooth. Orthodontic treatment with minimally surgical method is recommended in an adolescent patient with skeletal Class III malocclusion and anterior open bite.

Keywords: Ankylosed tooth, pediatric orthopedics, single-tooth osteotomy, corticotomy, skeletal Class III malocclusion

INTRODUCTION

Following traumatic tooth avulsion, the tooth must be quickly replanted and root canal therapy should be performed. Even if the avulsed tooth is successfully replanted, it can become ankylosed. When dental trauma results in bone ankyloses during growth, it can interfere with the vertical and anterior growth of alveolar bone, producing an open bite with unfavorable overjet that may lead to functional and esthetic issues.[1,2]

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It can be difficult to move the ankylosed teeth responsible for an anterior open bite and unfavorable overjet using only orthodontic treatment. Moreover, even with successful tooth movement, esthetic problems can remain due to the difference in alveolar bone height and anterior–posterior (AP) position difference.

In such cases, single-tooth osteotomy (STO) with/without corticotomy can be performed to simultaneously move the ankylosed tooth and the alveolar bone.[3] When an ankylosed tooth also limits the degree to which the adjacent teeth can be moved by orthodontic treatment, corticotomy can be performed to facilitate tooth movement.[4] Trauma-induced bony ankylosis that occurs during the growth period can hamper the vertical growth of alveolar bones. This condition results in an open bite that poses esthetic and functional problems.[1,2] A single-tooth dento-osseous osteotomy may be the optimal treatment plan.[2,3]

Recent advances enable surgical planning using simulations based on preoperative CT data, as well as easier and more accurate surgery performance using CAD/CAM-based equipment.[5] This study introduced a CAD/CAM-based method to assist surgeons in performing single-tooth dento-osseous osteotomies for delicate and elaborate surgery. In this method, a surgical guide was manufactured with CAD/CAM technology. The surgical guide was based on preoperative surgical simulation data. We showed that this method was highly conducive to successful single-tooth dento-osseous segmental osteotomy. Especially in this case report, a growing patient presented skeletal Class III malocclusion with an open bite due to bone ankylosis of a maxillary anterior tooth. A favorable outcome was achieved by performing orthopedic treatment combined with STO and corticotomy.

CASE PRESENTATION

A 13-year-old female patient visited the hospital with protrusive mandible and malocclusion, reverse overjet of anterior teeth, and an open bite. Five years earlier, the patient had experienced trauma causing avulsion of the right central incisor, which had been treated by reduction and endodontic treatment. The subsequent combination of palatoversion, lack of vertical eruption of the maxillary right central incisor, and insufficient vertical growth from the maxillary right canine to the maxillary left canine had resulted in malocclusion and an open bite (Figures 1 and 2). The patient was diagnosed as skeletal Class III with open bite due to tooth bony ankylosis.

Orthodontic and orthopedic treatments were planned to improve the patient's open bite and overall occlusion. After 2 years of orthopedic treatment using face mask, AP maxillary discrepancy was corrected. However, orthodontic treatment achieved little improvement in the positioning of the anterior

Figure 1. Pretreatment facial and intraoral photographs

Figure 2. Radiographs from the patient's initial diagnostic evaluation

maxillary teeth, or the vertical and AP position of the maxillary right central incisor (Figure 3).

It was decided that the patient should undergo STO, because correction of anterior alveolar bone and tooth position was needed for resolving the psychological problem due to the anterior open bite. A canine-to-canine labial corticotomy was also planned to improve the tooth axis and the vertical and AP position of the maxillary anterior teeth.

Preoperative digital simulation surgery was performed to minimize the risk of damaging the surrounding teeth during osteotomy, and to ensure movement of the ankylosed tooth to a functionally and esthetically favorable position (Figure 4). A three-dimensional (3D) reconstruction was generated from maxillary CT images, and osteotomy of the maxillary right central incisor was simulated. The simulation aimed to determine the precise location of the osteotomy relative to the root apices of the maxillary right central incisor, and with regard to its relationships with the left central incisor and the right lateral incisor.

A thickness of 0.7 mm was selected for the osteotomy line (STO and corticotomy), accounting for the thickness of the piezosurgical device to be used in the actual operation. The osteotomy line was planned to minimize injury to the nearby dental roots. Surgical simulation was also performed to determine the position of the corticotomy from the maxillary right to left canine. These osteotomy line and maxillary data were used with CAD/CAM technology to design and manufacture a surgical guide to be placed on the maxilla during the actual osteotomy procedure (Figure 4). The surgical guide was fabricated from a biocompatible material using a 3D printer (ProJet 3500 HDMax 3D Printer, 3D Systems, Inc., Rock Hill, SC).

In the operating room, the initial surgical incision was made near the maxillary incisor. Next, the surgical guide was placed on the alveolar bony area, and the guide fit and the osteotomy line position were checked. A piezoelectric saw blade was used to make a cut, following the groove in the guide to ensure a consistent position and angle as measured depth during the surgical simulation (Figure 5). For osteotomy to the left and right of the maxillary right central incisor and periapical osteotomy, the cut depth was determined with reference to the distance between the guide and the palatine bone, as measured during the simulation. For the corticotomy from the right to the left maxillary canine, the surgeon referred to the thickness of the cortex and the device in CT images, and relied on the manual sensation of cortical perforation to perform the corticotomy safely.

After the osteotomy, the segment was slowly moved inferiorly, taking care not to separate the tooth-bone segment from the palatal mucoperiosteal flap under verifying blood circulation (Figure 5). Following inferior movement of the tooth-bone segment, a xenogeneic bone (Geistlich Bio-Oss, Geistlich Pharma AG, Wolhusen, Switzerland) was grafted in the empty bone space. Upon confirming the stable position of the tooth-bone segment, the surgical wound was sutured with application of manufactured human plasma fibrinogen, completing the operation.

Figure 3. Intraoral photographs after 2 years of orthodontic treatment

After a 1-week postoperative latency period (Figure 6), postoperative orthodontic treatment was resumed to correct the patient's open bite, and the vertical and AP position and axis of the maxillary right incisor. Orthopedic and orthodontic treatment were completed 10 months after surgery, the open bite had improved, and favorable occlusion was obtained (Figures 7 and 8). The patient showed a favorable course of dental occlusion and mandibular growth without temporomandibular disorder (TMD) issues. There is no complication as severe gingival recession or relapse, 3 years after the STO and corticotomy surgeries (Figures 9 and 10).

All the above procedures were performed with patient consent. The patient also consented to the publication of the data in this study.

DISCUSSION

In patients with skeletal Class III malocclusion and ankylosed anterior tooth, if the anterior cross bite is properly improved through orthopedic treatment during the growth period and the leveling of anterior teeth can be maintained through STO, the possibility of the need for orthognathic and implant surgery after the termination of growth, can be significantly reduced. It may also be the best treatment in terms of socio-psychology or esthetics. Even for a growing patient, improvement in esthetics is of great social and psychological importance, and these procedures must be considered in the treatment of skeletal Class III patients with ankylosed teeth. The management of ankylosed tooth with STO in conjunction with orthognathic surgery is also an alternative treatment method.[6]

However, STO has a risk of postsurgical problems affecting the blood supply to the tooth-bone segment, which can result in complications such as gingival recession.[3,5] Even when the open bite is improved by surgery, gingival recession and adjacent tooth injury may lead to esthetic problems and a poor long-term prognosis.

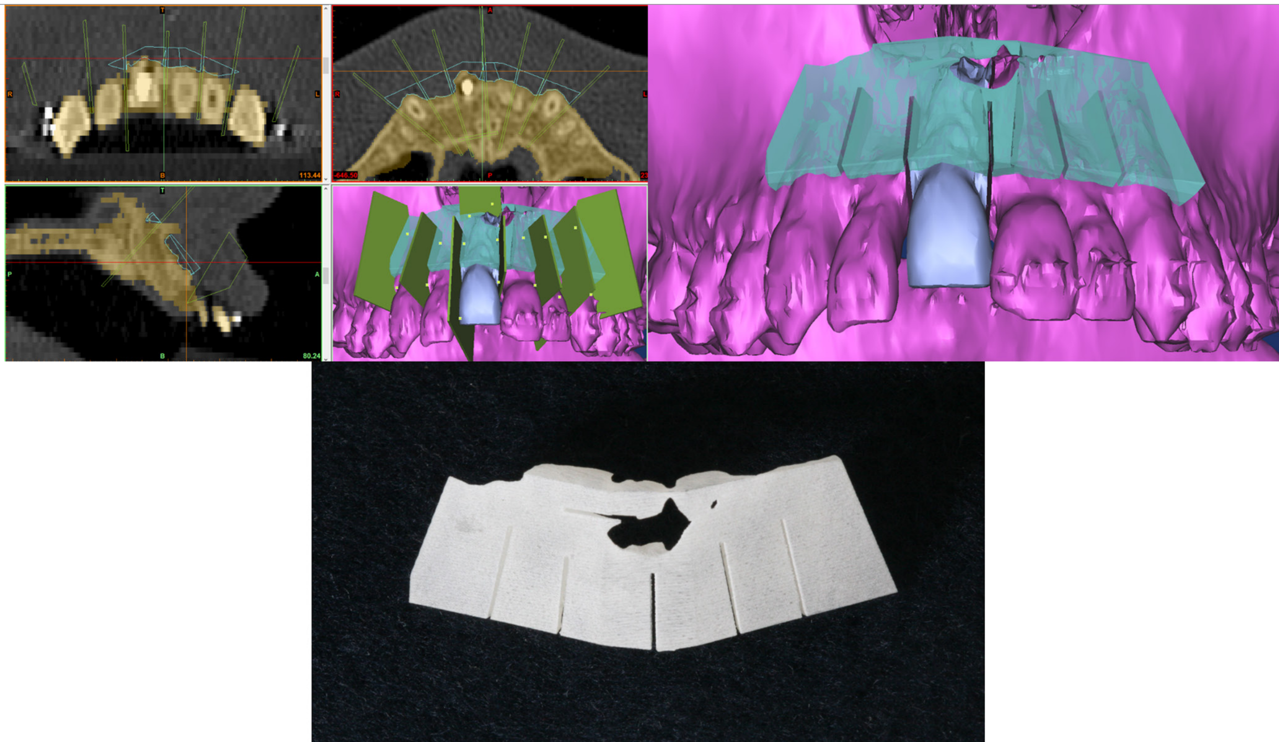

Figure 4. Surgical simulation and surgical guide 3D printing by biocompatible materials. The simulation aimed to determine the precise location of the osteotomy relative to the root apices of the maxillary right central incisor, and with regard to its relationships with the left central incisor and the right lateral incisor. The surgical guide was fabricated from a biocompatible material using a 3D printer.

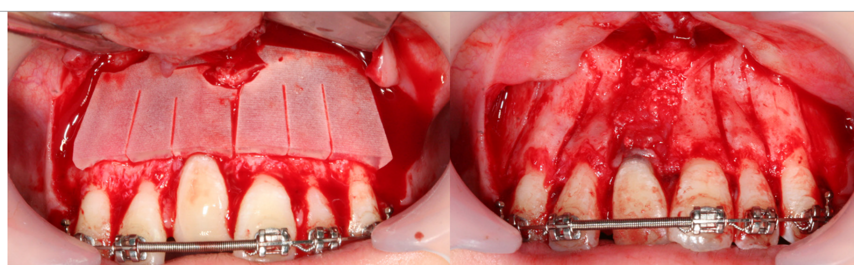

Figure 5. Operation of single-tooth osteotomy with corticotomy. The initial surgical incision was made near the maxillary incisor. Next, the surgical guide was placed on the alveolar bony area, and the guide fit and the osteotomy line position were checked. For osteotomy to the left and right of the maxillary right central incisor and periapical osteotomy, the cut depth was determined with reference to the distance between the guide and the palatine bone, as measured during the simulation. Following inferior movement of the tooth-bone segment, a xenogeneic bone was grafted in the empty bone space.

Figure 6. Postoperative intraoral photos and periapical radiograph

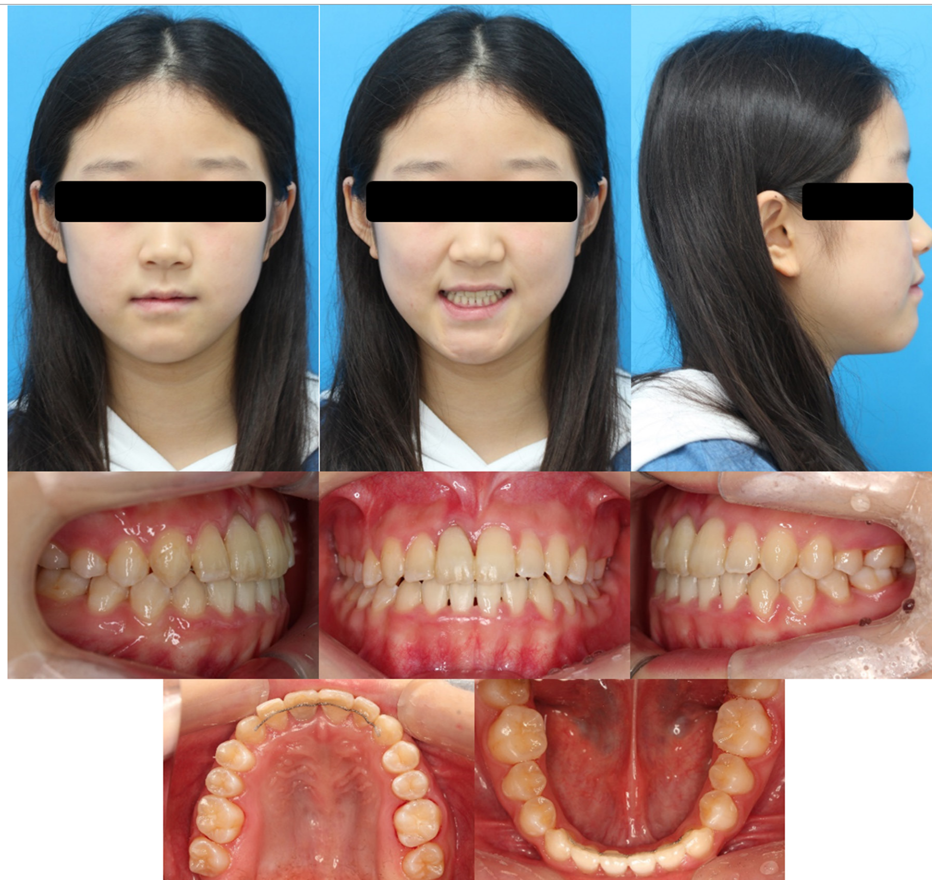

Figure 7. Post-treatment facial and intraoral photographs

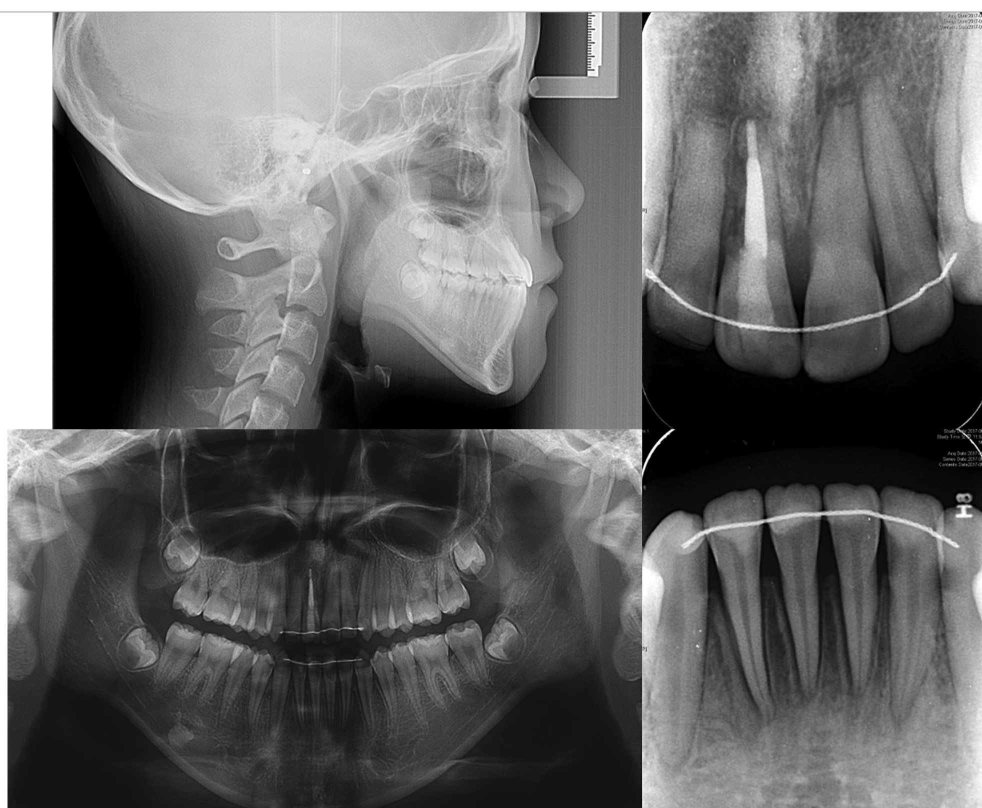

Figure 8. Post-treatment radiographs

Figure 9. Facial and intraoral photographs of 3 years' retention

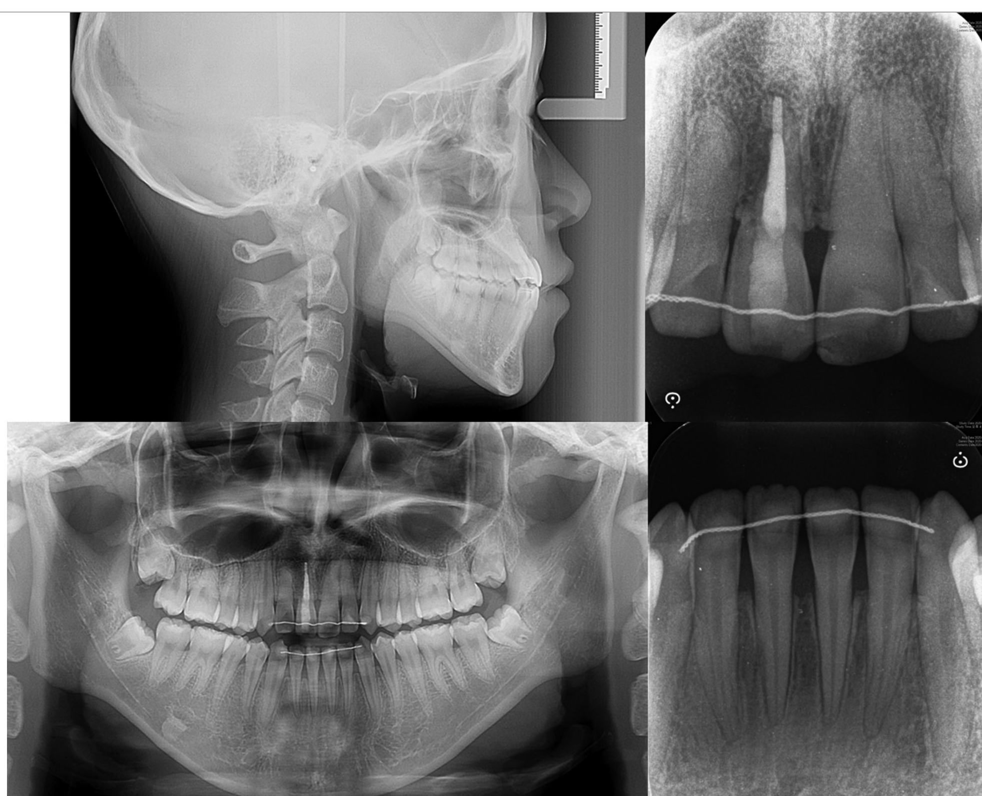

Figure 10. Radiographs 3 years after the completion of postoperative orthodontics

The ubiquity of CT imaging and the development of computer-based surgical simulations enable the determination of an ideal osteotomy position. Preoperative surgical simulation, and the use of surgical devices manufactured based on simulation data, can promote favorable outcomes and reduce dental injury and surgery duration.[5]

The patient in the present case required STO for the maxillary anterior teeth, with simultaneous corticotomy. Corticotomy on the premaxilla between both canines may have also been helpful to move the teeth adjacent to the ankylosed tooth. STO and corticotomy can be used in the patients in whom orthodontic movement by ordinary traction is difficult.[7]

To facilitate the forward movement of the teeth, corticotomies of the 6 maxillary anterior teeth were also performed in this case and they significantly helped to improve the overjet and overbite of this skeletal Class III patient. Even if the skeletal Class III malocclusion was improved by orthopedic treatment through face mask, patients with the ankylosed maxillary anterior teeth would be subject to tooth movement during orthodontic treatment. It can affect the anterior growth as well as the downward growth restriction of the ankylosed tooth area, and this growth restriction exacerbates the overjet. It is also more disadvantageous in patients with skeletal Class III malocclusion. Without STO, the alignment of the teeth would be impossible, and even if the skeletal malocclusion were relieved, the overjet and overbite of the ankylosed tooth area would not be improved, thus being very esthetically disadvantageous.

Computer-based preoperative simulation surgery and the use of manufactured surgical devices can promote favorable outcomes and reduce dental injury and surgery duration.[5] A guide for STO is small and simple to design using data from the simulation surgery. Using a 3D printer, such a guide can be manufactured with biocompatible materials. The application of a CAD/CAM surgical guide can reduce surgery time and trauma during surgery, which is expected to improve surgical outcomes. When using a CAD/CAM guide, it is essential to verify the guide's position during surgery, since deviating from the planned osteotomy position can cause damage to the surrounding teeth. The thickness of the osteotomy saw blade must be considered when determining the width of the groove in the surgical guide. The guide must be thick enough to resist the osteotomy. If the guide is too thin, it can break during surgery.

The osteotomy depth can be determined based on information from the surgical simulation regarding the distance from the superior aspect of the guide to the maxillary palate, thereby reducing damage to the palatine flap. It is thought that corticotomy can be safely performed by utilizing information about the cortex thickness obtained from CT images, as well as relying on the surgeon's manual sensation of cortical perforation. Piezoelectric devices are useful for STO and corticotomy.[8,9]

When STO is performed alone, the application of inappropriate traction force can break the cervix of the ankylosed tooth. Thus, it can be preferable to insert a screw into the tooth-bone segment and then to use the screw to apply traction to the whole segment.[10] Mini-implants can be also used for the tooth traction.[11] With this procedure, orthodontic traction must be applied to the tooth-bone fragment in cases showing dental root resorption. An alternative method was used in the present case. Rather than inducing bone regeneration by traction, the procedure was intended to achieve as much movement as possible during surgery, to minimize the bone defect by performing a bone graft, and to prevent the tooth-bone fragment from postoperatively returning to its original position. Further studies including a larger number of cases are needed to determine which method is better in STO.

The correction of anterior cross bite is properly improved through orthopedic treatment during the growth period, and the leveling of anterior teeth can be maintained through STO. This may also be the best treatment in terms of socio-psychology or esthetics, concerning the strong relationship between malocclusion and oral health-related quality of life.[12] It is reported that there is a strong relationship between malocclusion, especially the Class III group, and oral health-related quality of life in adolescent orthodontic patients.[13] The use of orthopedic appliances to correct Class III malocclusion in growing patients would not be considered as a risk factor for the development of TMD.[14] Through the current treatment method, the patient would not need not only the orthognathic surgery for Class III malocclusion but also alveolar bone augmentation on the anterior maxilla for dental implant.

In skeletal Class III growing patients, an orthopedic treatment can be considered depending on the amount of remaining growth, even though growth control is controversial. Therefore, orthopedic treatment and STO for improvement of alveolar bone height should be considered at an appropriate time in skeletal Class III patients with a traumatized anterior tooth.

In this patient, the skeletal disharmony was relieved through a face mask and the posterior occlusal relationship was improved. The occlusal relationship in the posterior teeth was overcorrected in consideration of the remaining pubertal growth, and an appropriate Class I occlusal relationship was obtained during the orthodontic treatment by fixed appliance.

In the present case, a young patient presented skeletal Class III malocclusion with an open bite due to ankylosis of an anterior maxillary tooth. This situation caused a severe esthetic problem and psychological stress for the patient. When orthopedic and orthodontic treatment alone failed to help, this issue was addressed by performing STO and corticotomy, with the use of a presurgical computer-based simulation, and a CAD/CAM surgical guide for the actual surgical procedure. After surgery, favorable orthopedic and orthodontic outcomes were achieved. These methods could be used to treat other young patients with reverse overjet and open bite due to traumatized ankylosed anterior teeth.

CONCLUSION

A favorable outcome was finally achieved by applying orthopedic treatment combined with STO and corticotomy for an anterior ankylosed tooth in the case where a young patient presented skeletal Class III malocclusion. CAD/CAM-based minimal surgery such as STO during orthodontic treatment immediately improves the adolescent patient's quality of life in terms of socio-psychology or esthetics. The patient's need for bone augmentation and dental implant on the anterior maxilla was reduced.

Orthodontic treatment with CAD/CAM STO is recommended in an adolescent patient with skeletal Class III malocclusion and open bite caused by traumatized ankylosed anterior teeth.

Informed Consent: Written informed consent was obtained from the patients who agreed to take part in the study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.H.K., J.Y.L.; Design - S.H.K., J.Y.L.; Supervision - S.H.K., J.Y.L.; Materials - S.H.K.; Data Collection and/or Processing - S.H.K., J.Y.L.; Analysis and/or Interpretation - S.H.K., J.Y.L.; Literature Review - S.H.K., J.Y.L.; Writing - S.H.K., J.Y.L.; Critical Review - S.H.K., J.Y.L.

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Review Article

Efficacy of the Cervical Vertebral Maturation Method: A Systematic Review

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Main Points

- The aim of the study is to evaluate whether the cervical vertebral maturation (CVM) method is effective in terms of predicting the growth spurt.
- Most of the previous studies have stated that the CVM method is an effective method for assessing skeletal maturity.
- The CVM method and skeletal analysis of the hand-wrist method do not show significant differences.
- No further radiographic investigations are required other than the lateral cephalogram.

ABSTRACT

Objective: The present systematic review was carried out to evaluate both qualitatively and quantitatively the effectiveness of the cervical vertebral maturation (CVM) method in predicting the pubertal growth spurt.

Methods: PubMed, PMC, Scopus, SciELO, Cochrane Central Register of Controlled Trials (CENTRAL), and Web of Science databases were searched. The research included every article published from 1970 to June 2019, featuring the keywords: ("cervical vertebrae" OR ("cervical" AND "vertebrae") AND ("orthodontics" OR "growth and development" OR ("growth" AND "development") OR ("growth"). The Preferred Reporting Items for Reporting Systematic Reviews and Meta Analyses (PRISMA) protocol was adopted, and quality assessments modified from the "Strengthening the Reporting of Observational Studies in Epidemiology" (STROBE) and the "Standards for the Reporting of Diagnostic Accuracy Studies" (STARD) were performed to conduct this systematic review.

Results: Initially, 1284 articles were found. All the articles were then examined, and 43 studies met the inclusion criteria. Sixteen articles had low-quality scores, 25 had moderate scores, and 2 had high scores. The results showed a moderate to high statistically significant correlation between the CVM and other maturation methods.

Conclusion: Overall, the CVM method can be considered an effective method and may be used with other skeletal indices for the radiographic assessment of skeletal maturity, and also to identify the growth peak in growing patients.

Keywords: Cervical vertebrae, skeletal maturation, growth spurt, systematic review, lateral cephalograms

INTRODUCTION

Timing is considered one of the most important factors for the success of an orthodontic treatment procedure. Recently, the issue of optimal timing has attracted the attention of both researchers and clinicians. Clinical research has shown that greater therapeutic effects are obtained when the mandibular growth peak is included in the treatment period. Therefore, the use of an effective biological indicator to estimate the pubertal growth spurt represents an effective diagnostic tool to treat patients with skeletal discrepancies.

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Typically, patients treated during the growth peak demonstrate significant skeletal effects, while patients treated during the pre-peak period show only dentoalveolar modifications.

Growth assessment is essential for functional orthodontic therapy, which performs its function best in the growing patient. For this reason, it is important to identify the growth peak of patients who undergo this treatment. In addition, other types of treatment should ideally be performed in certain growth stages, for example: the facial mask is ideal for use at a young age, that is, the cervical vertebral stage 2 (CVS2), while orthognathic surgery and implant positioning are not undertaken until growth ceases (CVS6). For this reason, the CVM stage can be a useful indicator in all ages and for a wide range of orthodontic treatments rather than just for functional treatment.

There are several biological indicators to estimate skeletal maturation such as chronological age, dental formula and tooth development, menarche in girls, and the change of voice in boys, height increase, non-invasive biomarkers taken from serum or from gingival crevicular fluid, and skeletal age assessed by radiography. However, the assessment of skeletal age is considered the best biological index related to the growth of facial bones. The classic and most generally used method for assessing skeletal age is the radiographic analysis of the hand-wrist bones, whose validity has been confirmed by numerous scientific studies.[1] However, the main disadvantages of this method are the additional radiograph of the hand necessary to perform the study, and the great difficulty in assessing the staging. Therefore, in recent years, the evaluation of the CVM has been increasingly utilized in determining the skeletal maturity of the growing patient. In 1972, Lamparski[1] introduced this method, allowing skeletal age estimation and eliminating the necessity for extra radiographic exposure since the cervical vertebrae were already recorded on lateral cephalograms taken as a diagnostic pretreatment record. Lamparski's method is predicated on the study of changes in size and shape of the 5 cervical vertebral bodies from the second to the sixth cervical vertebrae (C2-C6) and includes 6 phases of cervical vertebral stage development. However, the utilization of a lead collar to guard the thyroid during the execution of the x-ray can hinder the entire vision of the cervical spine. Thus, in 1995, Hassel and Farman[2] conceived a replacement CVM method, which evaluated the visible lateral profiles of C2, C3, and C4. Furthermore, Baccetti et al.[3] limited the number of cervical vertebrae analyzed during the evaluation of bone age, and published a CVM method to evaluate the maturation of the cervical spine. According to this method, only 3 vertebrae were evaluated—C2, C3, and C4—which are visible even with a protective collar for the thyroid.

Nevertheless, within the literature, there are conflicting results regarding the efficacy of the CVM method for the right identification of the growth spurt.[4,5] To get an accurate summary of the best available evidence when considering the CVM method, a scientific review of the literature seems necessary. Thus, the aim of this systematic review was to throw the spotlight on whether the CVM method is scientifically effective to determine skeletal maturity. The question of our research was: How effective is that the CVM method in terms of predicting the growth spurt?

METHODS

Information Sources

A global electronic database search was conducted to identify relevant publications. The following databases were searched: PubMed, PMC, Scopus, SciELO, Cochrane Central Register of Controlled Trials (CENTRAL), and Web of Science. All studies published from 1970 to June 2019 have been included in our research.

Search Study

The electronic search strategy focused on the following key words: ("cervical vertebrae" OR ("cervical" AND "vertebrae") AND ("orthodontics" OR "growth and development" OR ("growth" AND "development") OR "growth"). The Preferred Reporting Items for Reporting Systematic Reviews and Meta Analyses (PRISMA) protocol were adopted for this systematic review[6]; and the PROSPERO registration number of our review was: CRD42020155719.

Study Selection

At the first stage, 2 reviewers (LA and RG) screened the titles and abstracts of the retrieved records independently, duplicate exclusion was performed, and irrelevant articles were excluded. In the next phase, the complete texts of probably relevant papers were evaluated to determine whether they met the eligibility criteria. The inclusion criteria, supported by PICOS format, were: [1] cross-sectional and longitudinal articles in human studies that evaluate qualitatively and quantitatively the CVM method; [2] studies that compare the CVM method with other methods for assessing skeletal age, for example, the hand-wrist maturation (HWM) method, the middle phalanx of the third finger (MP3) method, body height, chronological age, and dental age; and [3] studies that evaluate at least 20 patients. The exclusion criteria were: [1] lack of a transparent description of inclusion/exclusion criteria; [2] studies with inadequate sample sizes; [3] letters to editor, opinion articles, reviews and meta-analyses; and [4] published articles not written in English.

Eligibility was independently assessed by the 2 authors, and any disagreements were resolved by discussion and consent or by a third expert author (MM) who was asked to arbitrate. Any elements that did not meet the inclusion criteria were excluded.

The PRISMA flowchart diagram for the study selection process has been reported (Figure 1).

Data Extraction and Quality Assessment of Selected Studies

Two review authors independently extracted data, consistent with a pre-set protocol. The extracted data included: first author, year of publication, study design, sample composition by sex and age, CVM evaluation method, standard method to evaluate skeletal age, CVM reproducibility statistical analysis, CVM

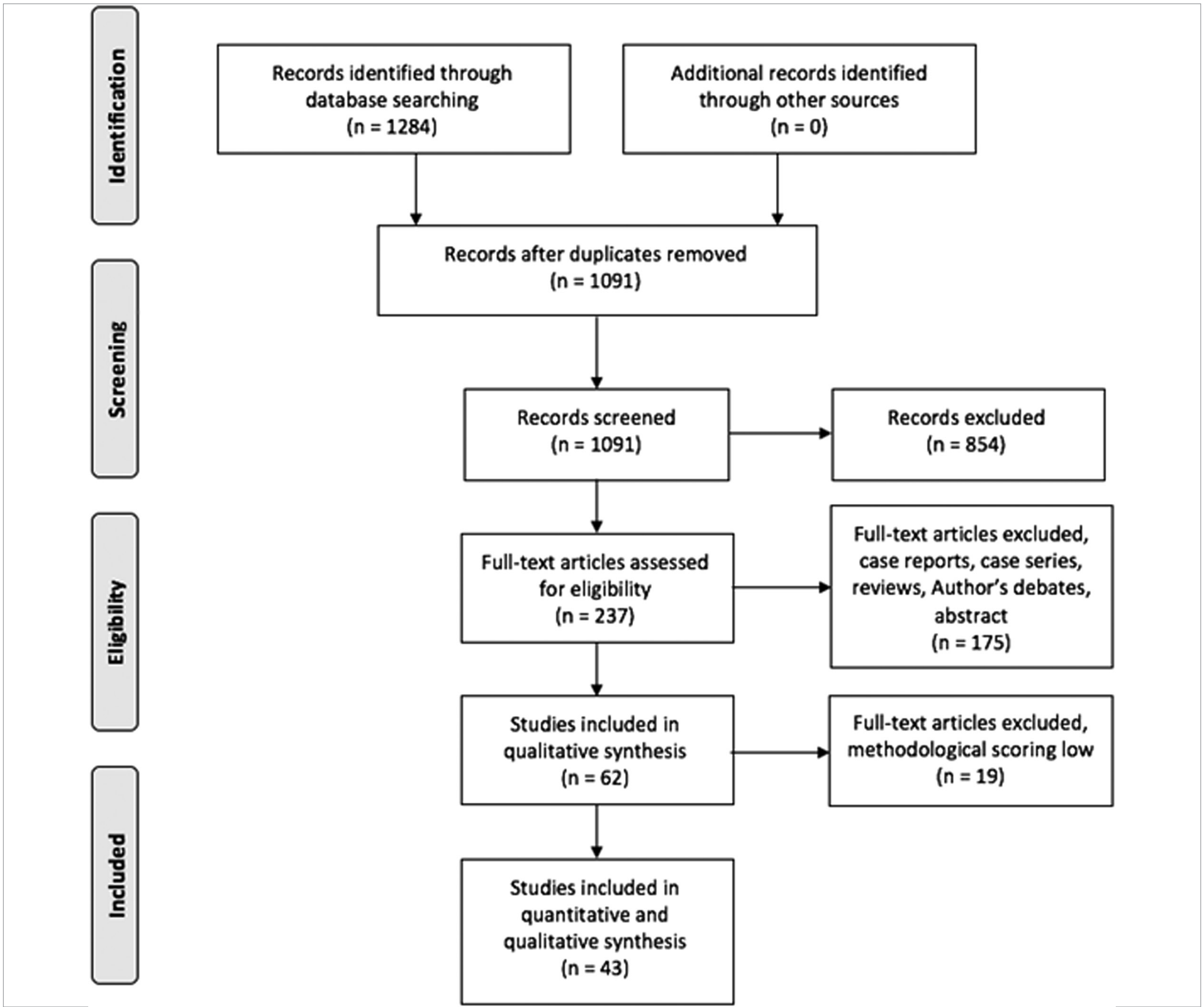

Figure 1. Flow Diagram of the literature search

correlation test versus standard method, CVM and standard method accuracy, and results.

If the data were not clear enough, the authors were contacted by e-mail. Any disagreements between the 2 authors were resolved by discussion and consent or consultation with a third expert author (MM).

Quality Analysis

The methodological quality of the selected articles was assessed using the assessment modified from the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE), and 7 Standards for the Reporting of Diagnostic Accuracy Studies (STARD) (Table 1).[8] When the 2 reviewers were not in agreement, a third investigator was called to succeed in reaching consensus. (MM). The kappa score measuring the extent of agreement was 0.89. Each criterion was assigned a point, if satisfied; no points, if not satisfied. Quality assessment scores ranged from 0 to 12. Articles were classified as "low quality" (score from 0 to 6), "moderate quality" (score from 7 to 10), or "high quality" (score from 11 to 12).

Synthesis Measures and Approach to Synthesis

Due to the heterogeneity between the studies included during this systematic review, particularly within the different methods of evaluating the cervical vertebrae and the lack of specific criteria for random selection, a meta-analysis could not be performed. A narrative summary was performed, illustrating the results of individual studies based on the groups evaluated.

RESULTS

The online database search yielded 1091 potentially relevant titles and abstracts after duplicates were sorted from a complete set of 1284 records. A total of 237 articles were reclaimed for

Table 1. Criteria for assessing quality components in the studies included

		Yes	No
1.	Is the objective clearly formulated?	1	0
2.	Are there key elements of study design early in the paper?	1	0
3.	Was the sample size calculated?	1	0
4.	Does the study report demographic characteristics of the study population?	1	0
5.	Were the sample selection criteria clearly described?	1	0
6.	Does the study describe specifications of material and methods involved including how and when measurements were taken?	1	0
7.	Was there a reliability assessment, with adequate level of agreement intraexaminer or/and interexaminer?	1	0
8.	Were measurements undertaken blindly?	1	0
9.	Does the study give details of methods of assessment (measurements) for each variable of interest?	1	0
10.	Was there a complete and adequate reporting of results, with self-explanatory tables and figures?	1	0
11.	Was there a statistical analysis appropriate for data?	1	0
12.	Was the *P* value stated or confidence intervals provided?	1	0

complete text evaluation, and 43 studies[2,5,9-49] that met the inclusion criteria, were selected from this analysis.

A methodological score was assigned to each study (Table 2). Sixteen articles had low-quality scores, 25 moderate scores and 2 high scores. The characteristics studies of moderate and high-quality scores have been reported (Table 3).

The 27 moderate and high-quality articles were categorized by topics as follows: 16 articles compared the CVM method with the HWM,[2,5,9,13-16,19,21,25-30,32] 2 articles compared the CVM method with chronologic age,[39,40] 1 article compared the CVM method with MP3,[42] 3 articles[46-48] compared the CVM method with dental age, 3 articles compared the CVM method with body height,[43,44,49] 1 article compared the CVM method with both chronologic age and dental age,[37] and finally 1 article compared the CVM method with both HWM and MP3.[24]

Cervical Vertical Maturation Versus Hand-Wrist Maturation

Seventeen moderate and high-quality articles compared the CVM method with the hand-wrist maturation analysis to determine skeletal maturity.[2,5,9,13-16,19,21,24-30,32] Six studies identified a reproducibility of the CVM method between 78% and 98%.[13-15,21,24,25] These studies used the Spearman correlation test or Cohen's Kappa statistic to determine reproducibility values. Thirteen studies described a significant correlation ($.00001 < P < .05$) between the 2 different methods.[2,13,15,16,19,21,24,26,28-30,32,34]

Cervical Vertebral Maturation Versus Chronologic Age

Three moderate-quality studies compared the CVM method with chronologic age.[37,39,40] According to 2 studies there was a statistically significant correlation between age and CVM method ($P < .001$).[37,40] One study, by Litsas et al.,[37] identified a stronger correlation for CVM stage IV for both males ($r = 0.554$) and females ($r = .68$) and a lower correlation for CVM stage III in males ($r = 0.433$; $P < .001$) and for stage II in females ($r = 0.393$; $P < .001$).

Cervical Vertebral Maturation Versus Middle Phalanx of the third finger

Two moderate-quality studies compared the CVM method with the MP3 method.[24,42] All the articles used the Cohen's Kappa statistic to evaluate the agreement between the 2 analyses and established a good relationship between the 6 phases of CVM and the 6 phases of MP3: 0.798 (in females) and 0.794 (in males) respectively.

Cervical Vertebral Maturation Versus Dental Age

Four moderate-quality studies compared the CVM method with dental age.[37,46-48] All articles showed that there was a statistically significant coefficient of correlation ($P < .05$) between dental age and CVM method, greater for males ($r = .703$) than females ($r = 0.499$).

Cervical Vertebral Maturation Versus Body Height

Three moderate-quality studies compared the CVM with body height.[43,44,49] Both articles stated that there was a good statistical correlation coefficient between the 2 methods ($P < .001$) and affirmed that the growth peak occurs between stage III and IV of CVM ($P < .001$), 100% in males and 87% in females.

DISCUSSION

This systematic review, including a comprehensive analysis of 2 high- and 25 moderate-quality studies, found that the CVM method can be considered as an effective method similar to the skeletal analysis of the hand-wrist method. In addition, our initial research question, namely "How effective is the CVM method in terms of predicting the growth spurt?" revealed the answer that the CVM method can be considered an effective tool in determining the growth spurt in growing patients.

The literature search initially uncovered 1284 publications, but only 43 quantitative studies were qualified for evaluation in this review. Such a finding is common when performing systematic reviews, as the initial and deliberate search of the literature was designed to include as many studies as possible in order not

Table 2. Quality assessment of selected studies

Articles	1	2	3	4	5	6	7	8	9	10	11	12	Total	Quality
Hassel and Farman[2]	0	0	0	1	1	1	1	0	0	1	1	1	7	Moderate
Beit et al.[5]	1	1	0	1	1	1	1	1	1	1	1	1	11	High
Mito et al.[9]	1	1	0	0	1	0	1	0	1	1	0	1	7	Moderate
Wong et al.[10]	1	0	0	1	0	0	1	0	1	0	1	1	6	Low
San Romàn et al. [11]	1	0	0	1	0	0	1	0	1	0	1	1	6	Low
Caldas et al.[12]	1	1	0	1	0	0	0	0	1	0	1	1	6	Low
Soegiharto et al.[13]	1	1	0	1	0	0	1	0	1	1	1	1	8	Moderate
Gandini et al.[14]	1	0	0	0	1	1	1	0	1	0	1	1	7	Moderate
Lai et al.[15]	1	1	0	1	0	0	1	1	1	1	1	1	8	Moderate
Pichai et al.[16]	1	1	0	0	0	1	0	0	1	1	1	1	7	Moderate
Kamal et al.[17]	0	0	0	1	0	0	0	0	1	0	1	1	4	Low
Stiehl et al.[18]	1	0	0	0	0	0	1	0	1	1	1	1	6	Low
Soegiharto[19]	1	1	0	1	0	0	1	0	1	1	1	1	8	Moderate
Byun et al.[20]	0	0	0	1	0	0	0	0	0	1	1	1	4	Low
Danaei et al.[21]	1	1	0	0	0	1	1	1	0	1	1	0	7	Moderate
Durka-Zająk et al.[22]	1	1	0	0	0	0	0	0	0	0	1	0	2	Low
Carinhena G et al.[23]	1	1	0	0	0	0	1	0	0	1	1	0	5	Low
Pasciuti et al.[24]	1	1	0	0	0	0	1	0	1	1	1	1	7	Moderate
Uysal et al.[25]	1	1	0	1	1	1	1	1	1	1	1	1	11	High
Chatzigianni et al.[26]	1	1	0	0	1	1	1	0	1	1	1	1	9	Moderate
Chang et al.[27]	1	1	0	1	1	1	0	0	1	1	1	1	9	Moderate
Heravi et al.[28]	1	1	0	1	1	1	0	0	1	1	1	1	9	Moderate
Mito et al.[29]	1	1	0	1	1	0	0	0	1	1	1	1	8	Moderate
Turkoz et al.[30]	1	1	0	1	1	0	0	0	0	1	1	1	7	Moderate
Varshosaz et al.[31]	1	1	0	1	0	0	0	0	0	1	1	1	6	Low
Litsas et al.[32]	1	1	0	1	1	0	1	0	1	1	1	1	9	Moderate
Flores-Mir et al.[33]	0	1	0	0	0	0	0	0	0	1	1	1	4	Low
Mahajan et al.[34]	0	0	0	1	0	0	0	0	0	1	0	1	3	Low
Altan et al.[35]	1	1	0	0	0	1	0	0	1	1	1	0	6	Low
Safavi et al.[36]	1	0	0	0	0	0	1	1	0	0	1	1	5	Low
Litsas et al.[37]	1	1	0	1	1	0	1	0	1	1	1	1	9	Moderate
Ramirez-Velásquez[38]	1	1	0	0	0	0	1	0	0	0	1	0	3	Low
Montasser et al.[39]	1	1	0	1	1	0	1	0	1	1	1	1	9	Moderate
Singh et al.[40]	1	1	0	1	1	1	1	0	1	1	1	1	10	Moderate
Prasad et al.[41]	1	0	0	0	0	0	0	0	0	0	1	0	2	Low
Ayach et al.[42]	1	1	0	1	1	0	1	0	0	0	1	1	7	Moderate
Franchi et al.[43]	1	1	0	0	1	0	1	0	1	1	1	1	7	Moderate
Hosni et al.[44]	1	1	0	1	1	1	1	0	1	1	1	1	10	Moderate
Felemban et al.[45]	1	1	0	1	0	0	0	0	0	1	0	0	4	Low
Kocasarac et al.[46]	1	1	0	1	1	1	1	0	1	1	1	1	10	Moderate
Camacho-Basallo et al.[47]	1	1	0	1	1	1	1	0	1	1	1	1	10	Moderate
Cossellu et al.[48]	1	1	0	1	1	1	1	0	1	1	1	1	10	Moderate
Montasser et al.[39]	1	1	0	1	1	1	1	0	1	1	1	1	10	Moderate

Table 3. Summary of the main characteristics of the 27 studies of moderate and high quality selected for the systematic review

Author/Year/Study design	Sample Size/Male-Female	Age	CVM Evaluation Method	Standard Method	CVM Reproducibility andStatistical Analysis	CVM Correlation TestVersus Standard Method	CVM and Standard Method Accuracy	Results
Hassel and Farman,[2] 1995, longitudinal study	220 (110 boys, 110 girls)	8-18 years	Hassel and Farman	Fishman (HWM)	Not evaluated	Not evaluated	Not evaluated	By using the lateral profiles of the second, third and fourth cervical vertebrae, it was possible to develop a reliable ranking of patients according to the potential for future adolescent growth potential.
Beit et al.,[5] 2013, cross-sectional study	730 (352 boys, 378 girls)	6-18 years	Not cited	Greulich and Pyle (HWM)	ANOVA	Pearson correlation coefficent	Not evaluated	The agreement between the HWM method and calculated skeletal age was modest and weaker than the agreement between the HWM and chronologic age.
T. Mito et al.,[9] 2002, longitudinal study	176 girls	7-15 years	Hassel and Farman	Tanner–Whitehouse (HWM)	*t*-test	Not evaluated	Not evaluated	Cervical vertebral bone age reflects skeletal maturity because it approximates bone age, which is considered to be the most reliable method for evaluating skeletal maturation.
Soegiharto et al.,[13] 2008, cross-sectional study	2167 (951 boys, 1216 girls)	8-17 years	Baccetti et al.	Fishman (HWM)	Cohen's Kappa	Not evaluated	ROC analysis	The CVM index and the SMI are valid methods to predict the pubertal growth peak.
P. Gandini, et al.,[14] 2006, longitudinal study	30(14 boys, 16 girls)	7-18 years	Baccetti et al.	Bjork Grave and Brown (HWM)	Not evaluated	Cohen Kappa concordant index	Not evaluated	The results show a concordance of 83.3% between the HWM method and the CVM method.
E. Lai et al.,[15] 2008, longitudinal study	709 (379 girls, 330 boys)	8-18 years	Baccetti et al.	NTUH-SMI method (HWM)	Spearman correlation coefficient	Spearman correlation test	Not evaluated	The Spearman's rank correlation (0.910 for males and 0.937 for females) confirmed a strong and significant correlation between CVMS and NTUH-SMI systems ($P < .001$).

S. Pichai, et al.,[16] 2014, longitudinal study	72	7-16 years	Baccetti et al.	Bjork Grave and Brown (HWM)	Not evaluated	Mann–Whitney test	Not evaluated	There was no significant difference between the HWM analysis and the CVM analysis for assessing skeletal maturation.
Soegiharto, et al.,[19] 2008, cross-sectional study	1422 (648 boys, 774 girls)	8-18 years	Baccetti et al.	Fishman (HWM)	Cohen's Kappa	Bland and Altman's method	ROC analysis	CVM shows a good intra-operator agreement. The K index is 0.85 for males and 0.97 for Indonesian females. The correlation index also shows a good correlation between the 2 methods of analysis.
S. Danaei et al.,[21] 2014, cross-sectional study	178 (90 girls, 88 boys)	8-17 years	Hassel and Farman	Fishman (HWM)	Cohen's Kappa	Not evaluated	Not evaluated	CVM method can be a valuable substitute for hand-wrist radiography in patients with short stature.
Pasciuti, et al.,[24] 2013, longitudinal study	100	6-18 years	Baccetti et al.	Tanner–Whitehouse (HWM) / Rajagopal e Kansal (MP3)	Cohen's Kappa	Not evaluated	Not evaluated	Complete agreement among the 3 methods was observed in 70% of the analyzed samples. The CVM method has the advantage of not necessitating an additional radiograph.
Uysal, et al.,[25] 2006, cross-sectional study	503 (213 boys, 290 girls)	5-24 years	Hassel and Farman	Bjork Grave and Brown (HWM)	Spearman correlation coefficient	Spearman correlation test	Not evaluated	CVM showed high reproducibility. The correlations between CVM and HWM maturation were 0.86 for sexes combined, 0.78 for male, and 0.88 for female subjects. All were significant at $P < .001$.
Chatzigianni, et al.,[26] 2009, longitudinal study	98 (40 boys, 58 girls)	8-17 years	Bookstein	Sempè (HWM)	*t*-test	Not evaluated	Not evaluated	Shape alone could not predict skeletal maturation better than chronologic age. C1 showed lower correlations with hand-wrist maturation.
Chang, et al.,[27] 2001, cross-sectional study	503 (244 boys, 259 girls)	8-18 years	Hassel e Farman	Fishman (HWM)	Wilcoxon test	Spearman correlation test	Not evaluated	The intra-judge and inter-judge reliability test indicated no significant difference for CVM ($P = .500$) and HWM ($P = .500$).

Table 3. Summary of the main characteristics of the 27 studies of moderate and high quality selected for the systematic review (*Continued*)

Author/Year/Study design	Sample Size/Male-Female	Age	CVM Evaluation Method	Standard Method	CVM Reproducibility andStatistical Analysis	CVM Correlation TestVersus Standard Method	CVM and Standard Method Accuracy	Results
Heravi et al.,[28] 2011, cross-sectionalstudy	47 (20 boys, 27 girls)	10-15 years	Hassel and Farman/ San- Roman/ Mito et al.	Tanner–Whitehouse (HWM)	Not evaluated	Pearson correlation test	Not evaluated	There was no significant correlation between CVM and HWM in 10-13-y.o. boys ($P < .05$).
Mito et al.,[29] 2003, cross-sectional study	40 girls	>7 years	Mito et al.	Tanner–Whitehouse (HWM)	*t*-test	Not evaluated	Not evaluated	The study produced a method to predict mandibular growth potential with latero-lateral x-rays only.
Turkoz et al.,[30] 2017, retrospective study	324 (167 girls, 157 boys)	7–17 years	Turkoz C. et al.	Greulich e Pyle (HWM)	Not evaluated	Pearson correlation coefficient	Not evaluated	The study developed a formula for evaluating the skeletal age that is reliable and applicable to both male and female subjects.
Litsas, et al.,[29] 2010, cross-sectional study	393	8–18 years	Hellsing	Chronologic age/Grave and Brown (HWM)	*t*-test	Spearman correlation coefficient	Not evaluated	The correlation coefficient between CVM and chronologic age is $r = 0.73$ for girls and $r = 0.72$ for boys
Litsas, et al.,[37] 2016, cross-sectional study	255 (145 girls, 110 boys)	8-18 years	Baccetti et al.	Chronologic age Demirjian et al. (dental age)	*t*-test	Spearman correlation coefficient	Not evaluated	Chronological and dental age showed a high correlation for both gender ($r = 0.741$ for boys, $r = 0.770$ for girls, $P < .001$). The strongest correlation was for the CVM Stage IV for both males ($r = 0.554$) and females ($r = 0.68$).
Montasser[39], 2017, cross-sectional retrospective study	Not evaluated	7-18 years	Hassel and Farman	Not evaluated	ANOVA	Not evaluated	Not evaluated	Racial differences in CVM II- III-IV-V stages between females and males have been highlighted.
Singh, et al.,[40] 2015, cross-sectional study	80 (40 boys, 40 girls)	10-19 years	Hassel e Farman	Chronologic age	Not cited	Pearson correlation coefficient	Not evaluated	The correlation coefficient CVM and chronologic age is $r = 0.855$ ($P < .001$).
Ayach et al.,[42] 2018, cross-sectional study	50 (21 boys, 29 girls)	8-16 years	Not cited	Perinetti et al. (MP3)	Cohen's Kappa	Spearman correlation coefficient	Not evaluated	The Spearman test reveals a significant correlation between MP3 and the fourth cervical vertebra.

L. Franchi et al.,[43] 2000, longitudinal study	24 (15 girls, 9 boys)	3-18 years	Lamparski	Body height	ANOVA	Not evaluated	Not evaluated	CVM appears to be an appropriate method for the appraisal of mandibular skeletal growth. This provides helpful indications concerning treatment timing of mandibular deficiencies.
S. Hosni et al.,[44] 2018, prospective study	108	8-18 years	Beccetti et al.	Body height	Cohen's Kappa	Not evaluated	Not evaluated	The peak in statural height growth velocity occurred at CVM stage III ($P < .001$). This study suggests that there is a significant relationship between CVM stage and statural height velocity.
Kocasarac et al.,[46] 2016, retrospective cross-sectional study	116 (43 boys, 73 girls)	8-28 years	Lamparski	Demirjian al. (dental age)	Cohen's Kappa	Spearman correlation coefficient	Not evaluated	A strong correlation between the mineralization of the third molar in males (r= 0,723) and weak for females ($r = 0.371$) was evaluated.
Camacho-Basallo et al.,[47] 2016, cross-sectional study	202 (104 boys, 98 girls)	12 years	Hassel e Ferman	Demirjian al. (dental age)	Not cited	Spearman correlation coefficient	Not evaluated	A correlation between the 2 methods of analysis was assessed, regardless of gender.
Cossellu, et al.,[48] 2014, cross-sectional study	500	10-20 years	Hassel e Ferman	Demirjian al. (dental age)	Cohen's Kappa	Spearman correlation coefficient	Not evaluated	A significant correlation between dental age and CVM was assessed (0.081-0.085).
Montasser,[49] 2019, longitudinal study	26 (14 boys, 12 girls)	9-15 years	Not cited	Not cited	*t*-test	Not evaluated	Not evaluated	CVM3 indicates the pubertal growth peak and CVM2 indicates that the peak is not yet reached.

HWM, hand-wrist maturation; CVM, cervical vertebral maturation; CVMS, cervical vertebral maturation stage; SMI, skeletal maturation index; TW3, Tanner–Whitehouse method; NTUH-SMI, National Taiwan University Hospital Skeletal Maturation Index; ROC, indicates Receiver Operating Characteristics; MP3, middle phalanx of the third finger.

to inadvertently miss or overlook any study. The selection was made systematically as illustrated in the Materials and Methods section. Sixteen studies were judged to be of low quality and therefore did not contribute to the evidence.

Only few studies included in our review used rigid parameters regarding the sample selection. Chang et al.[27] stated that the samples analyzed were chosen randomly; however, the specific criteria for random selection have not been described. Only Uysal et al.[25] established rigorous selection criteria, taking under consideration factors like lack of relative medical records, race, systemic diseases, and medical syndromes. Considering the influence of these co-factors on general growth and development, we believe that their strict selection criteria can contribute to the article substantial scientific evidence.

The scientific evidence of our sample was medium/high due to the presence of errors occurring in most of the studies analyzed, such as the lack of standardization in the procedures performed for data collection, the different age groups included in the studies, and the different methods of analysis of the cervical vertebrae. These factors can influence the results of our research. However, this should not be a major issue with regard to the quality of the studies. A detailed analysis of the articles and very selective inclusion criteria lead to a result which is useful internationally as scientific evidence. The study by Heravi et al.[28] showed different levels of correlation between 4 different CVM methods and therefore the same HWM (Tanner–Whitehouse) in a single sample. This result is consistent with several publications that identified different correlations between CVM and HWM using different methods of study of the cervical spine.[25,27] Therefore, the accuracy, correlation and reproducibility of this approach can be influenced by the method of analysis. In fact, there is a good sort of variety of CVM methods, including an easy qualitative analysis of the form and vertebral dimension, quantitative measures of the vertebral shape (some of which are limited to distances and ratios of height and width), depth of the lower concavity, and other more specific measurements.

In the literature, there are few studies that systematically analyze the efficacy of the CVM method. However, a review of the literature, similar to the present one, was presented by Santiago et al.[4] in 2012. Opposed to our study, the authors affirmed that the CVM method did not demonstrate a good correlation with the HWM method and its effectiveness could not be proved.

The longitudinal study is the best method to carry out research on craniofacial growth and development since it can provide a continuous comparison with respect to the development of the patient. However, most of the studies included in our review were founded on cross-sectional data that have limitations in terms of growth analysis. In fact, transversal sampling is relatively insensitive to individual variability, contrary to what happens in a longitudinal sample. Unfortunately, according to the study by Soegiharto et al.,[19] the difficulties of obtaining a large sample size tend to preclude this methodology.

Many radiological methods for evaluating skeletal age have been described and evaluated, and of all of them, hand-wrist radiography is believed the gold standard in the scientific literature. However, some authors recommend abstaining from hand-wrist radiography because of the extra x-ray exposition in growing patients.[3,13,14] For this reason, in recent years, the assessment of skeletal age based on vertebral morphology as shown by latero-lateral teleradiography has increasingly established itself in the clinical setting.

Anyhow, the evaluation of skeletal age based on the cervical spine has been critically examined by many authors and its clinical validity has been seriously questioned.[4,5,50] Most studies citing highly reproducible results for the CVM method (> 90%) used cervical vertebrae tracings and not actual radiographs during the spinal analysis steps, thus introducing bias in the results. The authors conducted a study to evaluate the reliability of CVM on a sample of 90 teleradiographs obtaining 62% of intra-observer agreement. This result corresponds to a “moderate” agreement. Nonetheless, the authors interpreted this result as poor proof of reproducibility. On the other hand, numerous articles included in our review reported a reproducibility of the CVM method as between 85% and 98% using actual radiographs of the patients.[13-15,21,24,25]

Several studies have described a significant correlation ($.001 < P < .05$) between HWM and CVM.[15,25,27] Other articles included in our review confirmed that bone age determined by the CVM method is an effective tool for the evaluation of skeletal maturity in the same way as hand-wrist radiograph, which represents the most established method of analysis in literature.[2,43] Thus, these studies showed a high correlation index between the 2 methods. Furthermore, Franchi et al.[43] and Baccetti et al.[3] proved the efficacy of the CVM method in predicting the pubertal growth spurt using hand-wrist radiographs as a concern. Nevertheless, these studies were conducted retrospectively, therefore their validity on the current population could be queried. However, a prospective study was performed recently on a current sample, which demonstrated that the CVM method was an efficient tool to predict the pubertal growth spurt and body growth.[44] Examining the high correlation between the CVM and the HWM methods indicated in the previously cited articles,[2,9,15,25,27,43] it can be inferred that CVM classification has the potential to replace HWM in assessing bone maturation and thus eliminate additional hand and wrist radiography which has been widely contested.[51,52]

According to Beit et al.,[5] chronological age is not a good indicator for assessing a patient's stage of development. In fact, there are many factors that influence a patient’s body growth and maturation. These factors include race, genetic conditions, climate, nutrition, hormonal disorders, and environmental influences. For this reason, Montasser et al.[39] recommend taking racial and sex differences into account when using CVM as an indicator of skeletal maturity.

One article included in our review showed that there was a statistically significant correlation between chronological age and skeletal maturity, determined by the analysis of the cervical vertebrae (r = 0.771).[37] All other studies regarding the correlation between dental age and the CVM method also demonstrated, through the Spearman correlation test, a statistically significant correlation between the 2 methods ($P < .001$).[46-48]

According to the study by Franchi et al.,[43] the accuracy of the CVM method provides useful indications in the identification of the mandibular growth peak, with the additional advantage of decreasing the exposure of patients to x-rays.

Some studies included in our review do not support the use of the CVM method if it is not used concurrently with other skeletal indicators for the precise determination of skeletal maturity.[26,28] However, most of the studies have stated that the CVM method is an effective method for assessing skeletal maturity ($P < .05$).[2,9,13-15,21,24,25,27,29,37,43,44,47,48]

CONCLUSION

With a moderate/high level of evidence:

- The CVM method can be considered as an effective tool to determine the growth spurt in growing patients
- The CVM method can be considered in the same way as the skeletal analysis of the hand-wrist method
- There is no clinical reason for submitting the growing patient to a further radiograph of the hand and wrist since the cervical vertebrae are already recorded on lateral cephalograms.

Peer-review: Externally peer-reviewed.

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Review

Orofacial Manifestations Associated with Muscular Dystrophies: A Review

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Main Points

- Patients with muscular dystrophies present alterations in growth and development as well as in orofacial morphology.
- Increased prevalence of malocclusions, of both skeletal and dental origins, characterize patients with muscular dystrophies.
- Different dentofacial characteristics are reported among patients with different types of muscular dystrophies.
- Further research is needed to clarify the orofacial phenotypic expression of muscular dystrophies.

ABSTRACT

The aim of this review is to evaluate the developmental, functional, and morphological aspects of the craniofacial complex in patients with myotonic dystrophy type 1 (DM1), Facioscapulohumeral muscular dystrophy (FSHD), and Duchenne muscular dystrophy (DMD). The degree of disease onset and severity varied from patient to patient, and most parameters indicated a greater degree of deterioration in older patients. It was found that all the muscular dystrophies studied showed altered craniofacial morphology, with malocclusion as the most consistent clinical characteristic. Particularly DM1 patients, who are the most studied, showed significant vertical aberration and post-normal occlusion. DMD patients are reported mainly with altered dental arch dimensions which influence functional capacities. Data for FSHD patients are very limited, but facial asymmetry and muscular weakness appear to be the most prominent findings. Patients with muscular dystrophies present deviations in growth and development as well as in orofacial morphology. Increased prevalence of malocclusions, of both skeletal and dental origins, characterize patients with muscular dystrophies. Different dentofacial characteristics are reported among patients with different types of muscular dystrophies. Further research is needed to clarify the orofacial phenotypic expression of muscular dystrophies.

Keywords: Genetic myopathies, myotonic dystrophy type 1, facioscapulohumeral muscular dystrophy, Duchenne muscular dystrophy, dentofacial morphology

INTRODUCTION

Muscular dystrophies are a group of hereditary degenerative diseases that affect the structure of skeletal muscles. They present related clinical symptoms, which include progressive dystrophic changes in muscle tissue. Moreover, they gradually destroy muscle cells, substituting them with connective tissue, resulting in progressive impairment of muscle strength and function. Muscular dystrophies are often characterized by atrophy in axial, facial, upper, and lower limb musculature. Other groups often affected by degenerative changes include the heart, respiratory, swallowing, and eyelid muscles.[1]

Various researchers have reported an impact of muscular involvement in the growth of the craniofacial complex.[2,3] The effects of a neuromuscular disease on the craniofacial muscles potentially disrupt both facial morphology

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and the functional aspects of dental occlusion. Although the effect of muscular weakness on the function of the craniofacial complex is reported in the literature, there are few systematic studies regarding the evaluation of such patients. Possible findings may contribute toward improvements in both the diagnostic and treatment regimens.[4]

The present review assesses the manifestations of the selected dystrophies through the prism of dentofacial science. The ultimate goal is to combine genetic testing, which verifies the presence of muscular dystrophy, with the orthodontic approach, which studies the effects on the function of the craniofacial complex, potentially leading to improved understanding of the etiology of such diseases to improve the quality of life of the patients.

MYOTONIC DYSTROPHY TYPE 1 (DM1)

Epidemiology

DM1 is one of the most common neuromuscular diseases in adults. The prevalence of the disease is 1:8000 births.[5]

Genetic Background

DM1 is inherited with an autosomal dominant pattern. The genetic locus of DM1 is localized in the long arm of chromosome 19.[6] The molecular basis of DM1 is proved to be caused by an unstable expansion of a trinucleotide repeat (CTG).[7] This repeat is localized in an untranslated area of the protein kinase gene of myotonic dystrophy (DMPK). This gene is expressed considerably in the heart, muscles, and to a lesser extent in the brain.[8]

The length of the trinucleotide repeat has been associated with the severity of the disease and the onset of the symptoms. The size of the repeats in the general population (normal individuals) mostly varies between 5 and 35 CTG. Patients with DM1 inherit a minimum of 50 repetitions, and in some cases, 2000 or more. The expansion of the trinucleotide repeat is found to be increased in consecutive generations of both female and male descendants.[9]

Clinical Picture

The dominant symptoms of DM1 include myotonia and progressive muscle weakness, especially in the face and furthermost areas of the upper and lower limbs. In addition, impairment has been reported in cardiac conductivity, smooth muscle system, hypersomnia, and cataract. In male patients, symptoms such as hair loss, testicular atrophy, infertility, and sexual dysfunction have been depicted.[10]

The weakness distribution is quite characteristic in DM1. In the early onset and frequent symptomatology, weakness of the facial musculature, eyelid ptosis, and impairment of the sternocleidomastoid muscle is included . Atrophy and degradation of temporal muscle lead to a concave shape of the temporal bone.[10] Furthermore, atrophy and degeneration of the masseter muscle have been diagnosed in patients with DM1 through an ultrasound examination.[11] Finally, the atrophy and weakness of pharyngeal muscles, resulting in a nasal tone in the patient's voice, affecting enunciation.[12]

FACIOSCAPULOHUMERAL MUSCULAR DYSTROPHY (FSHD)

Epidemiology

FSHD (MIM#158900), known as Landouzy–Dejerine disease, is one of the most common forms of muscular dystrophy, with an autosomal dominant pattern, with an estimated prevalence of 1:20 000. Although it was initially reported that most of the patients affected were male, the majority of asymptomatic patients were more frequently females. Therefore FSHD is believed to affect both genders equally.[13,14]

Genetic Background

FSHD is thought to result from the abnormal expression in muscle of a gene called DUX4. Typically, DUX4 is expressed only during early embryogenesis and in the cells that develop into sperm. However, when expressed in muscle tissue, DUX4 appears to be toxic.

Linkage studies have reported the genetic locus for FSHD in the DUX4 gene on the long arm's subtelomeric region in chromosome 4 (4q35).[15] Moreover, in this area, a repeating micro-satellite array was located. The size of this repeating array depends on the number of repeats named D4Z4 with a length of 3.3 kb (16). Individuals who suffer from FSHD1 carry a reduced number of 1-10 D4Z4 repeats, while non-patients have 11-100 repeats.[16-18]

Definitive diagnosis of FSHD is facilitated only through molecular genetic testing. This can be achieved with the Southern blot technique, by determining the number of repeating D4Z4 arrays, with the use of genomic DNA, which is digested with EcoRI and hybridized with the probe p13E11.39. Numerous reports have described an inverted relationship between the severity of the clinical symptoms and the number of D4Z4 repeats, because patients with a smaller number of repeat arrays exhibit a more severe phenotype. It is worthwhile mentioning that 5% of FSHD patients do not present deletions of D4Z4 arrays (non-linked FSHD) and are considered to be related to a second phenotype known as FSHD2.[17,19]

Clinical Picture

FSHD is described as a slow and gradually progressing muscular dystrophy, with onset in the second or third decade of life. The symptoms can develop at any age, from infancy through advanced age. The most prominent symptoms are weakness of the facial and shoulder muscles, followed by ankle and medial leg musculature weakness. There have also been reports of congenital and early onset cases, with various symptoms and degrees of severity.[20]

The disease shows gradual progression, due to which it is quite common that patients do not report all of their symptoms because they consider them to be caused by other diseases or injuries. In addition, although FSHD is inherited with an autosomal dominant pattern, an adverse family history does not exclude the existence of the condition. Between 10% and 30% of cases are caused by new (de novo) mutations and by asymptomatic gene carriers, which are frequently found in families with FSHD.[15]

DUCHENNE MUSCULAR DYSTROPHY (DMD)

Epidemiology

The estimated prevalence of DMD is estimated at 1:3300-4700 live male births.[21]

Genetic Background

DMD is an inherited myopathy, in which a genetic mutation is observed in the Xp locus, in the short arm of the X chromosome.[22] DMD is an X-linked disease caused by the absence of a protein named dystrophin, which is found more frequently in skeletal muscles and neurons in specific regions of the central nervous system.[23,24]

Clinical Picture

The initial DMD symptoms appear in childhood (usually around 5 years of age) with difficulty in walking. The most prominent symptoms include severe progressive muscle weakness in the pelvic, shoulder, upper, and lower limbs.[25]

METHODS

A literature search was conducted, including the online databases PubMed, Google Scholar, Scopus, Cochrane Library, MEDLINE, and Embase, by using the following search terms and their combinations: "Orofacial manifestations" AND "Muscular Dystrophies" OR "Genetic Myopathies," "Orofacial manifestations" AND "Myotonic dystrophy," "Orofacial manifestations" AND "Facioscapulohumeral muscular dystrophy," "Orofacial manifestations" AND "DMD." The references from the identified articles were incorporated to discover supplementary related publications.

Our inclusion criteria were the following:

1. Clinical studies based on humans only, including randomized control trials, clinical control trials, prospective or retrospective cohort studies, case reports, and case series.
2. Scientific articles written in the English language, and those in other languages were included if an English abstract was available.

Our exclusion criteria were in vitro or animal studies, and clinical outcomes based on questionnaires. Scientific articles in other languages than English, without an English abstract.

Clinical and Research Consequences

The total number of articles that met our inclusion criteria was 38. These articles were assessed and consequently reviewed.

Manifestations of DM1

The majority of research conducted regarding the manifestations of DM1 is currently related to the effects of the disease on craniofacial growth and development. Patients diagnosed with an early onset of DM1 present a hyperdivergent facial growth. A weak muscular system combined with a disrupted force equilibrium affects both the morphology of the craniofacial complex and the occlusion.[26,27] Patients with DM1 exhibit increased vertical facial growth compared to the normal population, narrower maxilla in the transverse dimension, and deeper palatal vault.[6,26,28] Kiliaridis et al.[26] reported an increased prevalence of malocclusion in DM1 patients, such as retrognathic maxilla and mandible, anterior open bite, and posterior crossbites. These results come in agreement with those reported by other researchers.[28,29]

Gazit et al.[30] described decreased strength in the orbicularis oris muscle, tongue thrusting, and oral breathing. Also, temporomandibular joint symptoms tend to affect patients with DM1, such as disc displacement, repeated locking during opening or closing of the mouth, and clicking sounds.

As reported by various researchers, the decreased biting force of DM1 patients advocates for impaired muscle strength due to the disease.[31] The involvement of facial musculature might be the reason for the mandibular clockwise rotation, either due to gravity or due to the absence of supra-hyoid muscle support.[32] This mandibular movement consequently affects the position of the tongue and head posture. In addition, new conditions are formed around teeth in the transverse dimension. The tongue, is positioned lower in the oral cavity, following the mandibular rotation, and cannot counterbalance the forces created by the disrupted facial musculature. These newly established conditions could affect the transversal position of the teeth, reducing palatal width and causing posterior crossbite. In combination with the decreased occlusal forces, the new postural position of the mandible might contribute to over-eruption of the maxillary posterior teeth. In this case, the palatal depth increases because of the overeruption. The lower jaw may rotate even more in the clockwise direction, contributing to the increase of the angle between the mandibular plane and the palatal plane.[33]

The increased prevalence of Angle Class II malocclusion in patients with DM1 is quite possibly linked to this mandibular posterior rotation. Moreover, changes in the mandibular plane angle are in concordance with Wolff's theory (1892), which states that bone shape and structure are related to the pressure of the functional forces applied by the muscles.[34] The facial and occlusal characteristics of DM1 patients resemble the "adenoid face," where the neuromuscular adaptation contributes to a secondary backward rotation of the mandible and changing of head posture to facilitate oral breathing. Notably, in myotonic dystrophy patients, the muscular weakness is the reason which leads to the backward rotation of the mandible, contributing to oral breathing, because these patients do not present impairment of nasal breathing.[35]

Difficulties in mastication and swallowing as symptoms of DM1 have been reported in the literature.[9,29,35-37] Kiliaridis et al.[26] described that DM1 patients need 2.5-fold more time for mastication and have 2.5-fold larger mastication cycles than healthy controls. Also, they had, on average, the half-maximal biting force compared with a healthy population.[26] Swallowing difficulties are related to myotonia, muscle atrophy, and weakness, xerostomia. In more detail, cases of choking during feeding, repeated swallowing attempts, and gastroesophageal reflux have been reported.[36] Harper et al.[9] highlighted the importance of palatal morphology in swallowing. In combination with the mandible and tongue position, this could precipitate aspiration of food into the bronchial tree.[9] This condition could be further aggravated by the degradation of pharyngeal and esophageal muscles.[9] According to Sjogreen, congenital and childhood-onset cases of DM1 exhibit reduced facial expressiveness, apprehension, feeding difficulties, and sialorrhea compared with healthy children of the same age.[38]

Finally, in a lateral cephalometric radiography study conducted by Fotinha et al.[39] in 2018, the following findings were reported in patients with childhood-onset and congenital form of muscular dystrophy. In the sagittal plane, the ANB angle was increased, and the SNPg angle was decreased in DM1 patients. In the vertical plane, in concordance with the literature, the mandibular plane angle (ML-SN) and the intermaxillary angle (ML-PP) were increased compared with a healthy population. Moreover, a difference was noted in the anterior cranial base angle (NSBa), which was reduced in DM1 patients. It is worthwhile mentioning that in 5 years, mandibular plane and intermaxillary angles did not decrease, in contrast with the same values in unaffected subjects.[39]

Manifestations of FSHD

One of the most characteristic symptoms of FSHD is the asymmetric weakness of facial muscles. The ones who are more frequently affected are orbicularis oris, orbicularis oculi, and the zygomaticus major muscle. Facial weakness is quite evident in 25% of patients, and often the participation of facial muscles is not recognized by up to 60% of the patients themselves. The patients themselves rarely report symptoms of facial weakness, and for that reason, it is quite essential that they are assessed by the examining clinician.[20,40]

The weakness in the orbicularis oculi affects the patient's ability to close his eyelids. Many tend to sleep with their eyelids partially open and experience a conjunctiva inflammation when they wake up. When a patient with a more severe phenotype is asked to closed his eyelids Bell’s phenomenon is apparent, which is an upward and outward movement of the eye. Furthermore, the lower weakness of orbicularis oculi may lead to “signe de cils,” which is characterized as the eyelashes’ inability to close entirely when the patient attempts to close his eyes tightly.[41,42]

The weakness in the orbicularis oris muscle results in evident asymmetry when in a rest position, which is quite prominent when the patient is asked to contract his lips or fill his cheeks with air. Tasks such as whistling, blowing a balloon, or consuming a liquid from straw could be quite challenging. In some cases, though, the upper lip might lose its mobility. The affection of zygomatic muscles contributes to the impaired ability to raise the corners of the mouth. Thus, when the patient tries to smile, his lips move in a horizontal direction producing the so-called “transverse smile,” characterized as a frowning grin. In many patients with a severe clinical picture of the disease, an extended weakness of the facial muscles may lead to a “myopathic face” absent of facial expressions.[41,42]

Finally patients with FSHD present poor oral hygiene, impaired masticatory functions, decreased mouth opening, high palatal vault, narrow dental arches, and a variety of malocclusions.[43]

Manifestations of DMD

After evaluating dental casts of DMD patients, Egli et al.[44] reported a higher prevalence of malocclusion compared with the healthy controls. First of all, anterior and posterior open bites were more frequent in DMD patients. The overbite was considerably decreased in individuals with DMD, supporting the increased tendency of anterior open bites. Moreover, changes in overjet and molar relationship were not found to be of significance. During the 2-year assessment of DMD patients by Egli et al.[44] the changes that were identified as most significant were in the transverse dimension, especially the increment of inter-molar, inter-premolar, and inter-canine mandibular widths. According to the assessment of cephalometric radiographs, the ANB angle was decreased over 2 years, while NSBa was increased. The SNB angle had also reduced, but not significantly.[44]

The maximal bite force of posterior teeth, as well as the labial force, were reduced in DMD patients compared to the control group.

The healthy population presented an increase in the previously mentioned categories during the 2 years in contrast with the individuals with DMD. Regarding the masseter muscle width, no significant differences were noted between the 2 groups.

Another study conducted by Morel et al.[45] reported a tendency for an Angle class III malocclusion in older DMD patients. Overjet exhibited a significant decrease during the growth of DMD patients compared with the healthy population.[45] In addition, the transverse dimension results were in concordance with those found from Egli et al.[44] The presence of posterior crossbites was increased in DMD patients. In both upper and lower arches, an increment of the inter-molar and inter-premolar widths was reported, as well as an increase in the mandibular inter-canine distance. It is worth mentioning that inter-molar distance was considerably increased in older patients, leading to the conclusion that individuals with DMD presented progressive wider mandibular dental arches. The angular measurements of ANB and lower incisor inclinations and the lower arch depth were reduced. Finally, the values of the mandibular plane angle (ML-SN) and intermaxillary angle (ML-PP) did not present any statistically significant difference between the DMD patient and the healthy control group.[44,45]

DISCUSSION

The reviewed literature demonstrates the orofacial manifestations of 3 of the most frequent types of muscular dystrophies.

According to the existing research, young patients with congenital or childhood-onset DM1 present a differentiated craniofacial morphology and an abnormal growth pattern compared to the average population. Particularly, differences at the sagittal level were observed with retrusion of the mandible as the more significant. In the vertical dimension, the mandibular plane and intermaxillary angles were significantly increased in DM1 patients and did not decrease during a longitudinal assessment in a 5-year period as documented in healthy controls.[26,28] A possible explanation could be that DM1 patients have a head posture similar to individuals who present oral breathing. This fact is consistent with the findings in patients with an oral breathing pattern, who had a smaller anterior cranial base angle (NSBa) similar to the DM1 patient group.[35] The craniofacial characteristics of DM1 can possibly be linked with weakness of the masseter muscles, which could impair the oral health as well as the function of the patients.[38] Individuals with such a clinical picture might present further deterioration of their oral functions, and their diet should be accordingly adjusted.[46] Furthermore, it has been described that populations with neuromuscular diseases, and especially with DM1, present an increased prevalence of malocclusions. This tendency for malocclusions could be correlated with the deviation of the vertical dimension during the craniofacial growth and development, which is affected by masseter and hyoid muscle impairment and leads to a backward head posture. Finally, it is worth mentioning that the facial muscles' weakness may lead to a clockwise rotation of the mandible, resulting in decreased occlusal forces in the posterior teeth region. This condition could initiate the overeruption of posterior dentition, causing a deeper palatal vault, reduced palatal width, and posterior crossbite, as it has been documented.[26] DM1 affects patients from a young age, and in some instances, the deviation of the vertical dimension of craniofacial morphology could be characterized as an initial sign of the disease.

Facioscapulohumeral muscular dystrophy exhibits a progressive, gradual muscular weakness, although disease severity differs even between members of the same family. If we compare other muscular dystrophies with FSHD, it has been described that the latter possess a steady progression course. Due to alternating periods of steady progression and rapid development of muscle weakness, the anticipation of disease course is challenging. The main factors contributing to a severe phenotype are the early onset of symptoms and short repeat arrays (10-20 kb).[47] Although life expectancy is not reduced, FSHD patients should seek genetic counseling to obtain information regarding possible upcoming disease flare-ups.[20] It is already known that FSHD affects the face, shoulders, back, and leg muscles.[41] Patients frequently do not recognize the disease's symptoms and signs, highlighting the necessity of neurological clinical examination and diagnosis confirmation through genetic testing.

It has been demonstrated from current research that DMD affects both orofacial function and structure.[48-50] Phenotypes of older patients deteriorated on a larger scale than younger ones, and cases of an open bite, reduced overjet, and posterior crossbite appear at a higher prevalence. Although a significant skeletal deviation with a tendency towards an Angle Class III skeletal malocclusion was reported in the sagittal dimension, at the vertical level, no statistically significant changes were noted, despite the decreased overbite. The fact that the ANB is decreased tends to confirm the Angle Class III pattern of skeletal growth. It could be hypothesized that the impairment of orofacial function contributed as an etiologic factor in the development of malocclusions.[48-50]

The increased tongue volume, as described by van den Engel Hoek et al.,[48,51] interferes with the posterior dentition's lingual surfaces, leading to an increased inter-molar distance more prominent in the lower than in the upper arch and causing a posterior crossbite. Likewise, the increased tongue volume that might lead to interferences between the posterior dentition could also contribute to development of an open bite.[49,52,53] Moreover, in most patients, there was a buccal inclination of the posterior teeth disrupting the lower arch's parabolic shape, probably due to increased tongue volume.[48,49] It needs to be mentioned that the research by Egli et al.[44] disagrees with the findings of Matsuyki et al.,[54] where it is stated that the mandible rotates backward during growth and the patient's skull can be characterized as dolichocephalic.[44,54] Furthermore, the biting force seemed to diminish, and labial strength remained constant in DMD patients through an extended period, in contrast with the control group, where it was increased. The reason behind this difference might be the progressive weakness of the skeletal muscles.

Nonetheless, the masseter muscle's width did not change, although it is proved that a correlation exists with biting force. A possible explanation for this contradiction could be the substitution of muscle tissue with adipose and connective tissue.[55] The preservation of right occlusion and adequate muscle function for the longest possible time could be vital for an acceptable quality of life of DMD patients.

At this point, we need to highlight that not all patients with muscular dystrophies present the same manifestations in their craniofacial morphology. Comparing the existing literature regarding the orofacial characteristics of DM1 with DMD patients, the latter show a higher tendency for malocclusions, including posterior crossbites due to wider mandibular arches, anterior and posterior open bites, and a tendency for an Angle Class III malocclusion. In the vertical dimension, DMD patients do not seem to present significant deviations. On the other hand, DM1 patients present a higher tendency of deviation of vertical growth, with a clockwise rotation and a retruded position of the mandible.[26,44,45] Although both diseases are characterized by muscular weakness, the posterior crossbite's etiologic factor in DM1 patients is the narrower maxillary arch. On the contrary, posterior crossbite develops in DMD patients due to the transversal widening of the mandibular arch, caused by the reduced tension of the masseter muscle close to the molar region. This, coupled with the oversized but decreased tonicity of the tongue, leads to disruption of the force equilibrium in the oral cavity resulting in the widening of the lower jaw.

The functional matrix theory highlights the muscles' influence on the growth and development of the craniofacial complex.[56] Their impairment by a neuromuscular disease could have adverse effects on the morphology and function of the craniofacial complex, necessitating additional investigations on the topic. Future findings may aid both in clinical diagnosis and treatment planning of a patient with muscular dystrophy. Improved knowledge regarding the phenotypic expression of neuromuscular diseases in the orofacial region may help to particularize genotype-phenotype studies resulting in a higher quality of evidence. For example, the evaluation of facial morphology by incorporating 3D stereophotogrammetry applied by Pucciarelli et al.[57] on patients with spinal muscular atrophy demonstrated the first group of manifestations on facial soft tissues.[57] Similar assessments of the craniofacial complex could be done in various neuromuscular diseases, including muscular dystrophies, aiming at the comparison of their manifestations, facilitating more precision in diagnosis, and differentiating overlapping characteristics that frequently portray these diseases.[57]

A thorough examination of muscle weakness is strongly suggested before the start of treatment because the progressive course of the disease could convolute treatment prognosis. Clinicians, particularly orthodontists, should be very careful when patients who present muscle weakness signs inquire about treatment. It is pretty often that patients with neuromuscular diseases are not aware of their condition, even though the orofacial manifestations of the disease appear before the systemic ones, particularly at young ages. Consequently, the recognition of early neurological symptoms may be critical to the diagnosis of an underlying neuromuscular disorder and establishing a more precise treatment and post-treatment retention plan.

CONCLUSION

The findings of the present review suggest that patients with muscular dystrophies exhibit alterations in growth and development as well as in dentofacial morphology. According to numerous reports, increased prevalence of malocclusions, of both skeletal and dental origins, are strongly associated with muscular dystrophies. Nevertheless, different orofacial morphological and functional characteristics are reported among patients with different types of muscular dystrophies. Finally, future research could aim at elucidating the genotypic–phenotypic correlation in patients suffering from muscular dystrophies.

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ERRATUM

In the article by Farhadifard et al., entitled “Plaque Removal Efficacy of 3 Cleaning Methods for Removable Orthodontic Appliances: A Crossover Randomized Clinical Trial” that was published in the September 2021 issue of the Turkish Journal of Orthodontics (Turk J Orthod. 2021; 34(3): 170-175. DOI: 10.5152/TurkJOrthod.2021.20079), co-author Sepideh Soheilifar's last name was erroneously written as Sohilifar.

The error has been corrected accordingly and updated in the journal’s archive. You may access the updated article via the link below.

https://www.turkjorthod.org/en/plaque-removal-efficacy-of-three-cleaning-methods-for-removable-orthodontic-appliances-a-crossover-randomized-clinical-trial-163930