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Original Article

Maxillary protraction to treat pediatric obstructive sleep apnea and maxillary retrusion: a preliminary report

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ABSTRACT

Introduction: Midface retrusion creates a size deficiency problem in the upper airway that has been improved in children using surgical midface advancement and orthopedic protraction of the maxilla. The results of these treatments have been mostly promising at enlarging the pharyngeal airway. Recently introduced bone anchored maxillary protraction (BAMP) uses implant inserted devices in the jaws to pull the maxilla forward against a backward pressure to the lower jaw. This is a pilot study that examines the use of BAMP as a strategy to treat maxillary retrusion, malocclusion and children with obstructive sleep apnea.

Methods: 15 children, ages 9–16 years with maxillary retrusion creating a skeletal malocclusion were treated with bone anchored maxillary protraction (BAMP) and the results were compared against an untreated control group. 8 children in the treatment group also had sleep disordered breathing/obstructive sleep apnea. All subjects had lateral cephalograms before and after BAMP therapy. The OSA cohort completed the pediatric sleep questionnaire (PSQ) and polysomnography prior to and at the end of BAMP.

Results: The majority of the OSA children (n = 5) showed improvement in their apnea-hypopnea index (AHI) and OSA symptoms after BAMP. Preliminary results of BAMP therapy show improvement in respiratory and airway parameters in OSA children with a highly significant change in the forward position of the upper jaw and enlargement in the nasopharyngeal to oropharyngeal junction as compared to an age and sex matched untreated control group. The outcomes were dependent on the age of treatment initiation and patient compliance.

Conclusions: This preliminary work suggests that bone anchored maxillary protraction may be considered as an adjunctive treatment option in adolescents for improving midface retrusion and sleep apnea, but further work is needed to explore this therapy.

1. Introduction

Anatomical deficiencies are a predisposing risk factor to the development of obstructive sleep apnea (OSA). The literature conclusively points to a deficient lower jaw as a risk factor, partly due to the attachment of the largest pharyngeal dilator, the genioglossus, to the lower jaw. While much attention is directed to the anatomy of the lower jaw, midface deficiency in adults was first described in 1984 [1] as a structural feature associated with obstructive sleep apnea, and later concluded by others [2–5]. Breathing problems can develop in maxillary retrusion as the maxilla is retropositioned and encroaching on the pharyngeal airway. The cranial skull base orientation can also be altered, creating a shallow nasopharynx. The maxilla is affected in width and length and there is narrowing of the nasal cavity from alterations in palatal width creating an increase in airway resistance [6] that is central to pediatric OSA. These palatal width discrepancies can present as a high arched narrowed palatal vault, prominent palatal shelves or increased palatal soft tissue.

The incidence of maxillary retrusion varies according to geography and ethnicity. There are no studies that examine the

Abbreviations: AHI, apnea-hypopnea index; OSA, Obstructive sleep apnea; BAMP, bone anchored maxillary protraction; PSG, polysomnogram.

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relationship of OSA to maxillary retrusion across the populations. In the general population, the incidence of maxillary retrusion has been cited as 1–26%, with a higher predominance in Asian populations [7]. Midface hypoplasia, or more currently termed midface retrusion, may present with posterior positioning and/or vertical shortening of the infraorbital and perialar regions of the maxilla, often resulting in an anterior dental crossbite and a concave facial profile. More often the term maxillary or midface hypoplasia is associated with premature fusion or synostosis of the facial and cranial sutures, as a feature of a complex craniofacial syndrome, where these children are at a high risk of developing OSA [8]. In the pediatric non-syndromic population maxillary retrusion has not been identified as a risk factor for OSA as it has in adults.

Often maxillary retrusion can be confused with the appearance of mandibular prognathia, but it can present as maxillary retrusion, mandibular prognathia or a combination of both. Treatment of the deficient midface in children was pioneered by the surgeon Delaire in 1976 [9] and orthodontist Petit in 1983 using an extraoral facemask appliance placed against the forehead and chin with elastic traction pulling the upper dentition forward in an effort to protract the maxilla. The appliance was anchored on the dentition and used to move the teeth and the maxillary complex, with resulting improvements on the occlusion. Often the concurrent tooth movement was greater than the skeletal protraction, limiting the usage of the appliance to children up to age [10,11].

Studies within the last decade have shown an accompanying improvement in the size of the airway with maxillary protraction. A recent meta-analysis of 6 studies concluded that maxillary protraction appliances can lengthen the nasopharynx and the posterior pharyngeal airway behind the maxilla [12]. However, these results were not always stable over the long term, with reported dentoalveolar relapse of 25–30%, and little mention on the stability of the skeletal orthopedic effect of the protracted maxilla or increase in posterior airway dimensions.

Bone anchored appliances for orthopedic and dental movement were introduced in 2008 [13]. Miniplate implant anchors are inserted directly into the maxilla to exert protracting loads to the upper jaw. The direct application of an applied load to the facial skeleton that bypasses the dentition offers distinct advantages over a removable facemask appliance. This pilot study examines the use of BAMP as a strategy to treat maxillary retrusion in children with OSA.

2. Materials and methods

2.1. Treatment group & OSA group

This pilot study is preliminary BAMP work on 15 children with maxillary retrusion and a resulting malocclusion using titanium bone anchored miniplate implants to protract the maxilla forward. All children were screened for OSA. 7 of these children presented for treatment for a class III malocclusion and did not report any symptoms of OSA so further OSA testing was not indicated. 8 of these children presented with a diagnosis of OSA, maxillary retrusion and a class III malocclusion. All treatment subjects, ages 9–16 years, had clinical evaluations and cephalometric radiographs taken to assess skeletal and dental positions prior to insertion of the bone anchors at time point T_0, and compared to progress cephalograms taken during treatment (time point T_1). For the 8 children with maxillary retrusion and OSA, current polysomnograms and pediatric sleep questionnaires (PSQ) [14] were obtained before BAMP was initiated (T_0) and after a defined treatment period (T_1).

A total of four bone anchored miniplates were placed in the upper and lower jaws. Two (2) bone maxillary bone anchors (either DePuy Synthes or Surgi-Tec, Bruges, Belgium) were placed on the infrazygomatic crest, fixated to the bony surface underneath the mucosa, as described by Cornelis et al. [15], and 2 mandibular bone miniplate anchors (either DePuy Synthes or Surgi-Tec, Bruges, Belgium) were placed on the surface of the anterior mandible underneath the gingival mucosa parallel to cortical bone and between the roots of the permanent canine and lateral incisor [15], as seen in Fig. 1.

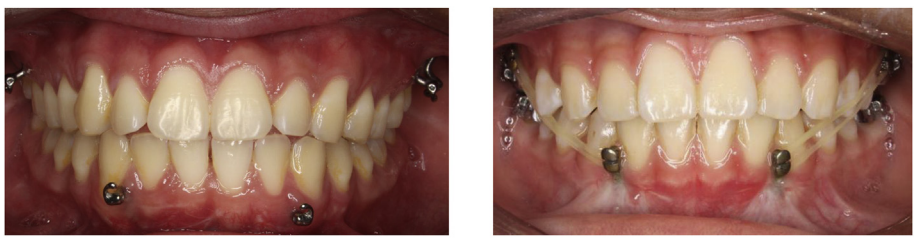
Elastic traction, averaging 200 g per side, was applied that attached the upper to the lower bone anchors. This created a forward and downward pull or protractive loading to the maxilla counterbalanced against a backward and upward directed loading to the mandible, as shown in Fig. 1B. A near constant load of elastic traction was applied on average of 20–22 h per day for a duration on average of 20 months ± 3.76 months.

2.2. Control group

6 untreated age and sex matched children with maxillary retrusion served as the control group. The observation period of 19 months ± 1.52 months for the control group was age matched to the treatment group. Within the control group only one of these six children had OSA and declined any OSA therapy. Two other children declined any BAMP therapy. Two other children had bone anchors placed, but the anchors loosened shortly after placement before any elastic traction was applied and the families declined replacement. One child had the bone anchors placed but refused to wear the elastic traction. Lateral cephalograms were taken to document growth changes during the observation period. A diagnostic PSG was taken for the OSA child in the control group, but since no treatment was rendered during the study time period, a follow-up PSG was not recommended.

2.3. Sleep variables

The 8 children with OSA had diagnostic and final full sleep studies (level 1 PSG) rendered anonymous, performed by certified specialists, and scored using the recommended criteria established



A B

Fig. 1. Placement of Bone Anchors. A: Mandibular anchors positioned between the permanent lateral incisor and cuspid and the maxillary anchor positioned under the zygoma but emerging at the upper first molar. Surgi-Tec type miniplate anchors. B: Elastic traction attaching the upper to the lower bone miniplate anchors, DePuy Synthes type.

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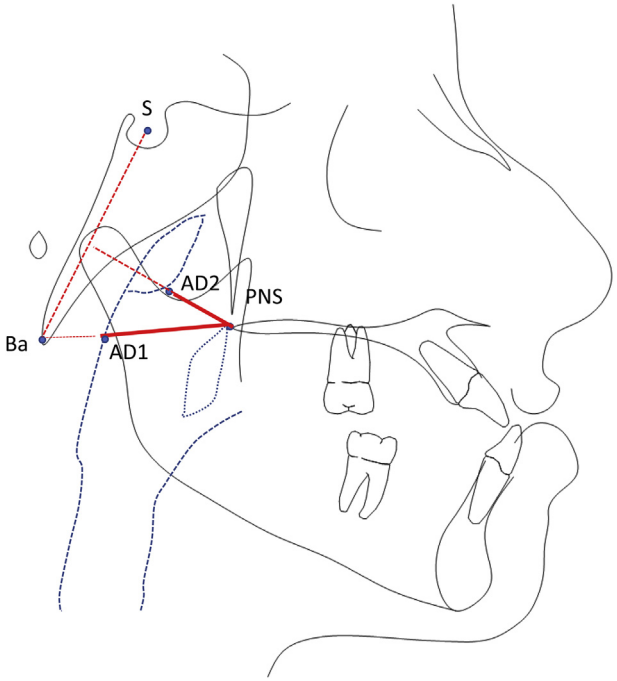

Fig. 2. Landmarks and determination of pharyngeal airway space, AD1, AD2 [19].

by the American Academy of Sleep Medicine (AASM) guidelines [16] for sleep and wakefulness and the respiratory variables: apnea, hypopnea, lowest oxygen saturation and calculation of an apnea-hypopnea index (AHI) based on the recommended criteria.

2.4. Cephalometric variables

Lateral cephalograms 2D (Planmeca Promax) were taken on all subjects. The cephalograms were analyzed using the Bjork [17] and Harvold analyses (1974) [18] and measurements of airway size were based on the work of Linder-Aronson & Henrikson (1973) [19], as illustrated in Fig. 2. Cephalometric measures included maxilla and mandible positions and lengths, incisor position and inclination, anterior and posterior facial heights, sagittal and vertical relationships and proportions and pharyngeal airway dimensions. Dolphin Imaging software™ (Patterson Technology, Chatsworth, CA, USA) was used to digitize and trace the cephalograms.

2.5. Statistical analysis

One-way ANOVAs were performed comparing the 3 groups: BAMP with OSA at T_0, BAMP without OSA at T_0, and Control at T_0 for the 5 measures of mid-face hypoplasia: Midface Length Co-A, Mx Length Co-ANS, SNA, ANB, ANPg There were no significant differences among the 3 participant groups for any of the 5 measurements of midface retrusion. Thus, we conclude that at baseline, all participant groups had comparable midface retrusion.

The treatment effects (cephalometry and respiratory values) between T_0 to T_1 were compared by 2-way analysis of variance. The entire BAMP treatment group as a whole was examined against the control group and the OSA subjects treated with BAMP were compared against the control group. Statistical analyses were performed with statistical significance set at $P \leq 0.05$. The results were expressed as means ± standard deviations. An analysis of variance with mixed model was first performed and then a post-hoc analysis comparing T_0 to T_1 for each subgroup was obtained.

Table 1A
Comparison of Cephalometric Data of 8 OSA children with maxillary retrusion treated with BAMP at baseline T_0 and after treatment T_1 versus untreated maxillary retrusion controls.

Parameter	Pre-treatment OSA (T_0)		Pre-Treatment Control (T_0)		Post-treatment OSA (T_1)		Post-Treatment Control (T_1)		Difference OSA (T1−T0)		Difference Control (T_1−T_0)		Significance
	Mean	SD	Mean	SD	Mean	SD	Mean	SD	Mean	SD	Mean	SD	
SNA	79.33	5.50	80.85	3.93	80.69	6.21	78.50	3.84	1.36	2.15	−2.35	2.08	0.0070 (**)
SNB	81.11	5.57	83.08	4.29	81.96	7.34	82.08	3.54	0.85	3.44	−1.00	3.97	0.37 (NS)
SNPg	81.50	5.23	84.07	4.96	82.63	8.20	82.98	4.16	1.13	3.55	−1.08	4.18	0.31 (NS)
SN-PP	8.85	5.41	7.30	7.37	7.01	7.50	9.35	5.06	−1.84	3.11	2.05	3.51	0.049 (*)
ANB	−1.79	2.42	−2.23	2.79	−1.26	3.09	−3.57	2.77	0.53	3.64	−1.33	4.52	0.41 (NS)
N-ANS	48.46	6.20	50.10	5.39	50.09	8.81	52.40	6.04	1.63	2.99	2.3	3.23	0.69 (NS)
ANS-Gn	61.49	8.79	65.20	12.86	62.86	9.12	66.35	13.58	1.38	2.55	1.15	1.07	0.84 (NS)
N-Gn	109.53	13.45	114.90	16.88	112.11	15.44	118.40	19.91	2.59	2.66	3.00	3.47	0.59 (NS)
ADP1	18.78	3.53	20.80	3.88	23.59	3.13	19.48	3.06	4.81	4.14	−1.32	3.32	0.012 (*)
ADP2	17.45	4.71	16.53	3.32	20.36	3.70	17.25	0.82	2.91	4.85	0.72	3.26	0.36 (NS)
Go-Pg	56.81	7.76	59.85	7.49	57.57	5.43	63.2	7.48	0.76	9.91	−1.32	3.32	0.41 (NS)
MP-OP	16.70	3.46	20.30	4.42	19.99	6.17	18.17	4.21	3.29	2.97	−2.13	1.77	0.0020 (**)
Co-A	73.71	3.49	74.90	6.01	77.19	4.26	76.62	6.64	3.48	3.08	1.72	5.66	0.47 (NS)
Co-ANS	78.76	2.94	79.37	5.33	82.99	4.43	81.83	5.30	4.23	4.55	2.47	5.64	0.52 (NS)
Co-Pg	103.18	4.58	107.57	8.49	106.96	4.15	110.90	7.30	3.79	4.04	3.33	6.85	0.88 (NS)

*$P < 0.05$; **$P < 0.01$; ***$P < 0.001$; NS not significant.
SNA - Sella-Nasion-Point A angle: Maxillary position.
SNB – Sella-Nasion-Point B angle: Mandibular position.
SNPg – Sella-Nasion-Pogonion angle: Mandibular position.
SN-PP – Sella – Nasion to Palatal Plane: Upper matrix rotation.
ANB – Point A – Nasion-Point B angle: Difference between.
N-ANS – Nasion-Anterior Nasal Spine: Upper facial height.
ANS-Gn – Anterior Nasal Spine – Gnathion: Lower facial height.
N-Gn – Nasion – Gnathion: Anterior facial height.
ADP1: Distance from Posterior nasal spine to the posterior pharyngeal wall, measure through basion.
ADP2: Distance from Posterior nasal spine (PNS) to the nearest adenoid tissue, measured though a perpendicular line to Sella-Basion from PNS.
Go-Pg – Gonion – Pogonion: Mandibular length.
Mp-OP – Mandibular Plane–Occlusal Plane: Lower Matrix rotation.
Co-A – Condylion – A point: Maxillary length.
Co-ANS – Condylion – Anterior nasal spine: Maxillary length.
Co-Pg – Condylion – Pogonion: Mandibular length.

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3. Results

3.1. Cephalometry & clinical evaluation

The subgroup of the 8 OSA subjects showed significant differences in the maxillary position and the pharyngeal length, with the results shown in Table 1A. Comparing the treatment effects of BAMP to natural growth as seen in the untreated control group, there was only 1 linear and 3 angular measurements that were significantly different. After BAMP, the nasopharyngeal airway (PNS-AD1) was larger and the angular position of the maxilla relative to the cranial base (SNA) was longer, and the relative positions of the dentition (SN-PP palatal plane to the cranial base and the MP-OP mandibular plane to the occlusal plane) differed when compared to the untreated controls. Linear measurements of maxillary and mandibular length (Co-A, Co-ANS, Co-Pg) were not different compared to the control, and this may partly be attributed to the ambiguity of landmark identification of condylion, Co.

These significant differences of the OSA subgroup were mirrored in the before and after cephalograms for the entire treatment group of 15 subjects, shown in Table 1B. The same variables found to be significantly different (SNA, ADP1. MP-OP) were again significantly different when comparing the entire BAMP treatment group to baseline T_0 and to controls baseline and T_1.

The malocclusion was improved in varying amounts in all subjects that consistently used the elastic traction. The subject in Fig. 3 with midface deficiency had little improvement with extraoral tooth borne facemask therapy when treated at a younger age, as shown in the jaw growth tracing in Fig. 4. BAMP treatment redirects mandibular growth vertically or posteriorly, but once the elastic traction ceases during puberty, forward lower jaw growth was evident, as seen in Figs. 5 and 6, with a return to the original growth pattern. Enlargement of the midface was also noticeable as the malar eminence was prominent and the inferior sclera exposure was eliminated, similar to prior results [20]. The facial and profile changes and occlusal changes are depicted in Figs. 3, 5 and 7. Both of these dolichofacial or leptoprosopic facial patterns were modified to a more mesofacial or mesoprosopic facial pattern after BAMP. (see Fig. 8).

3.2. Polysomnography

Of the 8 children with OSA undergoing BAMP, 5 showed an improved response on post treatment polysomnograms, as depicted in Table 2. In these very small groups, no significant changes were noted with ANOVA, however there were clear trends. The mean AHI for the OSA subgroup was 9.4 ± 6.3 at BAMP treatment initiation, and at T_1 the mean AHI decreased to 7.1 ± 5.9. For the responders, the AHI decreased from a mean of 9.86 ± 4.54 to 4.54 ± 1.77. The minimum oxygen saturation also improved with therapy, and in both the non-responders and the responders there was an increase in REM staging. The non-responders showed an AHI increase from 8.6 ± 3.8 to 11.23 ± 6.8. These subjects had no appreciable change in minimum oxygen saturation. Few significant findings were shown either at $P = 0.05$ or $p = 0.005$. The small number of subjects has to be taken into consideration. The small sample size precludes any definitive findings, but these results suggest BAMP as a possible treatment option for pediatric OSA that should be further investigated.

3.3. Complications

The most common complication using BAMP mechanotherapy was the premature loosening of the miniplate anchors, necessitating

Table 1B
Comparison of Cephalometric Data of all 15 children with maxillary retrusion treated with BAMP at baseline T_0 and after treatment T_1 versus untreated maxillary retrusion controls.

Parameter	Pre-treatment n = 15 (T_0)		Pre-Treatment Control (T_0)		Post-treatment n = 15 (T_1)		Post-Treatment Control (T_1)		Difference n = 15 (T1−T0)		Difference Control (T_1−T_0)		Significance
	Mean	SD	Mean	SD	Mean	SD	Mean	SD	Mean	SD	Mean	SD	
SNA	80.08	4.02	80.85	3.93	81.63	4.55	78.50	3.84	1.55	2.09	−2.35	2.08	0.0010 (**)
SNB	82.47	4.48	83.08	4.29	82.70	5.70	82.08	3.54	0.23	2.78	−1.00	3.98	0.43 (NS)
SNPg	82.70	4.23	84.07	4.96	83.18	6.16	82.98	4.16	0.49	2.70	−1.08	4.18	0.32 (NS)
ANB	−2.39	2.32	−2.23	2.79	−1.07	3.16	−3.57	2.77	1.32	3.20	−1.33	4.52	0.14 (NS)
N-ANS	48.62	5.30	50.10	5.39	51.05	6.93	52.40	6.04	2.43	2.69	2.3	3.23	0.93 (NS)
ANS-Gn	60.97	7.30	65.20	12.86	62.92	7.86	66.35	13.58	1.95	2.60	1.15	1.07	0.48 (NS)
N-Gn	109.26	11.28	114.90	16.88	113.35	12.70	118.40	19.91	4.09	3.26	3.50	3.52	0.72 (NS)
ADP1	18.17	5.05	20.80	3.88	22.61	4.88	19.48	3.06	4.43	3.55	−1.32	3.32	0.003 (**)
ADP2	16.70	5.50	16.53	3.32	19.85	4.07	17.25	0.82	3.15	4.34	0.72	3.26	0.23 (NS)
Go-Pg	59.24	7.39	59.85	7.49	57.77	6.44	63.20	4.48	−1.47	9.56	4.52	6.32	0.18 (NS)
MP-OP	17.43	3.57	20.30	4.42	18.73	5.58	18.17	4.21	1.3	3.68	−2.13	1.77	0.043 (*)
Co-A	73.30	3.80	74.90	6.01	77.05	4.05	76.62	6.64	3.75	2.45	1.72	5.66	0.43 (NS)
Co-ANS	77.77	3.67	79.37	5.33	81.37	4.40	81.83	5.30	3.61	3.51	2.47	5.64	0.58 (NS)
Co-Pg	102.06	5.06	107.57	8.49	106.88	5.53	110.90	7.30	4.82	3.38	3.33	6.85	0.63 (NS)

*$P < 0.05$; **$P < 0.01$; ***$P < 0.001$; NS not significant.
SNA - Sella-Nasion-Point A angle: Maxillary position.
SNB − Sella-Nasion-Point B angle: Mandibular position.
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SN-PP − Sella − Nasion to Palatal Plane: Upper matrix rotation.
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Co-A − Condylion − A point: Maxillary length.
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Fig. 3. Facial growth from age 9 to age 16. BAMP treatment initiated at age 13. BAMP continued until age 16.

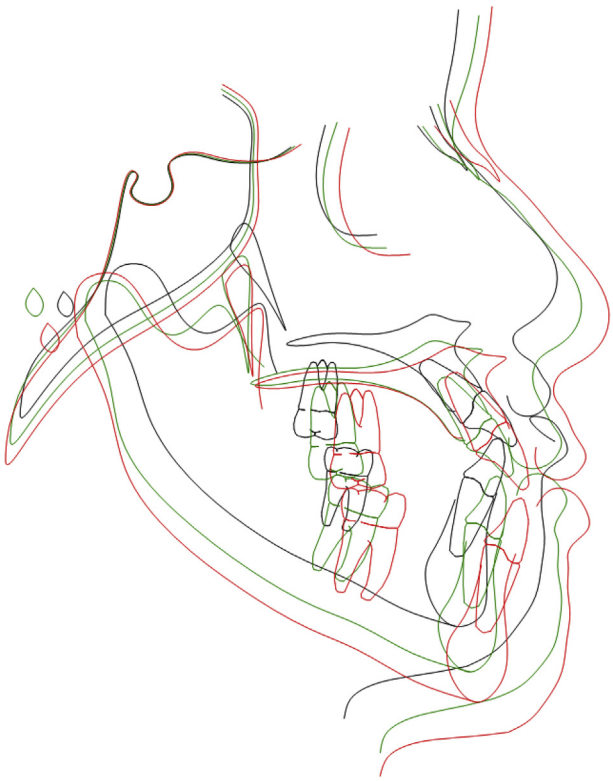

Fig. 4. Growth Superimpositions (method of Bjork) of subject in Fig. 2. Age 9 in black, Initiation of BAMP Age 13 in green, Age 14 in red. Little forward upper jaw growth until BAMP initiated as noted from the green to the red change in the maxilla. Continued vertical lowering of mandibular plane with some forward growth from ages 9 to 13 to 14.

replacement of the fixation device. This affected the outcome as it created delays in treatment. In the treatment group, 5 of the 15 subjects had initial loosening of one or more of the bone anchors, creating delays in loading of the bone anchor while in 2 of these 5 subjects full replacement of one or more of the bone anchors was needed. This agrees with other work that describes loosening of the anchors as an initial challenge to starting treatment [21]. 2 other patients had the bone anchors loosen before elastic traction was applied and they declined replacement and these subjects became part of the control group. 11 subjects showed unexpected tooth movement, both favorable and unfavorable. Depending on the orientation of the inserted anchor, 3 subjects had lingual tipping of the teeth as the anchor was inserted too close to the dentition, worsening the malocclusion in the transverse dimension, while 8 subjects showed favorable upper incisor uprighting, gradual space closure or resolution of crowding and other improvements in the occlusion. Compliance with full time elastic wear was completely dependent on the child engaging fully in treatment. Some patients were inconsistent in maintaining full time force application to the bone anchors during the nearly 2-year treatment course, potentially compromising the outcome.

4. Discussion

4.1. Timing of treatment – age of onset and duration

This is the first preliminary report of maxillary protraction as a strategy to treat OSA in patients with maxillary retrusion. While others have described the impact of maxillary protraction mediated through tooth borne removable appliances to increase the pharyngeal dimensions, this work examined a more direct loading of the facial skeleton via implants anchored to the upper and lower jaws. Tooth borne appliances and mechanics are limited by the concurrent tooth movement that accompanies the skeletal protraction. Extraoral tooth borne protraction devices can promote skeletal changes in younger children up to 10 years of age [11] and can be considered to treat this specific OSA pediatric population, but it is not effective during adolescence.

For the subjects with OSA, bone anchored protraction therapy in this study was implemented after other therapies were rendered. All the 8 children with OSA were diagnosed at an earlier age. Because the bone anchors are placed usually at 10 years or older, the subjects in this study had already been treated with other pediatric therapies, including adenotonsillectomy, bimaxillary expansion, allergy management, or myofunctional therapy.

Bone anchored force delivery offers possible treatment in the pre-teen and teenage population, which is especially important as there is increased growth velocity in adolescence. This is a critical

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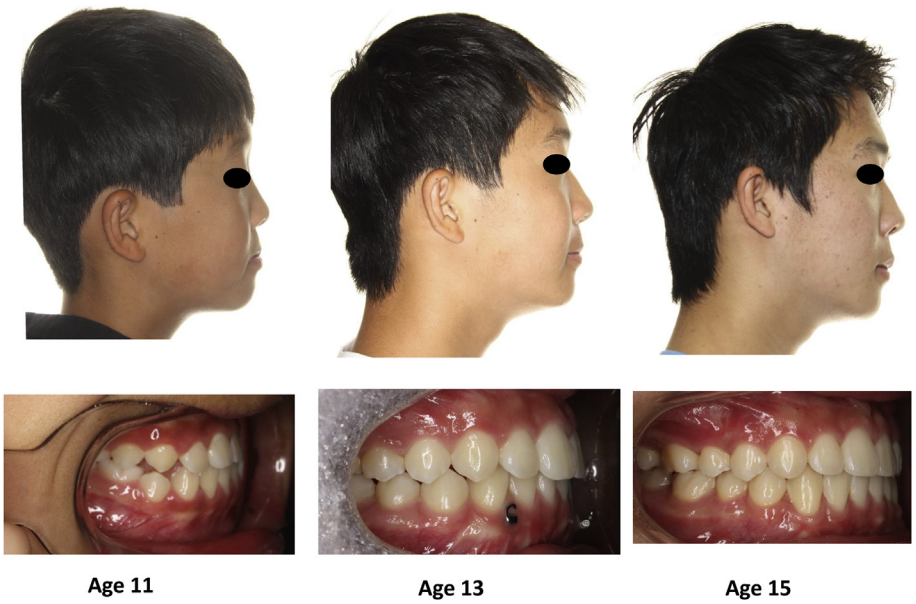

Fig. 5. Facial growth changes from age 11 years to age 15. BAMP initiated at age 11, and finished at age 13.

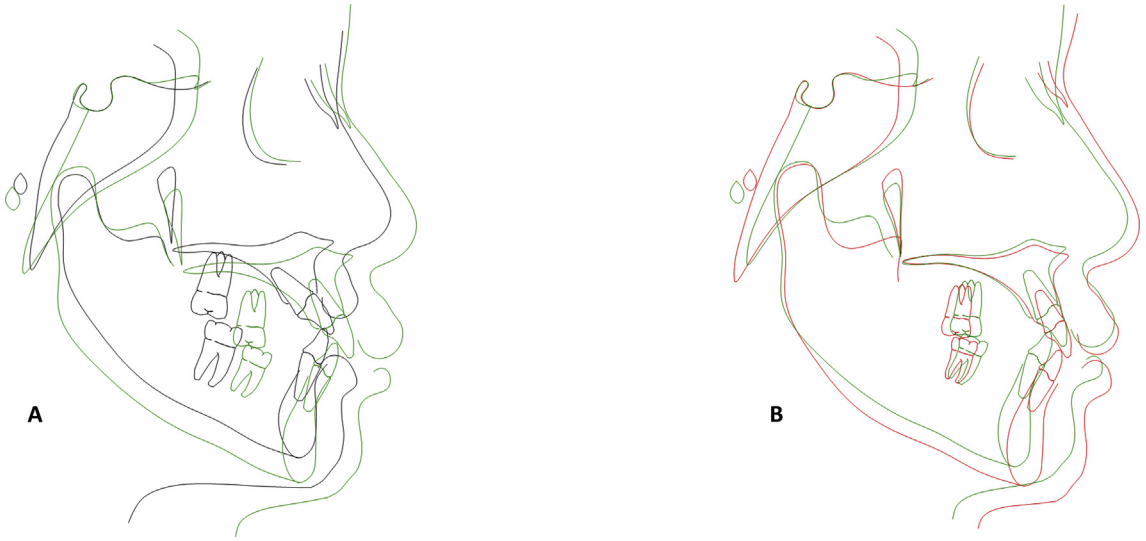

Fig. 6. Maxillomandibular growth changes of patient in Fig. 5. A: Jaw growth after wearing BAMP, age 11 (black)–13 (green). B: Jaw growth 2 years after cessation of BAMP. Refractory growth and return of original growth pattern, or rebound lower jaw growth, age 13 (green)–15 (red).

time for intervention as the soft tissue boundaries that define the pharyngeal airway are influenced by the position of the maxillomandibular complex. Although nasal resistance generally decreases with increasing age in the growing child, there are inconsistencies during the ages 10–13 and 15–17 and temporary increases between age 13 to 14, which mirror the ages of pubertal growth. The most optimal time for BAMP is dependent on the timing of dental eruption of the canines for insertion.

One of the goals of therapy was to maximally protract the upper jaw during active growth to enlarge the posterior airway. Treatment was interrupted if the bone miniplate anchors loosened, which occurred in about 30% of the patients, which is similar to the miniplate survival rate as reported in a recent multicenter trial [22]. Reinsertion of the anchors was not always timely and this created treatment delays. On average, the treatment time was 20 ± 3.76 months, which is longer than BAMP treatment for malocclusions. There is no definition of how long treatment should last if normal nasorespiratory function has not been restored, or even more fundamentally, what constitutes normal nasorespiratory function. It is possible that a longer treatment period and an earlier initiation than some of the older adolescents in our study could yield better outcomes.

4.2. Pharyngeal size

Prior cephalometric 2D and 3D CBCT studies on the effects of maxillary advancement on upper airway dimensions show mixed results, with only one of these studies [23] compared to controls. Even with the BAMP restriction or redirection of mandibular growth [21], BAMP has not been shown to hinder oropharyngeal

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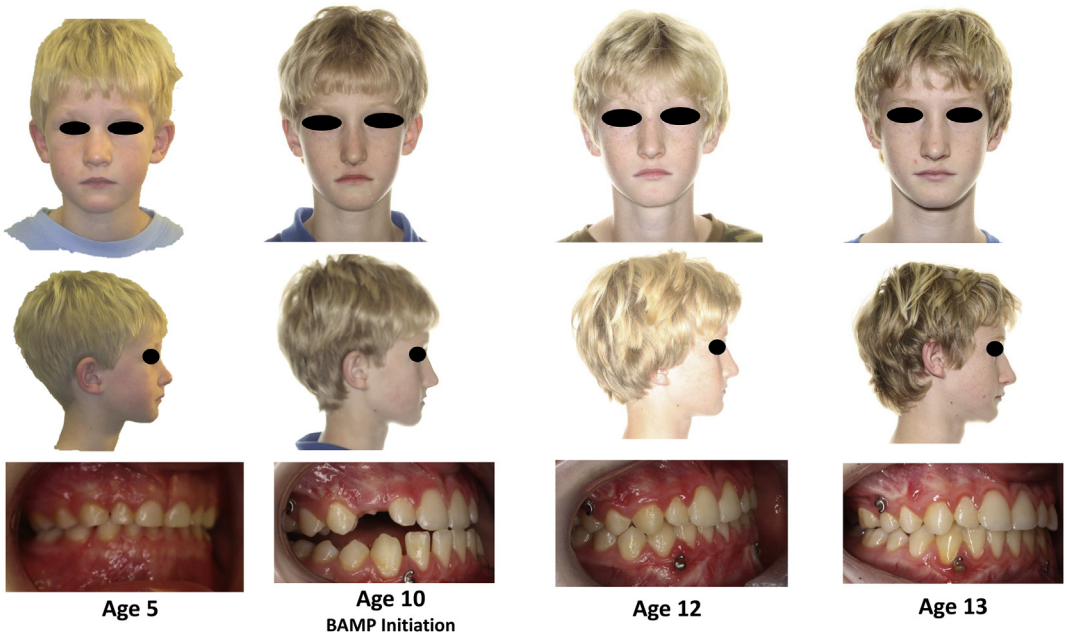

Fig. 7. Facial changes from age 5 to 13. BAMP therapy initiated at age 10 and ongoing beyond age 13.

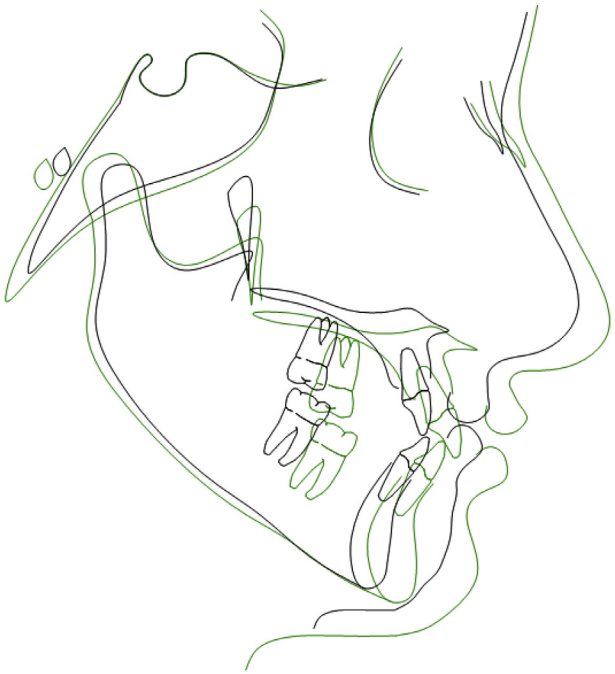

Fig. 8. Facial changes from age 10 (black) to 13 (green), wearing BAMP for 3 years.

airway development [23]. Prior investigations show either no volumetric change in airway measurements [23], or significant changes of both oropharyngeal and nasopharyngeal area dimensions [24,25] or significant improvement only in nasopharyngeal airway dimensions [26,27]. Few studies employed bone anchored traction, but most work is based on tooth anchored (Delaire or Petit facemask) mechanotherapy so the comparisons between tooth borne versus bone borne traction for airway enlargement may not be relevant. Characterizing size differences in the oropharyngeal or hypopharyngeal airway space may also not be accurate, as the pharynx is a collapsible tube with dimensions that are altered by neck flexion and head position so before and after images must be captured in precisely the same head position. In our 2D data, AD1 is a hard tissue landmark measurement that represents the transition between the nasopharynx and the oropharynx and is unaffected by head position. At $p < 0.05$ significance, the anterior-posterior length at the nasopharyngeal to oropharyngeal boundary increased with BAMP when compared to controls.

4.3. Maxillomandibular growth & dental occlusion

BAMP treatment created increases in maxillary lengths at $p < 0.01$. Surprisingly our data showed that the OSA subjects and the entire treatment group had no appreciable restriction of lower jaw growth as compared to the untreated controls despite the backward traction applied to the mandible. Posterior (clockwise) rotation of the mandible, vertical lowering of the chin point (Pg) and anterior facial height increases were evident in the both the

Table 2
PSG Data of 8 OSA children before (T_0) and after (T_1) BAMP.

Parameter	All subjects T_0 n = 8	All subjects T_1 n = 8	Responders (T_0) = 5	Responders (T_1) = 5	Non-responders (T_0) = 3	Non-responders (T_1) = 3
AHI	9.39 ± 6.30	7.05 + 5.85	9.86 ± 4.54	4.54 ± 1.77	8.6 ± 3.8	11.23 ± 6.8
O min Sat	90 ± 7.6	93 ± 1.4	89 ± 4	93 ± 2	92 ± 1	93 ± 1
Total Sleep Time (min)	361 ± 33	4 ± 33	348 ± 31	449 ± 44.3	385 ± 41	469 ± 54
Stage N1 (%)	9.2	8.9	11.7	14.3	3.9	7.7
Stage N2 (%)	40.8	44.0	47.1	44.2	29.6	43.1
Stage N3 (%)	29.5	23.4	26.9	17.4	36.4	29.2
Stage REM (%)	14.4	21.1	14.3	23.9	15.4	19.9

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entire treatment group and the OSA subgroup, but unexpectedly, these were not significantly different from the natural growth changes in the untreated control group, as depicted in Tables 1A and 1B, and in opposition to the work of others [21]. There were significant differences in the angular measurements of the maxillomandibular complex of the treatment group versus the control group, suggesting that dental eruption was interrupted, and dental extrusion and/or intrusion accounts for the changes in the occlusal plane, which differs from other BAMP studies [13,20–38]. Favorable occlusal improvements were also noted in the entire treatment group, showing the efficacy of BAMP in correcting malocclusions and supports the work of others that used this strategy to improve skeletal malocclusions [13,20–23,36–38].

4.4. Improvement in upper airway collapsibility and symptoms

Compliant patients that consistently applied traction to the upper jaw using elastic force showed anatomic changes that differed from controls. The polysomnographic data is mixed in this small sample size as not all patients showed improvements in respiratory values. All OSA subjects had varying degrees of moderate to severe OSA. While most of the children showed a positive trend toward improved AHI response to BAMP, 3 of the 8 OSA children showed worsening of AHI after treatment. The most optimal time for BAMP is cited as age 11 [13], coinciding with the eruption of the lower canine teeth. The 3 non-responders started BAMP at ages 13 or 14 while 2 of these 3 subjects were inconsistent with the elastic traction. This highlights both an age dependent response as their AHI increased indicating worsening of the OSA, although one of the responders was older than 14, and the necessity for patient engagement. Our small sample size precludes further statistical analysis. Based on these preliminary results, future studies with larger numbers of subjects is warranted. We believe that this technique is, however worth reporting as it has never been used in children with OSA with the presented morphometric changes. Children with this anatomic presentation are very vulnerable to CPAP usage as the applied mask pressure will exacerbate the maxillary deficiency.

The 2D data shows an increase at the nasopharyngeal/oropharyngeal transition area in the OSA group and entire treatment group as compared to controls. The oropharynx is the site of collapse in OSA. It is likely that n BAMP did not improve airway narrowing enough, or there were other sites of airway collapse that were not addressed. The responders of this study showed improvements in morning wakening, mood, daytime alertness and nasal breathing. None of the non-responders reported any improvement in symptoms, namely daytime performance, difficulty in morning wakening, restless sleep.

5. Conclusions

The application of bone anchored maxillary protraction may be an approach to treat children between the ages of 10–13 with maxillary restriction. This pilot study provides a preliminary framework to support future prospective studies. Our results must be balanced against the long-term effect of BAMP, as there is reported growth redirection of the mandible and this may impact the size of the hypopharyngeal airway space, although the results of this study do not support this notion. This preliminary work suggests that BAMP offers potential improvement for those OSA patients with maxillary retrusion through enlargement of the nasopharyngeal airway. Dental, skeletal and soft tissue alterations are noted. Although promising, our results should be interpreted with caution because of the small sample size. Ongoing work is focused on the surgical technique to minimize loosening of the miniplate anchors and better patient engagement and compliance. Further long-term 3D studies are required for more comprehensive analyses of airflow and volumetric changes.

Disclosure statement

This was not an industry supported study. The authors have indicated no financial conflicts of interest. This work was performed in clinical practice in Palo Alto, CA.

Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2018.12.005.

References

[1] Guilleminault C, Riley R, Powell N. Obstructive sleep apnea and cephalometric roentgenograms. Am Rev Respir Dis 1984;130:145–6. https://doi.org/10.1164/arrd.1984.130.1.145c.
[2] Lowe AA, Santamaria JD, Fleetham JA, et al. Facial morphology and obstructive sleep apnea. Am J Orthod Dentofacial Orthop 1986;90:484–91. https://doi.org/10.1590/2177-6709.20.6.060-067.oar.
[3] Lyberg T, Krogstad O, Djupesland G. Cephalometric analysis in patients with obstructive sleep apnoea syndrome. I. Skeletal morphology. J Laryngol Otol 1989;103:287–92. https://doi.org/10.1007/s00056-002-0057-x.
[4] Miles PG, Vig PS, Weyant RJ, et al. Craniofacial structure and obstructive sleep apnea syndrome – a qualitative analysis and meta-analysis of the literature. Am J Orthod Dentofacial Orthop 1996;109:163–72. https://doi.org/10.1016/S0889-5406(96)70177-4.
[5] Sforza E, Bacon W, Weiss T, et al. Upper airway collapsibility and cephalometric variables in patients with obstructive sleep apnea. Am J Respir Crit Care Med 2000;161:347–52. https://doi.org/10.1164/ajrccm.161.2.9810091.
[6] Hershey HG, Stewart BL, Warren DW. Changes in nasal airway resistance associated with rapid maxillary expansion. Am J Orthod 1976;69:274–84. https://doi.org/10.1016/0002-9416(76)90076-2.
[7] Hardy DK, Cubas YP, Orellana MF. Prevalence of angle Class III malocclusion: a systematic review and meta-analysis. O J Epi 2012;2:75–82. https://doi.org/10.4236/ojepi.2012.24012.
[8] Marcus CL, Brooks LJ, Draper KA, et al. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics 2012;130:714–55. https://doi.org/10.1542/peds.2012-1672.
[9] Delaire J. Le syndrome prognathique mandibulaire. Orthod Free J 1976;47:203–19. https://doi.org/10.4236/ojst.2014.47051.
[10] Younes M. Role of arousals in the pathogenesis of obstructive sleep apnea. Am J Respir Crit Care Med 2004;169:623–33. https://doi.org/10.1164/rccm.200307-1023OC.
[11] Wells AW, Sarver DM, Proffit WR. Long-term efficacy of reverse-pull headgear therapy. Angle Orthod 2006;76:915–22. https://doi.org/10.2319/091605-328.
[12] Ming Y, Hu Y, Li Y, et al. Effects of maxillary protraction appliances on airway dimensions in growing Class III maxillary retrognathic patients: a systematic review and meta-analysis. Int J Pediatr Otorhinolaryngol 2018;105:138–45. https://doi.org/10.1016/j.ijporl.2017.12.013.
[13] De Clerck HJ, Cornelis MA, Cevidanes LH, et al. Orthopedic traction of the maxilla with miniplates: a new perspective for treatment of midface deficiency. J Oral Maxillofac Surg 2009;67:2123–9. https://doi.org/10.1016/j.joms.2009.03.007.
[14] Chervin RD, Hedger K, Dillon JE, et al. Pediatric sleep questionnaire (PSQ): validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. Sleep Med 2000;1:21–32. https://doi.org/10.1016/S1389-9457(99)00009-X.
[15] Cornelis MA, Scheffler NR, Mahy P, et al. Modified miniplates for temporary skeletal anchorage in orthodontics: placement and removal surgeries. J Oral Maxillofac Surg 2008;66:1439–45. https://doi.org/10.1016/j.joms.2008.01.037.
[16] Iber C, Ancoli-Israel S, Chesson ALJ, et al. The AASM manual for the scoring of sleep and associated events: rules, terminology and technical specifications. 1st ed. Westchester, IL: American Academy of Sleep Medicine; 2007. https://doi.org/10.5664/jcsm.2172.
[17] Bjork A. The face in profile. An anthropological X-ray investigation on Swedish children and conscripts. Am J Phys Anthropol 1948;40:121–5. https://doi.org/10.1002/ajpa.1330060117.
[18] Harvold EP. The activator in interceptive orthodontics. St Louis: CV Mosby; 1974. p. 37–56.
[19] Linder-Aronson S, Henrikson CO. Radiocephalometric analysis of anteroposterior nasopharyngeal dimensions in 6- to 12-year-old mouth breathers compared with nose breathers. J Oto-Rhino- Laryngol Relat Spec 1973;35:19–29. https://doi.org/10.1159/000275083.

Please cite this article as: Quo S et al., Maxillary protraction to treat pediatric obstructive sleep apnea and maxillary retrusion: a preliminary report, Sleep Medicine, https://doi.org/10.1016/j.sleep.2018.12.005

[20] Nguyen T, Cevidanes LHS, Cornelius MA, et al. 3D assessment of maxillary changes associated with bone anchored maxillary protraction. Am J Orthod Dentofacial Orthop 2011;140:790–8. https://doi.org/10.1016/j.ajodo.2011.04.025.
[21] De Clerck H, Nguyen T, de Paula LK, et al. Three-dimensional assessment of mandibular and glenoid fossa changes after bone-anchored class III intermaxillary traction. Am J Orthod Dentofacial Orthop 2012 Jul;142(1):25–31. https://doi.org/10.1016/j.ajodo.2012.01.017.
[22] Van Hevele J, Nout E, Claeys T, et al. Bone anchored maxillary protraction to correct a class III skeletal relationship: a multicenter retrospective analysis of 218 patients. J Cranio-Maxillo-Fac Surg 2018 Oct;46(10):1800–6. https://doi.org/10.1016/j.jcms.2018.07.007.
[23] Nguyen T, DeClerck H, Wilson M, et al. Effect of Class III bone anchor treatment on airway. Angle Orthod 2015;85:591–6. https://doi.org/10.2319/041614-282.1.
[24] Hiyama S, Suda N, Ishii-Suzuki M, et al. Effects of maxillary protraction on craniofacial structures and upper-airway dimension. Angle Orthod 2002;72:43–7. https://doi.org/10.1043/0003-3219(2002)072<0043:EOMPOC>2.0.CO;2.
[25] Baccetti T, Franchi L, Mucedero M, et al. Treatment and post-treatment effects of facemask therapy on the sagittal pharyngeal dimensions in Class III subjects. Eur J Orthod 2010;32:346–50. https://doi.org/10.1093/ejo/cjp092.
[26] Oktay H, Ulukaya E. Maxillary protraction appliance effect on the size of the upper airway passage. Angle Orthod 2008;78:209–14. https://doi.org/10.2319/122806-535.1.
[27] Kilinc AS, Arslan SG, Kama JD, et al. Effects on the sagittal pharyngeal dimensions of protraction and rapid palatal expansion in class III malocclusion subjects. Eur J Orthod 2008;30:61–6. https://doi.org/10.1093/ejo/cjm076.
[28] Sayinsu K, Isik F, Arun T. Sagittal airway dimensions following maxillary protraction: a pilot study. Eur J Orthod 2006;28:184–9. https://doi.org/10.1093/ejo/cji095.
[29] Kaygisiz E, Tuncer BB, Yuksel S, et al. Effects of maxillary protraction and fixed appliance therapy on the pharyngeal airway. Angle Orthod 2009;79:660–7. https://doi.org/10.2319/072408-391.1.
[30] Barrera JE, Pau CY, Forest V, et al. Anatomic measures of upper airway structures in obstructive sleep apnea. World J Otorhinolaryngol Head Neck Surg 2017;3:85–91. https://doi.org/10.1016/j.wjorl.2017.05.002.
[31] Behlfelt K, Linder-Aronson S, McWilliam J, et al. Cranio-facial morphology in children with and without enlarged tonsils. Eur J Orthod 1990;12:233–43. https://doi.org/10.1093/ejo012.3.233.
[32] Harvold EP, Tomer BS, Vargervik K, et al. Primate experiments on oral respiration. Am J Orthod 1981;79:359–72. https://doi.org/10.1016/0002-9416(81)90379-1.
[33] Zettergren-Wijk L, Forsberg CM, Linder-Aronson S. Changes in dentofacial morphology after adeno-/tonsillectomy in young children with obstructive sleep apnoea–a 5-year follow-up study. Eur J Orthod 2006;28:319–26. https://doi.org/10.1093/ejo/cji119.
[34] Agren K, Nordlander B, Linder-Aronsson S, et al. Children with nocturnal upper airway obstruction: postoperative orthodontic and respiratory improvement. Acta Otolaryngol 1998;118:581–7. https://doi.org/10.1080/0001648985014766.
[35] Hultcrantz E, Larson M, Hellquist R, et al. The influence of tonsillar obstruction and tonsillectomy on facial growth and dental arch morphology. Int J Pediatr Otorhinolaryngol 1991;22:125–34. https://doi.org/10.10160/0165-5876(91)90032-7.
[36] Isono S, Shimada A, Utsugi M, et al. Comparison of static mechanical properties of the passive pharynx between normal children and children with sleep-disordered breathing. Am J Respir Crit Care Med 1998;157:1204–12. https://doi.org/10.1164/ajrccm,157.4.9702042.
[37] Donnelly LF, Surdulescu V, Chini BA, et al. Upper airway motion depicted at cine MR imaging performed during sleep: Comparison between young patients with and those without obstructive sleep apnea. Radiology 2003;227:239–45. https://doi.org/10.1148/radiol.2271020198.
[38] Marcus CL, Katz ES, Lutz J, et al. Upper airway dynamic responses in children with the obstructive sleep apnea syndrome. Pediatr Res 2005;57:99–107. https://doi.org/10.1203/01.PDR.0000147565.74947.14.

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