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Airway changes after fixed functional appliance treatment in children with and without morphological deviations of the upper spine: A 3-dimensional CBCT study.

Yousef Abdalla[a,b], Stavros Kiliaridis[c,d], Liselotte Sonnesen[a]

[a]Section of Orthodontics, Department of Odontology, Faculty of Health and Medical Sciences, University of Copenhagen, 20 Nørre Alle, DK-2200 Copenhagen N, Denmark. alson@sund.ku.dk

[b]College of Medicine and Dentistry, James Cook University, McGregor Rd, Cairns, Australia.

dr.yousef.abdalla@gmail.com

[c]Department of Orthodontics, School of Dental Medicine, University of Geneva, Switzerland.

[d]Department of Orthodontics and Dentofacial Orthopedics, University of Bern, Switzerland.

stavros.kiliaridis@unige.ch

Address for correspondence:

Liselotte Sonnesen

Professor and Director of Postgraduate Program

Section of Orthodontics, Department of Odontology

Faculty of Health and Medical Sciences, University of Copenhagen

20 Nørre Alle, DK-2200 Copenhagen N, Denmark

0045 35 32 66 70, alson@sund.ku.dk

ABSTRACT

Objectives: Functional appliances (FA) have a positive effect on the upper-airway volume and minimal cross-sectional area (MCA) in children. An association between morphological deviations of the upper spine (MDUS) and reduced treatment response was found in similar appliances used to treat adults with obstructive sleep apnoea. The aims of this study were to compare airway changes after FA treatment in children with and without MDUS and in controls and identify if MDUS cause a smaller upper airway in children.

Methods: Pre- and post-treatment CBCT scans were included from 21 children with MDUS and 42 without MDUS treated with a fixed FA, along with a matched control group (matched for chronological age, skeletal age, gender and mandibular inclination) who received orthodontic treatment for minor malocclusions without an FA. The influence of MDUS on changes in upper-airway volume and MCA were evaluated three-dimensionally on CBCTs using standardized, previously validated methods and mixed-effects linear regression.

Results: There was a significantly increased volume and MCA in the FA groups with and without MDUS compared to controls (p=0.003 – p=0.049) and in the FA group without MDUS compared with the MDUS group (p=0.008-p=0.011) after treatment. There was no significant pre-treatment difference in airway dimensions between the MDUS and non-MDUS FA groups.

Conclusion: The airway response with fixed FA is significantly reduced in MDUS children. MDUS caused no significant pre-treatment airway differences in children however, MDUS may be important in predicting airway changes in FA treatment.

Introduction

Morphological deviations of the upper spine (MDUS) can be categorized as fusion anomalies and posterior arch deficiencies.[1] MDUS appear in approximately 14% of healthy subjects[2], however they are significantly more prevalent in adult patients with severe skeletal malocclusion and disturbances in craniofacial morphology,[3–7] as well as obstructive sleep apnoea (OSA).[2] Furthermore, MDUS have been associated with head and neck posture in adults, with an increased curvature and greater inclination of the upper spine in individuals with MDUS compared with controls.[8] MDUS is also positively correlated with the cranial base angle.[8]

MDUS in class II children have been associated with an increased sagittal jaw relationship, retrognathia of the jaws and an increased vertical jaw relationship[9] and, in class III children, a greater dysplasia in the sagittal jaw relationship.[10] Additionally, variations in the dimension of the atlas bone have been correlated with dentofacial anomalies.[11]

Few studies have investigated the relationship between MDUS and the airway in three dimensions.[12] Although it has been shown that MDUS can be accurately detected with lateral cephalometric radiographs,[13] the upper airway is a complex three-dimensional structure. Consequently, its volume and degree of constriction can only be measured with three-dimensional imaging, such as cone beam computed tomography (CBCT).[14]

One CBCT study showed no association between MDUS the upper airway dimensions in adults with OSA.[15] In contrast, another study found that the symptoms of adults with OSA responded less favourably to treatment with mandibular advancement devices if they also have MDUS.[16] However, the sample size was relatively small and the MDUS were assessed with 2-dimensional radiographs. Recently, it has been reported that class II children with MDUS responded poorer to removable functional appliances (FA) treatment in the short and long term, compared to class II children without MDUS.[17]

An increased sagittal jaw relationship is a risk factor for small upper airways in class II children[18] and correcting the sagittal jaw relationship with FA treatment increases the upper airway volume and minimal cross-sectional area (MCA).[19,20,21] Additionally, class II malocclusions and mandibular retrognathia are associated with MDUS,[4,9] and MDUS have a negative effect on skeletal changes achieved with FA treatment in class II children.[17] Consequently, it is plausible that MDUS may also influence the upper airway changes achieved during FA treatment. The aims of this study therefore were to: 1) compare airway changes after fixed functional appliance (FFA) treatment in children with and without MDUS and in controls and 2) identify if MDUS cause a smaller upper airway in untreated children. The null hypotheses were that MDUS had no effect on the upper airway changes in children treated with FFA; and that MDUS were not associated with pre-treatment airway volume and MCA differences in children.

Methods

The present study was a retrospective study involving three groups: 1) a skeletal class II group with MDUS treated with FFA (Hanks Telescoping Herbst, American Orthodontics, Sheboygan, WI), followed by fixed appliances; 2) a skeletal class II group without MDUS treated with fixed FFA, followed by fixed appliances; 3) a matched skeletal class I control group without MDUS treated with fixed appliances only (no FFA) for minor class I malocclusions.

Pre- (T_0) and post-treatment (T_1) CBCT scans were obtained from a database of patients who received orthodontic treatment between 2006 and 2012. The scans were collected from a private practice in Australia, where guidelines do not prohibit routine use of CBCT in orthodontics. Based on observed airway changes in a previous study,[21] a power calculation determined that 18 subjects would be needed in each group to achieve a power of 80% ($\alpha = 0.05$). Once the inclusion and exclusion criteria had been applied (Table I), the final sample consisted of 21 patients in the FA with MDUS group and 42 patients in the FA without MDUS group.

A matched control group of 63 patients without MDUS with a class I sagittal jaw (ANB 0-5 degrees according to Bjork[22,23]) and molar relationship were selected to match the FA groups for chronological age (mean 12y, 0m (±1y,6m)), skeletal age (utilizing the Cervical Vertebral Maturation index according to Baccetti[24]), gender (M=31; F=32), mandibular inclination (within +/- 2^{o} (norm 28^{o}±6[22,23])) and time interval between the pre-treatment and progress scans (mean 1y,11m). The study was approved by the Danish Data Protection Board (ref: SUND-2015-57-0121).

Scan protocol

All images were acquired using an iCAT Next Generation CBCT machine (Imaging Sciences International, Hatfield, Pa). The following parameters were used: 120Kv, 5mA, 0.4mm voxel resolution, 8.9 seconds scan time 13cm X 16cm - 16cm X 22cm field of view. Patients were instructed to bite in maximum intercuspation during the scan. Scans were evaluated to ensure that the condyles were seated in the glenoid fossa and were excluded from the sample if the mandible was postured.

Airway assessment

Digital Imaging and Communications in Medicine (DICOM) data were analyzed under the same lighting conditions and by the same blinded investigator using a validated protocol, which has been described in a previous paper.[25] The airway margins are shown in Table II and Figure 1.

The appropriate threshold value was automatically determined by the software (Dolphin Imaging Software, version 11.5; Dolphin Imaging and Management Solutions, Chatsworth, Calif.) and manually adjusted for each dataset, if required, to ensure the correct threshold value (operator-adjusted threshold). The airway volume and MCA was then automatically calculated based on the established operator-adjusted threshold.

Upper spine morphology assessment

MDUS are considered to be morphological deviations defined by Sandham.[1] Fusion anomalies include fusion of one cervical vertebra with another at the vertebral bodies, articulation facet, neural arch or transverse processes and also occipitalization, which is defined as assimilation of the atlas with the occipital bone. Block fusion has been subsequently defined by Sonnesen and Kjær[3] as fusion of more than two units at the vertebral bodies, articulation facets, neural arch or transverse processes. Posterior arch deficiency includes partial clefts, which involves failure to fuse of the posterior part of the neural arch, as well as dehiscence, which is a failure of part of the vertebral unit to develop.[1] (Table III, Figure 2).

Two authors (YA and LS) independently assessed pre-treatment CBCT scans for MDUS in all three planes (Dolphin Imaging and Management Solutions, Chatsworth, Calif.) in a blinded fashion (Figure 2). If a morphological deviation was found, this was then confirmed on the post treatment scan using the same methodology. If there was any doubt as to the presence of MDUS, the spine was recorded as having normal morphology. Only patients who had MDUS confirmed on both scans were included in the MDUS group. Inter-observer agreement on the visual assessment of MDUS in CBCT scans using this methodology has previously been reported as very good (k=0.92) .[12]

Statistical analysis

The intra- and inter-group changes in the airway measurements between T_0 and T_1 were evaluated separately using a linear mixed effects model, which allowed for the longitudinal structure of the data. The fixed effects part of the models included the dependent variables volume and MCA and independent variables group and time as well as their interaction. The same model was used to determine pre-treatment airway differences between the FA with and without MDUS groups. Preliminary analyses for the mixed effects model and linear regressions were performed to ensure there was no violation of the assumption of normality, linearity and multicollinearity. Statistical analysis was carried out using Stata version 15 (StataCorp LLC, College Station, TX), and the level of significance was $p<0.05$.

Results

Of the 21 patients in the FFA with MDUS group, 9 (43%) had fusion anomalies, 10 (48%) had posterior arch deficiency and 2 (9%) had multiple deviations of the upper spine (Table III).

The results of the mixed effects regression model are shown in Table IV and Figure 3. The increase in airway volume and MCA in the control group over time was 1583mm^3 (13%) and 18.3mm^2 (14%) respectively; for the FFA with MDUS group the increase was 4695.9mm^3 (39%) and 47.4mm^2 (37%) respectively; and for the FFA without MDUS 7759.1mm^3 (65%) and 87.5mm^2 (68%) respectively.

When compared with controls, the FFA effect on volume and MCA in both FFA groups was statistically significant, and there was also greater change in airway of the FFA group without MDUS when compared to the FFA group with MDUS which was also statistically significant (FFA with MDUS: +3112.3mm^3 (Volume); +29.1mm^2 (MCA); p=0.003, p=0.049, respectively. Additional effect of FFA without MDUS compared to FFA with MDUS: +3063.2mm^3 (Volume); +40.1mm^2 (MCA); p=0.008, p=0.01, respectively; Table IV).

The pre-treatment airway volume and MCA were 361.0mm^3 and 2.4mm^2 lower, respectively, in the FFA with MDUS group when compared to the FFA without MDUS group, however these differences were not statistically significant (p=0.77, (CI -2037.0 – 2761.4); p=0.89, (CI -30.5 – 35.3), respectively; Figure 3).

Discussion

The present study analyzed the influence that MDUS have on airway volume and MCA changes in children treated with a FFA and compared these airway changes to those of a non- FFA control group. Additionally, the association between MDUS and a small upper airway in children was investigated. To the authors' knowledge, this has not previously been investigated.

The prevalence and pattern of MDUS in the present study varied in comparison to what has previously been reported on skeletal class II patients.[4,10] Although the prevalence of fusion anomalies is comparable, the present study showed a higher prevalence of posterior arch deficiency. One possible explanation is that previous studies have used lateral cephalometric radiographs, whereas the present study used CBCT. Consequently, posterior arch deficiency may be easier to diagnose using three-dimensional imaging. A previous study comparing the accuracy of diagnosing MDUS from CBCT and lateral cephalograms in patients with OSA found a greater number of deviations detected using CBCT, however the differences were not statistically significant.[12]

When the FFA group is subdivided into children with and without MDUS, both groups continue to show that the FFA has a significantly positive effect on the upper airway when compared to the non- FFA control group. However, the present study shows that, when treated with FFA, the increase in the upper airway volume and MCA of children without MDUS was significantly greater than in children with MDUS.

Although MDUS has been associated with several malocclusion traits,[3,4,9] only one previous study[17] investigating the effects of MDUS on orthodontic treatment could be found. Children with MDUS responded less favorably to treatment with a removable FA compared to children without MDUS, which is consistent with the findings of the present study.

No other studies looking at the influence of MDUS on the airway changes in children receiving orthodontic treatment were found. One study on adults[16] found that patients with OSA and MDUS may respond less favorably to treatment with mandibular advancement devices when compared to adults without MDUS, which is somewhat consistent with the findings of the present study. MDUS could therefore be important for predicting how the upper airway responds in children treated with FA.

The mechanism by which MDUS influences the airway response to FA treatment is still unclear but we propose a hypothesis that may be related to the early embryogenesis. The notochord runs the full length of the spine to sella turcica,[26] meaning that the upper cervical spine and posterior cranial base are derived from the same embryonic origin. Disturbances in the development of the upper spine could therefore also indicate morphological disturbances in the cranial base and the cranio-cervical angulation.[14,27] The jaws are attached to the posterior part of the cranial base, and the cranio-cervical angle has been linked with the airway changes seen in OSA,[2] meaning that disturbances in the development of the upper spine originating from the notochord may be directly or indirectly linked to jaws and airway development. A hypothesis is therefore proposed that disturbances in the early embryogenesis of the spine and posterior cranial base cause a disturbance in jaw and airway development which in turn leads to a reduced effect of FA on the airway.

Although the pre-treatment airway dimensions in the MDUS group were smaller than the non-MDUS group, the differences were not statistically significant. No comparable studies could be found on children, however the findings are consistent with a study on adults with OSA, which found no significant pre-treatment differences in airway dimensions in a group with MDUS compared with a group without MDUS.[15]

Conclusion

A fixed functional appliance results in an increase in the upper airway volume and minimal cross-sectional area compared with controls, however the response is significantly reduced in children with morphological deviations in the upper spine (MDUS). Although there were no significant pre-treatment differences in the airway volume and MCA in children with and without MDUS, MDUS may be an important marker to predict airway changes following functional appliance treatment in children.

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Figures

Figure 1. Margins for delineation of the upper airway. ***Superior***: line passing from the palatal plane (ANS – PNS) extending to the posterior wall of the pharynx; ***Inferior:*** line passing from the anterosuperior edge of the 4th cervical vertebra (C4) to menton (Me); ***Anterior*:** line passing from soft palate to menton (Me); ***Posterior*:** posterior wall of pharynx; ***Lateral**:* respective pharyngeal walls.[25]

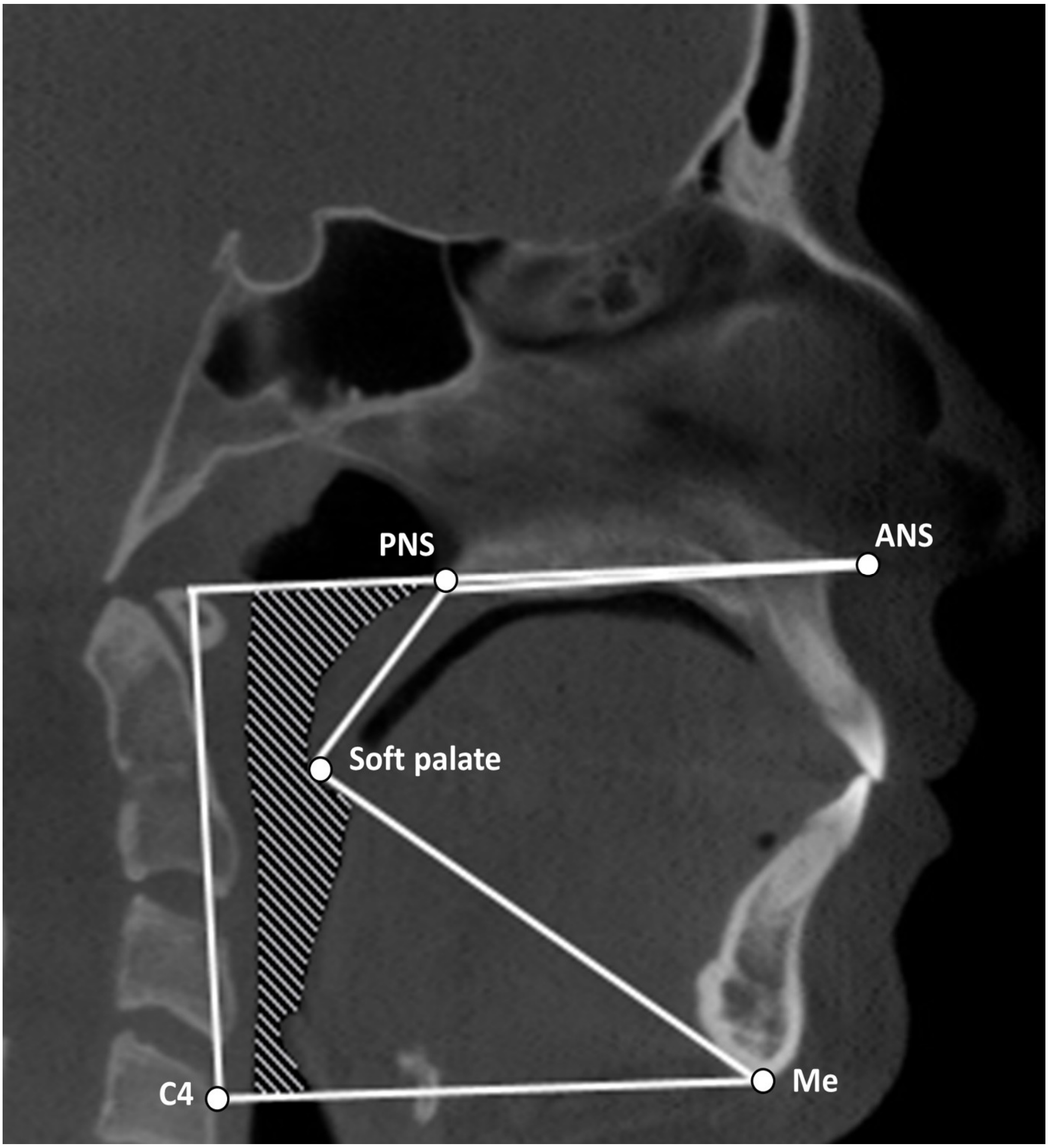


Figure 2. Examples of morphological deviations of the upper spine indicated by arrows; **A)** partial cleft on atlas (C1) in the sagittal, coronal and axial views; **B)** fusion of C2 and C3 in the sagittal, axial and coronal views.

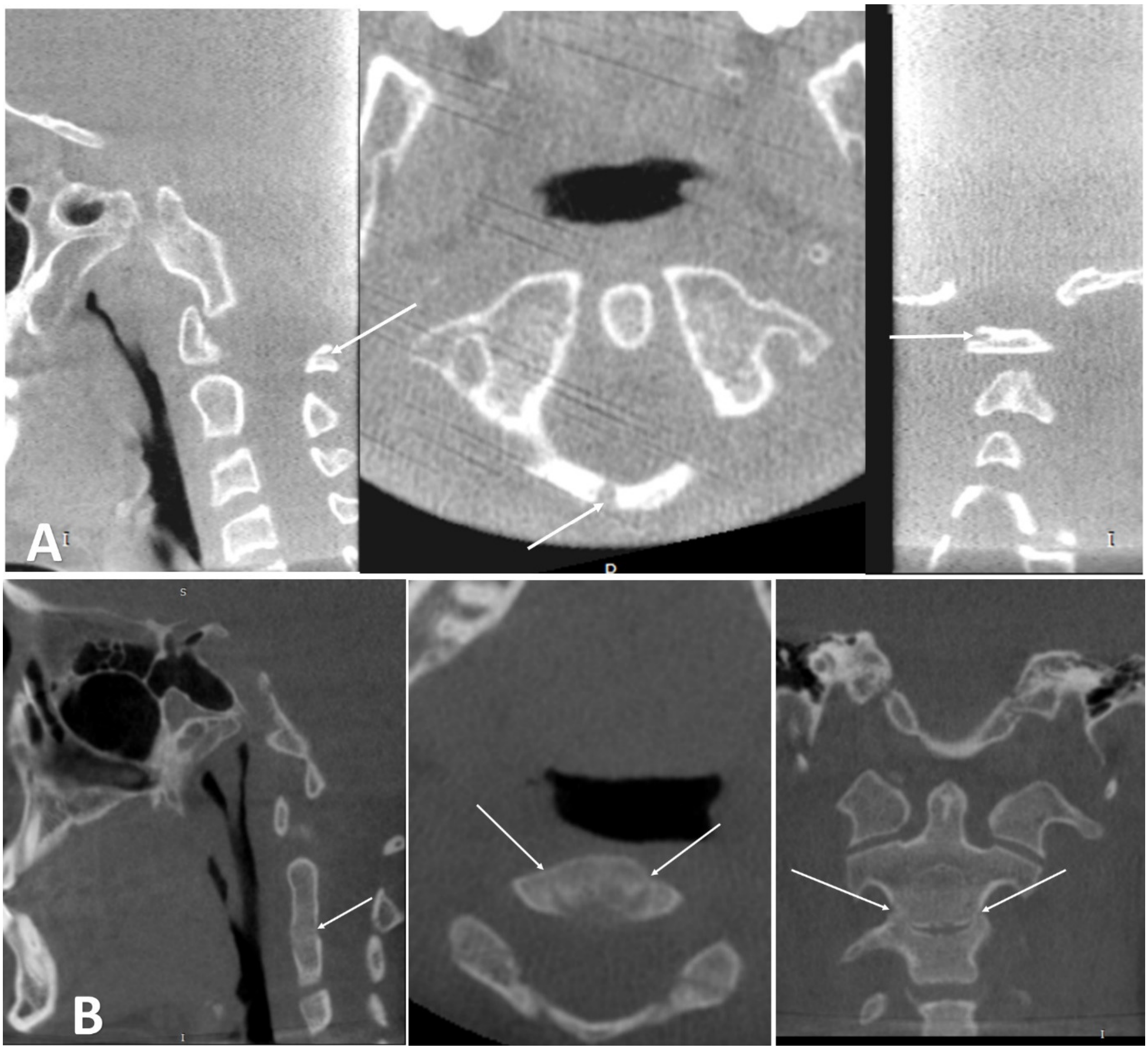
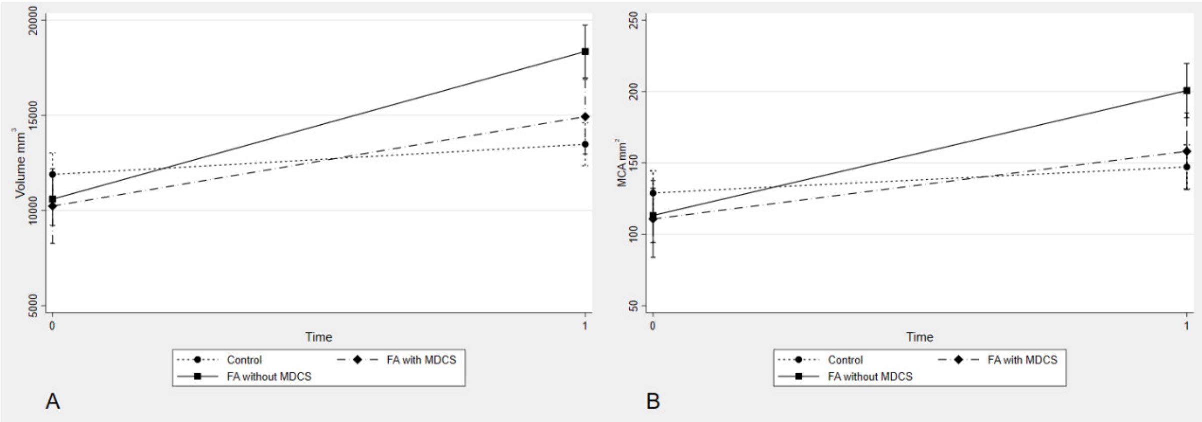


Figure 3. Change in **(A)** airway volume and **(B)** MCA in the three groups: FA without MDUS, FA with MDUS and control.



Tables

Table 1. Characteristics of morphological deviations of the upper spine (MDUS) in functional appliance (FA) treatment group with MDUS.

	n	***%***
Fusion anomalies	**9**	**43**
Fusion	4	19
Block fusion	1	5
Occipitalization	4	19
Posterior arch deficiency	**10**	**48**
Partial cleft	9	43
Dehiscence	1	5
Multiple deficiencies	**2**	**9**

Table II. Linear mixed effect modeling analysis for estimating the difference in airway volume and MCA between FA with MDUS, FA without MDUS and control groups over time (+/- denotes effect).

	Regression Coefficient	SE	*p* value	(95% CI)
Volume (mm^3)				
Intercept	11895.78	577.2	<0.001	(10764.5 – 1327.0)
Control group change T_0-T_1	+1583.6	514.9	0.002	(574.5 – 2592.7)
Additional effect of FA with MDUS compared to control group	+3112.3	1029.7	0.003	(489.0 – 2458.0)
Additional effect FA without MDUS compared to FA with MDUS group	+3063.2	1150.9	0.008	(807.5 – 5319.0)
MCA (mm^2)				
Intercept	128.9	7.9	<0.001	(113.3 – 144.4)
Control group change T_0-T_1	+18.3	7.3	0.013	(3.8 – 32.8)
Additional effect of FA with MDUS compared to control group	+29.1	14.7	0.049	(0.1 – 58.0)
Additional effect FA without MDUS compared to FA with MDUS group	+40.1	15.7	0.011	(9.4 – 70.8)