Relationship between growth of facial morphology and chronologic age in preschool children with obstructive sleep apnea

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ABSTRACT

Background: The purpose of this study was to evaluate the relationship between facial morphology using cephalometry and chronologic age in preschool children with obstructive sleep apnea (OSA).

Materials and Methods: From a group of lateral cephalometric radiographs taken of 35 children with OSA for diagnostic purposes, 15 were selected for the present investigation based on head position. The subjects consisted of preschool children with both OSA and primary dentition, all of them with a lowest documented SpO₂ <90% and a lowest 0 < AI. The control group included 15 preschool children with primary dentition and without enlarged tonsils or adenoids.

Results: The present findings reveal that, compared to the controls, OSA children had a narrower pharyngeal airway space related to OSA. In addition, we found that, although there were direct correlations between heights, nasal floor and corpus length, with chronologic age in the controls, there was no significant correlation between height, nasal floor length or corpus length with age in the OSA children.

Conclusion: Obstructive sleep apnea in children may be associated with growth disturbances of the nasal floor and corpus length.

Keywords: Children, growth, OSA, pharynx, weight.

INTRODUCTION

For some children, adenoid and tonsillar hypertrophy can cause an obstruction of the pharynx, particularly during sleep, which can result in breathing problems, sleep disturbances, and speech and swallowing difficulty.^{1–3} The facial patterns and dentition characteristics⁴ may contribute to the condition. Therefore, some individuals with genetically long-face syndrome and craniofacial growth disturbance may be more sensitive to the influence of environmental factors, such as enlarged tonsils, for example.⁵ In children, obstructive sleep apnea (OSA) is associated with mouth breathing and growth disturbances.^{1,2,6,7} Children with OSA might have space problems in the oropharynx, thus forcing the tongue to protrude forward, resulting in bite opening.^{5,7,8} However, children who breathe through their mouth are thought to suffer from positional changes, such as those in

the lower position of the mandible, the raised position of the head and hyoid bone, and the anterior inferior position of the tongue.^{7,9,10} It has been shown that a decrease in the size of the part of the maxilla or mandible occurs with growth in humans in the absence of nasal respiration or positional changes.^{9,10} Therefore, the purpose of this study was to evaluate the relationship between facial morphology and chronologic age in preschool children with OSA.

MATERIALS AND METHODS

Subjects

The test subjects included 15 preschool children with OSA and 15 controls, who visited a pedodontic and/or orthodontic

private clinic in Tokyo between September 2001 and March 2004. The OSA children were diagnosed by a pediatric otorhinolaryngologist. After introducing the medical doctors, informed consent was obtained from their parents at the time of study recruitment. The ethical committee at Meikai University approved this prospective study (A0818). The 15 OSA children with the lowest $\text{SpO}_2 < 90\%$ (mean $\pm$ SD: $86.1 \pm 1.8\%$) and the lowest $0 < \text{AI}$ (mean $\pm$ SD: 2.3 ± 1.5) were monitored at home during nocturnal sleep for three nights by their parents who were instructed in the use of a portable polysomnography machine (Apnomonitor III, Chest MI Co., Japan).¹¹ The oxyhemoglobin saturation measured by pulse oximetry was recorded as the SpO_2 . The lowest value of SpO_2 was denoted lowest SpO_2 . Apnea was operationally defined as the cessation of air flow lasting at least 10 s. The apnea index, which is the mean number of apneic episodes per hour of sleep, was then calculated. The 15 controls had no syndromic or non syndromic craniofacial abnormalities, neurologic diseases, or obesity. In addition, the controls were without enlarged tonsils and had no history of respiratory disorders, such as mouth breathing, apnea, or snoring. Although, controls had no skeletal discrepancies, they had malocclusion, such as an anterior cross bite or open bite. Children with OSA and controls were provided orthodontic treatment after their orthodontic diagnosis.

Cephalometry

Height and weight of the 15 OSA children and 15 controls were measured. Cephalometric radiographs were taken with all the subjects sitting upright in a chair. All the subjects were instructed to slowly breathe in through their nose without swallowing. To meet the requirement of having the Frankfort horizontal (FH) parallel to the floor, the subjects in whom the FH deviated by more than degrees from the bottom edge of the radiograph were excluded.²¹ In our previous study method, we observed no significant difference in the head position in relation to pharyngeal airway space.¹² As a result, all the 15 children with OSA (7 girls, 8 boys; 4.8 ± 0.8 years) and 15 controls (7 girls, 8 boys; 4.9 ± 0.8 years) were included. All cephalometric landmarks (Figure 1) were identified and digitized.

Measurements of pharyngeal airway space¹³

- ① D-AD1 (mm): The shortest distance from the PNS to the adenoid tissue measured along the line PNS–basion line (BA).
- ② D-AD2 (mm): The shortest distance from the PNS to the adenoid tissue measured along a line through the PNS perpendicular to the S–BA line.

- ③ D-PTV (mm): The shortest distance from a point on the pterygoid vertical (PTV) 5 mm above the PNS to the adenoid tissue.
- ④ Upper pharynx (mm): The nearest point on the adenoid soft tissue to the upper surface of the palatine velum.
- ⑤ Lower pharynx (mm): The linear shortest distance between the thyromental (TM) and the posterior pharyngeal wall.

Measurements of the facial skeleton¹³

- ⑥ Nasal floor length (mm): The distance between the anterior nasal spine (ANS) and posterior nasal spine (PNS).
- ⑦ Corpus length (mm): The distance between the XI and protuberance menti (PM).

Measurement of head posture¹²

Headposture (degrees): The angle between FH and the bottom edge of the radiograph.

Statistical analysis

The calculations were performed using a computerized cephalometric system, Win Ceph (Rise Co. Sendai, Miyagi prefecture, Japan). All measurements were repeated at an interval of at least 3 weeks, and the error of method was calculated using the formula; $S_i = \sqrt{\sum d^2 / 2n}$, where S_i is the error of method, d is the difference between the first and second measurements, and n is the number of double determinations. The greatest error of the method was found to be 0.75 (D-PTV) for a linear variable. The error variance of all variables was less than the biological variances. The differences in each variable were determined by performing a Mann–Whitney U -test. Before comparing between groups in regards to the pharyngeal airway space, the relationship between the head position and the pharyngeal airway space was investigated using Pearson's product moment correlation coefficient.

RESULTS

The preschool children with OSA had a lighter weight ($P < 0.05$) and a smaller body mass index (BMI) compared to controls ($P < 0.05$) (Table 1). The preschool children with OSA showed a narrower pharyngeal airway space (D-AD1; $P < 0.01$, D-PTV; $P < 0.01$ and upper pharynx; $P < 0.01$) and a more anterior tongue base position (lower pharynx; $P < 0.05$) (Table 2). There was no significant difference in the head position in relation to the pharyngeal airway space between the two groups (Table 3). A direct correlation was found between D-PTV ($r = 0.518$) and age in the controls;

Figure 1 Reference points and lines used in the cephalometric analyses.

ANS=the most anterior point of the bony nasal floor; BA=the most inferior posterior point of the occipital bone at the anterior margin of the occipital foramen; OR=the most inferior point on the lower border of the bony orbit; PM=the point where the curvature of the anterior border of the symphysis changes from concave to convex; PNS=the most posterior point of the bony hard palate; PO=the most superiorly positioned point of the bony external auditory canal; PT=the intersection point of the inferior border of the foramen rotundum with the posterior wall of the pterygomaxillary fissure; PTV=a line perpendicular to FH through the PT; S=the central point of the pituitary fossa (sella) of the sphenoid bone; XI=the point located at the geographic center of the ramus; the location of XI is placed geometrically in relation to FH and PTV using the following steps: 1) construct lines perpendicular to FH and PTV; 2) these constructed lines are tangents to four points (R1, R2, R3, and R4) on the borders of the ramus; 3) the constructed lines form a rectangle enclosing the ramus; 4) XI is located in the center of the rectangle at the intersection of the diagonals.

however, there was no significant correlation between pharyngeal airway space and age in the children with OSA (Table 4). Although, there were direct correlations between height ($r=0.682$), nasal floor length ($r=0.580$), and corpus length ($r=0.737$) with age in the controls, such correlation was not seen in the OSA children (Table 5).

Table 1 Body profiles of obstructive sleep apnea children and controls

	OSA children	Controls	P value
Height (m)	1.078 (0.105) ^a	1.081 (0.056)	0.5068
Weight (kg)	17.4 (4.048)	18.653 (1.686)	0.0152 ^{*b}
BMI ^c	14.790 (1.125)	15.993 (1.303)	0.0136 [*]

^aMean (SD), ^b* $P<0.05$, ^cBody mass index= kg/m^2 . BMI=body mass index; OSA=obstructive sleep apnea.

Table 2 Comparison of features of pharyngeal airway space, nasal floor, corpus length, and head posture between obstructive sleep apnea children and controls

	OSA children	Controls	P value
D-AD1 (mm)	13.52 (3.68) ^a	17.29 (3.42)	0.0084 ^{**b}
D-AD2 (mm)	10.65 (3.70)	12.03 (2.24)	0.0710
D-PTV (mm)	4.62 (4.22)	7.96 (2.29)	0.0023 ^{**}
Upper pharynx (mm)	3.22 (2.82)	6.95 (1.62)	0.0008 ^{**}
Lower pharynx (mm)	14.42 (3.76)	11.70 (1.93)	0.0152 ^{*c}
Nasal floor length (mm)	43.95 (3.37)	45.46 (2.80)	0.1403
Corpus length (mm)	54.51 (2.90)	55.97 (2.90)	0.2536
Head posture (deg.)	-2.13 (5.92)	1.23 (3.68)	0.1708

^aMean (SD), ^b*** $P<0.01$, ^c* $P<0.05$. PTV=pterygoid vertical; OSA=obstructive sleep apnea.

Table 3 Correlations between variables for pharyngeal airway space and degree of head posture within each group

	OSA children	Controls
D-AD1 (mm)	-0.155 (0.5889) ^a	-0.477 (0.0719)
D-AD2 (mm)	-0.134 (0.6395)	-0.274 (0.3297)
D-PTV (mm)	-0.091 (0.7527)	-0.355 (0.1982)
Upper pharynx (mm)	0.010 (0.9730)	-0.245 (0.3870)
Lower pharynx (mm)	-0.476 (0.0731)	-0.249 (0.3781)

^a r value (P value). PTV=pterygoid vertical; OSA=obstructive sleep apnea.

DISCUSSION

The OSA children and controls in this study were closely matched with respect to gender, chronological age and primary dentition. It is impractical to obtain multiple cephalographic records, for inhaling, not swallowing, not crying, particularly in a standardized head position.

There were no significant differences in head position in relation to the pharyngeal airway space or hyoid bone

Table 4 Correlation between variables for pharyngeal airway space and chronologic age within each group

	OSA children	Controls
D-AD1 (mm)	-0.242 (0.3921) ^a	-0.160 (0.5759)
D-AD2 (mm)	-0.102 (0.7224)	-0.434 (0.1078)
D-PTV (mm)	-0.052 (0.8568)	-0.518 (0.0472) ^{*b}
Upper pharynx (mm)	-0.099 (0.7309)	-0.325 (0.2431)
Lower pharynx (mm)	-0.170 (0.5513)	0.354 (0.2004)

^a*r* value (*P* value), ^b**P*<0.05. PTV=pterygoid vertical; OSA=obstructive sleep apnea.

Table 5 Correlation between variables of body, nasal floor, and corpus length with chronologic age within each group

	OSA children	Controls
Height (m)	0.411 (0.1300) ^a	0.682 (0.0039) ^{**b}
Weight (kg)	0.208 (0.4650)	0.412 (0.1265)
BMI	-0.409 (0.1320)	-0.391 (0.1522)
Nasal floor length (mm)	0.476 (0.0727)	0.580 (0.0219) ^{*c}
Corpus length (mm)	-0.409 (0.1320)	0.737 (0.0011) ^{**}

^a*r* value (*P* value), ^b***P*<0.01, ^c**P*<0.05. BMI=body mass index; OSA=obstructive sleep apnea.

position between the groups. Experimental studies demonstrated immediate head extension and changes in the postural electromyography (EMG) activity in the craniofacial muscles following obstruction of the nasal airway.¹⁴ The OSA patients exhibit an extended and forward natural head posture when compared with controls.^{15,16} This posture may be attributed to the compromised morphology and physiology of the upper airway and related structures observed in OSA patients that persist even when they are awake.^{16,17} Thus, from the method used in this study, we thought that the relationship between head posture and soft tissue or/and hyoid bone position should at least be evaluated in each research.

In this study, there was a direct correlation between pharyngeal airway space and chronologic age in the control group, but none in the OSA children. In general, lymphoid tissue develops rapidly after birth, reaches its maximum size in early childhood, begins to regress around 8–10 years of age and is usually completely atrophied by 12–14 years of age.¹⁸ In OSA children, excessive growth of the lymphoid tissue could lead to obstruction of the pharyngeal airway space, especially during sleep, which may cause breathing problems.^{1,2}

The cardinal features of children with OSA, excessive daytime sleepiness, poor school performance, abnormal behavior, enuresis, morning headache, abnormal weight, progressive development of hypertension, and loud snoring with apneic pauses have been described.^{1–3} In a review of

literature concerning children between the ages of 2–18 years, adenotonsillectomy was found curative in 75–100% of children with OSA, including those who were obese.¹⁹ However, more recent studies are not as enthusiastic. Of these cardinal features, abnormal weight is thought to be an important clinical finding in preschool children with OSA in regards to the relationship between growth disturbances and growth hormone secretion. Although, adolescents and adults secrete growth hormone during both awake and sleeping periods, prepubertal children secrete a majority of their growth hormone during sleep. OSA in its most severe form, is associated with growth retardation, and a reduction in growth hormone secretion.²⁰ The disruption of slow-wave sleep by frequent episodes of apnea has been thought to be the mechanism responsible for the suppression of growth hormone secretion.²¹ The treatment of OSA children results in a growth spurt as growth hormone secretion returns to normal and catch-up growth occurs.²⁰ Six-month postoperative follow-up study in a group of OSA children after adenotonsillar surgery showed a significant increase in height and weight.²² A lighter weight and a smaller BMI were noted in OSA children with hypertrophy of the tonsils and adenoid tissue in our study. In addition, they had a significantly larger value for the size of lower pharynx, i.e. the tongue base was in an anterior position. Although, direct correlations between height, nasal floor length, and corpus length with age were observed in controls, no significant correlation was observed in OSA children. This suggests that the OSA children had growth disturbances as a result of the decrease in the rate of growth hormone effects in the area of the facial skeleton, i.e. a small nasal floor length and a small corpus length, whereas controls had both normal body growth and normal facial growth. Particularly interesting were the correlations found between OSA, malocclusion, and maxillofacial malformations. In fact, many OSA patients show craniofacial abnormalities in both jaws as well as alteration in the skeletal structure of the respiratory dynamic space.²³ Micrognathia and nasal obstruction are among the risk factors for OSA. Also, chronic absence of active nasal respiration results in the reduced growth of the skull, thereby reducing skull size with body growth.²⁴ Our studies have earlier shown that, preschool children with OSA had a smaller nasal floor length (ANS–PNS length). Löfstrand-Tideström et al²⁵ too reported that the lower arch length is small in 4-year-old children with breathing obstruction. We here prove relationship between OSA and growth disturbances in the entire body and the facial skeleton in preschool children.

There is a limitation in this study. The study population was small. More cases need to be evaluated to confirm our results.

CONCLUSION

As a result of this study, it is important to note that although our evaluation based on mandibular shape or position does not necessarily distinguish between children with or without OSA, important differences can be found with regard to the pharynx and the children's weight. The growth disturbances related to height, weight, BMI, nasal floor length, and corpus length can be considered indicators of OSA children.

REFERENCES

1. Leach J, Olson J, Hermann J, Manning S. Polysomnographic and clinical findings in children with obstructive sleep apnea. *Arch Otolaryngol Head Neck Surg* 1992;118:741–4.
2. Kawashima S, Niikuni N, Lo C, Kohno M, Nakajima I, Akasaka M. Clinical findings in Japanese children with obstructive sleep apnea syndrome. Focus on dental findings. *J Oral Sci* 1999;41:99–103.
3. Wei JL, Bond J, Mayo MS, Smith HJ, Reese M, Weatherly RA. Improved behavior and sleep after adenotonsillectomy in children with sleep-disordered breathing. Long-term follow-up. *Arch Otolaryngol Head Neck Surg* 2009;135:642–6.
4. Hausund A, Bøe OE. Floating norms as guidance for the position of the lower incisors. *Angle Orthod* 1980;50:165–8.
5. Behlfelt K. Enlarged tonsils and the effect of tonsillectomy. Characteristics of the dentition and facial skeleton. Posture of the head, hyoid bone and tongue. Mode of breathing. *Swed Dent J Suppl* 1990;72:1–35.
6. Bonuck K, Parikh S, Bassila M. Growth failure and sleep disordered breathing: a review of the literature. *Int J Pediatr Otorhinolaryngol* 2006;70:769–78.
7. O'Ryan FS, Gallagher DM, LaBanc JP, Epker BN. The relation between nasorespiratory function and dentofacial morphology: a review. *Am J Orthod* 1982;82:403–10.
8. Kawashima S, Niikuni N, Lo C, Takahashi Y, Kohno M, Nakajima I, Akasaka M, Sakata H, Akashi S. Cephalometric comparisons of craniofacial and upper airway structure in young children with obstructive sleep apnea syndrome. *Ear Nose Throat J* 2000;79:499–506.
9. Linder-Aronson S. Adenoids, their effect on mode of breathing and nasal airflow and their relationship to characteristics of the facial skeleton and the dentition. *Acta Otolaryngol Suppl* 1970;265:5–132.
10. Subtelny JD. Oral respiration: facial mal-development and corrective dento-facial orthopedics. *Angle Orthod* 1980;50:147–64.
11. Kawashima S, Mikami Y, Khohara M, Sasaki Au, Otsuka Y, Mano M, Matsui S, Kanegae H. Reproducibility of portable polysomnography values in children with habitual snoring. *Orthod Waves* 2009;68:107–11.
12. Kawashima S, Peltomäki T, Laine J, Rönning O. Cephalometric evaluation of facial types in preschool children without sleep-related breathing disorder. *Int J Pediatr Otorhinolaryngol* 2002;63:119–27.
13. Ricketts RM, Roth RH, Chaconas SJ, Schulhof RJ, Engel GA. In: *Orthodontic Diagnosis and Planning: Their Roles in Preventive and Rehabilitative Dentistry*. Denver: Rocky Mountain/Orthodontics 1982.
14. Hellsing E, Forsberg CM, Linder-Aronson S, Sheikholeslam A. Changes in postural EMG activity in the neck and masticatory muscles following obstruction of the nasal airways. *Eur J Orthod* 1986;8:247–53.
15. Solow B, Ovesen J, Nielsen P, Wildschjødzt G, Tallgren A. Head posture in obstructive sleep apnea. *Eur J Orthod* 1993; 15:107–14.
16. Tangugsorn V, Skatvedt O, Krogstad O, Lyberg T. Obstructive sleep apnoea: a cephalometric study. Part I. Cervico-craniofacial skeletal morphology. *Eur J Orthod* 1995;17:45–56.
17. Özbek MM, Miyamoto K, Lowe AA, Fleetham JA. Natural head posture, upper airway morphology and obstructive sleep apnea severity in adults. *Eur J Orthod* 1998;20:133–43.
18. Songu M, Adibelli ZH, Tuncyurek O, Adibelli H. Age-specific size of the upper airway structure in children during development. *Ann Otol Rinol Laryngol* 2010;119:541–6.
19. Schechter MS. Section on pediatric pulmonology, subcommittee on obstructive sleep apnea syndrome technical report: diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics* 2002;109:69.
20. Goldstein SJ, Wu RHK, Thorpy MJ, Shprintzen RJ, Mario RE, Saenger P. Reversibility of deficient sleep entrained growth hormone secretion in a boy with achondroplasia and obstructive sleep apnea. *Acta Endocrinol (Copenh)* 1987;116:95–101.
21. Guillaume A, Benoit O, Gourmelen M, Richardet JM. Relationship between sleep stage IV deficit and reversible HGH deficiency in psychosocial dwarfism. *Pediatr Res* 1982;16: 299–303.
22. Stradling JR, Thomas G, Warley ARH, Williams P, Freeland A. Effect of adenotonsillectomy on nocturnal hypoxaemia, sleep disturbance and symptoms in snoring children. *Lancet* 1990; 335:249–53.
23. Pirelli P, Saponara M, Rosa C, Fanucci E. Orthodontic and obstructive sleep apnea in children. *Med Clin N Am* 2010;94: 517–29.
24. Schlenker WL, Jennings BD, Jeiroudi MT, Caruso JM. The effects of chronic absence of active nasal respiration on the growth of the skull: a pilot study. *Am J Orthod Dentofacial Orthop* 2000;117:706–13.
25. Löfstrand-Tideström B, Thilander B, Ahlqvist-Rastad J, Jakobsson O, Hultcrantz E. Breathing obstruction in relation to craniofacial and dental arch morphology in 4-year-old children. *Eur J Orthod* 1999;21:323–32.