Absence of facial type differences among preschool children with sleep-related breathing disorder

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The aim of this study was to find out whether there are specific facial types and a specific hyoid bone position in preschool children with sleep-related breathing disorder (SBD). A total of 69 children were divided into 4 groups based on the mandibular line/Frankfurt horizontal angle and apnea index. There were 19 children with hyperdivergent facial type and SBD and 19 children with neutral facial type and SBD, all of them with documented 0 < AI < 5. Ten children had hyperdivergent facial type and non-SBD, and 21 children neutral facial type and non-SBD. The present findings show that SBD may be associated with both hyperdivergent and neutral facial type. Furthermore, we could not find any specific hyoid bone position related to SBD, non-SBD, or to facial type. In conclusion, it is important to note that while evaluation based on facial type (mandibular shape or position) does not necessarily distinguish between children with SBD and children without SBD (non-SBD), important differences are found in the pharynx. Short nasal floor length, long soft palate, and particularly short upper pharyngeal width can be considered indicators of SBD. □ *Children; facial type; hyperdivergent face; neutral face; sleep-related breathing disorder*

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Generally, sleep-related breathing disorder (SBD) with obstructive sleep apnea has been found to occur in infants and children, and the relationship between craniofacial morphology and the disorder has been investigated with varying results (1–7). In children, SBD is commonly associated with hypertrophy of adenoids and tonsils, and anomalies that include morphological and/or functional narrowing of the airway or hypotonus of the pharyngeal airway musculature (6). In addition, children with SBD have a retrognathic and/or a micrognathic mandible, narrow pharyngeal airway space and inferior hyoid position (3, 4, 7–10)

The peak incidence of SBD in children is at approximately 4 years (11), by which age the craniofacial skeleton has attained 60% of adult size (3, 12). In healthy children, the area of the nasopharyngeal soft tissues increases from the age of 3 to 5 years (13). It has been suggested that specific morphologic features, such as a long facial type, might be present in certain preschool children, and that the facial type may even be modified by the early narrowed pharyngeal airway space (3, 11, 13). On the other hand, difference in facial type with SBD could be related to grade of the disorder, the pharyngeal airway space, or hyoid bone position.

The aim of this study was to evaluate whether there are hyperdivergent and neutral facial types and a specific hyoid bone position in preschool children with SBD.

Materials and methods

The subjects, whose characteristics are given in Table 1, consisted of preschool children who visited the Saitama Prefecture Children's Medical Center in Japan between April 1995 and March 2000. The children were informed by a pediatric otolaryngologist and a pediatric orthodontist that they would be admitted for a more detailed examination if they showed an apnea index (AI) in the range $0 < AI < 5$ on the polysomnograph (Apnomonitor II, Chest M.I.®). AI is the mean number of apneic episodes per hour of sleep, where the apneic episode is defined as the cessation of airflow through the nose and mouth for more than 10 s (2, 14). Children with sleep-related breathing disorder were monitored with a polysomnograph by their parents during nocturnal sleep for 3 nights. The AI value, calculated for 2 nights (excluding the 1st night) (7), ranged from $0.5 \leqslant AI \leqslant 4.8$ (mean 1.4 ± 0.9). A total of 38 preschool children (22 boys, 16 girls) constituted the SBD group ($0 < AI < 5$). The clinical manifestations of children with SBD included chronic mouth breathing and snoring during sleeping. In the control groups, there were 15 boys with non-SBD and 16 girls with non-SBD, with no enlarged tonsils, who did not snore and did not have a skeletal malocclusion, or any history of respiratory disorders, such as mouth breathing or sleep apnea. However, control groups had incisor crowding of maxilla and/or mandible. None of the 69 subjects was obese, and none had anatomic craniofacial

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Table 1. Comparison of facial characteristics of 69 children for division into four facial types: hyperdivergent facial type with sleep-related breathing disorder (SBD) or with non-SBD and neutral facial type with SBD or with non-SBD

	Hyperdivergent facial type		Neutral facial type	
	With SBD (*n* = 19)	With non-SBD (*n* = 10)	With SBD (*n* = 19)	With non-SBD (*n* = 21)
Mean age, yrs (*s*)	4.70 (1.51)	4.66 (0.71)	4.66 (1.45)	4.67 (0.88)
ML* (°)	31.1 < ML	31.1 < ML	23.3 ⩽ ML ⩽ 31.1	23.3 ⩽ ML ⩽ 31.1
Mean AI† (*s*)	1.5 (0.7)*	–‡	1.3 (1.1)*	–‡

* ML = mandibular line/Frankfort horizontal, angle (°).
† AI = Mean (*s*) of Apnea Index.
‡– = No examination.
§ There was no statistical significant difference in AI between hyperdivergent and neutral facial type.
s = Standard deviation.

abnormalities, neurologic, or cardiac disease. All children had a primary dentition and none had missing teeth, a crown decayed by dental caries, or a thumb-sucking habit.

During cephalometric registration, the children were sitting in a chair in an upright posture, keeping the dentition in natural occlusion; they were instructed to breath slowly through the nose (this was practiced) and not to swallow. The lateral cephalograms were taken by attempting to have the Frankfort horizontal (FH) parallel both to the floor and to the lower border of the radiograph. The radiographs of 69 children were selected from a larger sample, and were included if the head

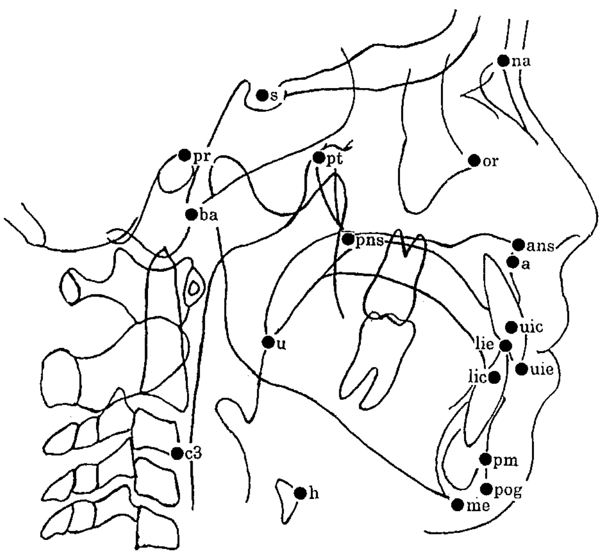

Fig. 1. Anatomical cephalometric landmarks: a, the deepest point on the concave outline of the upper labial alveolar process; ans, the most anterior point of the bony nasal floor; ba, the most inferior posterior point of the occipital bone at the anterior margin of the occipital foramen; c3, the most anteroinferior point on the corpus of the 3rd cervical vertebra; h, the most superior and anterior point on the body of the hyoid; lic, the cervical central point of the crown of the upper central incisor; lie, the incisal edge of the lower central incisor; me, the most inferior point of the symphysis; na, the most anterior point of the frontonasal suture; or, the most inferior point on the lower border of the bony orbit; pns, the most posterior point of the bony hard palate; pm, the point where the curvature of the anterior border of the symphysis changes from concave to convex; pog, the most prominent point of the chin; pr, the most superiorly positioned point of the bony external auditory canal; pt, the intersection point of the inferior border of the formen rotundum with the posterior wall of the pterygomaxillary fissure; s, the central point of the pituitary fossa (sella) of the sphenoid bone; u, the tip of the soft palate; uic, the cervical central point of the crown of the upper central incisor; uie, the incisal edge of the upper central incisor.

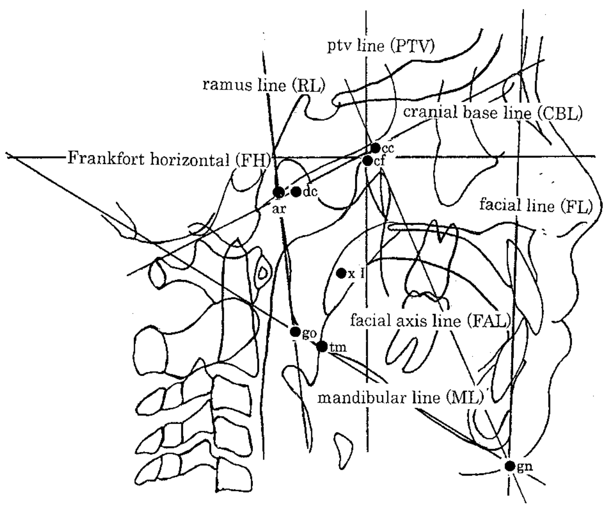

Fig. 2. Constructed cephalometric landmarks: ar, the intersection point between the contour of the external cranial base and the dorsal contour of the condylar process; cc, the intersection point between cranial base line (CBL, ba-na line) and facial axis line (FAL, pt-gn line); cf, the intersection point of the Frankfort horizontal (FH, or-pr) and ptv line (PTV, a line perpendicular to FH through pt); dc, the central point of the neck of the condyle on cranial base line (CBL, na-ba); gn, the intersection point of the mandibular line (ML, the lower border of the mandible) and the facial line (FL, the line connecting na and pog); go, the intersection point of the ML and the ramus line (RL, the tangent line on the posterior contour of the ramus ascendens through ar); tm, the intersection point between the surface of the tongue base and the lower contour of the mandible; xI, the point located at the geographic center of the ramus. Location of xI is placed geometrically to FH and PTV in the following steps: 1) By construction of lines perpendicular to FH and PTV; 2) these constructed lines are tangents to 4 points 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.

Table 2. Comparison of measurements of the craniofacial skeleton among hyperdivergent facial type with sleep-related breathing disorder (SBD) and with non-SBD, neutral facial type with SBD and non-SBD

	Hyperdivergent facial type		Neutral facial type		
	With SBD (*n* = 19) Mean (*s*)	With non-SBD (*n* = 10) Mean (*s*)	With SBD (*n* = 19) Mean (*s*)	With non-SBD (*n* = 21) Mean (*s*)	*P*
na-s-ba (°)	133.62 (4.34)	131.91 (4.32)	134.00 (5.26)	134.58 (3.78)	ns*
Facial depth(°)	81.97 (2.72)	83.90 (2.27)	84.90 (2.71)	84.00 (2.69)	0.040
Facial axis (°)	83.10 (4.92)	86.69 (1.62)	86.75 (2.99)	88.35 (3.24)	<0.001
Facial taper (°)	63.63 (4.42)	63.74 (1.22)	67.35 (3.27)	68.41 (3.38)	<0.001
Maxillary depth (°)	88.46 (2.85)	90.24 (2.83)	89.99 (3.01)	89.67 (3.00)	ns
Maxillary height (°)	57.70 (5.11)	56.02 (2.07)	56.17 (3.18)	54.32 (2.94)	ns
Palatal plane angle (°)	2.06 (2.01)	1.61 (3.24)	2.94 (2.55)	2.41 (2.33)	ns
Mandibular line angle (°)	34.73 (4.20)	32.71 (1.63)	27.74 (1.99)	27.67 (1.97)	<0.001
Mandibular arc (°)	26.35 (3.71)	28.50 (3.64)	29.02 (4.41)	29.04 (3.44)	ns
Posterior facial height (mm)	50.84 (4.01)	49.09 (3.11)	54.00 (4.83)	49.68 (3.86)	0.017
Gonial angle (°)	132.73 (4.48)	131.62 (3.30)	129.17 (4.04)	129.59 (3.85)	ns
Ramus position (°)	74.66 (3.29)	75.45 (1.66)	75.93 (3.73)	75.15 (3.51)	ns
ans-pns (mm)	41.99 (3.39)	44.86 (2.57)	43.68 (2.36)	45.11 (2.79)	0.019
Corpus length (mm)	55.55 (3.53)	56.62 (2.86)	56.26 (4.56)	55.32 (3.31)	ns
Facial convexity (mm)	5.94 (1.66)	5.40 (2.51)	5.11 (1.86)	4.76 (2.05)	ns
Lower facial height (°)	53.17 (4.01)	48.46 (2.23)	48.52 (3.18)	46.95 (2.76)	<0.001
Location of a (mm)	−0.94 (2.69)	0.63 (2.38)	1.16 (2.94)	−0.29 (2.54)	ns
Location of pogonion (mm)	−14.37 (5.41)	−10.46 (4.15)	−9.44 (5.09)	−9.95 (4.71)	0.046

* ns: no statistically significant difference.
s = Standard deviation.

position fell in the range of ±5 degrees between the bottom line of the radiograph and the FH (15). Another inclusion criterion was the facial type based on the angle between the mandibular line and FH (ML/FH), the hypodivergent facial type (ML/FH <23.3) excluded, whereas the neutral facial type (23.3 ⩽ ML/FH ⩽ 31.1) or the hyperdivergent facial type (31.1 < ML/FH) were included. By using these angles (range from 23.3 to 31.1, i.e. mean of 27.2 ± 1 *s* (standard deviation)), a standardized facial classification was devised from control values of children without ENT diseases (4). Thirty-eight children with SBD were divided into 2 groups: hyperdivergent facial type with SBD (*n* = 19) and neutral facial type with SBD (*n* = 19). Thirty-one children with non-SBD were divided into 2 groups: hyperdivergent facial type with non-SBD (*n* = 10) and neutral facial type with non-SBD (*n* = 21). All cephalograms were obtained before any oral, medical, or surgical intervention was undertaken.

Cephalometric analysis

The variables of the craniofacial skeleton, dentition, pharyngeal airway space, and hyoid bone position were measured by using cephalometric landmarks as illustrated in Figs 1 and 2. The magnification was the same for all radiographs and could consequently be disregarded.

Measurements of the craniofacial skeleton and dentition (4, 6, 15, 16): *na-s-ba:* the angle between na-s and s-ba; *facial depth:* the angle between the facial line (FL, n-pog) and the Frankfort horizontal (FH, or -po); *facial axis:* the angle between the facial axis line (FAL, pt-gn) and the cranial base line (CBL, ba-na); *facial taper:* the angle between the mandibular line (ML, go-me) and the FL; *maxillary depth:* the angle between the FH and na-a; *maxillary height:* the angle between na-cf and cf-a; *palatal plane angle:* the angle between the FH and ans-pns; *mandibular plane angle:* the angle between the FH and the ML; *mandibular arc:* the angle between the line xI-pm and

Table 3. Comparison of measurements of the dentition among hyperdivergent facial type with sleep-related breathing disorder (SBD) and with non-SBD, neutral facial type with SBD and non-SBD

	Hyperdivergent facial type		Neutral facial type		
	With SBD (*n* = 19) Mean (*s*)	With non-SBD (*n* = 10) Mean (*s*)	With SBD (*n* = 19) Mean (*s*)	With non-SBD (*n* = 21) Mean (*s*)	*P*
Incisor overjet (mm)	1.99 (1.85)	1.88 (1.28)	3.18 (2.08)	2.79 (1.47)	ns*
Incisor overbite (mm)	1.10 (1.11)	1.08 (0.96)	2.99 (3.23)	1.63 (1.39)	ns
Interincisal angle (°)	153.44 (8.75)	141.38 (12.59)	153.58 (13.09)	146.16 (10.48)	0.008
U1 to FH (°)	96.25 (6.49)	100.31 (5.26)	97.07 (8.79)	97.10 (4.77)	ns
L1 to ML (°)	76.36 (6.19)	83.45 (7.65)	80.33 (7.36)	86.74 (7.26)	0.001

* ns: no statistically significant difference.

Table 4. Comparison of measurements of pharyngeal airway space and hyoid bone position among hyperdivergent facial type with sleep-related breathing disorder (SBD) and with non-DBD, neutral facial type with SBD and non-SBD

	Hyperdivergent facial type		Neutral facial type		
	With SBD (*n* = 19) Mean (*s*)	With non-SBD (*n* = 10) Mean (*s*)	With SBD (*n* = 19) Mean (*s*)	With non-SBD (*n* = 21) Mean (*s*)	*P*
d-ad1 (mm)	13.53 (5.67)	15.59 (3.13)	11.95 (3.64)	16.55 (3.50)	0.006
d-ad2 (mm)	11.20 (4.73)	11.34 (2.45)	10.10 (3.45)	11.53 (2.01)	ns*
d-ptv (mm)	4.35 (3.18)	7.04 (1.90)	3.61 (2.83)	7.86 (2.74)	<0.001
Upper pharynx (mm)	2.67 (2.22)	6.54 (1.49)	2.34 (1.71)	6.25 (1.76)	<0.001
Lower pharynx (mm)	14.00 (3.66)	12.87 (2.10)	13.38 (3.39)	11.30 (2.48)	0.040
Soft palatal length (mm)	28.25 (2.40)	26.16 (2.35)	29.15 (2.68)	25.94 (1.65)	<0.001
me-h/c3-me (%)	51.33 (5.43)	52.18 (4.75)	48.06 (6.00)	51.26 (6.37)	ns
c3-h/c3-me (%)	49.56 (6.09)	49.58 (5.68)	53.90 (7.69)	49.60 (6.29)	ns

* ns: no statistically significant difference.

the line xI-dc; *posterior facial height:* the distance between go and cf; *gonial angle:* the angle between the ML and the ramus line (RL, ar-go); *ramus position:* the angle between the FH and cf-xI; *nasal floor length:* the distance between ans and pns; *corpus length:* the distance between xIand pm; *facial convexity:* the distance from a to FL; *lower facial height:* the angle between ans-xI and xI-pm; *location of a:* the distance between point a (a) and the line from the nasion perpendicular to the FH; *location of pogonion:* the distance between point pogonion (pog) and the line from the nasion perpendicular to the FH; *incisor overjet:* the shortest horizontal distance between the midpoint of the upper incisal edge (uie) and the midpoint of the lower incisal edge (lie); *incisor overbite:* the shortest vertical distance between uie and lie; *interincisal angle:* the angle between the lines uie-uic (the cervical central point of the upper central incisor) and lie-lic (the cervical central point of the lower central incisor); *U1 to FH:* upper incisor inclination to the FH = the angle between uie-uic and the FH; *L1 to ML:* lower incisor inclination to the ML = the angle between the lie-lic and the ML.

Measurements of the pharyngeal airway space (4, 13, 15, 17, 18) and the hyoid bone position (15, 19, 20): *d-ad1:* the shortest distance from pns to the adenoid tissue measured along the line pns-ba; *d-ad2:* the shortest distance from pns to the adenoid tissue measured along a line through pns perpendicular to s-ba; *d-ptv:* the shortest distance from a point on the pterygoid vertical line (ptv line) and 5 mm above the intersection point between the perpendicular ptv line through the pns and ptv line, to the adenoid tissue; *upper pharynx:* the shortest distance point on adenoid soft tissue to the upper surface of the palatine velum; *lower pharynx:* the shortest distance between tm and the posterior pharyngeal wall; *soft palatal length:* the distance between pns and u (tip of soft palate); *me-h:* the distance between me and h (most superior and anterior points on the body of the hyoid); *c3-h:* the distance between c3 (most anteroinferior point on the corpus of the third cervical vertebra) and h; *c3-me:* the distance between c3 and me.

The distances between the 3rd cervical vertebra, the menton, and the hyoid bone were indicated by the standardized c3-me distance (15, 20). The purpose of the standardized hyoid bone position is to examine the balance of the hyoid bone position: *me-h/c3-me (ratio):* the c3-me distance divided by the me-h distance; *c3-h/c3-me (ratio):* the c3-me distance divided by the c3-h distance.

All cephalometric hard and soft tissue landmarks were identified and digitized from the mean values obtained by two pediatric orthodontists. The calculations were performed by means of the computerized Win Ceph, cephalometric system (Rise Corporation®). All measurements were rounded to the nearest angle and 0.6 mm.

Statistical analysis

The difference in AI between hyperdivergent and neutral facial type with SBD was determined using the Mann-Whitney U-test. The differences in each variable among the 4 groups were determined with the Kruskal-Wallis test. The differences between each group were calculated with Mann-Whitney's U-test after the Kruskal-Wallis test was done.

Results

There was no difference in AI between hyperdivergent and neutral facial type with SBD (Table 1).

The results (mean ± *s*) of the craniofacial skeleton (Table 2) and dentition (Table 3), and of the pharyngeal airway space and hyoid bone position (Table 4), revealed differences among the four groups, such as findings of facial depth, facial axis, facial taper, mandibular line angle, posterior facial height, nasal floor, lower facial height, location of pogonion, interincisal angle, lower incisor to the mandibular line (L1 to ML), pharyngeal airway space (d-ad1, d-ptv, upper pharynx, lower pharynx), and soft palatal length. These variables were selected for comparison with the Kruskal-Wallis test between each group. No difference in hyoid bone position was found among the 4 groups. Comparisons among the groups are given in Table 5.

Table 5. Comparison of selected measurements of the craniofacial skeleton, dentition and pharyngeal airway space between hyperdivergent facial type with sleep-related breathing disorder (SBD) and with non-SBD, neutral facial type with SBD and non-SBD

	P	
	Hyperdivergent facial type with SBD and non-SBD	Neutral facial type with SBD and non-SBD
Facial depth	ns	ns*
Facial axis	0.017	ns
Facial taper	ns	ns
Mandibular line angle	ns	ns
Posterior facial height	ns	0.008
ans-pns	0.025	ns
Lower facial height	0.001	ns
Location of pogonion	ns	ns
Interincisal angle	0.006	ns
L-1 to ML	0.021	0.007
d-ad1	ns	<0.001
d-ptv	0.007	<0.001
Upper pharynx	<0.001	<0.001
Lower pharynx	ns	0.043
Soft palatal length	ns	<0.001

* ns: no statistically significant difference.

Hyperdivergent facial type with SBD compared to hyperdivergent facial type with non-SBD displayed, for example, a short length of the nasal floor (ans-pns; $P < 0.05$), a long anterior lower facial height (facial axis; $P < 0.05$, lower facial height; $P < 0.01$), a large interincisal angle (interincisal angle; $P < 0.01$) with retroclined lower incisors (L1 to ML; $P < 0.05$), and a narrow pharyngeal airway space (d-ptv; $P < 0.01$, upper pharynx; $P < 0.001$).

Neutral facial type with SBD compared to neutral facial type with non-SBD displayed a long posterior facial height (posterior facial height; $P < 0.01$), retroclined lower incisors (l-1 to ML; $P < 0.01$), a narrow pharyngeal airway space (d-ad1; $P < 0.001$, d-ptv; $P < 0.001$, upper pharynx; $P < 0.001$, lower pharynx; $P < 0.05$), an anterior tongue base position (lower pharynx; $P < 0.05$), and a long soft palate (soft palatal length; $P < 0.001$).

Discussion

SBD with obstructive sleep apnea is common in both adults and children, the prevalence for children being 1%–3% (21, 22), and for adults 2%–4% (23). Standards for evaluation of polysomnographic values in children with SBD are different from those used in adults. It is important to realize that children with symptomatic obstructive sleep apnea often have low apnea indices compared to adults (8, 24–26). While AI > 1 is considered to be a statistically significant difference (14), it is not apparent what level can be regarded as clinically significant. In the present study, children with AI > 0.5 were included, since we consider that children with AI < 1 also show deviation from normal.

The characteristic craniofacial skeleton in children with both hyperdivergent facial type and SBD, compared to hyperdivergent facial type with non-SBD, displays a statistically significantly longer anterior lower facial height and shorter length of the nasal floor. A similar trend was also found in the length of the nasal floor and lower facial height in children with both neutral facial type and SBD compared to neutral facial type with non-SBD, although no statistical significance was reached. On the other hand, statistically significantly longer posterior facial height was found in neutral facial type with SBD children compared to neutral facial type with non-SBD ones. Reportedly, characteristic SBD in preschool children is associated with a long anterior lower facial height, such as found in hyperdivergent facial type with SBD (4, 7), while we found that neutral facial type with SBD is particularly associated with a long posterior facial height.

Both hyperdivergent and neutral facial type with SBD were associated with hypertrophy of adenoids and tonsils. Some authors have noted that as a result of chronic nasal obstruction, mouth breathing and SBD, the altered posture of the mandible during growth leads to development of the adenoid face and/or the long face (27–33). Finally, it seems that both in children with hyperdivergent and neutral facial type the mandibular form and position may contribute to an increase in intraoral volume in order to facilitate a smooth mouth breathing (25). However, adaptation of the mandible seems to occur in different ways. In hyperdivergent facial type, anterior facial height is increased, while in normal face type, any increase in intraoral volume is created by any increase in posterior face height.

It seems that children with either hypertrophy of adenoids and tonsils (30) or obstructive sleep apnea syndrome (4), and children with both hyperdivergent and neutral facial type with SBD have a large interincisal angle with retroclined lower incisors.

The soft palate was statistically longer in the children with neutral facial type with SBD than in the children with both neutral facial type and non-SBD. In addition, the tongue base position associated with neutral facial type with SBD was more anterior than that in neutral facial type with non-SBD. Here again a trend was found towards a longer soft palate and a more anterior tongue position in children with hyperdivergent facial type with SBD compared to hyperdivergent facial type with non-SBD, although no statistical significance was reached. Concerning the length of the soft palate in adults and children, our findings are in agreement with previous investigations (34–37). Furthermore, adult SBD patients have shown reduced minimum airway dimensions behind the soft palate and tongue (38), while the relationship between the soft palatal length and the tongue base position might consequently be used to indicate the airway space. It is probable that a long soft palate persists after the preschool period.

Regarding the hyoid bone in adult normal subjects with hyperdivergent facial type, the distance between the mandibular line and the hyoid bone position is reportedly increased (39). Similarly, the hyoid bone position was low in children with obstructive sleep apnea syndrome (7). In the present study, the distances between the third cervical vertebrae, the mandible and the hyoid bone showed a constant value among the groups. Adamidis & Spyropoulos (39) have stated that a difference in the position and inclination of the hyoid bone between children with normal occlusion and mandible protrusion was cephalometrically demonstrable. Nevertheless, Hosoda et al. (20), reporting on the hyoid bone position in malocclusions, have demonstrated that this position is constant in children with skeletally normal occlusion and maxillary and mandibular protrusion; it is determined by the musculature and does not depend on the occlusion. The results of the present study suggest that the hyoid bone position is not related to facial type.

In conclusion, the present findings show that SBD may be associated with both hyperdivergent and neutral facial type. Furthermore, we could not find any specific hyoid bone position related to SBD or non-SBD or to facial type. It is important to note that while evaluation based on facial type (mandibular shape or position) does not necessarily distinguish between children with SBD or non-SBD, important differences are found in the pharynx. Short nasal floor length, long soft palate, and particularly short upper pharyngeal width can be considered as indicators of SBD. Therefore, preventive measures should be considered in preschool children with sleep-related breathing disorder.

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