# Obstructive Sleep Apneic Patients Have Craniomandibular Abnormalities

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Summary: One hundred fifty-five unselected obstructive sleep apneic patients seen in succession had cephalometric roentgenograms and polygraphic recordings performed. These patients were compared to a group of 41 subjects who had consulted orthodontists for malocclusion and had no clinical indication of sleep apnea. The cephalometric landmarks were also compared to those published as normative data in the literature. The limits of "normalcy" were conservatively defined as mean  $\pm 2$  standard deviations. Only two obstructive sleep apneic patients had normal cephalometric landmarks and 150 of the 155 patients had at least two significantly different landmarks from the normative data in the literature. The common findings were a retroposition of the mandible, a different cranial base flexure with a nasion-sella-basion angle more acute than expected, and a displacement of the hyoid bone to a lower position than expected. These combined changes reduced the space occupied by soft tissues anchored on the skull and mandible, and the length of the soft palate was increased. Key Words: Obstructive sleep apnea syndrome-Cephalometric roentgenograms.

Obstructive sleep apnea syndrome (OSAS) has attracted significant attention in the past 10 years because of its deleterious effects on daytime alertness and cardiovascular functions. The clinical symptoms of the syndrome are well established, but the underlying abnormalities associated with development of obstruction of the upper airway during sleep are still subject to controversy. Treatments should be aimed at the causative abnormalities rather than the symptoms. The understanding of the factors linked with the appearance of OSAS may also lead to its prevention.

Rojewski et al. (1), using video-endoscopy, mentioned that OSAS patients may present a "disproportionate anatomy" consisting of a large base of tongue, large soft palate, shallow palatal arch, narrow mandibular arch, and mandibular deficiency. Riley et al. (2), studying cephalometric roentgenograms in 10 patients, reported that two areas of obstruction could be identified: above the base of the tongue, associated with a large soft palate, and behind the base of the tongue, associated with retroposition of

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the lower mandible and a large distance between the mandibular plane and hyoid bone. Rivlin et al. (3), using cephalometric roentgenograms made of nine patients, found that there tended to be posterior displacement of the anterior mandibular landmark, and compared with normative values, mandibular size was smaller. Mandibular retroposition was found to be not statistically significant.

In this study, we systematically measured the cephalometric landmarks of 155 unselected obstructive sleep apnea patients seen in succession in a sleep disorders clinic by one of the investigators during a 6-month period.

#### **METHODS**

### Definitions of patient population

All subjects were referred to a sleep disorders clinic for loud snoring, excessive daytime sleepiness, or disrupted nocturnal sleep. They were all examined by the same investigator and underwent the same basic experimental protocol, which included cephalometric roentgenograms and 1 night of polygraphic monitoring. The variables monitored during the polygraphic recording were electroencephalogram, electro-oculogram, chin electromyogram, and electrocardiogram (modified V2 lead). Respiration was monitored by strain gauges or uncalibrated inductive respiratory plethysmography; airflow was measured by thermistors, and oxygen saturation by ear oximetry (Biox). Respiratory events and sleep stages were scored according to standard definitions (4), based upon findings obtained from respiratory, airflow, oximetry, and other channels. The respiratory disturbance index (RDI), which takes into account the number of abnormal breathing events per hour of sleep, was calculated (4). Patients were included in the study if their RDI was equal to or greater than 10. One hundred fifty-five patients fulfilled these clinical and polygraphic criteria. They were subdivided into 142 men with a mean age of  $49.5 \pm 11.5$  years (range 21–74 years), and 13 women with a mean age of  $50.4 \pm 10.6$  years (range 32-68 years). The mean Body Mass Index (BMI) (5) of the total population was  $32.1 \pm 5.7$  (normal < 30). The BMI among men was  $30.5 \pm 5.6$  (range 19.8–57.5), and the BMI for women was  $36.4 \pm 7.6$  (range 21.6-47.5).

# Cephalometric roentgenogram studies

Lateral cephalometric roentgenograms were obtained with the Wemer cephalostat. The technique of Riley et al. (2) was used. Tracings of the roentgenograms from all subjects were performed on an acetate sheet by a single investigator (Figs. 1 and 2). The following cephalometric landmarks previously determined in normal populations and cited in textbooks and referenced articles were obtained: S: sella, the center of the hypophyseal fossa (sella turcica); N: nasion; A: subspinale, the deepest point on the premaxillary outer contour between the anterior nasal spine and the central incisor; B: supramentale, the deepest point on the outer mandibular contour between the mandibular incisor and the pogonion (Pg); ANS: anterior nasal spine, the most anterior part of the nasal floor; PNS: posterior nasal spine, the most posterior part of the contour of the hard palate; Gn: gnathion, the most inferior point in the contour of the chin; Go: gonion, the most posterior and inferior point on the convexity of the angle of the mandible; MP: mandibular plane, a plane constructed from the gnathion through the gonion; H: hyoid, the most anterior-superior point on the body of the hyoid bone; Ba: basion, the midpoint of the anterior border of the foramen magnum (Fig. 1).







measurement from sella (S) to nasion (N) to point B (supramentale)], GoGn-SN [angle ] measurement formed by the intersection of a line passing through Go (gonion) and Gn (gnathion) and another line through N (nasion) and S (sella)], NSBa [also called cranial base flexure (angle formed by the intersection of lines drawn from nasium to sella and 2 sella to basion)], MP-H [distance from mandibular plane (MP) to hyoid bone (H)], PNS-P [distance from posterior nasal spine (PNS) to the tip of the soft palate (P), not  $\overset{\circ}{\otimes}$ shown in figure] (Fig. 2).

The posterior airway space (PAS), defined by Riley et al. (2) as the space behind the  $\gtrsim$ base of the tongue (2) and limited by soft tissues (i.e., more difficult to clearly delineate than a bony landmark), was also measured for comparison purposes. g

# Definition of normative values and selection of "control" cephalometric roentgenograms from well-defined populations

The normative values with which the study group was compared were those pub- $\frac{1}{2}$ lished in textbooks on craniofacial anatomy and referenced articles (6-14). However,  $\aleph$ since mandibular retroposition had been mentioned in previous articles dealing with OSAS and cephalometric roentgenograms (2,3), we attempted to obtain cephalometric roentgenograms from control subjects. As control subjects, we selected individuals who had consulted orthodontists for mild to moderate malocclusion or overiet but who had no clinical indication of OSAS. None of these individuals had a history of loud snoring, excessive daytime sleepiness, or disturbed nocturnal sleep. This control population included 41 subjects: 29 men, mean age 28.6  $\pm$  14.0 years (range 16-59 years), and 20 women, mean age 27.7  $\pm$  9.8 years, (range 16-50 years). From these 49 subjects, we created a subgroup (control II) based on age, i.e., eliminating subjects between 16 and 20 years of age since there were no patients in that age group (and mandibular growth may have not been completed at 16 years). We also eliminated subjects whose findings were not within the two limits of standard deviations for data published in the literature. "Control group II" included 17 subjects: seven men, mean age  $37.8 \pm 13.9$  years (range 23-59 years), and 10 women, mean age  $31.2 \pm 9.8$  years (range 20-50 years).

### Statistical analysis

Analysis of variance and Student's t tests were used for comparison between groups. Separate variance T or pooled variance T was used depending on the results of the Levene test for equality of variances (15). The Bonferroni criteria for significance levels were used when multiple simultaneous comparisons were made (15).

#### RESULTS

The means and standard deviations of each of the measured angles and dimensions for the OSAS patient group, and the orthodontic treatment subjects, as well as normative data from the literature and control group II, are presented in Table 1.

The limits of "normalcy" were conservatively defined as mean  $\pm 2$  standard deviations, using normative values published for SNA, SNB, GoGn-SN, NSBa angles, and MB-H and PNS-P distances (6-14). We found that only two OSAS patients out of 155 simultaneously fulfilled these six criteria of "normalcy." Allowing one measurement out of the six outlined above to be outside the limits of "normal" resulted in only three additional patients being added to this group.

Except for two individuals, the entire group of unselected OSAS patients presented at least one abnormality of craniomandibular anatomy, and 150 out of 155 had at least two landmarks significantly different from the norms.

In contrast, when a similar analysis was performed on subjects sent for orthodontic treatment, 41 out of 49 subjects were within 2 standard deviations of the mean for all considered variables (by definition, control group II was within 2 standard deviations). Each angle or dimension was treated by analysis of variance, following the conservative approaches indicated above. There were no statistically significant differences for angle SNA among OSAS patients, normative values in the literature, orthodontic subject group, or control group II. There were, however, significant differences for each of other measurements, particularly when the results were compared with normative values from the literature. These data are presented in Table 1.

Particularly noteworthy is the finding that OSAS patients presented statistically significant craniomandibular landmark differences when compared with normative published values. The same was true when a control group, initially defined by taking into consideration the normative data from the literature (control group II), was compared with the OSAS patient group. Even when we compared our OSAS patient group to subjects with orthodontic problems, and those with a potential risk of developing OSAS, most landmarks (with the exception of SNB) were statistically different from those noted in OSAS patients (Table 1).

The different analyses indicated that OSAS patients had the following characteristics:

(a) a normally positioned maxilla,

(b) a retroposition of the mandible,

(c) different cranial base flexure with a nasion-sella-basion angle smaller than expected (i.e., more acute).

Cephalometric variables	OSAS patients			Literature data		Orthodontic treated subjects			Control group II			Significanaa
	x	SD	N	x	SD	x	SD	N	$\overline{x}$	SD	N	levels
SNA angle (degrees)												
Total population	81.1	4.1	154	82	2	81.5	2.5	49	83.0	1.7	17	NS
Men	82.1	4.2	141			80.5	2.3	29	80.8	1.5	7	NS
Women	80.1	3.7	13			82.5	2.8	20	84.5	2.5	10	NS
SNB angle (degrees)												
Total population	77.5	4.3	155	80	2	78.5	2.5	49	79.4	2.6	17	a,d
Men	79.0	4.2	142			78.7	2.6	29	80.1	2.3	7	j
Women	76.0	4.3	13	-		78.4	2.4	20	78.9	2.8	10	j
GoGn-SN (degrees)												
Total population	35.1	7.4	155	36	6	33.8	6.4	33	33.8	7.3	17	NS
Men	33.3	7.5	142			31.8	6.0	19	30.0	5.1	7	NS
Women	36.9	7.4	13	-		35.9	6.8	14	36.5	7.6	10	NS
NSBa (degrees)												
Total population	130	6.5	155	137	5.7	132.5	5.3	33	131.9	4.0	17	c,e
Men	128.7	6.4	142			131.9	5.5	19	131.7	3.3	7	h
Women	131.4	6.8	13			132.6	4.4	14	132.0	4.6	10	NS
MP-H (mm)												
Total population	25.0	6.8	154	15.4	3	14.6	4.5	49	14.5	4.1	17	c.f,i
Men	26.6	6.7	142			15.5	4.9	29	16.1	3.6	7	i
Women	23.4	7.0	12			13.7	4.2	20	13.3	4.3	10	i
PNS-P (mm)												
Total population	45.7	6.5	155	37	3	36.8	7.5	49	37.5	6.1	17	c.f,i
Men	46.7	6.7	142			37.3	9.1	29	36.4	4.4	7	h,e
Women	44.7	5.7	12			36.3	5.6	20	38.2	7.2	10	h,d
PAS (mm)												
Total population	5.1	3.2	151	11	1	10.6	3.4	49	10.8	4.0	17	c,f,i
Men	5.3	3.3	138			11.2	2.8	29	12.4	3.5	7	f,i
Women	5.0	3.0	13			9.9	3.6	20	9.6	4.2	10	f,i

TABLE 1. Cephalometric variables in different subject groups

OSAS, obstructive sleep apnea syndrome; PAS, posterior airway space; NS, not significant. See Methods for definition of other abbreviations.

<sup>*a*</sup> p < 0.05, OSAS patients vs. literature data.

Sleep, Vol. 9, No. 4, 1986

<sup>b</sup> p < 0.01, OSAS patients vs. literature data.

c p < 0.001, OSAS patients vs. literature data.

<sup>*d*</sup> p < 0.05, OSAS patients vs. control group.

e p < 0.01, OSAS patients vs. control group.

f p < 0.001, OSAS patients vs. control group.

 $^{8}$  p < 0.05, OSAS patients vs. orthodontic subjects.

<sup>h</sup> p < 0.01, OSAS patients vs. orthodontic subjects.

 $^{i}$  p < 0.001, OSAS patients vs. orthodontic subjects.

 $^{j}$  p < 0.05, male vs. female patients with OSAS.

<sup>k</sup> p < 0.01, male vs. female patients with OSAS.

 $^{1}$  p < 0.001, male vs. female patients with OSAS

JAMIESON ET AL

The combined effect of (b) and (c) reduces the space occupied by soft tissues anchored on the skull and mandible.

(d) a displacement of the hyoid bone to a lower position than expected,

(e) significant increase in the length of the soft palate.

The total group of 49 orthodontic subjects appeared to have cephalometric measurements intermediate in value between OSAS patients and control group II (defined by the 2 standard deviations limit from normative data published in the literature). However, when compared with OSAS patients, the orthodontic subjects had (a) a higher position of the hyoid bone, (b) a shorter soft palate, and (c) in men, a larger cranial base angle.

Comparison between men and women presenting with OSAS indicated that women had a more pronounced retroposition of the mandible (p < 0.05) and had a less steep mandibular plane (a smaller GoGn-SN angle).

#### COMMENTS

To date, this is the largest group of unselected OSAS patients subjected to systematic cephalometric analysis. The study was planned to cover a large enough patient population to ascertain the frequency of craniofacial abnormalities in OSAS in a reasonable period of time. One investigator only, blind to clinical findings, performed all the cephalometric analyses.

The orthodontic treatment control group does not represent the general population. It was selected because none of the individuals presented clinical indications of OSAS, and because we have not subjected a large group of normal controls to our method of cephalometric analysis. Although it is possible that some of these subjects may develop OSAS at a later age, the absence of clinical findings of OSAS at the time of this study enabled us to analyze the validity of our hypothesis: that OSAS patients tend to have craniomandibular abnormalities.

The results were more or less as expected. The majority of our population was male, as OSAS is much more prevalent in men than in women. Women with OSAS presented a statistically more significant retroposition of the mandible than men with OSAS. Associated with the more posterior position of the mandible, as indicated by angle SNB, is the backward displacement of the attached soft tissue. We attributed little value to the GoGn-SN angle difference between OSAS men and women. A statistical difference was found when control group II women were compared with control group II men, but the retroposition of the mandible alone is not necessarily sufficient to induce OSAS, as seen in our patients undergoing orthodontic treatment. In addition to mandibular retroposition, our OSAS patients had a more acute cranial base angle. Teleologically, one would expect prograthism to develop as a result of this to maintain a good airway. Finally, OSAS patients have a hyoid bone position that is lower than expected. Wickwire et al. (16), investigating the effects of experimental mandibular osteotomy on tongue position, have demonstrated that posterior displacement of the mandible has an immediate impact on the placement of the tongue and hyoid bone. One may question if the more pronounced cranial base flexure with a more acute nasion-sella-basion angle (NSBa) and retroposition of the mandible are not primary in the low hyoid position noted in OSAS. The difference in hyoid position between OSAS patients and patients undergoing orthodontic treatment must be emphasized.

Individuals without OSAS but subject to orthodontic treatment have an SNB angle

#### A. JAMIESON ET AL.

similar to OSAS patients. As a group, however, their cranial base angle is less pronounced, allowing a larger pharyngeal space and a more normal position of the hyoid bone. We hypothesize that the more acute NSBa angle, i.e., a more acute cranial base flexure with the associated lower position of the hyoid bone, leads to abnormal development of the hypopharyngeal soft tissues associated with lower position of the hyoid bone. Men and women with Oorner groups. In the same manner, alterations of the soft tissue may and conjoint effect of mandibular retroposition and acute cranial base flexures (i.e., the length of the soft palate and the position of the hyoid bone were significantly different in OSAS patients when compared with the findings noted in orthodontically treated patients with similar retroposition of the mandible who had no signs of OSAS and no in oranial base flexure). We had hoped that the area limited anteri-the "retromandibular triangle" (Fig.

orly by the posterior ramus of the mandible, i.e., the "retromandibular triangle" (Fig. 2, triangle x-y-z), would delineate an area whose measurement would be helpful for distinguishing OSAS patients from other subjects with retroposition of the mandible, but such was not the case. In conclusion, our study indicates that OSAS patients typically present a combination of craniomandibular abnormalities. It seems that one abnormality alone (such as a moderate change in SNB angle, or low hyoid position) may not be sufficient to lead to OSAS, but that more complex relationships among craniomandibular elements may be involved. The experimental work performed by orthodontists (16) has demonstrated that depending on bone placements, soft tissues will be affected, particularly the soft palate and the secondary hyoid bone location. Since the hyoid serves as one of the two anchors of the tongue muscles that anteriorly limits the upper airway in the hypopharynx, this may represent a significant predisposition to the development of obstructive apneas during sleep. These reported findings may be important in the investigation of patients with OSAS and in planning therapeutic approaches. Acknowledgment: Supported by the General Clinical Research grant 00070 funded by the National Institutes of Health. Andrew Jamieson is supported by a U.S. Public Health Service International Research Fellowship 1 FO5 TWO3648-01. **REFERENCES** 

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