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Case Report

Surgical treatment for sleep apnea: changes in craniofacial and pharyngeal airway morphology in a child with achondroplasia

Kieko Sato[1,2], Naoko Niikuni[1,2], Ichiro Nakajima[1,2], Tetsuo Shirakawa[1,2] and Hideaki Sakata[3]

[1]Department of Pediatric Dentistry, Nihon University school of Dentistry, Tokyo, Japan
[2]Division of Oral and Craniomaxillofacial Research, Dental Research Center, Nihon University school of Dentistry, Tokyo, Japan
[3]Department of Otolaryngology, Saitama Children's Medical Center, Saitama, Japan

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Abstract: Here, we report the case of a male child with achondroplasia who was diagnosed with obstructive sleep apnea and underwent adenoidectomy and tonsillectomy. By analyzing lateral cephalograms, we evaluated the craniofacial and pharyngeal airway morphology immediately before surgery (age, 5 years 6 months) and 1 year 2 months after surgery (age, 6 years 8 months). Adenoidectomy and tonsillectomy dilated the pharynx and improved the craniofacial and pharyngeal morphologies, apparently thus improving the sleep apnea. (J. Oral Sci. 49, 173-177, 2007)

Keywords: achondroplasia; child; sleep apnea; craniofacial morphology; pharyngeal airway morphology.

Introduction

Achondroplasia (AP) is an autosomal dominant disease characterized by low stature, morphological abnormalities such as cranial and maxillo-facial hypogrowth, and functional disorders such as sleep-disordered breathing (1-5). In a previous study, to evaluate the status of sleep-disordered breathing in children with AP, we conducted a questionnaire survey of breathing during sleep, and found higher incidences of snoring and apnea during sleep in children with AP than in healthy children (6). Also, in a previous investigation of craniofacial and pharyngeal morphologies in AP patients with sleep-disordered breathing, we observed narrowing of the upper airway, retruded position of the chin, and the dolicofacial pattern with an increase in lower facial height, suggesting that the craniofacial and pharyngeal morphologies in AP induce snoring and apnea during sleep (7). However, there is no established treatment to improve the craniofacial morphology of children with AP.

In the present case study of a male patient with AP, who was diagnosed with obstructive sleep apnea and underwent adenoidectomy and tonsillectomy, we used analysis of lateral cephalograms to compare craniofacial and pharyngeal airway morphology before surgery (age, 5 years 6 months) and 1 year 2 months after surgery (age, 6 years 8 months).

Correspondence to Dr. Kieko Sato, Department of Pediatric Dentistry, Nihon University School of Dentistry, 1-8-13 Kanda-Surugadai, Chiyoda-ku, Tokyo 101-8310, Japan
Tel: +81-3-3219-8106
Fax: +81-3-3219-8353
E-mail: onodera-k@dent.nihon-u.ac.jp

Case Report

The patient was brought to the Otolaryngology Outpatient Service, Saitama Children's Medical Center, with chief complaints of snoring and frequent several-second apnea during sleep. The patient had a systemic disorder, AP, and was diagnosed with obstructive sleep apnea. Because pharyngeal and palatine tonsillar hypertrophy was observed, bilateral tonsillectomy was performed. Using standard lateral cephalograms, the craniofacial and pharyngeal morphologies were compared before bilateral tonsillectomy and 1 year 2 months after surgery. The patient's age was

5 years 6 months at the time of surgery, and was 6 years 8 months at 1 year 2 months after surgery. For presentation of this case report, we obtained approval from the Ethics Committees of Saitama Children's Medical Center and Nihon University School of Dentistry. We explained the study objective and contents in writing and orally to the patient and his parents, and obtained their informed consent.

Cephalometric evaluation

The methods used to analyze craniofacial and pharyngeal morphologies are shown in Figs. 1, 2 and 3, respectively. The standard analytical values of craniofacial and pharyngeal morphologies were derived from Ricketts analysis (8), McNamara analysis (9), and Deberry-Borowieskin analysis (10).

1) Measurements of the craniofacial morphology and the pharyngeal airway (Figs. 1, 2):
①facial axis: angle between the facial axis plane and BA-NA plane
②facial depth: angle between the facial plane and the FH plane
③mandibular plane angle: angle between FH plane and the mandibular plane
④lower face height: angle between ANS-Xi plane and Xi-Pm plane
⑤mandibular arc: angle between Pm-Xi plane and Xi-DC plane
⑥corpus length: distance between Xi point and Pm point
⑦A-to-McNamara line: distance between the McNamara line and point A
⑧pogonion-to-McNamara line: distance between the McNamara line and the pogonion
⑨nasal floor length: the distance between the ANS point and PNS point.
⑩D-AD1: the distance between the PNS point and the nearest adenoid tissue, measured along the PNS-Ba plane
⑪D-AD2: the distance between the PNS point and the nearest adenoid tissue, measured along a line from the PNS plane perpendicular to the S-Ba plane
⑫upper pharynx: the shortest distance from the upper surface of the palatine velum to the adenoid tissue
⑬lower pharynx: the distance between the intersection point where the tongue base meets the lower contour of the mandible and the posterior pharyngeal wall.

2) Landmarks and lines (Fig. 3):
A: the deepest point on the concave outline of the upper labial alveolar process, extending from the anterior nasal spine to the prosthion
ANS: the most anterior point at the sagittal plane on the bony hard palate
Ar: the intersection point of the ramus plane (a tangent lin on the posterior contour of the ramus) and the occipital born bas-fond interior margin
Ba: the most inferior posterior point of the occipital bone at the anterior margin of the occipital foramen
Gn: the intersection point of the mandibular plane and the facial plane
Go: the intersection point of the mandibular plane and the

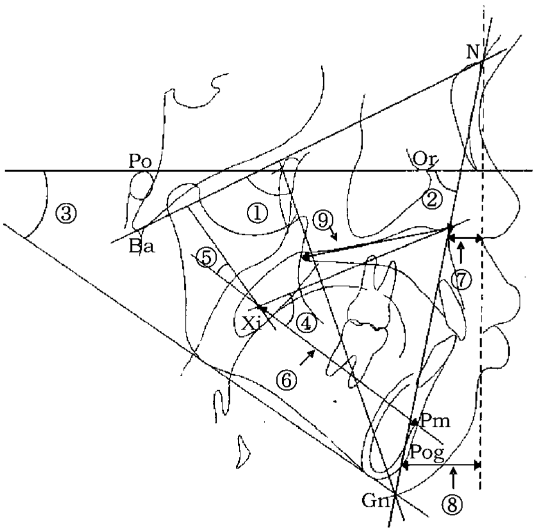


Fig. 1 Measurements of the craniofacial morphology.
①facial axis ②facial depth ③mandibular plane angle ④lower face height ⑤mandibular arc ⑥corpus length ⑦A to McNamara line ⑧pogonion to McNamara line ⑨nasal floor length

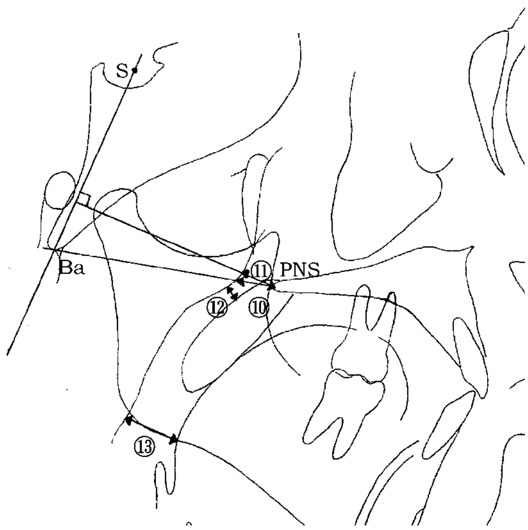


Fig. 2 Measurements of the pharyngeal airway.
⑩D-AD1 ⑪D-AD2 ⑫upper pharynx ⑬lower pharynx

ramus plane

N: the most anterior point of the frontonasal suture

Or: the most interior 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 at the sagittal plane on the bony hard palate

Po: the most superior point on the radiolucency of the external and internal auditory meati; it is located posterior to the mandibular condyle and posterior clivus

Pog: the most prominent point of the chin

PT: the intersection point of the inferior border of the foramen rotundum and the posterior wall of the pteryomaxillary fissure

S: the center of the pituitary fossa of the sphenoid bone

Xi: a point located at the center of the ramus; the location of Xi is keyed geometrically to the FH and PTV plan in 4 steps: 1) planes are constructed perpendicular to the FH and PTV planes; 2) these constructed planes are tangent to points (R1, R2, R3 and R4) on the borders of the ramus; 3) the constructed planes form a rectangle enclosing the ramus; 4) Xi is located in the center of the rectangle at the intersection of its diagonals.

facial axis plane: line connecting the PT and Gn points

facial plane: line connecting the Na and pog points

frankfort horizontal (FH) plane: line connecting the Po and Or points

McNanara line: line, perpendicular to the FH plane, that passes through N

mandibular plane: line tangent to the lower border of the mandible

Pterygoid vertical (PTV) plane: line, perpendicular to the FH plane, that passes through the PT

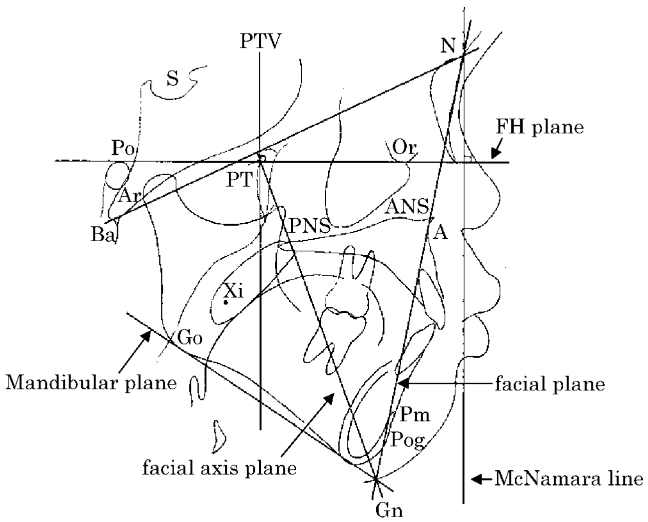


Fig. 3 Landmarks and lines.

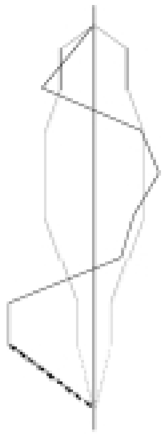
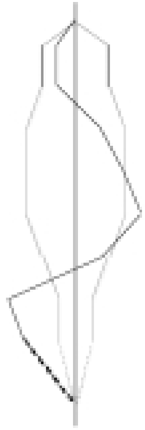
Results and Discussion

The results of analyses of craniofacial and pharyngeal morphologies are shown in Table 1.

A recent study indicates that the characteristics of craniofacial and airway morphology of AP can induce sleep apnea (7). Other studies indicate that adenoidectomy of a child is an effective means of improving sleep apnea, daytime sleepiness and quality of life (11,12). However, there have been no reported studies of craniofacial and airway morphology after adenoidectomy and tonsillectomy. Therefore, in the present study, we compared craniofacial and pharyngeal morphologies before and after adenoidectomy and tonsillectomy in a child with AP diagnosed with sleep apnea accompanied by tonsillar hypertrophy.

In the present comparison of preoperative craniofacial morphology using standard values, both the maxilla and mandible (point A, point pog) were retruded, and the lower facial height was high. The preoperative lower facial height was +1 s.d., relative to the standard value, but the postoperative lower facial height was within the standard range. At point A, the preoperative value was -4 s.d. relative to the standard value, but the postoperative value (-3 s.d.) was close to the standard value. At point pog, the preoperative value was -4 s.d., relative to the standard value, but the postoperative value (-2 s.d.) was close to the standard value. The items that differed significantly between preoperative and postoperative measurements in the present study were the same items that differed significantly between AP patients and healthy subjects in previous studies. Tanimoto et al. (13) evaluated association between snoring and craniofacial morphology in subjects aged 6 to 15 years, and reported that more marked retrusion of the mandibular position was associated with a higher frequency of snores during sleep at night. In the present study, the patient's sleep apnea was improved after surgery. We speculate that this improvement in sleep apnea was due to the change in the mandibular position to a more anterior position. These results suggest that adenoidectomy and tonsillectomy lead to protrusive restoration of craniofacial morphology. In the present patient, all postoperative analytical values of the laryngo-nasal and oropharyngeal regions were larger than the preoperative values. Studies show that in children, respiratory impairment during sleep inhibits somatotropin secretion related to body growth

Table 1 Standard values of analysis of craniofacial and pharyngeal morphologies, and measured values

Measurement	aged 5 years and 6 months (pre-operation)				aged 6 years and 8 months (post-operation)			
	standard data	s.d.		case data	standard data	s.d.		case data
Craniofacial morphlogy								
facial axis (deg)	86.0	3.0		84.1	86.0	3.0		84.2
facial depth (deg)	84.8	3.0		80.7 (-1s.d.)	85.1	3.0		83.5
mandibular plan angle (deg)	30.8	4.0		34.8	30.6	4.0		32.5
lower facial hight (deg)	49.0	4.0		55.2 (+1s.d.)	49.0	4.0		53.0
mandibular arc (deg)	23.8	4.0		27.0	24.1	4.0		31.0 (+1s.d.)
corpus length (mm)	57.0	3.0		59.0	58.5	3.0		61.0
point A (mm)	1.0	2.0		-9.0 (-4s.d.)	1.0	2.0		-6.0 (-3s.d.)
point pog (mm)	-8.0	2.0		-18.0 (-4s.d.)	-7.5	2.0		-10.5 (-2s.d.)
nasal floor length (mm)	–	–		41.0	–	–		45.2
Pharyngeal airway								
D-AD1 (mm)	–	–		6.8	–	–		8.2
D-AD2 (mm)	–	–		5.8	–	–		7.2
upper pharynx(mm)	–	–		2.5	–	–		4.0
lower pharynx(mm)	–	–		6.5	–	–		7.0

s.d.: standard deviations, -: no data

(14). In the present case, we speculate that the improvement in the sleep disorder, as a result of adenoidectomy and tonsillectomy, led to some improvement of the AP-associated growth impairment.

In summary, in the present case, adenoidectomy and tonsillectomy in a child with AP dilated the pharynx and changed the craniofacial morphology, leading to improvement of sleep apnea.

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