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Chapter

The Development of Oral Functions in Children: A Clinical Study of Stomatognathic Dysfunction

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and Taketo Yamaguchi*

Abstract

In most countries that have reached an aging society, the feeding function among the elderly population has declined and become a serious problem. Therefore, understanding the development of human oral function is required to address this problem. However, only a few research studies have reported oral motor functions and dysfunctions in children. Our chapter describes the relationship between oral motor functions (chewing, swallowing, and breathing) and maxillofacial morphology in children. In addition, case studies on children with cerebral palsy and sleep aspiration disorders will also be introduced. This study would also like to clarify the significance of human oral function development from infancy in pediatric dentistry.

Keywords: child, oral function, obstructive sleep apnea, cerebral palsy, achondroplasia

1. Introduction

Oral diseases include dental caries, periodontal (gum) disease, tooth loss, oral cancer, orodental trauma, and normal and birth defects such as cleft lip and palate [1]. These diseases have been known to be preventable by oral care [2]. Recently, most countries with aging societies implemented dental care significantly and strategically preventing age-related decline in oral functions [3, 4].

Oral health is a fundamental component of overall health. All children and youth should have access to preventive and treatment-based dental care. However, some children have difficulty availing of oral care and dental treatment due to their systemic disease or disability [5, 6]. Thus, caring and supporting for their oral functions remain unclear. Particularly, only a few studies have been conducted on the causes of oral dysfunctions in children with developmental motor dysfunction (i.e., cerebral palsy [CP], muscular dystrophy, etc.) and congenital malformation (achondroplasia, ectodermal dysplasia, etc.).

To clarify the relationship between systemic symptoms and oral dysfunction, symptoms of sensory-motor dysfunctions in CP and congenital malformation in achondroplasia have been examined, respectively.

Therefore, this chapter reviews the findings of previous studies and discusses the importance of establishing clinical research on oral dysfunctions in children with congenital diseases.

The contents of this chapter are as follows:

- Development of eating behavior
- Sensory-motor dysfunctions in the patients with cerebral palsy
- Obstructive sleep apnea in the children with achondroplasia

This chapter is believed to include useful content as a reference for clinical research on oral functions in children with disabilities.

2. Development of eating behavior

In this section, we briefly review the eating behavior development from the neonatal period in humans.

Oral functions consist of eating (breastfeeding and mastication), swallowing, and pronunciation/speech. The key to eating function development is the transition from breastfeeding to mastication behavior. Breastfeeding is an eating function composed of primitive reflexes, whereas mastication is a learning behavior composed of voluntary movement and chewing rhythm. In this section, we briefly describe the feeding function changes from the fetal to the neonatal to the weaning period.

2.1 Breastfeeding/suckle

The human jawbone is already formed around the 6th week of embryonic development. Moreover, the tooth germ formation of the primary teeth also begins around the 7th week. The calcification of the primary teeth begins around the 4th month. Since birth, a primitive reflex enables a newborn to suckle [7].

A newborn's oral cavity is sensitive and reflexive. For example, the sucking reflex is easily elicited by stimulating (tactile/chemical) the palate with the nipple.

- Rooting reflex: The reflex to seek the nipple
- Captive reflex: The reflex to catch the nipple
- Sucking reflex: The reflex to suck the mother's nipple with a fixed rhythm

The intermaxillary space and the presence of a sucking fossa in the palate are suitable forms for breastfeeding. During sucking, the tongue presses the nipple against the sucking fossa at the center of the palate (**Figure 1A**). The intermaxillary space is found in the anterior alveolar ridge of the upper and lower jaws before the primary tooth eruption. A newborn baby can use that space to hold the nipple (**Figure 1B**).

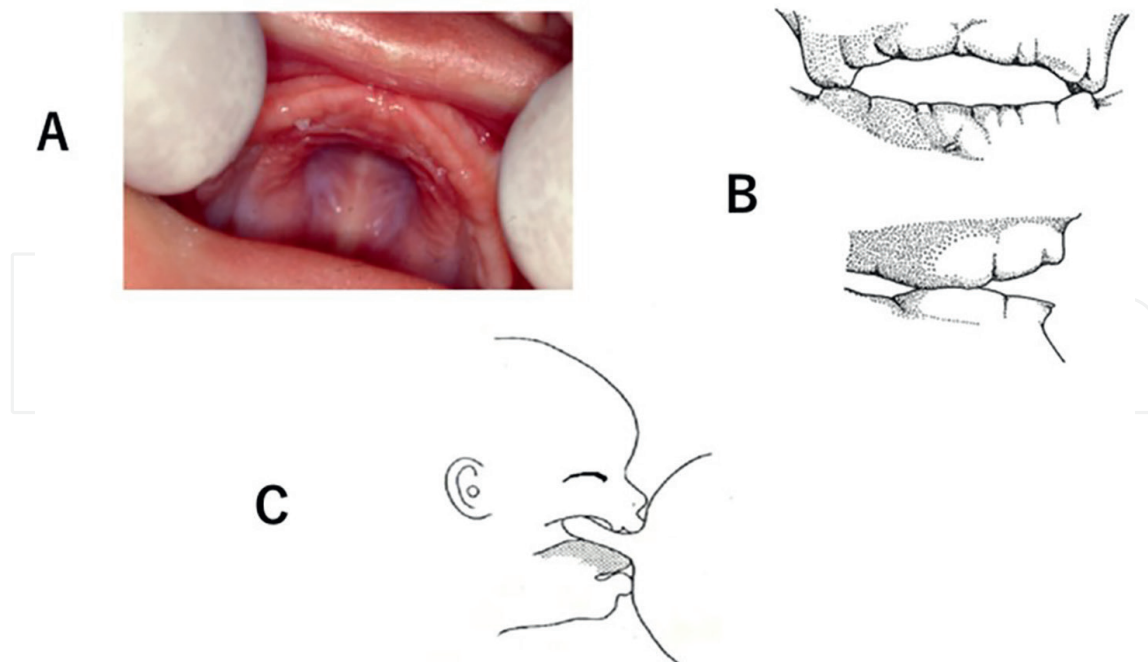


Figure 1. Features of oral morphology for breastfeeding in newborns. A: sucking fossa, B: intermaxillary space, C: Tongue movement during breastfeeding.

The baby makes peristaltic movements from the tip of the tongue to the base of the tongue, creating negative pressure in the oral cavity and ingesting milk (**Figure 1C**).

Recently, Shiv Shankar Agarwal et al.'s cross-sectional retrospective study reported that children breastfed for <6 months had an almost twofold increased probability of developing sucking habits and non-spaced dentition than those who breastfed for >6 months duration [8]. Their data suggest the possibility that nonnutritive sucking habits may act as a dominant variable in the relationship between breastfeeding duration and occurrence of convex facial profile and disocclusion in deciduous dentition.

Furthermore, the study also evaluated the effects of breastfeeding and bottle feeding methods, such as bottle and cup feeding, on infant masticatory muscle activity, indicating that masticatory muscle activity is relatively high during breastfeeding. In any case, the oral function development in humans is thought to involve the suckling condition.

2.2 Mastication/chewing

Transitioning from sucking to mastication is a characteristic eating behavior change in mammals including humans. Tooth eruption and the associated stimulation from the surrounding tissue of the periodontal membrane are believed to be associated with the initiation of mastication (**Figure 2**). The weaning period is very important for the beginning of mastication.

At 5–6 months post-birth (weaning period), the upper and lower anterior teeth erupt, making it easier to separate the lip and tongue movements.

The changes observed during this period are as follows:

1. Expansion volume of the oral cavity.
2. Easier up and down movement of the tongue.

3. Mature swallowing with closing lips.
4. Crushing food with the tongue and alveolar ridge.
5. Crushing food with the teeth.

The upper and lower second primary molars erupt around age 2 years and 6 months, and the occlusion of 20 primary teeth is completed around 3 years (**Figure 3**).

With the second primary molar eruption, the masticatory force is believed to be increased. Biting force is further increased when the first permanent molars are occluded at 6 years, resulting in the establishment of mastication corresponding to growth during school age.

Recently, Nabeel et al. conducted a systematic review of jaw movements, bite force, and electromyograms of mastication from children aged <6–18 years [9].

They demonstrated that after 12 years, a significant increase in bite forces and electromyogram (EMG) activities occurred, and the frontal jaw pattern became similar to that of adults, suggesting that mastication gradually improves with the development of orofacial structures and was mainly influenced by a dental eruption.

As described in this section, an oral or stomatognathic function can be considered to be closely associated with nervous and muscular development as well as maxillofacial, oral cavity, and tooth growth.

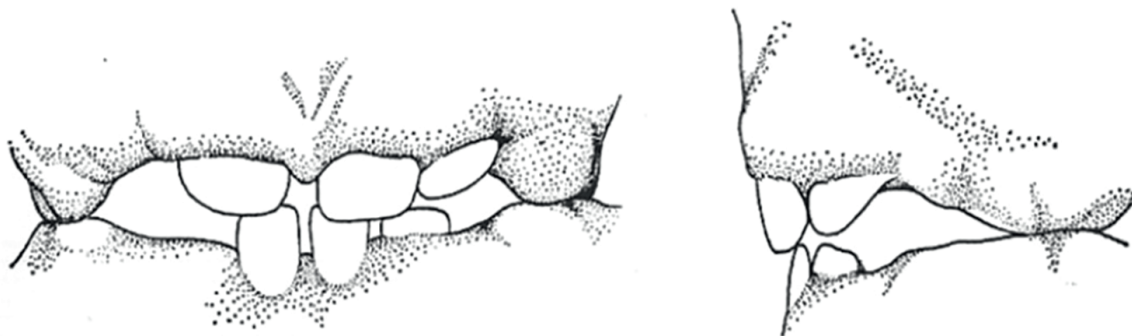


Figure 2.
Eruption of primary incisors.



Figure 3.
Primary dentition occlusion.

3. Cerebral palsy (CP)

CP is a nonprogressive disorder that affects the motor system that moves the body due to a defect in the central nervous system (CNS). The defect might be before or at the time of or after the time of child's delivery. [10]. The manifestation of CP is varied and depends upon the subtypes of CP. Motor system is the predominantly affected system with altered muscle tone, abnormal monosynaptic and polysynaptic reflexes, altered motor control and motor learning. The defects don't limit itself to the limbs and the trunk but also affect the face muscles, jaw muscles, which are closely associated with the head and neck muscles. Patients with CP were known to have lower feeding function than healthy subjects [11]. All clinical studies in this section were performed with ethics committee approval.

3.1 Masticatory efficiency and bite force

Nakajima et al. examined the relationships between masticatory efficiency, bite force, and masticatory rhythm in children with CP, comparing them with those in healthy children [12].

Table 1 shows the results of the comparison of masticatory efficiency and maximum bite force between children with CP and healthy children. The age ranges of children with CP and healthy children were 6–15 (mean age, 10) years and 6–15 (mean age, 10) years, respectively. Adenosine triphosphate granules were used as mastication samples, and the amount of pulverization was measured based on absorbance.

	Masticatory efficiency	Maximum biting force
Children with Cerebral Palsy	0.167 ± 0.135	32.2 ± 20.8
Normal Children	0.545 ± 0.355	51.1 ± 16.0
Significant difference	**	**

Adapted from reference [12].

Table 1.
 Comparison between children with cerebral palsy and healthy children in masticatory efficiency and maximum biting force.

As shown in **Table 1**, children with CP have significantly lower masticatory efficiency and bite force than healthy children of the same age. In the healthy children group, a significant correlation was also obtained with a correlation coefficient of 0.568 between the two indices ($p < 0.01$). Conversely, in children with CP, no significant correlation was observed with a correlation coefficient of -0.173 ($p > 0.05$).

These findings indicated that the muscle strength of mastication in CP may not be sufficiently developed to crush the mastication sample, as compared with their healthy counterpart.

Electromyogram (EMG) of the masticatory muscles (temporal and masseter muscles) during the masticatory efficiency measurement was recorded in the experiment. EMG data showed that children with CP had an unstable chewing rhythm compared to healthy children, suggesting an oral-facial sensation dysfunction that controls mastication.

3.2 Orofacial sensation

Patients with CP tend to develop accentuated involuntary muscle tonus in orofacial muscles during mastication. The muscle tonus abnormality is considered to affect oral sensation and a factor in reducing the eating function of patients with CP.

Therefore, Yoshida et al. investigated abnormalities in the lower-jaw-position sensation in patients using a lower-jaw-position discrimination test [13].

In that study, the mandibular position sensation was measured for adults with CP (CP group) and healthy adults (control group) using the following method.

This test was performed based on the extent of mouth opening to estimate a sensation associated with elongation of muscle spindles in masticatory muscles.

In the test, the participants were asked to hold a 10.0-mm metal rod (reference mouth opening) between the upper and lower incisors for several seconds and memorize its thickness. Thereafter, they were asked to hold another 0.5-mm-thick or thin metal rod and to answer whether these rods were “larger” or “smaller” than the standard mouth opening to count the wrong answers to examine the rate of miss estimate (RME).

Figure 4 shows the comparison between both groups based on RME data.

The RMEs of patients with CP were higher than those of healthy participants for jaw opening magnitudes, suggesting that some abnormalities exist in the mandibular sensation afferent system from the peripheral to CNS in patients with CP.

This phenomenon might be explained by the excessive excitation of the gamma motor nerves of muscle spindles, considering that the RME is extremely high at rods lower than the reference rod.

In other words, it is conceivable that patients with CP remain more sensitive to oral sensation than healthy participants.

Morimoto et al. reported that vibrating stimuli in healthy adults decreased the mandibular position sensation and considered that this phenomenon was caused by the regulation of the mandibular position sensation by muscle spindles [14]. Therefore, we also investigated the RME of mandibular position sensation by applying vibration stimulation to determine whether muscle sensory abnormalities in patients with CP are related to muscle spindles.

Figure 5 shows the comparison between both groups based on RME data after the vibration stimulation. No significant differences were observed between two groups for any interincisal distances ($p > 0.05$).

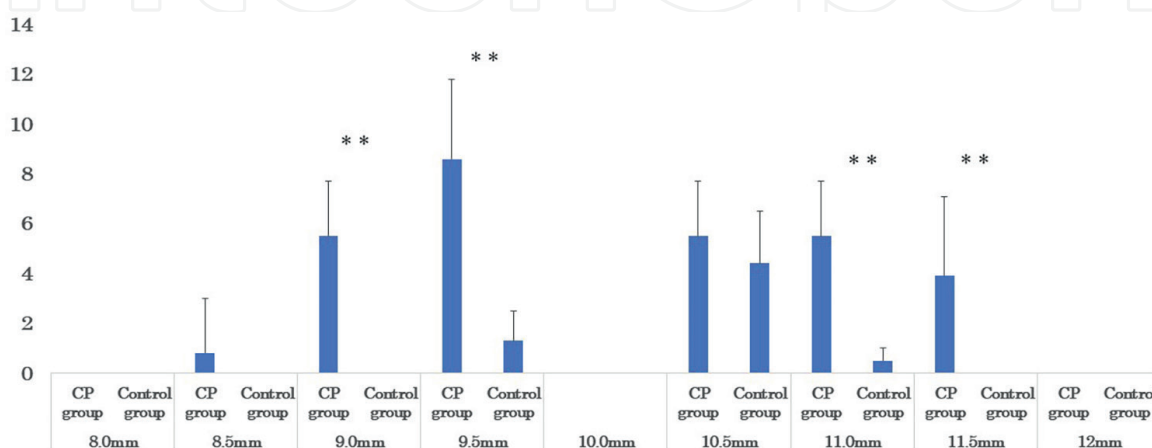


Figure 4. Comparison between CP and control groups in R.M.E. for jaw position sense. Adapted from reference [13].

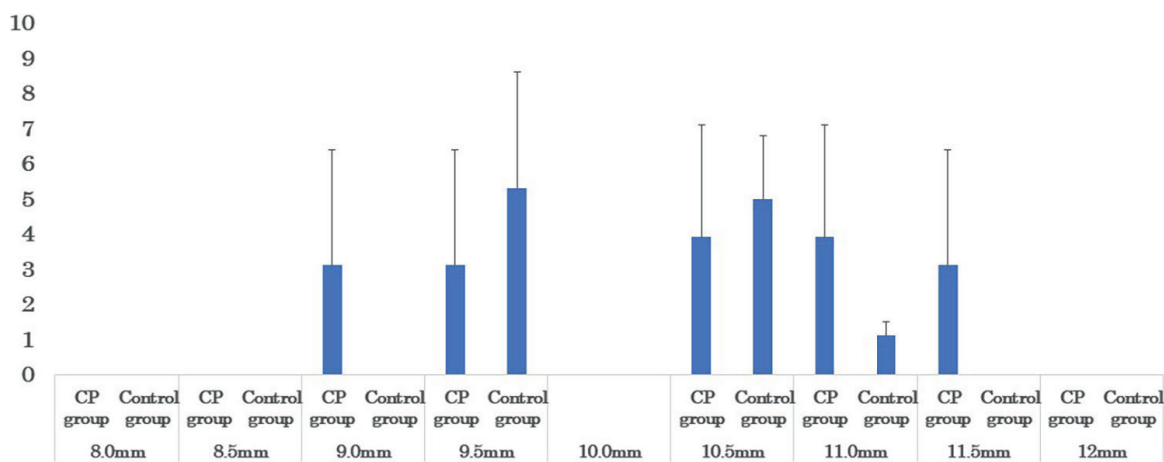


Figure 5. Comparison between CP and Control groups in R.M.E of jaw position sense after vibration stimulation Adapted from reference [13].

When the discrimination ability of patients with CP was compared before and after the stimulus application, it was found to be significantly higher after the stimulation than before the stimulation when the interincisal distance was smaller (9.5 mm) than that with the reference stick ($p < 0.05$).

Conversely, in healthy participants, it was significantly lower after the stimulus than before the stimulus when the interincisal distance was smaller (9.5 mm) than that with the reference stick ($p < 0.05$).

Generally, it is known that following the application of vibration stimulus to voluntary muscles including the masticatory muscle and these muscles exhibit tonic vibration reflex (TVR) in which the muscles slowly shrink [14].

Vibration stimulation increases the excitability of muscle spindles and increases the impulse of GIa afferent nerve fibers. As the next step, monosynaptically connected alpha motor neurons are excited, causing continuous muscle contraction.

Assuming that TVR was expressed in both CP and control groups in the experiment, the result may suggest that the effect of TVR-induced muscle sensation was different between the two groups.

It was noteworthy that the discrimination ability in patients with CP increased by the vibration stimulation. Recently, vibration therapy is increasingly used to reduce signs and symptoms associated with this developmental disability [15] These findings may support the effectiveness of these vibration therapies for orofacial muscles to improve oral functions in patients with CP.

3.3 Dental treatment and oral health care

Oral care and dental treatment are very important for patients with CP to maintain oral function. In dental clinics, patients with CP tend to accentuate involuntary muscle tonus in orofacial and other muscles when they must hold their jaw open, such as during teeth cleaning and dental treatment.

This abnormal muscle activity causes muscle fatigue and mental stress for patients.

Clinically, drug-induced sedation is used for patients with CP; however, no studies have examined its effects in detail. Therefore, to investigate how to control the muscle tonus, the authors investigated the effects of laughing gas (N₂O) inhalation sedation on orofacial muscle tonus using EMG as an index [16]. In this study, the mean frequency of orofacial muscle EMG discharge was measured with other sedative indexes,

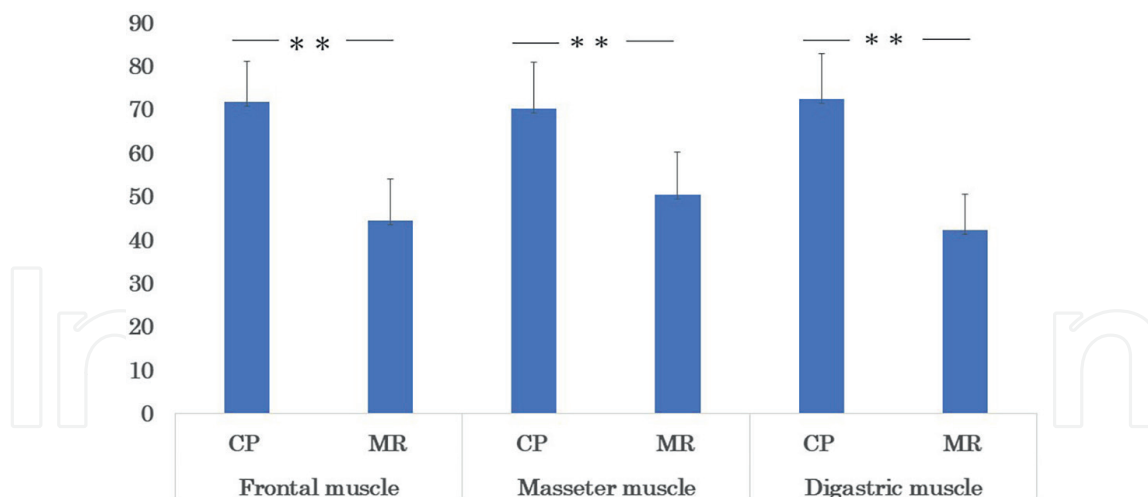


Figure 6. The comparison of reduction rate of mean frequency between CP and MR groups during inhalation of N₂O. Adapted from reference [14].

blood pressure, and heart rate. The study patients were 15 patients with CP and 15 with mental retardation (MR) as the control. By forcing the jaw opening for dental treatment, the enhanced level of the mean EMG frequency was higher in patients with CP than in those with MR. After N₂O inhalation, the mean frequency was significantly reduced in both patients with CP and MR.

Figure 6 shows the comparison of the reduction rate of mean frequency in patients with CP and MR. As shown in **Figure 7**, N₂O selectively suppressed the muscle tonus in patients with CP.

In general, γ motor neuron blocking with phenol is a commonly used method to suppress accentuation of the limb skeletal muscles in patients with CP.

Therefore, hyperactivities of γ motor neurons may be involved in the increased muscle tonus of the orofacial area in patients with CP. The presence of muscle spindles has also been confirmed in the masseter muscle. However, the present study demonstrated that N₂O also suppressed the muscle tonus in the frontalis and digastric muscles, which are thought to have no muscle spindles innervated by γ motor neurons.

Therefore, these results indicated the possibility that N₂O suppressed the functions of the upper central nervous system related to the accentuation of the muscle tonus.

4. Obstructive sleep apnea in achondroplasia children

Sleep-related breathing disorders (SRBDs) during childhood are known to cause growth failure because of disturbed sleep and growth hormone secretion rhythm [18].

Furthermore, sleep-disordered breathing has also been found to affect brain development [18].

There is strong evidence that childhood SRBDs are associated with behavioral and emotional regulation, scholastic performance, sustained attention, selective attention, and alertness deficits. Failing to treat SDB appears to leave children at risk for long-term neurobehavioral deficits. Thus, a research platform has been created to validate the diagnosis and treatment of sleep apnea during childhood.

Obstructive sleep apnea (OSA) is classified as one of the SRBDs occurring when there are recurrent episodes of upper airway collapse and obstruction during sleep associated with arousals with or without oxygen desaturations.

Achondroplasia (hereinafter referred to as AP) is an autosomal dominant genetic disease with an incidence rate of 1 per 10,000 people.

There are abnormalities such as growth failure [19]. OSA symptoms have been reported in children with AP [20]. However, the relationship between OSA symptoms and maxillofacial morphology in children with AP is not yet sufficiently clarified. Therefore, A fact-finding survey on OSA symptoms for achondroplasia was implemented [21], and factors of maxillofacial morphology that cause apnea were analyzed using cephalometric X-ray data [22].

4.1 Respiratory symptoms and oral findings

A questionnaire survey was performed on a total of 30 children with AP (AP group), comprising 20 preschool and 10 school-aged children. The control group consisted of healthy kindergarten, primary school, and junior high school children to compare the incidence of snoring, apnea, mouth breathing, and malocclusion. Data from the control group were also obtained from the results of a questionnaire survey of kindergarten, primary school, and junior high school children, a survey of physical growth of preschool children, and a survey by the Japanese Society of Pediatric Dentistry. Except for the height and weight at birth, children aged 1.5 and 3 years were significantly smaller than those in the control group, except for the birth weight in the AP group.

Table 2 shows the incidences of snoring, apnea, mouth breathing, and cross/open bite at both preschool and school ages in the AP and control groups, respectively.

Group	Symptoms	Groups	Incidences	Significant difference
Preschool	Snore	Achondroplasia	95	**
		Control	5.9	
	Apnea	Achondroplasia	45	**
		Control	1.9	
	Mouth breathing	Achondroplasia	85	**
		Control	0.5	
	Cross-open	Achondroplasia	65	**
		Control	9.1	
School	Snore	Achondroplasia	90	**
		Control	60.6	
	Apnea	Achondroplasia	20	**
		Control	2.7	
	Mouth breathing	Achondroplasia	90	**
		Control	27.4	
	Cross-open	Achondroplasia	60	**
		Control	9.2	

Adapted from reference [22].

Table 2.
Symptoms in children with achondroplasia.

At preschool age, the incidences of snoring, apnea, mouth breathing, and cross/open bite were significantly higher in the AP group than those in the control group, respectively ($P < 0.01$). At school age, the incidences of mouth breathing and cross/open bite in the AP group were significantly higher than those in the control group, respectively ($P < 0.01$). The incidence of snoring and apnea was higher than those in the control group, respectively ($P < 0.05$).

As a result of comparing the two groups, the AP group showed significantly higher incidences of snoring, apnea, mouth breathing, and reverse or open bite in infants and schoolchildren than those in the control group.

4.2 Craniofacial morphology (CFM)/airway morphology (AWM) in children with AP and healthy children

Figure 7 shows the measurement points of cephalometric photography.

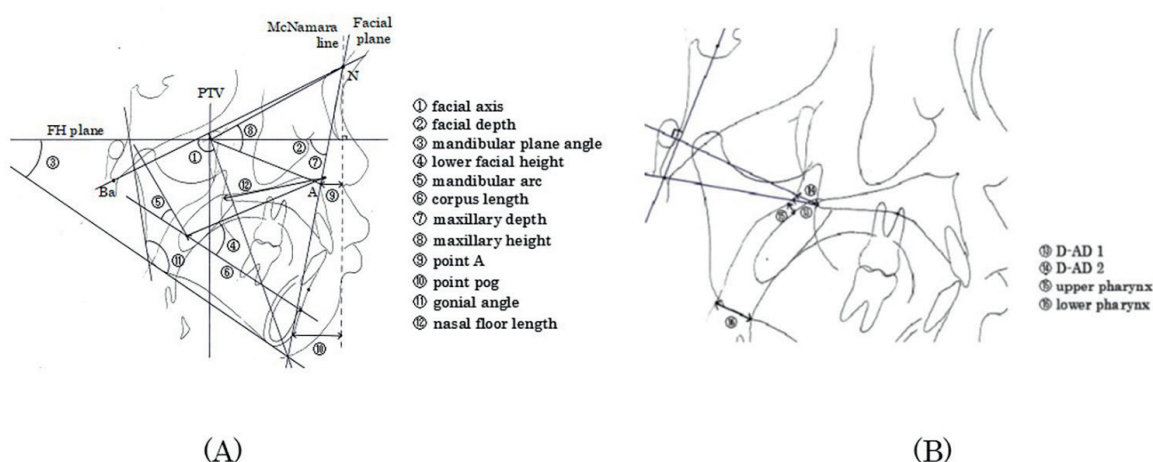


Figure 7. Measurement items (A) Maxillofacial morphology (B) Airway morphology. Adapted from reference [17].

Table 3 shows the comparison between children with AP and healthy groups in the measurement items of craniofacial/airway morphology.

In CFM, values for facial depth, nasal floor length, point A, point pog, and saddle angle were significantly lesser ($p < 0.01$), and those for mandibular plane angle and gonial angle were remarkably greater ($p < 0.01$) among AP group compared to the healthy. In AWM, D-AD1, D-AD2, and upper pharynx values were significantly lesser ($p < 0.05$ among AP group).

These findings have been reported as common features in the unique facial features of children and adults with OSA [23].

The AP group showed CFM/AWM that was characteristic of upper airway stenosis, a retruded chin position, and a greater mandibular plane angle because of partial early ossification of the cranial bones and a greater lower facial height because of a greater mandibular angle. This suggests that that AP group will frequently encounter sleep snoring and sleep apnea compared to the healthy.

4.3 Diagnosis and treatment

In general, pediatric patients with achondroplasia undergo surgical procedures such as adenoidectomy and tonsillectomy in the field of otorhinolaryngology.

		Achondroplasia		Healthy		p-value	
		Mean	S.D.	Mean	S.D.		
Craniofacial morphology	Significantly lower measurements	Facial depth	80.7	3.95	84.77	13.72	**
		Nasal floor length	43.03	3.37	48.2	1.97	
		Point A	-4.67	3.7	0.84	2.37	
		Point pog	-16.84	6.79	-8.62	3.31	
	Significantly higher measurements	Saddle angle	104.97	32.88	127.93	3.77	
		Mandibular plane angle	36.13	4.1	27.78	5.28	
		Gonial angle	135.61	6.8	126.27	7.45	
Airway morphology	Significantly lower measurements	D-AD1	8.3	3.43	17.08	4.34	**
		D-AD2	6.6	3.02	11.1	2.88	
		Upper pharynx	2.09	1.27	5.83	1.95	

Adapted from reference [17].

Table 3.
Morphological features of children with achondroplasia.

Sato et al. reported one case that adenoidectomy and tonsillectomy dilated the pharynx and improved the craniofacial and pharyngeal morphologies, apparently thus improving the sleep apnea [24].

A new diagnosis and treatment system for sleep apnea patients will be expected under the collaboration between pediatric dentistry and otolaryngology.

5. Conclusions

Clinical studies on developmental motor dysfunction are still limited. Further research developments that enable statistical review are warranted.

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Conflict of interest

The authors declare no conflict of interest.

Notes/thanks/other declarations

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