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REVIEW

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Orthodontic treatment in children to prevent sleep-disordered breathing in adulthood

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Abstract The purpose of this article is to review human craniofacial growth and development, especially the growth of the mandible, to clarify the relationship between obstructive sleep apnea (OSA) syndrome and craniofacial abnormality, and finally, to propose the hypothesis that negative pressure produced in the chest of the OSA child inhibits the growth of the mandible. Recently, the development of diagnosis and treatment of OSA syndrome has progressed rapidly; however, the prevention of OSA syndrome was merely seen. Craniofacial abnormality is reported as one of the causes of OSA syndrome. If craniofacial abnormality is determined only by genetics, it is difficult to manage the craniofacial skeleton to prevent OSA syndrome. The role of epigenetic factors on craniofacial growth and development is still controversial. However, if we stand on the functional matrix hypothesis, we can manage not only growth of the mandible but also the craniofacial skeleton as a whole. The author proposes the hypothesis that the negative pressure produced in the chest inhibits the growth of the mandible even if the patients have a capacity for growth and development; therefore, if this negative pressure disappears because of the removal of the tonsil and/or adenoids or by an orthodontic treatment to make a patency of the airway, the mandible may grow normally, and we can prevent or reduce a number of OSA syndromes in the future.

Keywords Obstructive sleep apnea · Craniofacial growth and development · Functional matrix hypothesis · Negative pressure in the chest · Growth of the mandible · Orthodontic treatment · Airway patency · Prevention

Introduction

In 1997, Dr. Donald H. Enlow (anatomist, author of *Handbook of Facial Growth* [1]) wrote this introduction for these articles of Dr. Melvin L. Moss [anatomist, advocator of functional matrix hypothesis (FMH)] [2–5]:

> *This series of four articles is a cohesive and constructive perspective of "where we are now after all the dust has settled." But, there is another important and I think key feature and that is a discussion of functional matrix-type studies (by deferent names, perhaps) in other biologic disciplines that otherwise we probably would be quite unaware of. This in itself is a most noteworthy contribution, because most of us, in both the basic and clinical orthodontic sciences, are really not aware of advances in other relevant fields. We can learn! Then, at the end, there is a look at the future, and this goes conceptually beyond anything we presume to understand today. In all, Dr. Moss's assessment of his own work as a revision is, I think, more of a scholarly elaboration, based on a broad quiltword of biologic understanding, now gleaned from a variety of other specialties...*

One point I would have liked Dr. Moss to have addressed in greater depth in the final pages is how the functional matrix is involved in its own growth and development on how it is controlled. That is how much genome and how do the provocative ideas of complexity and self-organization play into this?

Guilleminault and Pelayo [6] reported that the nasal obstruction and mouth breathing influence facial growth, which may further lead to difficulty in breathing while asleep. Clinical signs of sleep-disordered-breathing children include abnormal paradoxical inward motion of the chest.

There has been a lot of research on craniofacial growth and development; however, we still do not know how much genome and how the provocative ideas of complexity and self-organization play into this, as Dr. Enlow mentioned.

The purpose of this article is to summarize data and theories about the role of the functional matrix on craniofacial growth and development, to show the possibility that orthodontic treatment can affect the size of the airway, and to propose the hypothesis of mandibular growth.

Craniofacial growth and development

Essentially, the words growth and development have different meanings in biologic phenomenon. Watson and Lowrey [7] noted that growth is used mainly to describe the enlargement of a form, and development is used as the differentiation, the enlargement of the function and the complications. Todd [8] defined growth as the enlargement of size, and development is the process to maturity. However. growth and development have a relation with each other and are related to the same biologic phenomenon together—the authors use "growth and development" as one word.

Slavkin [9] noted that ideas, in a scientific sense, about the nature of growth and development appear to have started with Hippocrates in the fifth century B.C. Soon thereafter, Aristotle defined the basic question that would fuel this intellectual adventure for several millennia to follow (Aristotle, 1526). Aristotle asked, "do all parts of embryos come into existence at the same time, or do they appear in some sequence of events?" He asked whether everything is preformed and predetermined from the beginning, or is growth and development the resultant of many sequential processes that are integrated (somehow) into the forming organism? Aristotle rejected preformation and favored epigenesis!

Studies on the growth of the jaws and eruption of the dentition by Hunter [10] are widely credited as the first scientific research on craniofacial growth.

Heredity vs environmental factors on growth and development controversies

Controversies over the predetermining role of heredity and the effects of the environment on craniofacial growth and development have existed for more than 50 years. Do sutures of the craniofacial skeleton, mandibular condyle, and nasal septum work as the "growth center" on the craniofacial growth and development? There are numerous experimental studies to support each theory, such as the sutural theory by Weinmann and Sicher [11]. It suggests the sutural growth and development, and mandibular condyle has the ability of autonomy for craniofacial growth and development.

The nasal septum theory by Scott suggests that nasal septum growth and development is the pacemaker of craniofacial growth and development, especially of the midfacial area. The functional matrix theory by Moss purports that body functions (vision, listening, breathing, conversation, mastication, deglutition, digestion, etc.) affect the growth of the cartilage and bones as secondary changes.

The sutural theory

The sutural theory is identified most closely with the works of Weinmann and Sicher [11], two famous anatomists, whose textbooks on skeletal growth, *Bone and Bones*, became the standard textbook on the skeletal growth for orthopedics and dentistry.

According to the sutural theory, much like epiphyses of long bones, the connective tissue and cartilaginous joints of the craniofacial skeleton are the locations at which intrinsically regulated primary growth of bone take place. According to this view, growth of the cranial vault is caused by the intrinsic pattern of expansive proliferative growth by sutural connective tissue that forces the bones of the vault apart.

Importantly, for considerations of growth modification and dentofacial orthopedics, the sutural theory supported and reinforced the concept that growth of the face and jaws was essentially immutable. The sutures, as well as the cartilages of the craniofacial skeleton, were essentially the locations of centers of bone growth at which the inherited pattern of craniofacial form and facial type, however determined, were expressed, and the pattern cannot be changed [12].

An alternative analysis of early suture development and growth put forward by Scott and Pritchard et al. [13, 14] concluded that the osteogenic layers within the suture are actually continuations of the osteogenic layer periosteum and dura within the cranial vault and of the periosteum in facial sutures. As a result, according to these investigators, sutural growth should be considered as a specialized form of periosteal growth rather than akin to cartilaginous

growth. Finally, several experimental studies involving vital dyes [15] and surgical manipulation [16, 17]of cranial sutures in appropriate animal models clearly indicated that although sutures were major sites of craniofacial skeletal growth, they played no determining role in that growth.

Each of these lines of evidence led many investigators to question the validity of the sutural theory.

The nasal septum theory

Scott proposed the nasal septum theory as the single and unified theory of craniofacial growth [18–20]. Scott assumed that the primary controlling factors in craniofacial growth and development are found only in the cartilage and the periosteum, and that the sutures are secondary and passive. He viewed the cartilaginous sites throughout the skull as primary centers for growth. Sutural growth may be further altered by local and environmental factors. According to the nasal septum theory, sutures play little or no direct role in the growth of the craniofacial skeleton. The essential primary elements directing craniofacial skeletal growth are the cartilages found within the cranial base and, in particular, the anterior extension of the chondrocranium, the nasal septal cartilage. Scott concluded that the nasal septum is most active and important for craniofacial skeletal growth prenatally and postnatally. During that time, the anterior–inferior expansive growth of the nasal septal cartilage drives the midface downward and forward. The cranial base synchondroses were thought to have a longer-lasting effect on craniofacial growth. Finally, Scott asserted that the cartilages of the mandibular condyles behave similarly to cranial base and nasal septal cartilages and directly determine the growth of the mandible as they "push" the mandible downward and forward.

However, Koski [21] reported that there were qualitative differences between the epiphyseal cartilage, articular cartilage, condylar cartilage, and synchondrosis when they transplanted those cartilages either subcutaneously or intracerebrally to see the existence of the autonomy for craniofacial growth and development. Koski [21] noted that the most interesting differing features would seem to be the cell type of the proliferating layer, the rapid transformation of the cartilage cells into hypertrophied cells, and the growth-promoting potential of the condylar and synchondrosal cartilages, when transplanted in the subcutaneous tissue, appeared to be clearly less than that of the epiphyseal end-cartilage of a long bone.

The functional matrix theory

Moss [22] noted the following:

> *It is now established beyond doubt that the mandibular condylar cartilages are not primary sites of mandibular growth. They are the loci at which secondary, compensatory periosteal growth occurs. The bilateral removal of mandibular condylar cartilages in growing experimental animals, as in man, does not inhibit either the spatial translation of the now acondylar complex of contiguous mandibular functional cranial component*

Also, he showed the cephalometric superimposition for 7 years of the mandibular growth of a patient subsequent to complete condylectomy (Fig. 1).

Enlow [23] noted that Moss had presented his idea of the functional matrix, which hypothesize that the growth of the skull is quite secondary, as being determined largely by the growth and function of functional matrices. Moss's ideas are based on the theory of functional cranial components originated by Van der Klaauw. According to Van der Klaauw [24], the skull is made up of units the size, shape, and position of which are determined primarily by their functions. Moss's functional matrix refers to adjacent structures related to the presence and functions of Van der Klaauw's functional components. Moss asserts that the growth of the functional component, irrespective of their ossification mechanism, is entirely dependent on the growth and function of the functional matrices. Moss denies any intrinsic regulatory control in the growing bony tissues themselves. Control of the bone growth is by either local epigenetic factors or, additionally, environmental factors.

Abnormal pattern of craniofacial growth

Enlow [25] noted that the basic underlying causes of the severe congenital types of dysplasias are virtually unknown at present. However, severe congenital types of dysplasias tend to have obstructive sleep apnea (OSA); it is reported that 45% of people with Down's syndrome had OSA [26].

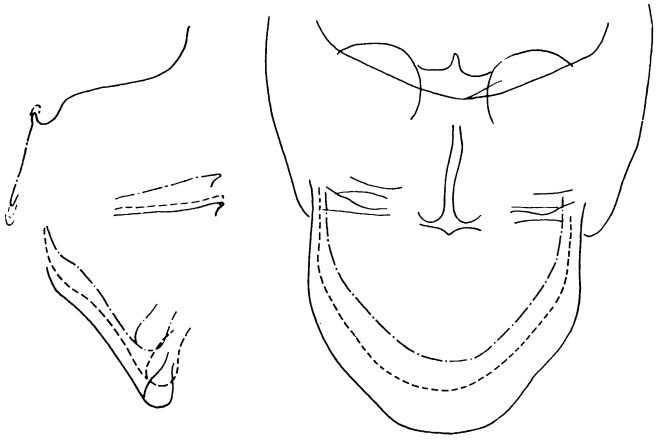

Fig. 1 Cephalometric superimposition of a patient subsequent to complete bilateral condylectomy revealed the mandibular growth for 7 years (adapted from Moss and Salentijn [22])

Enlow [25] also noted the following:

(1) Craniostenosis is such a condition. It is the premature closure of cranio vault sutures.
(2) Scaphocephaly results from early closure of the sagittal suture of the scull roof, and a long, narrow calvaria is produced with a prominence of the frontal and occipital areas. The cranial base may also be affected, and this then can be passed on to cause deformities of the face. During its development, the coronal sutures may also close (brachycephaly) prior to the completion of brain growth.
(3) Pierre Robin syndrome is a relatively rare condition that involves severe mandibular hypoplasia and a cleft palate (not lip).
(4) In Crouson's disease, the eyes bulging as a consequence of premature synostosis and a half-open mouth are all present.
(5) Apert's syndrome is a congenital malformation in which the top of the head is pointed (acrocephaly) in conjunction with early fusion of the coronal and sagittal sutures.

Many of the severe congenital types of dysplasias have the problem of genes, like trisomy21 of Down's syndrome. However, closure (fusion) of the suture and cleft palate may be the key words for understanding why patients of one syndrome look alike rather than their brothers or sisters.

Growth of the mandible

Ferraro [27] stated that the craniofacial structure has a cephalocaudal gradient of growth. The midface has more postnatal growth potential than the cranium, and the mandible has more growth potential than the midface.

Enlow [23] notes that there is more variability in mandibular growth than maxillary growth since the mandibular relationship with the neurocranium is not as intimate as the maxillary relationship.

Nezu and Nagata [28] reported that three fourths of Japanese maxillary protrusion cases were of the mandibular retrusion variety. This diagnosis was made from the reference line of McNamara to the point pogonion [McNamara line is the line perpendicular to Frankfurt Horizontal (FH, Po-Or) line from Nasion].

Thus, in this chapter, we will focus on the growth of the mandible.

Growth of the mandible (Enlow's ground bone sections)

Enlow has developed and used extensively methods for studying bone deposition, resorption, and the process of remodeling in prepared ground sections (Fig. 2). Enlow showed the method of studying remodeling from ground bone sections as follows [29].

The sequence of remodeling changes that produced the cortical arrangement seen in photomicrograph A is shown schematically in B, C, and D. Prior to the lateral drift, as seen in stage B, the cortex is composed of inner (endosteal) and outer (periosteal) zones. Simultaneously, new bone is added at surface 1, removed from side 2, added to surface 3, and resorbed on side 4 as shown in C. The composite result is a drift movement of this entire region of the bone in the direction indicated by the arrows in C. The final stage schematized in D is comparable with the actual photomicrograph shown in A (Fig. 2).

Thus, Enlow found not only typical growth of the craniofacial skeleton but also of the mandible. Enlow and Harris [30] noted that growth directions of the mandible involving periosteal resorption are indicated by arrows pointing into the bone surface, and growth directions involving periosteal deposition are represented by arrows

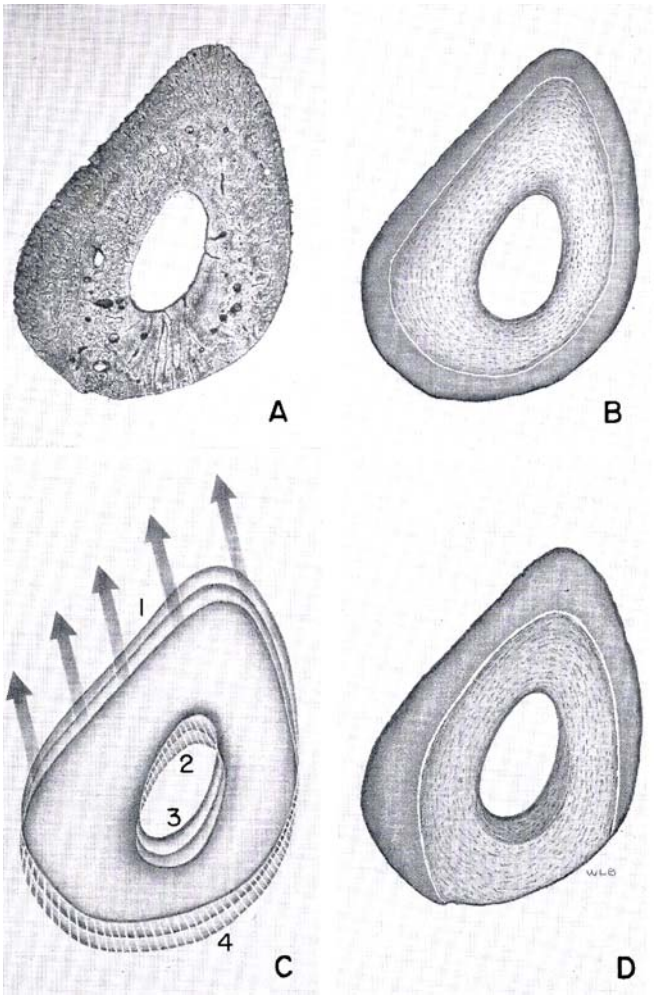

Fig. 2 Enlow's method of studying remodeling from ground bone sections (adapted from Enlow [29])

pointing out of the bone surface (Fig. 3). Enlow pointed out the average growth directions of the mandible.

Growth of the mandible (Bjork's implant)

Bjork [31] has devised an ingenious method of implanting tiny bits of tantalum into growing bone of animals and human beings (Fig. 4). These serve as reference markers during serial cephalometric analysis. The method allows precise orientation of the serial cephalograms and provides information concerning the amount and site of bone growth. Bjork found out that the growth of the mandible occurred mainly in the condyle and ramus region, like Enlow's findings; however, Bjork and Skieller [32] found out that an individual has different growth directions of the mandible (Fig. 5). We have to think about why individuals have different growth patterns.

Craniofacial pattern of OSA

The craniofacial characteristics of OSA patients have been reported as follows:

(1) Short anterior cranial base [33–35, 43]
(2) Less obtuse cranial base flexture angle [36, 37]
(3) Retropositioned mandible [33, 36, 38–41, 43]
(4) Small mandible [34, 35, 41, 42]
(5) Small maxilla [34, 35, 43]
(6) Steep mandibular plane [38, 44]
(7) Long soft palate [33, 36, 40, 45]
(8) Decreased airway space [34, 36, 38, 40, 44]

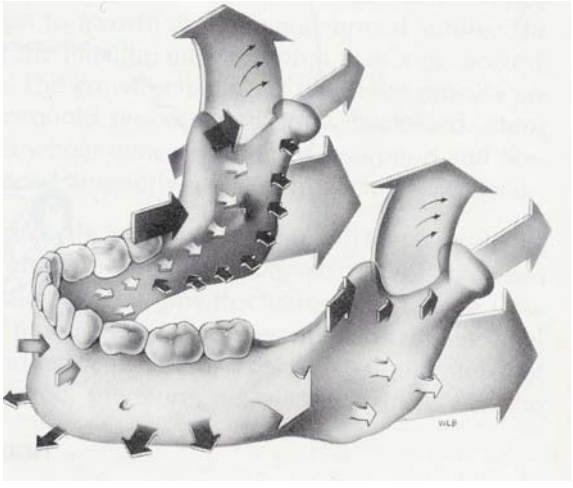

Fig. 3 Summary diagram of the growth of the mandible. Growth directions involving periosteal resorption are indicated by *arrows* pointing into the bone surface, and growth directions involving periosteal deposition are represented by *arrows* pointing out the bone surface (adapted from Enlow and Harris [30])

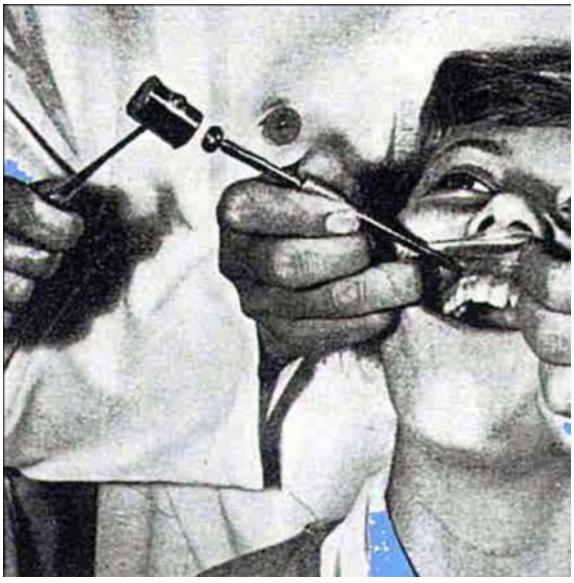

Fig. 4 Bjork has devised an ingenious method of implanting tiny bits of tantalum into growing bone of human beings (adapted from Bjork [31])

(9) Lowered position of hyoid bone [36, 40, 43, 44, 46]
(10)Increased anterior facial height [33, 38, 40, 43, 44].

We [40] reported that the craniofacial pattern of OSA patients was a Dolico facial pattern (increased anterior facial height) by the cephalometric analysis of Ricketts, and OSA patients have retropositioned mandible, long soft

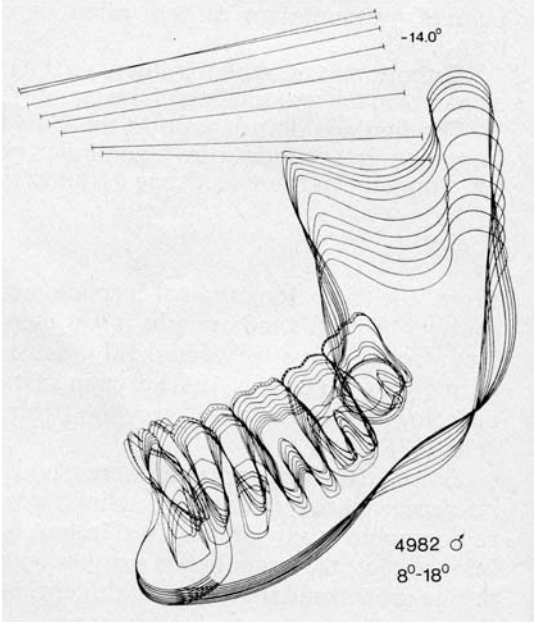

Fig. 5 Bjork found out that individual has different growth directions of the mandible (adapted from Bjork and Skieller [32])

palate, decreased airway space, and lowered position of the hyoid bone. However, we [47] analyzed cephalograms of OSA patients and controls by Ricketts analysis and Downs–Northwestern analysis. We found out through Ricketts analysis that OSA patients had a Dolico facial pattern (increased anterior facial height) and a tendency to have a retropositioned mandible, long soft palate, decreased airway space, and lowered position of the hyoid bone. However, using Downs–Northwestern analysis, we did not find any significant differences between OSA patients and controls analyzing the same cephalograms of Ricketts analysis. The reason why the difference in the result happened is the difference in the analysis and reference points.

Ricketts [48] noted that point basion (Ba) was selected as reference for the occipital bone. Pterygoid (Pt) point represented the central point on the sphenoid bone. Nasion (N), on the frontonasal suture (on the frontal or superior side), was chosen as a point to separate the facial bones from the calvaria. Historically, in roentgenographic cephalometry, the center of sella turcica (point S) has been employed, possibly due to the ease of observation in the lateral film. It was considered a biologic point because it represented the pituitary gland. A line from S to N was employed as the anterior cranial base (SN) and became the most commonly accepted reference plane with point sella registered. Sella was particularly questioned when it was recognized that the anterior portion of the face moved forward with growth from the coronal suture complex which crosses that line representing the anterior cranial base. Computer analysis showed that it did not behave in an organized order in a polar phenomenon in the facial growth constellation, and therefore, SN can be misleading in facial typing as well as longitudinal description. Therefore, Ricketts gave up using sella as a reference point and started to select Pt, N, and Ba points to analyze the cephalograms.

Analyzing the cephalograms by the Ricketts method to clarify the characteristics of craniofacial skeleton of OSA syndrome should be recommended.

Craniofacial pattern of child OSA

We [40, 49] reported that the craniofacial pattern of child OSA was intermediate of adult OSA and adult control (Table 1). All the data showed intermediate values from the adult control to adult OSA, which suggest that the continuation of OSA makes the craniofacial pattern worsen year by year (Table 1).

Functional matrix hypothesis (FMH)

Moss developed the functional cranial analysis from the philosophy of functional cranial component of van der Klaauw by thinking of what the growth is, how they grow, and how can craniofacial growth and development be measured [23].

Table 1 Cephalometric variables

	Control (adult) (n=26)	Patient (child) (n=29)	OSAs (adult) (n=31)
FX (°)	84.3±4.8	81.8±3.1	79.8±4.7
FD (°)	87.3±3.4	84.8±3.2	86.4±4.3
MP (°)	27.6±5.7	31.9±5.6	29.4±7.0
LFH (°)	50.8±4.7	54.6±5.3	55.7±5.4
MA (°)	27.7±5.1	21.2±4.8	24.3±5.8
TFH (°)	64.8±5.2	68.4±4.5	69.8±6.3

FX Facial axis angle, BA-CC-GN; *FD* facial depth angle, crossing of facial plane to facial height; *MP* mandibular plane angle, crossing of mandibular plane to facial height; *LFH* lower facial height angle, ANS-XI-PM; *MA* mandibular arc angle, crossing of condylar axis to corpus axis; *TFH* total facial height angle, crossing of corpus axis to cranial base plane (adapted from Kikuchi et al. [40, 49])

Moss noted that a decade's study of the regulatory roles of intrinsic (genomic) and extrinsic (epigenetic) factors in cephalic growth evolved into the FMH [50], and also noted that the principal FMH concepts are either generally known or easily available [22, 50–57].

The functional matrix theory can be explained as follows:

There are many relatively independent functions at the craniofacial skeleton. Functions (visions, smell, listen, breath, conversation, mastication, deglutition, digestion, etc.) need the proper soft tissues to accomplish the function, which need the proper skeletal elements to support and protect the soft tissues. Therefore, soft tissue means not only muscles and tendons but also glands, nerves, vessels, teeth, and sinuses. One set of soft tissue and skeletal element relating the one function is called functional cranial component. Total skeletal element is skeletal unit, and total soft tissue is functional matrix. Each skeletal unit exists relatively independently because the function of the soft tissue is different. The generation, growth, and maintenance of one skeletal unit depend upon the amount and quality of the functional matrix related to the skeletal unit. For instance, the primary growth-promoting potential to the skeletal unit is the functional matrix, and the bone grows as a secondary reaction.

There are two types of functional matrix: one is the periosteal matrix, and the other is the capsular matrix. Muscle is a good example of the periosteal matrix. For instance, the size of the coronoid process (microskeletal unit) is dependent on the strength of the temporalis muscle (periosteal matrix). The brain is a good example of the capsular matrix. The size of the cranium (macroskeletal unit) is dependent on the size of the brain.

The periosteal matrix works on the bone directly, like bone resorption and deposition, and the capsular matrix works on the bone indirectly. This is the difference between the periosteal and capsular matrices.

For example, the mandible is a combination of relatively independent functional cranial components each composed of a functional matrix and a skeletal unit.

The relationships between functional matrix and skeletal unit of the mandible are the temporalis muscle vs coronoid process, masseter and internal pterygoid muscles vs gonial angle, teeth vs alveolar bone, condylar cartilage vs condyle, and the mandibular nerve and vessels vs the basal bone of the mandible. These skeletal units are called microskeletal units (Fig. 6) [58].

Kikuchi et al. [59] reported that experimental work to examine if there are functional matrix relations between masseter muscle and gonial angle region. Kikuchi cut the masseteric nerve only on the right side of the rats. The result supported the functional matrix hypothesis that the size of gonial angle region of the denervated side was reduced (Fig. 7).

Normal bone growth, as well as the maintenance of osseous form, is primarily a reflection of the mechanical requirements of the matrix. Briefly, the associated fascial viscera, ligaments, and muscles of the temporomandibular joint are a matrix to carry out the function of mastication. The condylar process exists only as a skeletal unit supporting this functional matrix. The growth of any skeletal unit is secondary and adaptive to change in the functional matrix.

Orthodontic treatment and airway

Moyers [60] noted that orthodontic therapy is directed to abnormal occlusion of the teeth, growth of the complex of craniofacial bones, and function of the orofacial neuromusculature, which alone or in combination may cause any of the following: (a) impaired mastication, (b) unfortunate facial aesthetics, (c) dysfunction of the temporomandibular articulation, (d) susceptibility to periodontal disease, (e) susceptibility to dental caries, and (f) impaired speech due to malpositions of the teeth.

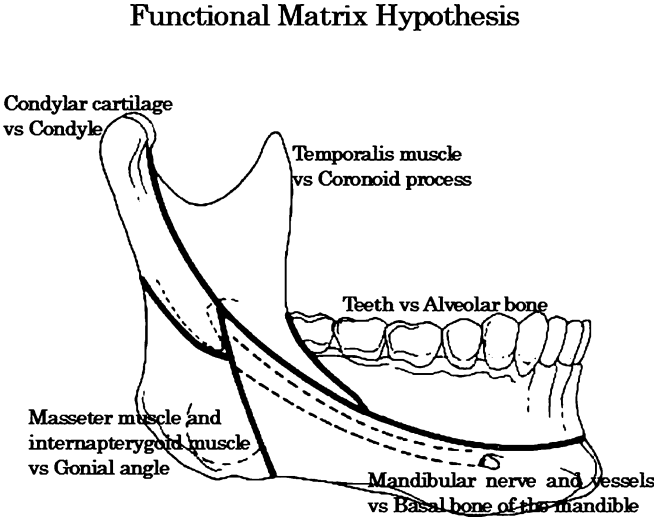

Fig. 6 The relationship between functional matrix and skeletal unit of the mandible are temporalis muscle vs coronoid process, mass masseter and internal pterygoid muscles vs gonial angle, teeth vs alveolar bone, condylar cartilage vs condyle, and mandibular nerve and vessels vs basal bone of the mandible (adapted from Hanada and Ohyama [58])

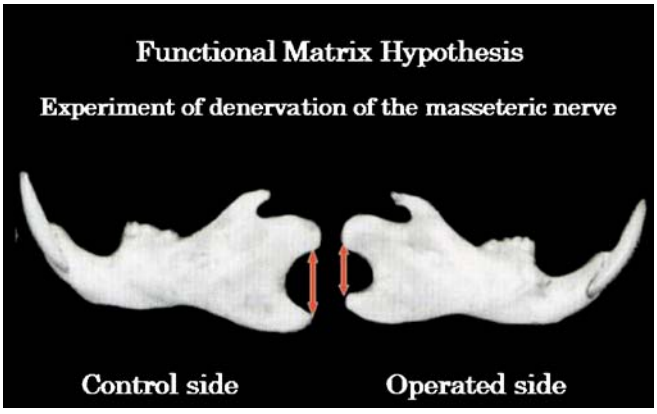

Fig. 7 Result of experimental work after cutting the masseteric nerve only on the right side of the rats. Size of gonial angle region of the denervated side was reduced (adapted from Kikuchi et al. [59])

However, many of the orthodontic patients came to our office for the purpose of getting a good profile. Because of that, the orthodontist tends to treat the patients only for the purpose of aesthetics, and the diagnostic system tends to change to apply only to the patients' need. Consequently the most important missing diagnosis is the airway. Nevertheless, breathing is the most important action for human beings to live; we forgot the airway to make a diagnosis of the orthodontic patients. If we stop our breathing for 10 min, many of us would surely be dead. For that purpose, we have to build in the airway an analysis to make the orthodontic diagnosis. I would like to show you the orthodontic cases, which suggested that orthodontic treatment influenced the size and the function of the airway.

Cases 1 and 2

These patients were 12 years and 11 months and 11 years and 9 months sisters. Therefore, they still have a growth possibility. They complained about their maxillary protrusion. The elder sister was treated by extraction of five teeth because she had one congenitally missing tooth and one root-resorped tooth. However, the younger sister was treated by nonextraction with Herbst appliance. Treatment term was 3 years and 11 months for the elder sister and 3 years and 2 months for the younger sister. Both sisters had a typical muscle strain on the chin when they closed their lips together (Fig. 8).

When we superimposed the cephalograms at the baseline of these sisters, there were a few differences between the sisters before treatment, like the nose of the elder sister

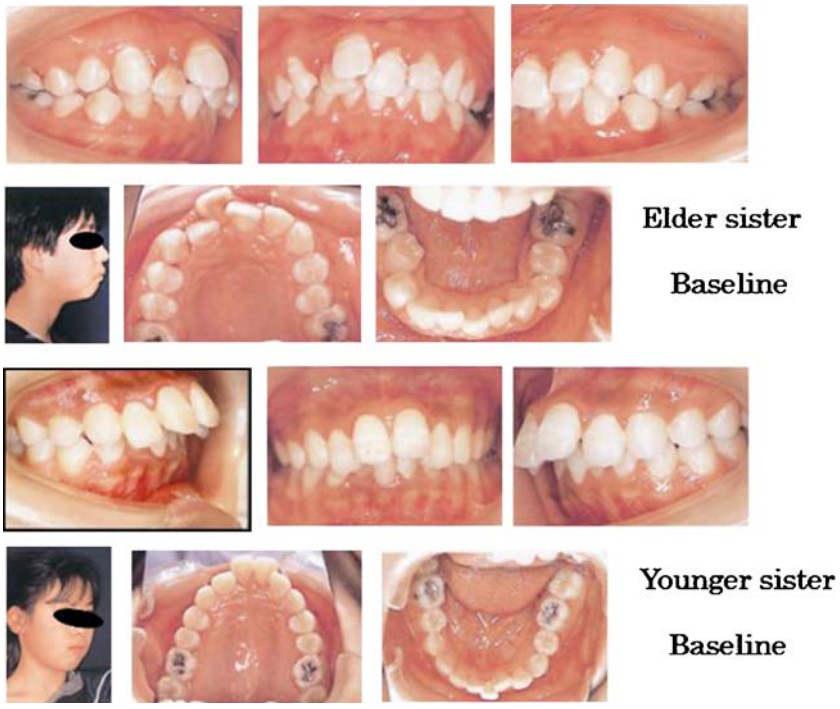

Fig. 8 Cases 1 and 2, oral and facial photos of the baseline. Chief complaint was maxillary protrusion. Note the typical muscle strain on the chin

was bigger than that of the younger sister. However, the superimposition of the cephalograms of both cases revealed that these sisters were almost the same. Please pay attention that their lower pharynxes were of the same width at the baseline (Fig. 9).

As a result of the orthodontic treatment of the sisters, the muscle strain on the chin disappeared. Both sisters looked better and had a satisfactory result. There was little difference between the sisters from their appearance (Fig. 10).

However, when we superimposed the cephalograms of the baseline and the result of the elder sister, she grew only

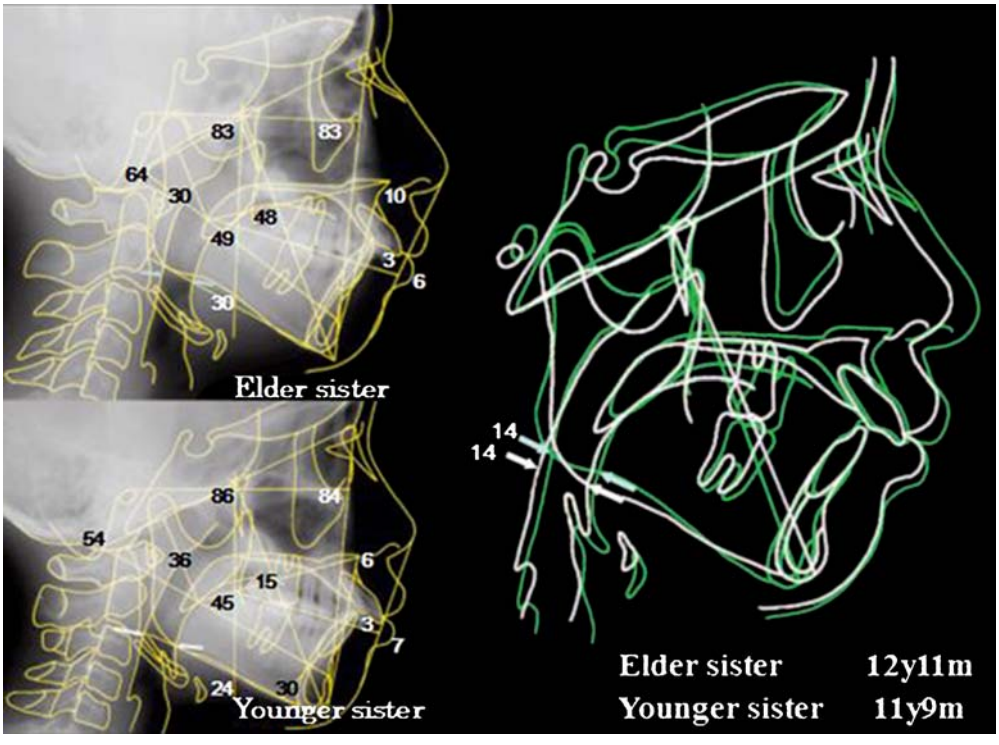

Fig. 9 Cases 1 and 2, superimposition of the cephalograms at the baseline of these sisters. These sisters have almost the same craniofacial skeletons. Note that their lower pharynxes (airway size) were the same width at the baseline

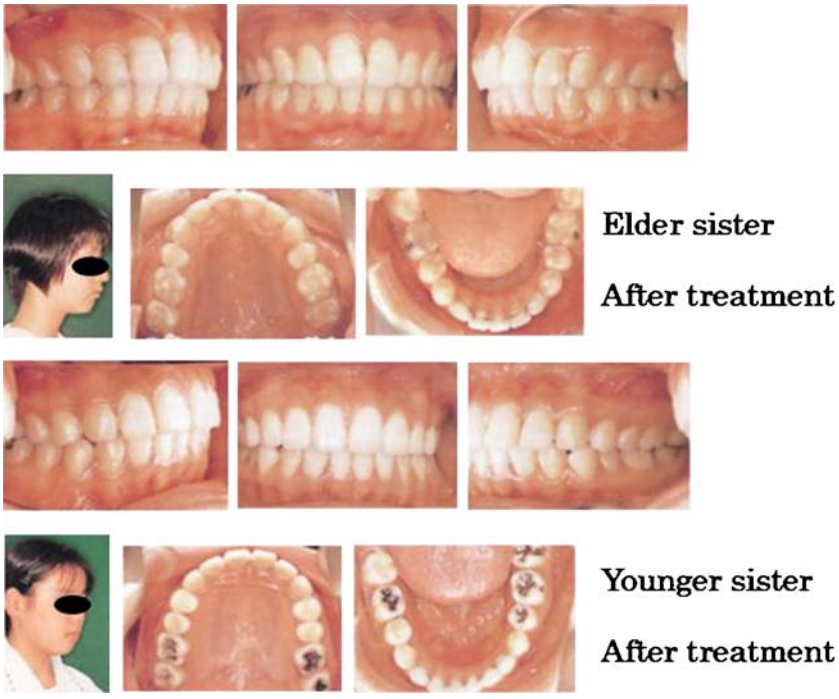

Fig. 10 Cases 1 and 2, oral and facial photos after the treatment. Chief complaints disappeared, and the typical muscle strain on the chin was gone. They were satisfied with the result, and there was little difference between the sisters from their appearance

at the nose, and the other part did not grow at all, and the width of the lower pharynx was reduced from 14 to 10 mm. When we superimposed the cephalograms of the baseline and the result of the younger sister, the mandible of the younger sister grew a lot. As a consequence, the width of the lower pharynx was increased to 17 from 14 mm. The lower pharynx size of the sisters were the same before the treatment; however, total difference in the lower pharynxes on the cephalograms became 7 mm after the treatment (Fig. 11).

The result of the treatment looks almost the same from the appearance; however, there were big differences between the sisters inside the face that was the most important structure for human beings: the size of the airway. Orthodontic treatment may influence the size of the airway. However, we do not have clear evidence at this moment yet. We have to collect our data as soon as possible.

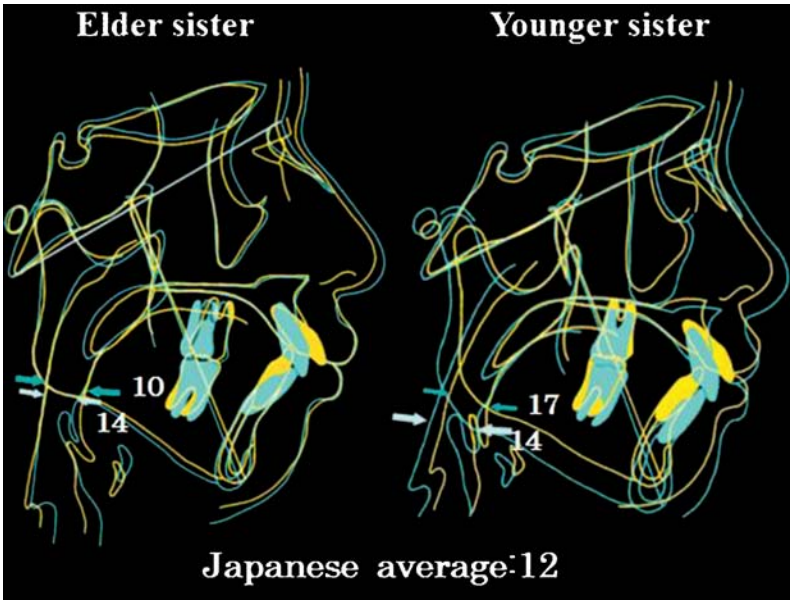

Fig. 11 Cases 1 and 2, superimposition of the cephalograms of the baseline and the result. Mandible of elder sister did not grow at all. Lower pharynx of elder sister was reduced to 10 from 14 mm (*left*). Mandible of younger sister grew a lot. As a consequence, the width of the lower pharynx was increased to 17 from 14 mm (*right*). Total difference became 7 mm after the treatment

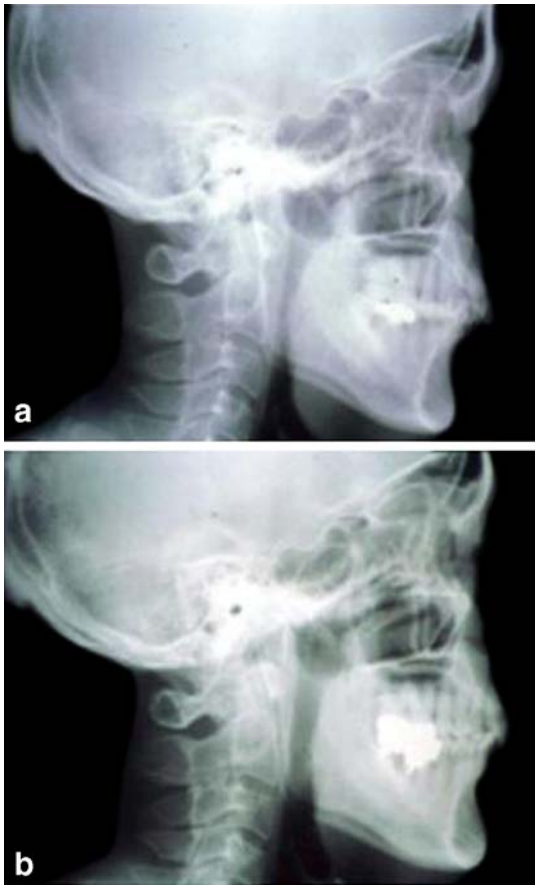

Fig. 12 Case 3, cephalograms of the baseline (**a**) and the result (**b**). Note the differences of the airway (adapted from Miyao et al. [61])

Case 3 (courtesy of Dr. Miyao)

Chief complaints—daytime sleepiness, snoring, and sleep apnea.

Gender, male; Age, 51; and body mass index (BMI), 21.2.

Medical history: He got the jaw injury at 22 years of age, and his jaw was misshapened. He started snoring loudly at the age of 45 and witnessed apnea at the age of 49. He was diagnosed with OSA [apnea–hypopnea index (AHI) 26.3] based on polysomnography (PSG).

Orthodontic diagnosis: Angle's classification class 2, division 1 (a protruded maxilla in reference to the mandible associated with flared maxillary incisors) (Fig. 12).

Orthodontic treatment: Multi-bracket treatment, conducted to expand the arches' distance between the left and right molars by 9 mm, and overjet and overbite were improved. Consequently, the oral space was enlarged, and the tongue and the mandible were moved anteriorly. The AHI according to PSG changed from 26.3 to 9.3/h. Patient's data from cephalograms (Fig. 12) and polysomnography at the baseline and after the orthodontic treatment are listed on Table 2.

Table 2 Results of pre- and post-treatment cephalometric analyses, PSG, and BMI

Cephalometric analyses	Control	Pre-treatment	Post-treatment
SNA (°)	81.4±2.9	87.1	88.3
SNB (°)	78.4±3.0	79.6	82.5
ANB (°)	3.0±1.0	7.5	5.8
Overjet (mm)	3.2±1.2	10.2	4.5
Overbite (mm)	1.5±1.1	9.9	5
PNS-P (mm)	39.2±3.1	47	47
MPT (mm)	11.2±1.6	10	11
Mp-H (mm)	14.0±5.4	27	23
SPAS (mm)	14.7±2.0	4	7
PAS (mm)	16.1±2.3	5	12
Distance between the left and the right lower molar (dental model, mm)		27	36
PSG			
Arousal index(/h)		45.5	38.3
REM (%)		19.3	20.9
Stage 1 (%)		43.8	21.7
Stage 2 (%)		36.8	47.7
Stages 3 + 4 (%)		0	9.7
Lowest SpO_2 (%)		73	91
ODI (/h)		15.3	5.4
BMI (kg/m^2)		21.1	21.7

SNA Angle of protrusion of the superior alveolar base, *SNB* angle of protrusion of the inferior alveolar base, *ANB* difference in the angle of protrusion between the superior and inferior alveolar bases, *SPAS* thickness of the airway behind the soft palate along a line parallel to the Go-B point plane, *PAS* thickness of the airway along a line extending through the Go-B point plane through P (adapted from Miyao et al. [61])

We may change the airway patency by the orthodontic

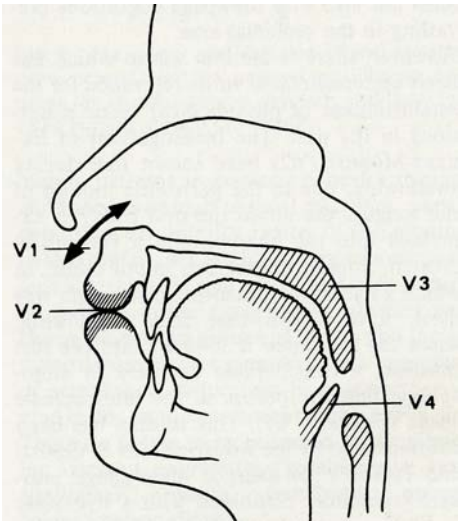

Fig. 13 There are four valves in the dentofacial skeleton such as V1 nasal valve, V2 lip valve (so-called lip seal), V3 tongue and soft palate valve (so-called oral seal), and V4 epiglottis valve (adapted from Frankel and Frankel [62])

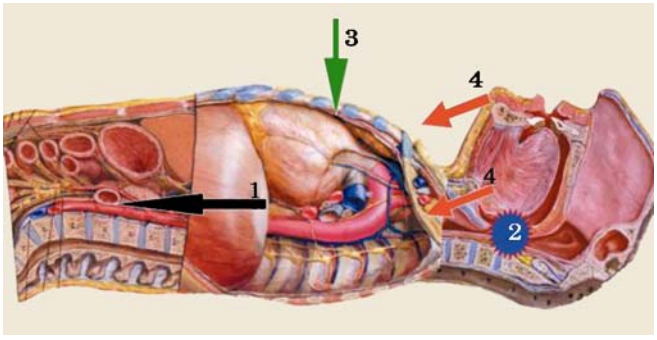

Fig. 14 Negative pressure produced in the chest inhibits growth of the mandible. *1* indicates diaphragm moves downward to inhale; *2*, obstruction at pharyngeal region; *3*, negative pressure produced in the chest (esophagus); and *4*, this negative pressure pulls the tongue and mandible downward and backward

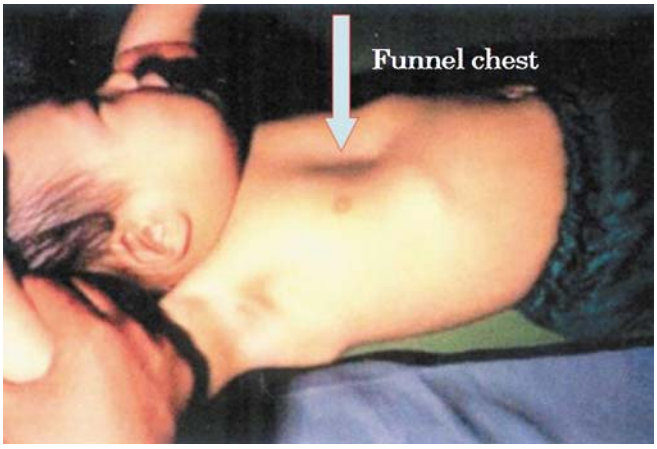

Fig. 15 Negative pressure produced in the chest may cause a funnel chest (courtesy of Prof. S. Miyazaki http://www.hyssa.com/shiga/shiga/)

treatment even if the patient is an adult, like in this case. Miyao et al. concluded that some patients can expect a cure or diminishment of OSAs with orthodontic treatment [61].

Importance of the oral seal to breath

Frankel and Frankel [62] noted that the impact of the "space problem" in the physiology of the orofacial complex has been emphasized by Moss [54, 56]. He maintains that a proper functioning of either the digestive or the respiratory systems depends upon a functionally adequate patency of the oral and the nasopharyngeal passageways. This assumption receives significant support from the work of Bosma [63], who emphasized the impact of adequate space conditions in the physiology of the orofacial complex, with particular emphasis on airway maintenance. The life-sustaining functions of breathing, as well as the intake and transport of food, are dependent on the adequate patency of these spaces and the proper functioning of the various valves, as seen in Fig. 13.

Frankel emphasized that there are four valves in the dentofacial skeleton, such as the V1 nasal valve, V2 lip valve (the so-called lip seal), V3 tongue and soft palate valve (the so-called oral seal), and V4 epiglottis valve, and we can maintain physiologic atmospheric pressure in the oral and nasopharyngeal spaces in the presence of a competent oral seal. Frankel emphasized that the life-sustaining functions of breathing as well as the intake and transport of food are dependent on the adequate patency of these spaces and the proper functioning of these valves.

Frankel also noted that Bosma [63] has emphasized that the posterior soft tissue barrier formed by the soft palate and the tongue is an important factor for airway maintenance. The positional stabilization of the pharyngeal airway is the initial manifestation of the distinctive coordination of posture. The physiological relevance of the infantile performance of stabilization about the airway is identified by its impairment in Pierre Robin syndrome. When these afflicted children are lying on their backs, the apposition of the tongue to the palate cannot take place because of the hypoplasia of the dorsal portion of the palate mostly associated with cleft. The infantile mechanism of pharyngeal airway maintenance fails to operate, and the tongue falls downward and backward, occluding the pharynx. In a prone position, the tongue approximates the palate by the force of gravity, permitting nasal portal respiration. Bosma holds that mandibular retrusion in Pierre Robin syndrome may be interpreted as an appropriate component of a regional failure of a positional function, which emphasizes the physiological relevance of the muscular barrier separating the oral cavity from the pharyngeal space.

Negative pressure produced in the chest inhibits growth of the mandible

I would like to propose the hypothesis that the mechanism of thoracic-negative pressure inhibits the growth of the mandible backward and downward.

When the obstruction occurs to the patient at the pharyngeal area, the patient wants to move down the diaphragm to intake the air; accordingly, the negative pressure is produced in the chest. We can check the negative pressure produced in the chest of the OSA patient by the esopharyngeal pressure monitor. We can see the deformity of the chest when the patient inhales, which is opposite the manner of breathing. Mechanism of the negative pressure inhibits the growth of the mandible as follows (Fig. 14):

(1) Diaphragm moves downward to inhale,
(2) Obstruction at pharyngeal region,
(3) Negative pressure produced in the chest (esophagus), and

(4) This negative pressure pulls the tongue and mandible downward and backward.

If we stand on this hypothesis and the functional matrix hypothesis, it is easier to interpret why an OSA patient tends to have the Dolico facial pattern (long face) and lower-positioned hyoid bone, longer soft palate, and narrow airway. These may be the result of the negative pressure in the chest. This negative pressure may cause a funnel chest (Fig. 15). Castiglione et al. [64] reported that 82% of 23 children affected by chronic upper-airway obstruction were pectus excavatum, and 82% were enlarged tonsils and adenoids.

If this negative pressure disappears because of the removal of the tonsil or adenoids, or by the orthodontic treatment to make a patency of the airway, the mandible may grow normally, and we can prevent or reduce a number of sleep apneas in the future.

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