

Cephalometric Analysis of the Velopharyngeal Muscular Triangle as a Possible Prognostic Factor for Velopharyngeal Closure in Submucous Cleft Palate

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Abstract

Objective: To elucidate the anatomical characteristics of submucous cleft palate (SMCP), we analyzed the velopharyngeal (VP) structures focusing on the relationship between the position of posterior pharyngeal wall (PPW) in the VP muscles and VP closure acquisition in SMCP patients.

Methods: Cranial landmarks for cephalometric analysis were identified in a study of two cadavers, and the area of the velopharyngeal muscular triangle (VPM-triangle), which was bordered by the origin of the levator veli palatini muscle, the origin of the palatopharyngeal muscle, and the insertion of both muscles, was defined. We then cephalometrically measured the VP structures of 14 SMCP patients (SMCP group) and the position of the PPW within the VPM-triangle. As a comparison group, 20 healthy Japanese children (control group) and 20 patients who underwent palatal repair for cleft palate (postoperative CP group) were enrolled. In addition, we analyzed the correlation between VP closure and position of the PPW within the VPM-triangle in the SMCP group.

Results: The SMCP group exhibited shorter velar length, greater pharyngeal depth and greater height. In the control and postoperative CP groups, the part of the PPW within the VPM-triangle was located near to the motion vector of the levator veli palatine muscle, while it was located significantly more posteriorly in the SMCP group. The PPW of the poor VP closure subgroup of the SMCP group tended to locate more posteriorly than those of the favorable VP closure subgroup and the control group.

Conclusions: The VP forms of the SMCP group were characterized by a shorter velum, a deeper and higher pharynx, and a more posterior PPW than the motion vector of palatal muscles. A positional discrepancy of the velopharynx including the PPW position relating to the direction of the motion of the VP muscles may influence the difficulty of VP closure achievement in SMCP patients.

Keywords: Cleft palate; Submucous cleft palate; Velopharyngeal closure; Velopharyngeal muscular triangle; Cephalometric analysis; Palatoplasty; Cleft palate speech

Introduction

Submucous cleft palate (SMCP) is a congenital condition, in which significant defects of the secondary palate occur in the absence of an actual opening into the nasal cavity. It was first reported by Roux [1] in 1825 and was named SMCP by Kelly [2] in 1910. Calnan [3] described the pathogenesis, clinical behavior, differential diagnosis, and treatment of SMCP. The main symptoms of the condition include abnormal hypernasality and articulation disorders due to velopharyngeal incompetence (VPI) [4].

There have been many reports regarding the postoperative acquisition rate of velopharyngeal (VP) closure after palatal repair in patients with SMCP, but patients often display poor speech outcomes [5-8]. Many previous authors have discussed mechanisms responsible for the difficulty of achieving favorable VP closure in SMCP patients,

and they include delayed surgical intervention, neurological deficiency, and anatomical abnormalities such as a short velum, a deep or high pharynx, or a large nasopharyngeal gap [9-12]. Kaplan [13] indicated the short palate and the deep nasopharynx in SMCP patients. Sommerlad et al. [12] classified SMCP into various types according to the severity of the associated anatomical abnormalities and found that patients with less severe anatomical abnormalities achieved less satisfactory speech results. Thus, although it is understood that anatomical conditions, e.g., in the craniopharyngeal region, can contribute to the difficulty of VP closure achievement in the presence of SMCP, there still remain questions regarding why the acquisition of favorable VP closure is difficult even in cases involving small cleft, and the specific anatomical features that influence the difficulty of VP closure.

When we consider the VP closure mechanism, VP closure is controlled by synchronous and three-dimensional velopharyngeal tissue movements. The latter movements consist of upward and backward movements of the soft palate, inward movement of the lateral pharyngeal wall, and forward movement of the posterior

pharyngeal wall (PPW). These motions, especially the backward and upward movements of the soft palate, are mainly controlled by the coordinating mechanism of the levator veli palatini and palatopharyngeal muscles [14,15]. Podvinec [16] explained the function of the soft palate by demonstrating the synthesized motion vectors of the levator veli palatini and palatopharyngeal muscles, and suggested that a discrepancy in craniopharyngeal growth might cause tonic contraction of the soft palate in an abnormal direction. The authors previously examined the craniopharyngeal morphology of cleft palate (CP) patients with persistent VPI and reported that anatomical discrepancies of the upper pharynx, such as a wide base and counterclockwise rotation of the pharyngeal triangle, which included the cranial base, cervical vertebrae, and posterior maxilla, were related to persistent VPI after palatal repair [17]. From the above findings, it can be hypothesized that a positional discrepancy of the synthesized motion of the levator veli palatini and palatopharyngeal muscles and PPW due to congenital craniopharyngeal growth abnormalities might make it difficult to achieve VP closure in SMCP patients.

The purpose of this study was to elucidate the reasons why it is difficult to achieve VP closure in SMCP patients. We analyzed the VP structures of SMCP patients focusing on the positional relationship between PPW and the velopharyngeal muscles. Then, we examined the relationship between these factors and VP closure acquisition in order to discuss possible prognostic factors associated with VP closure in SMCP patients.

Methods

Subjects

Fourteen patients with submucous cleft palate (SMCP), who were diagnosed and treated at the Department of Oral and Maxillofacial

Surgery, Kagoshima University Medical and Dental Hospital (Kagoshima University Hospital), were enrolled and subjected to cephalometric analyses of their VP structures (SMCP group). The patients included 6 males and 8 females, and their age at the time of the cephalometric assessment ranged from 3 years and 2 months to 11 years and 8 months (mean: 6 years and 7 months) (Table 1). A diagnosis of SMCP was made when a patient exhibited Calnan's triad: bifid uvula, translucency of the midline of the soft palate, and a V-shaped defect of the posterior edge of the hard palate [3]. Before the operation, a nasopharyngeal fiberoptic examination was performed in all patients to ensure midline defect of the nasal surface of the soft palate representing the incomplete union of palatal muscles. All of the patients were Japanese and belonged to a consecutive series of patients that visited our outpatient department between 2006 and 2013. Patients whose clinical symptoms were suggestive of 22q11.2 deletion syndrome were excluded. In addition, other syndromic patients and those with mental retardation were also excluded. A mental development test based on a questionnaire examining exercise, social skills, and language was performed, and the patients that presented with significant delays (more than one year) were considered to be mentally retarded.

| | SMCP | Control | Postop CP |
|--|--------------------------------|-------------------------------|------------------------------|
| No of Subjects | 14 | 20 | 20 |
| Sex distribution | | | |
| Male | 6 | 10 | 9 |
| Female | 8 | 10 | 11 |
| Age (mean) at Cephalometric assessment | 3 yrs 2 months-11 yrs 8 months | 4 yrs 6 months-6 yrs 2 months | 4 yrs 0 months-6yrs 9 months |

Table 1: Study populations for the cephalometric analysis.

Palatal repair was performed with the modified V-Y palatoplasty procedure, which allowed conservation of the periosteum in the anterior part of the maxilla [18]. The palatal muscles were sufficiently repositioned as they were turned sideways, producing a levator sling. To extend the nasal mucosa of the soft palate, the nasal mucosa of the soft palate was extended using a large Z-plasty and a free mucosal graft obtained from the buccal area.

As a comparison group, 20 healthy age- and sex-matched Japanese children, who had previously undergone cephalometric radiograph examinations at the Department of Pediatric Dentistry, Kagoshima University Hospital, were also enrolled in this study (control group). They included 10 males and 10 females, and their ages ranged from 4 years and 6 months to 6 years and 2 months (mean: 5 years and 7

months). Furthermore, as references, 20 patients with cleft palate (CP) solely, who had already achieved favorable VP closure after undergoing palatal repair at our department using the same palatal repair procedure, were enrolled (postoperative CP group). The patients included 9 males and 11 females, and their ages at the time of the cephalometric assessment ranged from 4 years to 6 years and 9 months (mean: 4 years and 7 months). The patients in the postoperative CP group seemed to be younger than those in the SMCP group, but there were no significant differences in the sex ratio or age among the three groups ($p=0.09$ (SMCP vs. Control), 0.13 (Control vs. CP), and 0.08 (SMCP vs. CP), respectively).

This study was approved by the clinical research ethical review board of Kagoshima University Graduate School of Medical and Dental Sciences (#93).

Comparison of cephalometric measurements of VP structures

Lateral cephalometric radiographs that were obtained in a resting position were used for the analyses of craniopharyngeal morphology. The lateral cephalometric radiographs of the subjects in the SMCP group were taken before the palatoplasty was performed. The dimensions of the craniopharynx were measured by a single examiner (M.T.) to eliminate interoperator error and any operator-based bias. The skeletal landmarks and measurements were derived from tracings of the lateral cephalograms by drawing the S-N plane to create the X-axis, and projecting a line that ran perpendicular to this plane through point S to create the Y-axis. The reference baseline and cephalometric landmarks are illustrated in Figure 1.

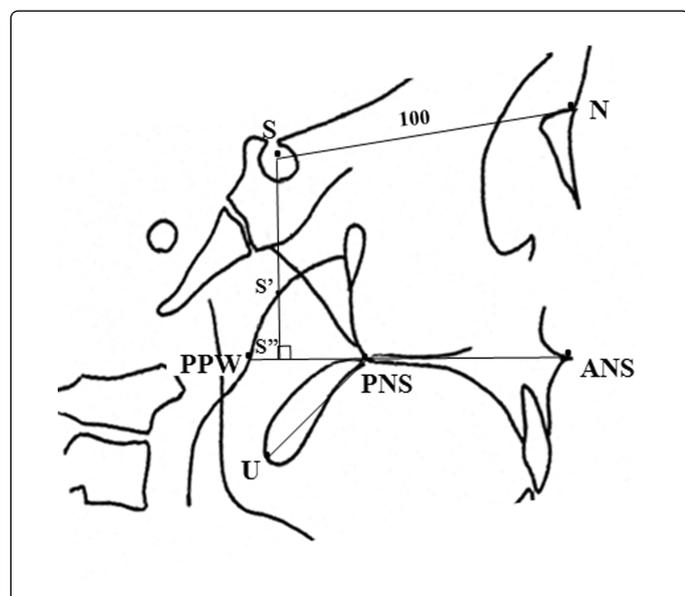


Figure 1: Cephalometric landmarks and velopharyngeal measurements. S=sella: the midpoint of the sella turcica; N=nasion: the most anterior point of the nasofrontal suture; ANS=anterior nasal spine: the tip of the anterior nasal spine; PNS = posterior nasal spine: the tip of the posterior nasal spine; PPW=posterior pharyngeal wall: the margin of the posterior pharyngeal wall at the junction of the palatal plane; S'=the top of the upper pharyngeal space: the point at the junction of the line running perpendicular to the S-N plane and the posterior pharyngeal wall; S''=the bottom of the upper pharyngeal space: the point at the junction of the line running perpendicular to S-S' and the palatal plane. Velar length (PNS-U), pharyngeal depth (PPW-PNS), and pharyngeal height (S'-S'') were measured.

The measurements included velar length (PNS-U), pharyngeal depth (PNS-PPW), and pharyngeal height (S'-S''), and the ratio of velar length to pharyngeal depth ($PNS-U/PNS-PPW \times 100$) was also calculated. As the age and sex distribution (and hence, craniofacial size) of the subjects in the CP and control groups was uneven, all craniopharyngeal dimensions were standardized relative to the length

of the anterior cranial base (S-N), which was given a value of 100 (S-N revised).

Identification of anatomical landmarks relating to the motion vectors of the palatopharyngeal muscles

Prior to the analysis of the cephalometric analysis of VP structure, we identified landmarks that corresponded to the origin (L) and insertion (M) of the levator veli palatini muscle and the origin (P) and insertion (M) of the palatopharyngeal muscle using two cadavers (Figure 2a).

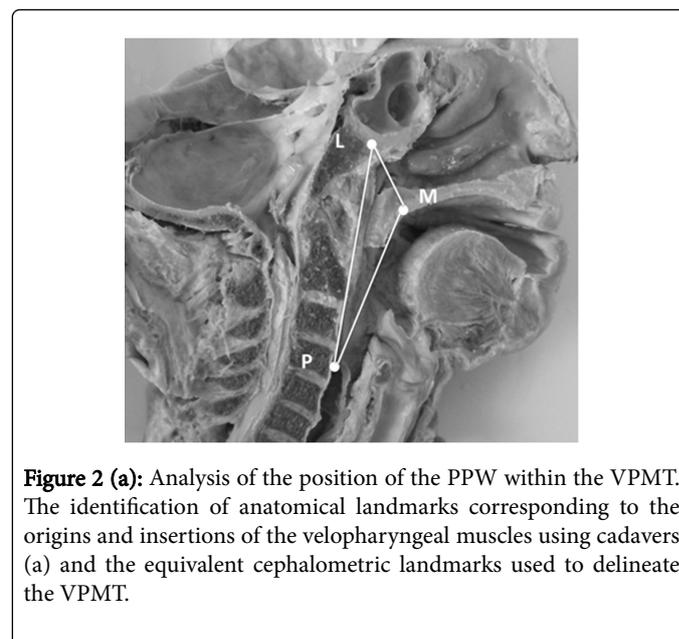


Figure 2 (a): Analysis of the position of the PPW within the VPMT. The identification of anatomical landmarks corresponding to the origins and insertions of the velopharyngeal muscles using cadavers (a) and the equivalent cephalometric landmarks used to delineate the VPMT.

The corresponding lateral cephalogram landmarks for the VPM-triangle were defined as follows (Figure 2b):

L: The junction of the bottom of the sphenoid bone and the anterior border of the mandibular condyle

P: The center of the anterior border of the 4th cervical vertebra

M: The anterior 1/3rd of the soft palate

V: The junction of the virtually synthesized motion vector M-L, the motion vector M-P, and the posterior border of the VPM-triangle (L-P).

To analyze the anteroposterior position of the PPW within the VPM-triangle (the PPW to VPM-triangle ratio), the anteroposterior position of the point PPW', which was located at the junction of an extended version of L-PPW and the virtually synthesized vector M-V, was calculated using the following formula:

$$\text{Anteroposterior position of PPW} = M-PPW' / M-V \times 100$$

The PPW to VPM-triangle ratio was compared among the SMCP, CP, and control groups.

The skulls of two soft embalmed cadavers were dissected at the midline. Through the sagittal plane of each hemi-face section, the origins and insertions of the levator veli palatine and palatopharyngeal muscles were identified. We then chose cephalometric landmarks that corresponded to these origins and insertions. The triangle produced by the projection of points L, M, and P on the sagittal plane of the hemi-

Comparison of the anteroposterior position of the PPW within the VPM-triangle among the three groups

Figure 3 schematizes the mean craniopharyngeal structure measurements obtained in the three groups superimposed on the S-N plane (X-axis) and the line perpendicular to it (Y-axis).

The form of the VPM-triangle and the position of the PPW within the VPM-triangle varied among the three groups. The VPM-triangle of the SMCP group was anteroposteriorly wider than that of the control group. The VPM-triangle of the postoperative CP group was anteroposteriorly narrower, vertically longer, and rotated counterclockwise compared with those seen in the other two groups.

Regarding the position of the PPW, the height of the PPW was almost the same in all three groups. The anteroposterior position of the PPW differed among the three groups (Figure 3). The PPW was situated near to the line running perpendicular to S-N in the postoperative CP group, and the PPW was much closer to the motion vector line L-M in the control and postoperative CP groups than in the SMCP group.

| | SMCP (n=14) | Control (n=20) | Postop. CP (n=20) |
|---------------------------|---------------|----------------|-------------------|
| PPW to VPM-triangle ratio | 47.87 ± 21.95 | 39.94 ± 16.82 | 33.35 ± 12.26 |

Table 3: Comparison of the PPW to VPM-triangle ratio between the SMCP, control, and postoperative CP groups

Statistical analyses of the mean (and SD) PPW to VPM-triangle ratio revealed that the ratio of the SMCP group (47.87 ± 21.95) was significantly greater than that of the postoperative CP group (33.35 ± 12.26, p<0.05) and tended to be greater than that of the control group (39.94 ± 16.82) (Table 3). The above findings suggest that the PPW was positioned close to the motion vector of the levator veli palatini muscle in the healthy subjects and CP patients who had already acquired favorable VP closure, while it was situated more posteriorly within the VPM-triangle and further away from the synthesized motion vector of the levator veli palatini and palatopharyngeal muscles in the SMCP patients.

| | SMCP (n=13) | | Control (n=20) |
|---------------------------|----------------------------|-----------------------|----------------|
| | Favorable VP closure (n=8) | Poor VP closure (n=5) | |
| PPW to VPM triangle ratio | 44.77 ± 20.93 | 56.81 ± 24.10 | 39.94 ± 16.82 |

Table 4: Comparison of the PPW to VPMT ratio between the patients that exhibited favorable and poor VP closure in the SMCP and control groups

The PPW to VPM-triangle ratio of the poor VP closure group tended to be greater than those of the favorable VP closure group and healthy children; however, the differences were not significant.

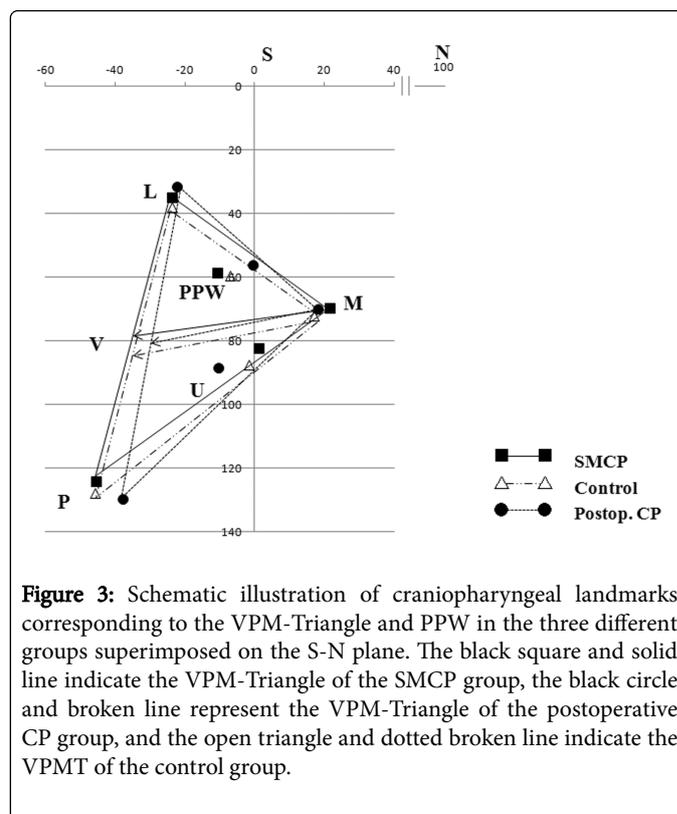


Figure 3: Schematic illustration of craniopharyngeal landmarks corresponding to the VPM-Triangle and PPW in the three different groups superimposed on the S-N plane. The black square and solid line indicate the VPM-Triangle of the SMCP group, the black circle and broken line represent the VPM-Triangle of the postoperative CP group, and the open triangle and dotted broken line indicate the VPMT of the control group.

Analysis of the relationship between the PPW to VPM-triangle ratio and postoperative VP closure in the SMCP group

Speech assessments were conducted in 13 patients in the SMCP group. Because in one patient VP closure function was not stable in speech evaluation, we excluded from this evaluation. The hypernasality assessments produced the following results: normal: 4 patients, slight: 4 patients, moderate: 4 patients, and severe: 1 patient. Nasal emission was classified as follows: normal: 3 patients, slight: 5 patients, moderate: 4 patients, and severe: 1 patient. Regarding the subjects' postoperative VP closure status, favorable VP closure was observed in 8 subjects (4 VPC and 4 borderline VPC), and poor VP closure was seen in 5 subjects (4 borderline VPI and 1 VPI). The mean and SD PPW to VPM-triangle ratios of the favorable VP closure group, poor VP closure group, and healthy children were 44.77 ± 20.93, 56.81 ± 24.10, and 39.94 ± 16.82, respectively (Table 4).

Discussion

As mentioned above, the acquisition of favorable VP closure is difficult in SMCP patients, even in those with small cleft. Actually, it is clear that the favorable VP closure acquisition rate for SMCP patients

(61.5%) in this series was markedly lower than those for patients with other cleft types, when we compare these findings with the postoperative acquisition rates of VP closure after palatoplasty at our department between 2006 and 2012 [18]. This investigation was an attempt to characterize the velopharyngeal structures that may influence VP closure acquisition in SMCP patients, focusing on the coordinating mechanism of the levator veli palatini and palatopharyngeal muscles and the location of the posterior pharyngeal wall. If possible, we also hoped to identify craniopharyngeal morphological markers that could be used as possible indicators of speech outcomes following palatal repair for SMCP.

Several conclusions are thought to be warranted from our data. The first major conclusion is that the craniopharyngeal structures of SMCP patients are characterized by a short velum and a deep and high pharynx, when compared with those of age-matched healthy children and CP patients. On the other hand, although all of the CP patients were examined at the postoperative stage, the craniopharyngeal forms were quite different from those of the other subjects. For example, they demonstrated significantly longer velum and significantly shallower and smaller (in height) pharynx than the SMCP patients and healthy subjects. The above differences in the craniopharyngeal forms of the 3 groups suggest that SMCP and CP may not represent a severity continuum of anatomical soft palate abnormalities.

There have been several reports regarding the velopharyngeal morphology of SMCP patients, but there are several different opinions regarding the craniopharyngeal structures of SMCP patients. Weatherly-White [19] reported that not all SMCP patients exhibited speech disorders; therefore, VPI might be caused by not only a short palate but also the degree of development of the velar muscles. Fujita et al. [9] examined the cephalograms of 20 SMCP patients with VPI and stated that SMCP patients with VPI had significantly shorter velar. Kaplan [13] also indicated that the hard and soft palates were shorter and the nasopharynx was deeper in SMCP patients. On the other hand, Harita et al. [10] reported that a deep pharynx did not affect the risk of VPI in SMCP patients based on videofluoroscopic measurements of the velar length to pharyngeal depth ratio in 13 patients. Our present results support Fujita and Kaplan's view that a shorter velum and a deeper pharynx are morphological characteristics of SMCP and suggest that these characteristics might affect the postoperative acquisition of VP closure.

Our second major conclusion is that the VPM-triangle, which is produced by the virtually synthesized motion vectors of the levator veli palatini and palatopharyngeal muscles, was characterized by a wide-based triangle and a posteriorly located PPW in the SMCP group. Prior to this study, we hypothesized that the cause of contradiction; less severe anatomical abnormalities (a smaller cleft) resulted in less satisfactory speech results in SMCP patients, might be caused by both anatomical (morphological) factors and the functional disruption of the velopharyngeal muscles. Therefore, we focused on the VPM-triangle produced by the synthesized motion vectors of the levator veli palatini and palatopharyngeal muscles because of several reasons. The first, it is well known that velar activity and VP closure together with the synthesized motion vector of the levator veli palatini and palatopharyngeal muscles [16]. Braithwaite [20] reported that the VP closure mechanism involves coordinated movement of the levator veli palatini, palatopharyngeal, and superior constrictor pharyngeal muscles, and especially the action of the former two muscles, which have the same insertion in the soft palate. The second, congenital craniofacial malformations, including cleft lip and palate, with growth

disturbances and disharmony in the maxillofacial region could be a factor in VPI [21]. The authors [17] compared the pharyngeal structures of CP patients with/without VPI and found a short velum and counterclockwise rotation and wide base of the pharyngeal triangle in those with persistent VPI. These results suggested that the rotation of the pharyngeal structure could affect the difficulty of achieving sufficient VP closure in patients with CP.

Figure 4 provides a schematic illustration of the differences in the position of the PPW within the VPM-triangle between the control or postoperative CP group and the SMCP group. In the control and postoperative CP groups, the PPW took a more anterior position in the VPM-triangle and was located close to the motion vector of the levator veli palatini muscle. When the levator veli palatini muscle constricts, the nasal surface of the soft palate can easily come into contact with the pharyngeal wall in healthy subjects (shown by the solid line in Figure 4a). Furthermore, in the postoperative CP group, in which significantly smaller pharyngeal height values were recorded, VP closure could also be achieved easily by the action of the levator veli palatini muscle (shown by the dotted line in Figure 4a). However, in the SMCP group, the PPW was located more posteriorly, as the pharynx was high and deep (Figure 4b), therefore, greater soft palate strength and flexibility might be required to enable contact between the soft palate and pharyngeal wall in SMCP patients.

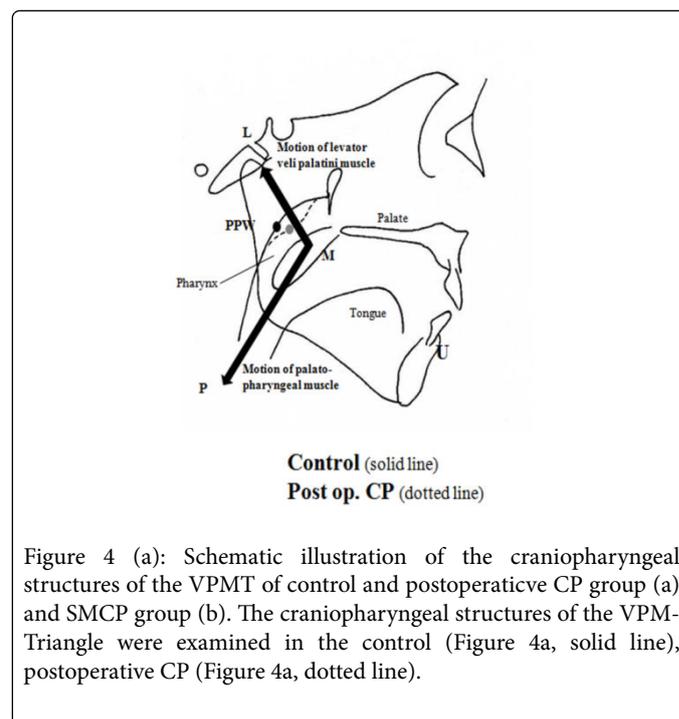
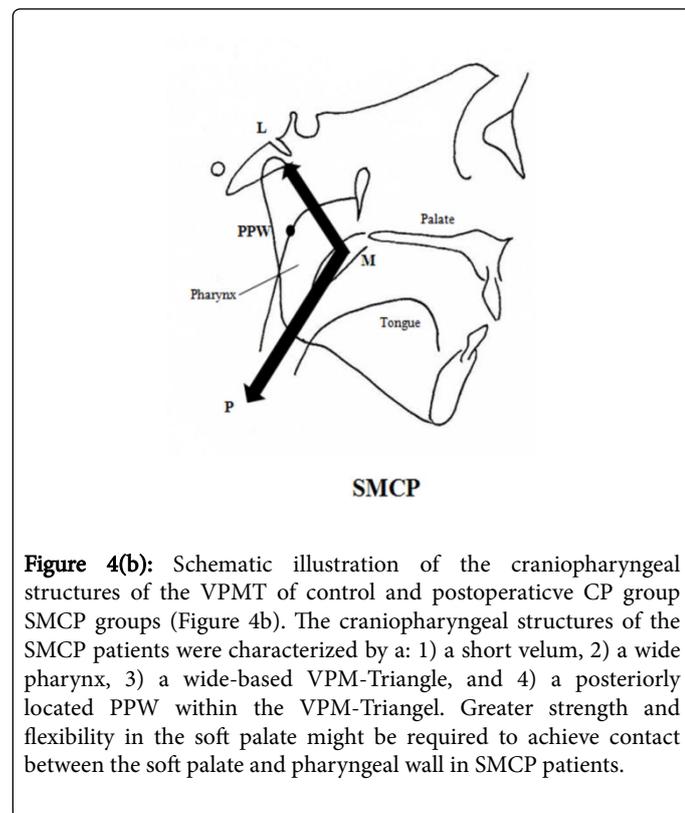


Figure 4 (a): Schematic illustration of the craniopharyngeal structures of the VPMT of control and postoperative CP group (a) and SMCP group (b). The craniopharyngeal structures of the VPM-Triangle were examined in the control (Figure 4a, solid line), postoperative CP (Figure 4a, dotted line).

Our final conclusion is that the anteroposterior position of the PPW within the VPM-triangle might be a prognostic factor for favorable VP closure in SMCP patients. In the present study, the PPW was located more posteriorly within the VPM-triangle and further away from the motion vector of the levator veli palatini muscle in the SMCP group, especially in the poor VP closure subgroup. The results suggest that a positional discrepancy of the PPW relative to the direction of motion of the velopharyngeal muscles due to the disharmony of the velopharyngeal development might be one cause of the difficulty of achieving VP closure in SMCP patients. However, the significant difference was not detected in the position of PPW due to the small

sample size of the present study. Therefore, further studies are necessary to develop clinically useful prognostic factors for SMCP.

the direction of the motion of the velopharyngeal muscles may influence the difficulty of VP closure achievement in SMCP patients.



This study had several limitations that should be taken into consideration when interpreting our results. The first is that the anatomical VPM-triangle landmarks, which were used in pediatric case, were established using adult cadavers. Obviously, the craniopharyngeal morphologies of adults and children might differ. However, it is very difficult to obtain child cadavers, and the mechanism responsible for closing the VP space is considered to be the same in both adults and children. Therefore, the three anatomical landmarks, L, M, and P, identified during the examinations of the adult cadavers were considered to be sufficiently reliable markers for this study. In addition, it would have been preferable to compare the preoperative VP structures of the SMCP and CP groups in order to elucidate the morphological characteristics. However, it was not possible to collect preoperative cephalograms for patients with CP who had not undergone palatal repair until the age of 6 years in Japan. Furthermore, since previous studies indicated the differences in pharyngeal form between infants and young children due to variations in the size of the adenoids and maxillary growth [14], we used age-matched the subjects that took part in the cephalometric analyses. Despite these limitations, the present study described the mechanism responsible for a poor speech prognosis in SMCP patients and provided a possible morphological marker for predicting postoperative VP closure in SMCP patients.

Conclusions

The velopharyngeal forms of the SMCP group were characterized by a shorter velum, a deeper and higher pharynx, and a more posterior PPW than the motion vector of palatal muscles. A positional discrepancy of the velopharynx including the PPW position relating to

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