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Preliminary Study in Relationship Between Cleft Lip and Palatal Bony Defect in Submucous Cleft Palate

Chang Jiang Du, MD, Xia Zhou, MD, and Lian Ma, MD, PhD

Submucous cleft palate, presenting as varying degrees of palatal bony defect, can be difficult to detect in its early stage. The connection between submucous cleft palate and cleft lip has been noticed by clinicians but are rarely reported in literature.

Aims: To investigate the correlation between the degree of deformity of palatal bony defect and that of cleft lip.

Patients and Methods: Thirty-four patients with unilateral (n = 23) or bilateral (n = 11) cleft lip presenting with submucous cleft palate were included. Patients were divided into 3 groups according to the degree of malformation of cleft lip (microform, incomplete, and complete). The length and width of palatal bony defect was then measured from the palatal computer tomography.

Results: In patients with unilateral cleft lip, the proportions of microform cleft lip, incomplete cleft lip, and complete cleft lip were 17.4%, 60.9%, and 21.7%, respectively. In patients with bilateral cleft lip, there were 3 cases with microform and 1 case with incomplete cleft lip on both sides. No correlation was found between the length or relative width of palatal bony defect with the side (P length = 1.000; P relative width = 0.262) or the severity (P length = 0.605; P relative width = 0.254) of cleft lip.

Conclusions: The form of cleft lip presenting with submucous cleft palate varies, and there was no correlation with the length or relative width of palatal bony defect. Advanced imaging techniques for children with cleft lip may assist the early diagnosis of submucous cleft palate.

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Submucous cleft palate (SMCP) was a relatively rare variant of cleft palate with a reported prevalence of 1:1250 to 1:6000.[1,2] Kelly[3]described the main symptoms in SMCP as speech problems caused by velopharyngeal incompetence. Calnan[4] described SMCP as the classic anatomical triad of bifid uvula, a translucent zone in the midline of the soft palate and bony notch of the posterior border of the hard palate.

Early in 1954, Calnan[4] firstly reported the asymptomatic SMCP patients, and more clinicians were aware of that part of SMCP patients without any treatment appear no speech problems. It was controversial to perform operation in early age which was not available to predict velopharyngeal function.[5] In these very young patients, it was necessary to apply alternative methods and techniques for the estimation of velopharyngeal function, which was also considered the predictive index of velopharyngeal function and therefore was very important for the appropriate therapeutic schedule planning for children with SMCP. By assessing the threedimensional computer tomography (3D CT) image and nasoendoscopy, it was found that the velopharyngeal function was correlated to the palatal bony defect in SMCP patients.[6]

Kono[7] reported thirteen percent patients accompanied with cleft of primary palate were diagnosed SMCP. In 1999, Gosain et al[8] found that the prevalence of classic SMCP was 150 to 600 times in patients accompanied with isolated cleft lip than the reported prevalence in the general population. It was recommended that clinicians should pay more attention to the patients accompanies with isolated cleft lip.[8,9] There was rare research about the correlation between the characteristics of cleft lip and palatal bony defect in SMCP patients accompanied with cleft lip. The aim of this study was to investigate the correlation between the degree of deformity of palatal bony defect and that of cleft lip in patients with SMCP.

MATERIALS AND METHODS

Patients

This was a retrospective single-center archive study, approved by Peking University School Hospital of stomatology, China, with principles following the Declaration of Helsinki.

Consecutive 34 SMCP patients accompanied with cleft lip from January 1, 2013 to December 31, 2020 in Peking University Hospital of stomatology were included. All patients had complete records of preoperative CT scan and facial photography. Based on review of medical records and facial photographs, the patients were grouped according to the type of cleft lip: bilateral and unilateral, also the degree of cleft lip: microform cleft lip, incomplete cleft lip, and

FIGURE 1. Classification for cleft lip based in the severity of the cleft. (A) Microform cleft lip. (B) Incomplete cleft lip. (C) Complete cleft lip.

complete cleft lip.[10] The designations and associated descriptions are as follows:

Microform cleft lip: It may appear as a vertical Cutaneous groove and muscular depression rather than a philtra ridge, a notched vermilion mucosa, discontinuous vermilion-cutaneous junction, mild nasal deformity (ie, flattened nostril rim and displaced ala), or any combination (Fig. 1A).

Incomplete cleft lip: The severity of which was between the microform and complete cleft lip. Cleft goes beyond the margin of the lip, but not penetrated the nostril (Fig. 1B).

Complete cleft lip: Cleft penetrated the nostril (Fig. 1 C).

Data Collection

All patients were scanned using a 16-slice CT scanner (GE Bright Speed Elite; GE Healthcare; Boston, Massachusetts, USA) to obtain morphometric CT data of the bony palate and maxillary structures. Computer tomography scans without contrast (thickness 1.25 mm, ≥200 mA, 120–140 kV) were obtained in the sequence described above. A 3D CT reconstruction of the patient's head was created using Carestream PACS 11.0 software with bone density. The areas below the mandibular dentition were cut out to superimpose the palate alone. Each occlusal view of the hard palate was captured from the 3D model.

Visual Inspection

By assessing the severity of the palatal bony defect, it was divided into 2 types: palatal bony defect not involved the incisive foramen (Fig. 2A) and palatal bony defect involved the incisive foramen (Fig. 2B).

Digimizer Version 4.2.6.0 was used to fix point and measure in the palatal photography, the specific standards were shown in Figure 3. The width of the palatal bony defect was the length of (a)-(a′) and the width of the hard palate was the length of (b)-(b′). (a)-(a′)/(b)-(b′) was the ratio of the width of the palatal bony defect to the width of the hard palatal plate, which was defined as the relative width of palatal bony defect. The evaluation of width was based on whether the (a)-(a′)/(b)-(b′) exceeded 1/3.

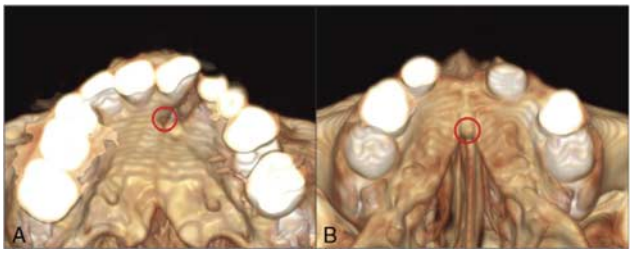

FIGURE 2. Two types of palatal bony defect shown by 3D reconstruction from CT scan images. (A) Palatal bony defect not involving the incisive foramen. The red circle represents the location of the incisive foramen. (B) Palatal bony defect involving the incisive foramen. The red circle represents the location of the incisive foramen. 3D, three-dimensional; CT, computed tomography.

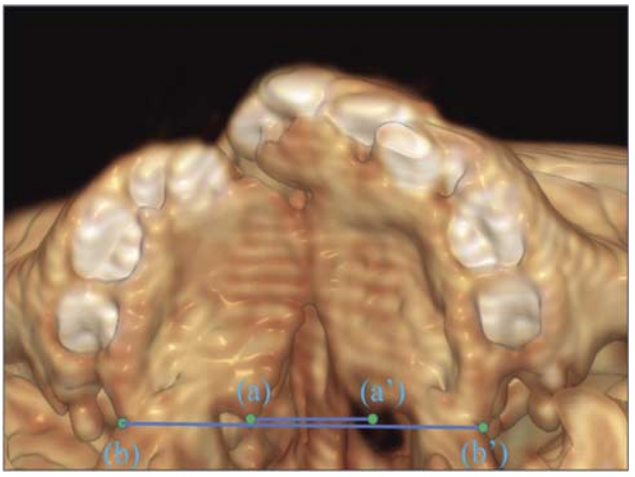

FIGURE 3. Measuring the relative width of palatal bony defect referring to hard palate. Points (a) and (a^0) denote the posterior points of the fissure edges of the palatal bony defect on both sides. The linear distance between (a) and (a^0) is expressed as (a)-(a′). Points (b) and (b′) represent the outermost point at the junction of the lateral pterygoid plate and the maxillary tuberosity on both sides. The linear distance between (b) and (b′) is expressed as (b)-(b′).

Statistics Analysis

All the data analyses were performed using the IBM SPSS 26.0. Chi-squared test to investigate the correlation between the length or relative width of palatal bony defect with the type (bilateral and unilateral) or the severity (microform, incomplete and complete) of cleft lip. $P < 0.05$ was regarded as being statistically significant. For the bilateral cleft lip, the side with more severe was considered as the degree of cleft lip malformation.

RESULTS

Basic Information

Among the 34 patients consisted of 32 males (94.1%) and 2 females (5.9%). The average age was 6 years old. Twenty-three patients were unilateral (67.6%) and eleven patients were bilateral (32.4%), a total of 45 affected sides.

Analysis of Cleft Lip

Among the 23 unilateral cleft lip patients, the distribution of microform cleft lip, incomplete cleft lip, and complete cleft lip was as follows: 17.4% (4/23), 60.9% (4/23), and 21.7% (5/23). In 11 patients with bilateral cleft lip, there were 4 cases with the same type of cleft lip in both sides (3 patients with microform cleft lip of both sides, 1 patient with incomplete cleft lip of both sides). The results were as shown in Supplementary Digital Content, Table 1, http://links.lww.com/SCS/D707.

Analysis of Palatal Bony Defect

Twenty-two patients were assessed as palatal bony defect not involving the incisive foramen, accounting for 64.7% of the total patients. The rest twelve patients were assessed as palatal bony defect involving the incisive foramen. Twenty-one patients were assessed as the ratio of the width of the hard palatal bony defect to the width of the hard palatal plate less than one-third, accounting for 61.8% of the total patients. The rest thirteen patients were assessed as the ratio of the width of the palatal bony defect to the width of the hard palatal plate more than one-third.

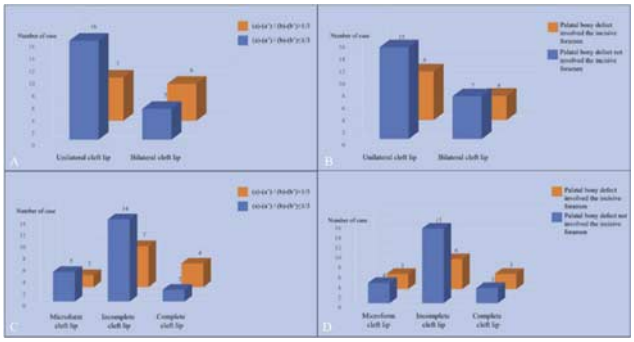

FIGURE 4. Group comparison of basic morphologic characteristics (defect length and relative width) among patients with palatal bony defect accompanied with different types of cleft lip. Group-wise difference was analyzed by conducting a Chi-squared test. (A) Comparison of the relative width in palatal bony defect accompanied with bilateral and unilateral cleft lip. (B) Comparison of the length in palatal bony defect accompanied with bilateral and unilateral cleft lip. (C) Comparison of the relative width in palatal bony defect and severity of accompanied cleft lip. (D) Comparison of the length in palatal bony defect accompanied with cleft lip at different levels of severity.

Correlation Between Cleft Lip and Palatal Bony Defect

The results showed no correlation was found between the length or relative width of palatal bony defect with the types-unilateral and bilateral (P_{length}= 1.000; *P*relative width = 0.262) (as shown in Supplementary Digital Content, Table 2, http://links.lww.com/SCS/D707 and Supplementary Digital Content, Table 3, http://links.lww.com/SCS/D707). There was also no correlation found out between the length or relative width of palatal bony defect with severity which was expressed by microform, incomplete and complete (P_{length}= 0.605; $P_{relative}$ width = 0.254) of cleft lip (as shown in Supplementary Digital Content, Table 4, http://links.lww.com/SCS/D707 and Supplementary Digital Content, Table 5, http://links.lww.com/SCS/D707). The correlation results were demonstrated clearer in Figure 4A-D.

DISCUSSION

Submucous cleft palate was diagnosed later than the cleft palate on account of its malformation with minor indication. Most patients were diagnosed when they admitted into hospital for speech problems, which may result in unsatisfactory prognosis. The importance of early diagnosis was self-evident. Some studies shown that the prevalence of SMCP was much higher in isolated cleft lip than the general population, so investigators suggested that more comprehensive clinical examinations of the palate should be performed in patients with cleft lip.[7,8] In our current study, we have reported several patients diagnosed with microform cleft lip who had as well developed severe palatal bony defect, highlighting the necessity of additional examination of the palate in patients with cleft lip. Further, for those were not cooperative with the examination or difficult to diagnosis account for no evident signs of SMCP, advanced imaging techniques such as CT may assist the early diagnosis of SMCP.

The SMCP patients who will appear speech problems need receive treatment as early as possible, but this proportion was difficult to identify in the early age. In 1954, Calnan[4] described the differences in the extent of palatal bony defect during operation. Sommerlad et al[11]reported the similar research in 2004. With the popularization of CT examination and other imaging techniques, it was possible to observe the palatal deformity clearly and directly. Clinical studies have found that the degrees between soft tissue defect and bony defect were not completely consistent in incomplete cleft palate patients.[12] In 2015, Ren et al[13]found the significant variability in palatal bony defect in SMCP patients by observing CT images and 3D reconstructions. By assessing the 3D CT image and nasoendoscopy, it was found that the velopharyngeal function was correlated to the palatal bony defect in SMCP.[6] The research about the correlation between the palatal bony defect and cleft lip deformity may aid in the treatment for clinicians.

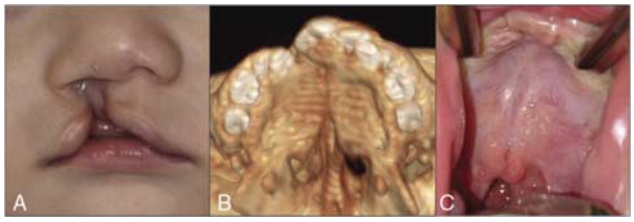

FIGURE 5. Photography and 3D reconstruction results from CT scans in a patient with submucous cleft palate accompanied with complete cleft lip. (A) Facial photography showing unilateral cleft lip on the right side. (B) Threedimensional CT reconstruction showing the palatal bony defect not involving the incisive foramen. (C) Intraoral photography showing the clinical manifestation of the case. 3D, three-dimensional; CT, computed tomography.

Among the 34 patients, 8 (23.5%) patients were admitted into hospital for unclear speech, 25 (73.5%) patients were diagnosed with SMCP when they came into hospital for repair for cleft lip, secondary deformity, or cleft alveolar. One (3%) patient came into hospital for the morphological abnormality of palate. It was important to exam the palate in patients with cleft lip more careful, because it was a precious opportunity for these patients to come into hospital early. Although there was controversy about the optimal treatment timing and the necessity of preventive surgery, doctors could follow up patients closely and give timely intervention when necessary.

By comparing the degree of deformity of cleft lip and that of palatal bony defect, we found that there was no correlation between these 2 types of malformation. Patients accompanied with slight cleft lip may appear severe palatal bony defect. Conversely, patients accompanied with severe cleft lip may appear slight palatal bony defect as in Figure 5A-C. The pathological mechanisms and the process of embryonic development of cleft lip and palatal bony defect may be independent, which means the virulence genes related to the development of the defect may be distinct. Our findings provide some information that may boost the further exploration of the pathological mechanism of the deformity, and may play a guiding role in the mapping of pathogenic genes in future.

Among the 34 patients, male patients accounted for 94.1% (32/ 34). In some reports, the number of male patients was more than female, but the ratio of male to female was lower than this study.[5] There may be bias in the selection of samples, but such a disparity in the ratio of male to female suggested that the sex may have an impact on the pathogenesis of cleft lip and palate.

In this retrospective study, some SMCP patients accompanied with cleft lip may be missed diagnosis. To expand the samples' quantity, more detailed examinations should be paid to the patients with cleft lip. In the present research, we grouped our patients into multiple subtypes based on the degree of deformity of cleft lip and palatal bony defect assessed using the 3D reconstructed CT image. Future studies are necessary where

computerized analysis should be introduced to quantitatively determine the characteristics of different deformation subtypes, and to further explore the correlation between cleft lip and palatal bony defect explicitly.

Patients with SMCP may be accompanied with cleft lip and the characteristics of cleft lip varies. There was no correlation between the characteristics of cleft lip and palatal bony defect in patients with SMCP, indicated that the mechanisms of cleft lip and cleft palate may be independent.

In this retrospective study, we collected and analyzed the clinical data of 34 SMCP patients accompanied with cleft lip. We applied a rigid patient inclusion criterion, resulting in a moderated study sample size for a variety of distributions. Future research is suggested to enlarge the size of our current patient cohort. Besides, the current classification standard for cleft lip and palatal bony defect categorizes the degree of deformity into different levels which does not precisely describe the abnormality in the shape of cleft lip and cleft palate. To further explore the correlation between cleft lip and palatal bony defect, more studies are required where data mining and machine learning techniques can be implemented to quantitatively determine the characteristics of different subtypes of the malformation. Finally, in the present research we performed manual measurement of palatal bony defect by reading the palatal bony view of the 3D-reconstructed model. This research may benefit from the computational analysis of the 3D model where topological features such as area and length of the palatal bony structure can be automatically extracted.

CONCLUSIONS

The form of cleft lip presenting with SMCP varies, and there was no correlation with the length or relative width of palatal bony defect. Advanced imaging techniques for children with cleft lip may assist the early diagnosis of SMCP.

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