

Original Research

The Relationship between Submucous Cleft Palate and a History of Nasal Regurgitation in Patients during Infancy

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Abstract

Objectives: Submucous cleft palate is a congenital disorder resulting in velopharyngeal insufficiency. Nasal regurgitation is one of the symptoms of submucous cleft palate. This study aims to investigate a relationship between submucous cleft palate and a history of nasal regurgitation in patients during infancy.

Methods: This retrospective study includes patients diagnosed with submucous cleft palate at our hospital for 20 years, from 1998 to 2017. Based on the clinical records of these patients, we investigated sex, age at referral to our facility, age at surgery, presence of nasal regurgitation, method of surgery, association of syndromes and mental retardation, and transition of velopharyngeal function.

Results: There were fifty-seven patients referred to our facility. Patients with episodes of nasal regurgitation were referred to clinics later than those without nasal regurgitation. The timing of surgery was also the same. The velopharyngeal function of patients with nasal regurgitation was worse than that of those without nasal regurgitation.

Conclusions: A history of nasal regurgitation cannot predict submucous cleft palate. However, patients with submucous cleft palate with nasal regurgitation had worse velopharyngeal function than those without nasal regurgitation. An increased understanding of the relationship between submucous cleft palate and nasal regurgitation and further study will predict submucous cleft palate and determine a good timing for surgical interventions.

Keywords

submucous cleft palate, nasal regurgitation, velopharyngeal function, surgical indication

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<https://doi.org/10.53045/jprs.2022-0046>**Introduction**

Submucous cleft palate (SMCP) is a congenital disorder resulting in velopharyngeal insufficiency (VPI) without an apparent cleft palate¹⁾. VPI caused by SMCP is associated with an abnormal muscle union across the soft palate²⁻⁴⁾. Calnan reported that bifid uvula, palate muscle diastasis (zona pellucida), and bony defect (notching) of the posterior hard palate comprised the triad of abnormal physical characteristics associated with SMCP⁵⁾. The incidence of SMCP in the normal population was 0.02%-0.08%⁶⁾. Since detecting the triad is challenging in clinical practice, SMCP is often undetected in infancy. Patients with SMCP commonly do not receive a diagnosis until hypernasality and articulation disorder caused by VPI become apparent. Patients suspected of

SMCP are referred to the clinic at a mean age of approximately 3-5 years^{1,7-11)}. However, it is also common for patients to remain undiagnosed with SMCP, even as they grow up.

The surgical indication for SMCP is determined by the presence of VPI⁴⁾. However, patients with SMCP diagnosed by physical examination occasionally present with VPI. The surgical timing for SMCP is also controversial⁴⁾. Some reports recommend early surgeries to improve the velopharyngeal function (VPF)^{5,11-13)}, whereas others recommend delayed surgery until a speech evaluation is completed and a diagnosis of VPI is made^{4,6,8,9,14-17)}.

Nasal regurgitation is among the symptoms of VPI caused by SMCP^{1,16)}. Detecting SMCP by physical examinations may be challenging, as identifying hypernasality and speech

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Table 1. Information of Included Patients.

| Nasal regurgitation | Number of patients | Sex | | Age at referral months (mean \pm SD) | Number of operated patients | Age at surgery months (mean \pm SD) | Syndrome | ID |
|---------------------|--------------------|------|--------|--|-----------------------------|---------------------------------------|-----------------|-----------------|
| | | Male | Female | | | | | |
| Exist | 29 | 16 | 13 | 44.03 \pm 35.72 [†] | 25 ^Σ | 59.56 \pm 33.87 [‡] | 10 ^Φ | 16 [‡] |
| None | 21 | 9 | 12 | 30.14 \pm 25.35 [†] | 12 ^Ψ | 46.83 \pm 25.60 [‡] | 2 ^{§Φ} | 4 [‡] |

^ΣFurrow method, 21 patients; Pushback method, 1 patient; Pharyngeal flap, 3 patients.

^ΨFurrow method, 11 patients; Pharyngeal method, 1 patient.

[‡]22q.11.2 deletion syndrome, 8 patients; Moebius syndrome, 1 patient.

Haidu-Cheney syndrome, 1 patient.

[§]22q.11.2 deletion syndrome, 1 patient; Treacher-Collins syndrome, 1 patient.

[†]Student's t-test: $p = 0.121$ [‡]Student's t-test: $p = 0.23$

^ΦFisher's exact test: $p < 0.01$ [‡]Fisher's exact test: $p < 0.01$

disorder takes time. However, nasal regurgitation can be easily noticed in infancy by clinicians and the patient's family.

We considered that patients with SMCP with nasal regurgitation are more likely to require surgeries than those without nasal regurgitation. Thus, the history of nasal regurgitation must be explored among patients with SMCP to identify the appropriate treatment plans. The present study aimed to investigate the following two hypotheses: patients with SMCP with nasal regurgitation are referred to hospitals and operated earlier than those without nasal regurgitation, and patients with SMCP with nasal regurgitation have worse VPF and require surgical interventions. We also sought to explore the relationship between SMCP and nasal regurgitation.

Methods

Study characteristics and patient inclusion criteria

Our study protocol obtained institutional review and was approved by the Ethics Committee of our hospital (approval number: 2020-024). All patients diagnosed with SMCP at any age at our hospital during a 20-year period from 1998 to 2017 were enrolled. We retrospectively reviewed the clinical records of patients with SMCP and then collected data on sex, age at referral to our facility, age at surgery, presence of nasal regurgitation, method of surgery, association of syndromes and mental intellectual disability (ID), and transition of VPF. Patients who could not be evaluated for VPF because of severe ID or auditory disturbances were excluded from this study.

The diagnostic criterion of SMCP

The diagnostic criterion was the presence of at least one abnormal physical finding of Calnan's triad by oral examination.

The evaluation of VPF and surgical indication

Two experienced speech therapists with >10 years of experience evaluated the presence of VPF using a combined approach of analyzing speech (hypernasality and nasal emission) and blowing examination based on the Japanese standard test established by the Cleft Palate Speech Committee of the Japanese Association of Communication Disorders¹⁸⁻²⁰⁾.

VPF was categorized into the following four degrees: normal, good (borderline competency), fair (borderline insufficiency), and poor (insufficiency). We performed surgery on patients with fair or poor VPF. Moreover, we operated on patients presenting with an apparent muscle diastasis of the soft palate before their VPF could be evaluated. There was no standard decision on which procedure to perform among surgeons; instead, the surgical procedure was decided by the surgeons.

To compare the VPF before the surgical interventions between patients with or without nasal regurgitation, we categorized the patients with VPF into the following two groups: patients with normal and good VPF (nonindication for surgeries), and patients with fair and poor VPF (indication for surgeries).

The transition of VPF (before and after the operations) was investigated in patients operated with the Furlow method and those evaluated with VPF before surgeries. Post-operative VPF was evaluated at 6 months to 1 year after surgery.

Statistics

We used Fisher's exact test to investigate the statistical differences and defined $p < 0.05$ as significantly different. To determine "age at referral" in months and "age at surgery" in months, the Student's t-test was used, and $p < 0.05$ was defined as significantly different.

Results

Fifty patients (25 men and 25 women) were enrolled in our study. The patients' mean age at referral was 38.2 months (range, 1-176 months). The number of operated patients was 37. Altogether, 32 patients were operated on using the Furlow method, one by the Pushback method, and four by using a pharyngeal flap. The patients' mean age at surgery was 55.4 months (range, 11-178 months). Twenty-nine patients presented with a medical history of nasal regurgitation. Twenty patients presented with ID, and 12 patients presented with syndromic diseases. These results are shown in **Table 1**. There were no significant differences in the "age at referral" and "age at surgery" between patients with and without a history of nasal regurgitation. **Table 2**

Table 2. The Results of VPF before Surgery.

| Nasal regurgitation/VPF | Normal | Good | Fair | Poor | Undeterminable |
|-------------------------|--------|------|------|------|----------------|
| Exist | 2 | 0 | 9 | 15 | 3 |
| None | 8 | 1 | 5 | 3 | 4 |

Nonoperated patients were included; the VPF of these patients was evaluated at the age when VPF could be evaluated for the first time.
Undeterminable; patients operated on at the age when VPF could not be evaluated.

Table 3. The Results of VPF before Surgical Interventions.

| Nasal regurgitation/VPF | Normal + Good | Fair + Poor |
|-------------------------|---------------|-------------|
| Exist | 2 | 24 |
| None | 9 | 8 |

Patients with nasal regurgitation presented with significantly worse VPF (Fisher’s exact test: $p < 0.01$).

Table 4. Pre/postoperative VPF.

| Nasal regurgitation/VPF | | Normal | Good | Fair | Poor |
|-------------------------|---------------|--------|------|------|------|
| Exist | Preoperation | | | 7 | 11 |
| | Postoperation | 5 | 2 | 8 | 3 |
| None | Preoperation | | | 5 | 3 |
| | Postoperation | 4 | 4 | | |

Patients operated on using Furlow method were included.

and 3 show the results of the VPF evaluation before the surgery. The data of patients who were operated at an age when VPF could not be evaluated are not presented in Table 3. Patients with nasal regurgitation showed significantly worse VPF (Fisher’s exact test: $p < 0.01$) and required surgical interventions.

The transition of VPF (before and after surgeries) is shown in Table 4 and 5. Patients operated on using the Pushback method and a pharyngeal flap were excluded because there were only five of them, and it was difficult to compare the data among three surgical procedures; thus, we included only the data of patients operated on by the Furlow method in both tables. The VPF parameters in Table 5 were converted from those in Table 4 because the postoperative VPF of the patients was fair or poor, and we were required to consider other speech therapy interventions to allow for an easy interpretation of the surgical effects. Postoperative VPF of patients with nasal regurgitation was significantly worse than that of those without nasal regurgitation (Fisher’s exact test: $p < 0.01$).

Discussion

The “age at referral” and “age at surgery” between patients with and without a history of nasal regurgitation (Table 1) did not significantly differ, indicating that nasal regurgitation is not associated with early hospitalization and SMCP diagnosis. Patients with nasal regurgitation had significantly worse VPF than those without nasal regurgitation, and the effect of surgical interventions was also significantly

Table 5. Pre/postoperative VPF.

| Nasal regurgitation/VPF | | Normal + Good | Fair + Poor |
|-------------------------|---------------|---------------|-------------|
| Exist | Preoperation | 0 | 18 |
| | Postoperation | 7 (39%) | 11 |
| None | Preoperation | 0 | 8 |
| | Postoperation | 8 (100%) | 0 |

This table was derived from Table 4. The parameters of VPF were converted to “normal + good” and “fair + poor.” Postoperative VPF of patients with nasal regurgitation was significantly worse than that of those without nasal regurgitation (Fisher’s exact test: $p < 0.01$).

worse in patients without nasal regurgitation. This shows that a history of nasal regurgitation is associated with surgical intervention among patients with SMCP, and their response to surgical intervention is poor (Table 3, 4 and 5).

Several limitations exist in this study. First, our diagnostic criterion of SMCP exhibits at least one abnormal physical finding of Calnan’s triad by oral examination. This criterion might not be correct. Different reports use different criteria. In some reports, SMCP was diagnosed by determining the presence of two or three findings in Calnan’s triad^{2,6,14,15,17}. Some reports also emphasized the diastasis of the velum muscle structure as a diagnostic criterion^{4,10,21-24}. Additionally, the concept of subspecific SMCP, termed occult SMCP, was proposed. Occult SMCP accounts for 10%-20% of all SMCP^{7,8}. However, diagnosing occult SMCP is difficult because it does not demonstrate the same apparent physical findings of Calnan’s triad^{18,25,26}. Rather, its definitive diagnosis is made by detecting soft palate muscle diastasis (abnormal levator muscle insertion into the posterior border of the palate) during surgery^{25,27} or by clinical examination of the soft palate via nasopharyngoscopy^{7,8,26}. Thus, detecting occult SMCP only through an oral examination is difficult for physicians¹². Moreover, the diagnosis is very difficult in infants. Considering these diagnostic difficulties, we created our diagnostic criterion for SMCP. However, this might lead to inconsistencies when assessing VPF among patients with SMCP. To accurately assess VPF, the precise diagnostic criterion of SMCP is needed.

The second study limitation is that we included patients with ID and syndromes. Third, four surgeons were present in our study. Fourth, three operation methods were used in our study. Furthermore, there was no standard criteria for adopting a surgical procedure. The scarcity of patients with SMCP prevented us from excluding patients with ID and syndromes and limiting patients operated by a single surgeon. Owing to these limitations, comparing several exami-

nation items is difficult in our study. These might affect our results. To ensure a precise assessment, future studies must involve patients with SMCP without ID and any syndromes under a single surgeon.

The timing of referral to hospitals

We hypothesized that patients with SMCP with a history of nasal regurgitation would be hospitalized earlier than those without a history of nasal regurgitation. However, our result was not consistent with our hypothesis. Nasal regurgitation is one of the symptoms of SMCP. This phenomenon can easily be detected, and we hypothesized that detecting nasal regurgitation would lead to early operations. Patients with episodes of nasal regurgitation were referred to clinics later than those without nasal regurgitation. Additionally, the timing of surgeries was delayed. The timing of referral and surgery (Table 1) did not significantly differ. The families of patients later referred to clinics might not remember their child's episode in detail; thus, families could forget the episode of nasal regurgitation. Park reported that nasal regurgitation disappeared by 1 year of age in all patients²⁸. This could be related to the nonrecognition of nasal regurgitation. Nasal regurgitation was reported to appear in approximately 20%-30% of all patients with SMCP^{1,7}. This may be fewer than the actual number of patients with SMCP with nasal regurgitation. Furthermore, clinicians who are unfamiliar with cleft lip and palate treatment do not always recognize the relationship between SMCP and nasal regurgitation. Widely promoting the understanding of this relationship among clinicians could lead to an increase in early referrals of patients with SMCP to suitable facilities. Moss suggested that nasal regurgitation and slow feeding rate combination strongly indicate SMCP⁶. Although we could not investigate feeding time, clinicians should pay attention to nasal regurgitation and feeding time.

Relationship between nasal regurgitation and VPF

We hypothesized that patients with SMCP with nasal regurgitation had worse VPF and required surgical interventions. This hypothesis concurred with our study results. The patients in our series presenting with episodes of nasal regurgitation in infancy also presented with worse VPF than those without nasal regurgitation episodes. Eighty-six percent of patients with nasal regurgitation required surgery to improve their VPF, with only 39% of these patients exhibiting improvements in VPF beyond "good" and "normal." All patients without episodes of nasal regurgitation who underwent surgery (i.e., patients whose VPF before surgery was undeterminable were excluded) exhibited improvements postoperatively. The postoperative VPF of patients with nasal regurgitation was significantly worse than that of those without nasal regurgitation (Table 5). From these results, nasal regurgitation could be considered a surgical indication and indicate a refractory response.

Our hypotheses implied that the early detection of SMCP can lead to early surgical interventions and improvement of VPF. However, controversial opinions exist regarding the

surgical timing in patients with SMCP. Oji recommended an early operation to improve VPF¹². McWilliams mentioned in their study that 44% of patients diagnosed with SMCP did not require an operation¹⁴. Furthermore, speech outcomes were the same between the patients operated at < 4 years and > 4 years of age. Some studies reported that only 5%-10% of patients with SMCP become symptomatic (with VPI)^{8,15,17,25}; thus, early surgeries are not always recommended. Therefore, the following two main opinions exist: Recommendations for early surgical intervention, and recommendations for delaying surgery until a speech evaluation is completed.

However, we need to detect SMCP patients early to operate on them during infancy. The most common reasons for referral to the clinic are speech disorders and anatomical abnormalities. However, speech evaluation remains underdeveloped in infancy; therefore, the early detection of anatomical abnormalities is important for early surgical interventions. However, detecting anatomical abnormalities in infancy is difficult for clinicians. Dam mentioned that in 83% of patients receiving an oral operation in their facility, the bifid uvula was undocumented¹.

The existence of different diagnostic criteria for SMCP and inclusion criteria of patients with or without ID and syndromes among reported studies created confusion about the results of the surgical interventions. Thus, several opinions about the timing of surgical interventions among SMCP patients, and several diagnostic criteria of SMCP are present. To investigate the best timing of surgical interventions among patients with SMCP, a unified diagnostic criteria for SMCP is needed to identify patients with SMCP and to precisely determine the effect of surgeries.

Our study data suggested that a history of nasal regurgitation in infancy indicated surgical intervention among patients with SMCP. However, a history of nasal regurgitation did not result in early hospitalization among patients with SMCP. To understand the relationship between nasal regurgitation, the timing of referral to hospitals, the relationship between nasal regurgitation and VPF, and the necessity for surgical interventions, we must conduct a similar study on several patients with unified diagnostic criteria of SMCP. The number of patients referred to the clinic early in infancy must be increased, allowing pediatricians to elucidate the relationship between nasal regurgitation, early detection of SMCP, and severity of SMCP.

Conclusion

A history of nasal regurgitation was not associated with the early detection of SMCP. However, clinicians might not be familiar with the relationship between SMCP and nasal regurgitation, and an increased understanding of this relationship could allow SMCP detection early. Furthermore, a history of nasal regurgitation may be a key factor in determining the surgical requirements and may indicate the difficulty of VPI treatment. Further clinical investigations are needed using unified diagnostic criteria of SMCP.

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Ethical Approval: This study was approved by the Chiba Children's Ethical Committee (Approval number: 2020-024)

Informed Consent: Informed consent was not required for this study because this study does not include any pictures, names, and other patients' identifiable information.

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