

## Case Report

## Surgical Premaxillary Setback with Vomerine Ostectomy for Complicated Bilateral Cleft Lip

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*Department of Plastic and Reconstructive Surgery, Tokyo Medical University, Tokyo, Japan***Abstract**

Important factors that affect the initial repair of complete bilateral cleft lip (CBCL) include not only the width of the cleft but also the distance between the premaxilla and alveolar bone. We report a case of a patient with CBCL in whom favorable morphological and aesthetical results were achieved by surgical premaxillary setback. The patient was a 5-month-old boy with CBCL, who had a severely protruding premaxilla. Despite repeated lip adhesion before lip closure surgery, wound dehiscence occurred, and therefore, we performed premaxillary setback with vomerine ostectomy. The bone was removed posterior to the vomero-premaxillary suture, and the premaxilla was indirectly stabilized by bilateral mucosal bridging. The procedure for the vomer worked well, and his premaxilla was moved back enough to undergo surgery. The premaxillary setback is thought to be a reasonable option to treat CBCL in patients with a protruding premaxilla. The subsequent long-term follow-up of the midfacial development of these patients is necessary.

**Keywords**

bilateral cleft lip, premaxillary setback, vomero-premaxillary suture (VPS), vomerine ostectomy

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## Introduction

In the initial repair of complete bilateral cleft lip (CBCL), it is favorable to align the premaxillary segment with the lateral alveolar segments. When there is severe protrusion of the premaxilla, the tension on the repaired lip can become strong, which may lead to the inability to perform aesthetic reconstruction of the nasolabial components or functional closure of the orbicularis oris muscle. Not only is a small width of the cleft but also the short distance between the premaxilla and alveolar bone crucial for a successful surgery. In some cases, preoperative taping or devices are used to reduce the cleft width, but they may be ineffective in CBCL patients with extremely protruding premaxilla.

In this study, we report a case of a patient with CBCL in whom favorable morphological and aesthetical outcomes were achieved by surgical premaxillary setback followed by DeHaan's method<sup>1)</sup> for cleft lip closure.

## Patient and Methods

*Case*

In this study, we examined a 5-month-old boy with CBCL, who had a severely protruding premaxilla (**Figure 1**). In a fetal screening test at 21 weeks of gestation, the patient was identified as having anophthalmia and cleft lip and palate. He was referred to our hospital at 27 weeks of gestation and was born at 38 weeks; he was then admitted to the neonatal intensive care unit.

The patient was born with several comorbidities: i.e., severe neonatal paralysis, arteriovenous patency (spontaneous closure), multiple malformations (anophthalmia, nasal defects, obstruction of the external auditory canal, low auricle, and congenital subglottic stenosis), vesicoureteral reflux, left cryptorchidism, and gastroesophageal reflux. He was tracheotomized at 14 days of age owing to subglottic stenosis. Vesicoureteral reflux was about grade 4 to 5 on the left and grade 2 on the right, and the patient was considered to require surgery on a standby basis. Although gastroesophageal reflux was present, he showed steady weight gain with en-



**Figure 1.** Preoperative findings.  
Severe protrusion of the premaxilla was observed.

teral nutrition. After consulting with the pediatrician, it was determined that his gastroesophageal reflux and vesicoureteral reflux did not require immediate surgery, so the plan was to perform cleft lip surgery first. The cleft lip surgery was postponed once owing to a urinary tract infection; thus, we decided to perform voiding cystourethrogram for vesicoureteral reflux after the cleft lip surgery. Systemic examination revealed no other abnormalities, such as cardiac malformations. He did not have any family history of congenital anomalies.

### Methods

Before the operation, computed tomography (CT) imaging was performed to plan the surgery. Because of the patient's multiple facial malformations, it was necessary to understand the association between the bones to be removed and the base of the skull.

Under general anesthesia, the procedure was started with vomerine osteotomy. A part of the bone posterior to the vomero-premaxillary suture (VPS) was removed. The lower end of the nasal septum was approached from the oral cavity and was incised, and the exposed vomer was then chipped away. To correct the angle of the premaxilla, a wedge-shaped osteotomy was performed, and acquirement of mobility was confirmed. The position of the premaxilla was corrected in the posterior direction by manual manipulation and fixed with steel wire to the alveolar bone (**Figure 2**). After sufficient retraction of the premaxilla was achieved (**Figure 3**), lip repair was performed using DeHaan's technique followed by rhinoplasty. Finally, his lip was closed with favorable aesthetic outcomes (**Figure 4**). The patient did not have any major complications during and after the surgery.

### Discussion

In some previous studies, various devices and/or surgical techniques have been reported to correct protruding or de-

formed premaxilla in patients with CBCL<sup>2-9</sup>.

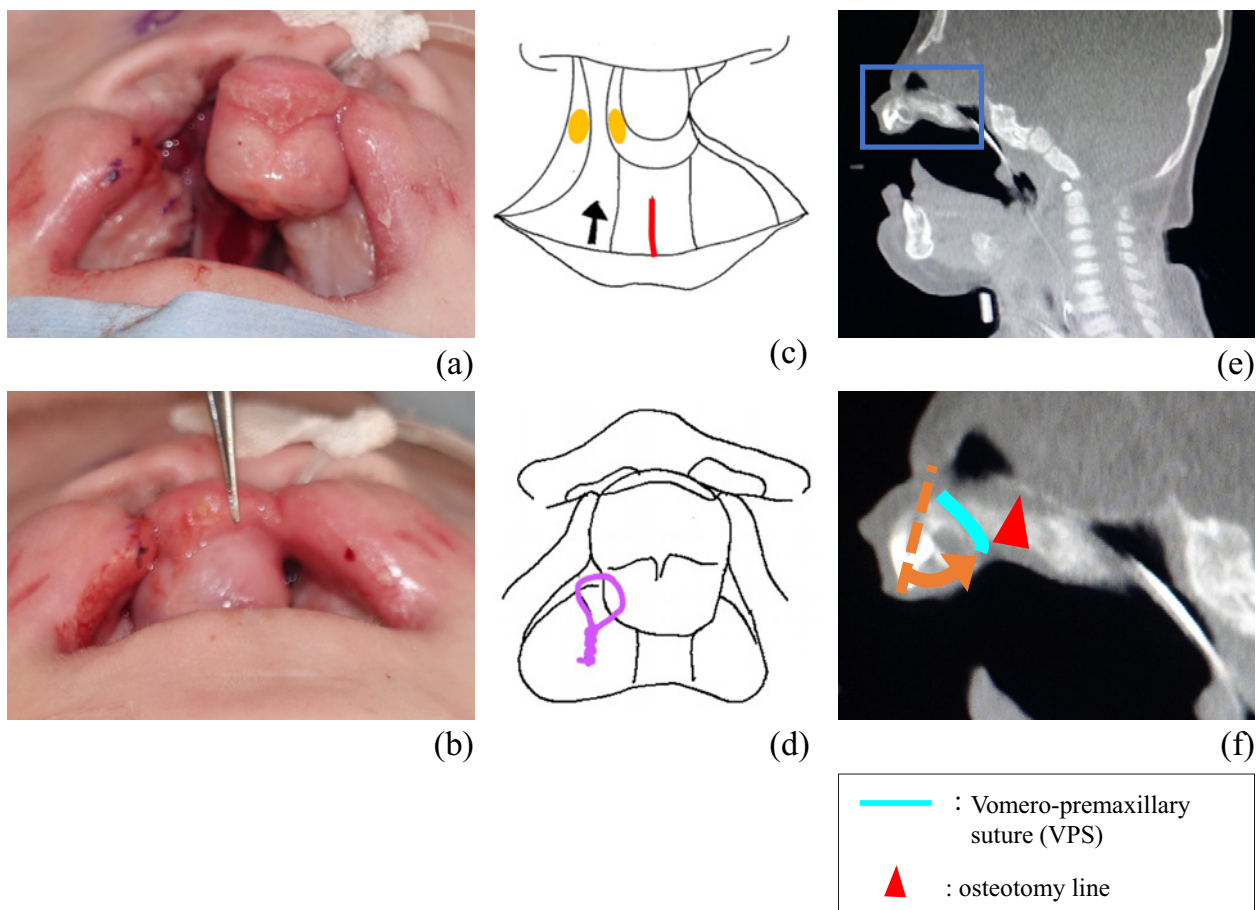
For example, Chung et al. performed the intraoperative "rapid premaxillary molding" technique to patients with complete bilateral cleft lip and palate and obtained satisfactory results<sup>2</sup>. Their method is as follows: intermittent manual compression to the premaxilla is applied until mobility is achieved. During the process, care should be taken not to apply excessive pressure to prevent fracture of the premaxilla or vomer. After the premaxilla is sufficiently retracted, the lips and nose are formed.

In our present patient, the premaxilla was initially retracted manually and fixed with a wire needle. However, the tension between the premaxilla and lateral lip was too tight, which resulted in wound dehiscence twice. To overcome this situation, we performed premaxillary setback by vomerine osteotomy.

Before discussing osteotomy, it is important to consider embryology. The anterior part of the hard palate is of primary palatal origin, whereas the posterior part is of secondary palatal origin (**Figure 5**). Therefore, from the perspective of embryology, it is deemed reasonable to separate the anterior approach from the posterior one with the VPS as the reference spot.

There have been some previous reports of vomerine osteotomies using a similar technique to premaxillary setback. In 2018, Park reported that they performed premaxillary setback on 12 patients with bilateral cleft lip and palate, approaching from anterior or posterior to the VPS, depending on each patients' situation<sup>7</sup>. On the same year, Almas also reported that they were able to perform premaxillary setback on 41 patients, approaching from posterior to the VPS<sup>8</sup>. Furthermore, in 2015, Fakihi-Gomez reported that they performed premaxillary setback approaching from anterior to the VPS in four patients<sup>9</sup>.

Comparing osteotomy approaches performed from anterior and posterior to the VPS (**Table 1**), it was found that correcting a premaxillary angle when approached from anterior to the VPS is technically more difficult. In addition, an



**Figure 2.** Premaxillary setback procedure.

(a, c) The lower end of the nasal septum was excised, and the exposed vomer was drilled, and a part of it was removed. (e, f) To correct the angle of the premaxilla, a wedge-shaped osteotomy was performed from behind the VPS. (b, d) The position of the premaxilla was corrected in the right posterior direction by manual manipulation and fixed with a wire.

anterior approach to the VPS may inhibit blood flow in the premaxilla. On the other hand, a posterior approach may lead to midfacial growth disorder as the VPS is thought to be a region involved in midfacial development, and surgical manipulation may result in growth retardation. Regarding whether the VPS is actually involved in midface development, Friede et al. performed a study<sup>11)</sup>, wherein metallic implants were inserted on both sides of the VPS in eight infants with unilateral cleft lip and palate at the time of the first surgery. The patients were followed roentgencephalometrically until the age of 3 years old. Growth between the vomer and premaxilla was recorded in every patient. It was noted that the horizontal growth between the pins was greater than the vertical growth. Therefore, based on the risk of each patient, surgeons must choose the appropriate osteotomy line.

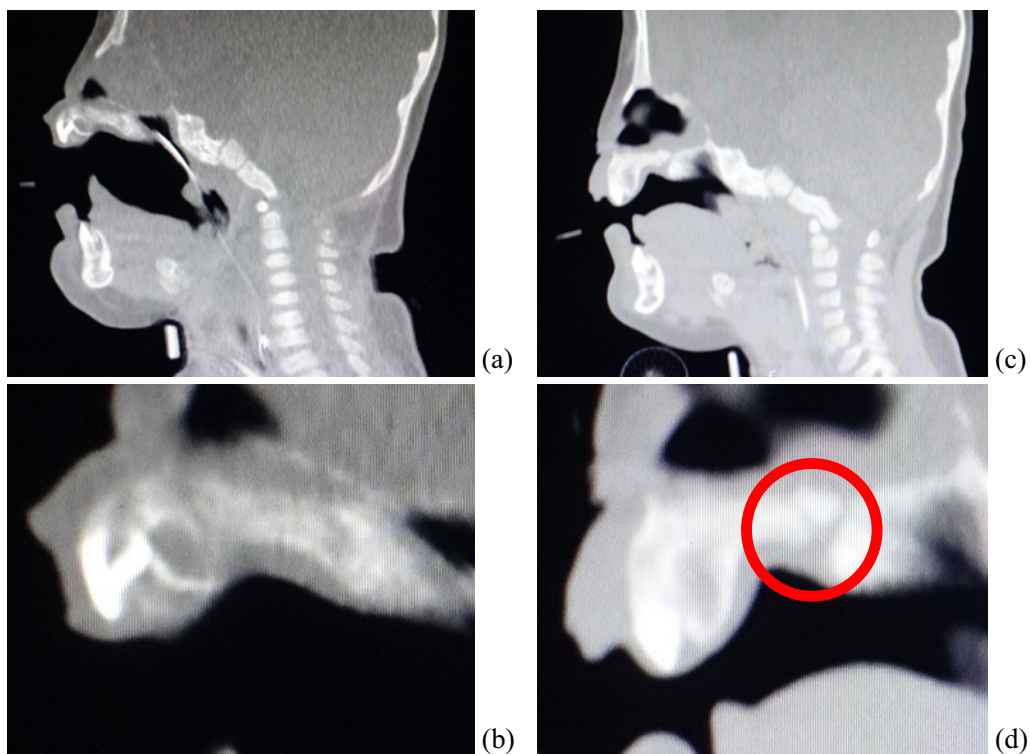
In our present patient, as the distance between the premaxilla and alveolar bone was wide, surgical premaxillary setback was thought to be the most favorable method. Taking blood flow into consideration, the choice of a posterior approach to the VPS is deemed reasonable. As the patient's prognosis was expected to be unfavorable, owing to his food intake disturbance and tracheotomy, this approach was considered to be the optimal at this time, even if it had the risk

of future midface hypoplasia. As the VPS may be involved in the development of the midface, it is necessary to carefully follow up the patient's midface growth.

Before surgery, the patient's premaxilla protruded 12 mm more than the right lateral segment and 8 mm more than the left lateral segment. The projection of the premaxilla was measured from the alveolar arch of the lateral segment to the premaxillary arch on CT images. It has been reported that patients with bilateral cleft lip with a protruding premaxilla of more than 10 mm achieved noninferior lip symmetry after premaxillary setback with vomerine osteotomy as patients with less severe original deformities<sup>12)</sup>.

This study has limitations. For one, this only reports on one case; thus, we need to accumulate more cases in which the patient has a protruding premaxilla that appears to be difficult to repair via the usual process. In addition to protrusion of the premaxilla, facial deformities, such as hypoplasia of the lateral segments, may also be a factor that made repair difficult in this patient. However, we believe that this report is valuable, as there are only a few reports, to date, of surgeries performed on patients with both cleft lip and anophthalmia<sup>13)</sup>. Further studies should be performed in the future on the combined factors to determine what combinations of factors affect surgery.





**Figure 3.** Comparison of preoperative and postoperative CT images. Preoperative (a, b) and postoperative (c, d) images are shown. The premaxilla was markedly retracted after surgery (c, d). Vomerine osteotomy was performed in the area indicated by the red circle in (d).



**Figure 4.** Postoperative findings. Favorable shape of the lips without any conspicuous scars was achieved.

## Conclusions

In this study, we examined a case of a patient in which favorable lip appearance was obtained by surgical premaxillary setback followed by DeHaan's method. As per our results, bilateral cleft lip with anterior protrusion of the premaxilla may have a favorable outcome when the premaxilla is retracted prior to cleft lip repair. As the VPS is thought to be a region associated with the development of the midface, extra caution must be exercised to make sure not to injure the VPS during surgery. Long-term follow up that is focused on the development of disorders in the VPS is hence neces-

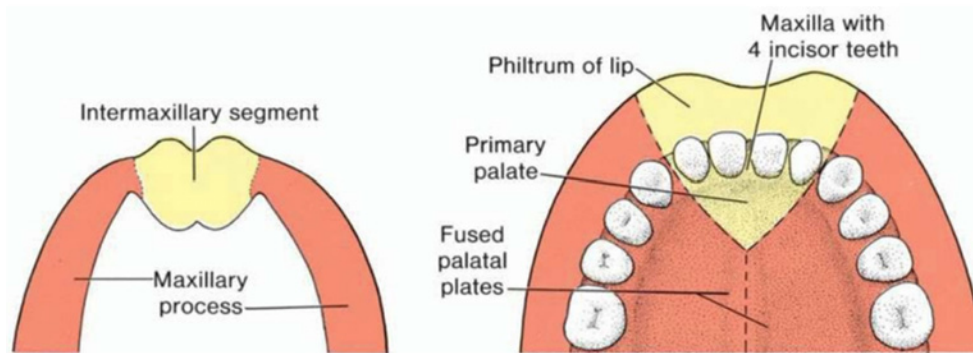
sary.

**Author Contributions:** Y.Od., Y.Oj., N.A., M.A., Y.I., and H.M. provided care to the patient and performed the surgery. Y.Od. collected and analyzed the previous papers. Y. Od. and Y.Oj. designed the study and wrote the manuscript. H.M. supervised the study.

**Conflicts of Interest:** There are no conflicts of interest.

**Ethical Approval:** Written informed consent was obtained from the patient.

**Consent to Participate:** The patient's parents provided their written informed consent to participate in this study.



**Figure 5.** Embryology of palates.

The anterior part of the hard palate is of primary palatal origin, whereas the posterior part is of secondary palatal origin. The figure was from *Langman's Medical Embryology*<sup>10</sup> and was modified by the author with permission from *Lippincott Williams & Wilkins*.

**Table 1.** Comparing the Pros and Cons of the Anterior and Posterior Approaches.

Osteotomy line	Anterior to the VPS	Posterior to the VPS
Correction of the premaxillary angle	Difficult	Not difficult
Blood flow disturbance in the premaxilla	Likely to occur	Less likely to occur
Midfacial developmental disorder	Less likely to occur	Likely to occur

**Consent for Publication:** The patient's parents provided their written informed consent for the publication of this study including patient's photo release permission.

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