Surgical Management of Median Cleft Lip Extending as Far as Alveolus Using Bone Grafting

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Abstract

Median clefts, which run through the midline of the upper lip, and alveolus are considered a type of craniofacial cleft. A Tessier number 0 median cleft lip is the rarest, and there are few reports concerning its surgical management. Here, we describe the case of a 6-year-old boy who presented at our hospital with a median cleft and alveolus of the upper lip with a shortened bifid frenulum. Inverted-V and Z-plasty incisions were performed for the median cleft and iliac bone grafting for the midline alveolus. At 1 year 5 months later, the postoperative course was uneventful, and proper approximation of the orbicularis oris muscle and vermilion were achieved, with a symmetrical Cupid’s bow and labial tubercle and philtrum.

Key words: Median cleft lip — Tessier number 0 cleft — Inverted-V incision — Z-plasty incision — Iliac bone grafting

Introduction

Craniofacial clefts are rare, with an incidence of approximately 1.4 to 4.9 per 100,000 live births5). One type of such cleft, the median, runs through the midline of the upper lip. Median clefts in the upper lip have been reported to occupy between 0.43% and 0.73% of all craniofacial clefts5). This type of cleft is caused by malfusion of the frontonasal processes, or migration of the mesoderm at the midline6). Clinically, it presents in a variety of forms, including notching of the upper lip, vermilion, and nose, in addition to as an alveolar bone defect. A number of techniques have been reported in the repair of mild, or moderate true median cleft lip1,8,9). Few studies have investigated the surgical management of median cleft lip, however. Here, we report the surgical management of a case of median cleft lip and alveolus in which the postoperative course was uneventful. A sym-
metric philtrum and Cupid’s bow on the vermillion, as well as a good labial tubercle, were obtained.

**Case Presentation**

The patient was a 6-year-old boy who presented at Tokyo Dental College Suidobashi Hospital in May 2014 with the chief complaint of a maxillary median diastema. The patient was normal weight at birth, and there was no evidence of any other physical anomaly. The median diastema had been examined by the family dentist when the boy was aged 5. This dentist lacked the knowledge necessary to diagnose his facial malformation, however. The patient had no family history of clefts. On examination at our hospital, a median cleft of the upper lip and alveolus with a shortened bifid frenulum were identified. The vermillion was separated at the midline. The peak-to-peak distance of the Cupid’s bow was 12 mm, with 3 mm of epithelium at the midline (Fig. 1a, b). A defect at the alveolar midline was observed on preoperative computed tomography (CT) images (Fig. 1c). A general physical examination including an electrocardiogram revealed nothing abnormal. An intraoral examination revealed open bite. The occlusal relation was Class II with +7 mm overjet and −4 mm overbite. The diagnosis was a Tessier number 0 cleft, and the patient was subsequently referred to the Department of Orthodontics for occlusal management in July 2014.

Repair of the median cleft lip and alveolar bone graft were performed under general anesthesia in July 2017. The cleft lip and alveolus were reconstructed using the following method. An inverted-V incision was made intraorally over the white roll, vermillion, and the labial mucosa, extending just beyond the cleft margin (Fig. 2). Epithelium around the midline was removed. The orbicularis oris muscle was undermined and repositioned.
using 5-0 bio-absorbable sutures. The skin was sutured in a straight line using 6-0 nylon sutures. The frenulum was released using a Z-plasty incision (Fig. 3). The Z-plasty flaps of the frenulum were sutured with 5-0 bio-absorbable sutures. Bone grafting was performed on the alveolar midline defect (Fig. 4) by harvesting cancellous bone from the iliac bone. The patient was satisfied with the surgical results. The Cupid’s bow was properly aligned, with equal height bilaterally; the vermilion form was satisfactory; and the fullness and continuity of the orbicularis oris were maintained (Fig. 5).

After 1 year 5 months, in November 2018, postoperative 3D-CT images confirmed successful bone grafting at the midline. The surgical region healed successfully, and there were no complications such as infection, dehiscence, or hypertrophic scar (Fig. 6).

**Discussion**

A median cleft is defined as one running vertically through the midline of the upper lip. This type of cleft is a rare anomaly, and the exact developmental origin is unclear. It is believed, however, to be caused by malformation of the medial nasal prominences during the third week of gestation. Although the present patient’s family dentist was unable to diagnose the malformation, number 0 clefts are actually the most common form of craniofacial cleft. Craniofacial clefts are rare, with an incidence of about 1.4 to 4.9 cases per 100,000 live births. Midline cleft lips and noses are designated number 0 to 14 under the Tessier classification. They are rare, however, and are considered an anomaly, occurring in only between 0.37% and 0.73% of all clefts.

Median clefts may be classified according to any of several systems. In 1937, Veau identi-
fied 3 types of median cleft: a notch of the lip; a cleft extending as far as the columella; and a defect resulting from atrophy of midline facial structures\textsuperscript{19}. DeMyer described 2 groups of facial anomalies associated with either orbital hypotelorism or hypertelorism\textsuperscript{3}. In the present case, the patient had a median cleft of the upper lip and alveolus with a shortened bifid frenulum. No orbital hypertelorism was observed.

Radiographic images play a very important role in the diagnosis of alveolar disorders and bone defects, and preoperative and postoperative 3D-CT images are also required for an accurate evaluation. In the present case, the boy was 8 years old at the time of surgical

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Fig. 4 Bone grafting performed on midline defect of alveolus
Iliac bone was used for bone grafting.

Fig. 5 Closure of skin and mucosa
Closure of skin and mucosa with natural lip was achieved.
intervention. Generally, soft tissue defects should be repaired when a child is 1-year old. The optimal timing for bone grafting remains to be established, however.

In the present patient, reconstruction of the Cupid’s bow, labial philtrum, vermillion, and alveolar bone defect were considered when developing the treatment plan. Very few studies have reported the surgical techniques required for median clefts. Some have reported using a V-Y flap, an inverted-V incision, a Z-plasty incision, or Pfeifer incision during its surgical management, however\(^1\). Although a V-Y flap can be used to extend a shortened bifid frenulum, it is insufficient for reconstruction of the labial tubercle. A Pfeifer incision is made in short-curved waves around the defect, which help expand the length and width of the tissue when sutured to form a straight line. The results can be unpredictable, however.

In the present case, an inverted-V incision and a Z-plasty incision were made to reconstruct a mild true midline cleft. For mild-to-moderate defects involving the philtrum, Millard recommended an inverted-V incision at a 90-degree angle, 2 mm above the mucocutaneous white roll on each side of the cleft in order to lengthen the skin at the midline of the Cupid’s bow\(^7\). In the present case, the Z-plasty incision helped expand the length of the midline tissue, making it possible to achieve a Cupid’s bow and vermilion with a natural appearance. It is important to identify the areas affected by a facial defect, as this will allow optimal selection of surgical techniques required for proper management. While combining various techniques, however, it is also important to consider the bloodstream of the flap. To the best of our knowledge, no long-term studies to date have investigated median cleft lip and alveolus. Studies including long-term follow-up are needed to further establish the validity of such combinations of surgical techniques.

Fig. 6 Postoperative follow-up at 1 year 5 months
a, b: Scar is unnoticeable.
c: Successful bone grafting for midline defect of alveolus.
References


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