Incidence and Contributing Factors for Palatal Fistula after Primary Palatoplasty in a Medical Collaboration Program in Madagascar

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Abstract: We have been conducting medical collaboration programs for cleft lip and palate in the Republic of Madagascar for seven years, from 2011 to 2017. The purpose of this study was to investigate the incidence of fistula after primary palatoplasty in the Republic of Madagascar, which is a developing country. A further objective was to determine if the group of patients in Madagascar had an increased incidence of palatal fistula compared to a similar group of patients in Japan, and to examine the factors that might be involved in any increase. We conducted a survey of the cleft type, age at time of surgery, and fistula incidence in 44 patients (28 males and 16 females) in Madagascar. The age at the time of surgery was 11 months to 29 years (average, 7 years and 8 months), and the cleft types were 0 class I, 14 class II, 18 class III, and 12 class IV, by the Veau classification. We used a modified two-flap palatoplasty for palatal closure. In addition to the selection of surgical technique, the factors which are suggested to influence the incidence of fistula include the experience of the surgeon, and the extent of the cleft. The incidence of postoperative fistula complication in these patients was zero.

In medical collaboration settings in developing countries, there are factors which may delay wound healing, such as poor oral hygiene, poor nutrition, and instability of the flap blood flow. Understanding such factors in surgery is important to avoid palatal fistula. We report that a palatal fistula incidence rate of 0% can be achieved by avoiding those factors which contribute to fistula formation.

Key words: cleft palate, palatal fistula, two-flap palatoplasty, developing country, Madagascar

Introduction

Cleft lip and palate are the most frequent anomalies among congenital malformations of the face. There is a racial difference in the frequency of occurrence, with about 1 case in 500 births in Asians. The incidence of only cleft palate is about 1 case in 600–700 births. The surgical goals of palatoplasty are complete closure, velopharyngeal competence and normal maxillary growth. Of these three, the latter two are viewed as medium- to long-term goals, but complete

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closure of the palate, that is, the presence or absence of a fistula after palatoplasty, is useful as a short-term indicator of the outcome\textsuperscript{1}. In addition, the presence of a palatal fistula causes speech problems, problems with food and drink coming out of the nose and becomes a negative factor in oral hygiene. It has been suggested that a palatal fistula after repair is related mainly to the extent of the cleft and the experience of the operating surgeon\textsuperscript{2}.

Medical collaboration programs for cleft lip and palate in developing countries have been implemented by several developed nations. However, palatoplasties performed in developing countries as part of a medical collaboration program have a significantly higher incidence of fistula than those performed in developed countries, even when performed by the same surgeon\textsuperscript{3}. Certainly, controlling the increased fistula incidence is clinically very important in developing countries, and considering the factors related to the increase is essential for achieving this outcome.

We have been conducting a medical collaboration program for cleft lip and palate in a developing country, the Republic of Madagascar, for seven years, from 2011 to 2017. The purpose of this study was to investigate the incidence of fistula after primary palatoplasty in the Republic of Madagascar. In addition, we examined whether the fistula incidence was increased compared to a group of patients in Japan, and we considered the factors that might be involved in any increase.

**Materials and methods**

**Patients**

The study participants were 44 consecutive patients who underwent primary palatoplasty for cleft palate from among 133 patients with conditions related to cleft lip and palate who underwent surgery at the Ave Maria Hospital in Antsirabe, Madagascar in about one week of each year from May 2011 to September 2017. The patients included 28 males and 16 females, and the age at surgery ranged from 11 months to 29 years, with an average age of 7 years and 8 months. The age distribution of the 44 patients is shown in Fig. 1.

![Age distribution at the time of primary palatoplasty](image)

**Fig. 1.** Age distribution at the time of primary palatoplasty. Surgery was performed at 1–2 years of age (the optimal time for primary palatoplasty) in 17 patients (38.6%), and at 3 years of age or more in about 60% of patients. The average age at primary palatoplasty was 7 years and 8 months.
The types of cleft lip and palate in the 44 patients, according to the Veau classification, are shown in Table 1.

Palatoplasty for cleft palate

All surgeries for cleft palate were performed under general anesthesia, according to the modified two-flap palatoplasty method reported by Tosa et al. (Fig. 2). In the hard palate, the oral mucosa is divided into three layers: nasal cavity mucosa, muscle, and oral mucosa. In the nasal mucosa, a Z-plasty is performed. The nasal mucosal periosteal flap is sutured using a Z-plasty. The muscle is sutured by an intravelar veloplasty, and the oral mucosa is sutured with 5-0 vicryl.

Table 1. Veau classification and distribution of cleft type

<table>
<thead>
<tr>
<th>Class</th>
<th>Extent of cleft</th>
<th>No. of Patients (%)</th>
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<tbody>
<tr>
<td>I</td>
<td>Soft palate only</td>
<td>0 (0)</td>
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<tr>
<td>II</td>
<td>Hard and soft palate</td>
<td>14 (31.8)</td>
</tr>
<tr>
<td>III</td>
<td>Unilateral cleft lip and palate</td>
<td>18 (40.9)</td>
</tr>
<tr>
<td>IV</td>
<td>Bilateral cleft lip and palate</td>
<td>12 (27.3)</td>
</tr>
<tr>
<td>Total</td>
<td></td>
<td>44 (100)</td>
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Fig. 2. Illustration of the modified two-flap technique by Tosa et al. (a) Design of the modified two-flap technique. (b) The soft palate is divided into three layers of nasal cavity mucosa, muscle, and oral mucosa. In the nasal mucosa, a Z-plasty is performed. (c) The nasal mucosal periosteal flap is sutured using a Z-plasty. (d) The muscle is sutured by an intravelar veloplasty, and the oral mucosa is sutured with 5-0 vicryl.
mucosa was incised about 3 mm from the alveolus on the lateral part, and the cleft was incised with a No. 15 scalpel at the boundary of the oral mucosa and nasal mucosa. The soft palate was divided into three layers of nasal mucosa, muscle, and oral mucosa. In the nasal mucosa, Z-plasty was performed, and the muscular layer was sutured in three layers according to the intravelar veloplasty technique. Composite muscles (levator and other muscles) were moved back as far as possible toward the posterior position and sutured at the center of the cleft to form a muscular sling. The oral mucosa was completely sutured to the anterior hard palate.

Follow-up system after palatoplasty

The follow-up period after palatoplasty averaged 3 years and 9 months, and ranged from 3 months to 6 years and 6 months. The postoperative palatal condition was always checked before individual patients were discharged. Thereafter, the palatal condition was rechecked by our team in subsequent years and by a Japanese nun qualified as a midwife working at the Ave Maria Hospital.

Location of palatal fistula

The Pittsburgh fistula classification system (seven types from type I to type VII) was used to classify the location of the fistula (Fig. 4). A type I fistula is a bifid uvula. A type II fistula is in the soft palate. A type III fistula is at the junction of the soft and hard palates. A type IV fistula is in the hard palate. A type V fistula is at the incisive foramen, at the junction of the primary and secondary palates; this designation is reserved for Veau IV clefts. A type VI fistula is in the lingual-alveolar region. A type VII fistula is in the labial-alveolar region.

Size of palatal fistula

The size of fistulas was graded as small (1–2 mm), medium (3–5 mm), and large (5 mm or more), according to the report of Muzaffar et al.

Statistical analysis

The patients’ sex, age at time of surgery, and type of cleft palate according to the Veau classification were analyzed statistically using the Fisher test, with a hazard ratio of 5% taken as a significant difference.

Results

No patient was returned to the operating room due to bleeding that could not be controlled immediately after surgery. No patient received a blood transfusion.

Follow-up system after palatoplasty

Postoperative follow-up was accomplished for all 44 patients, and none of the 44 patients had complications resulting in fistula formation, and thus the incidence of postoperative fistula compli-
Fig. 3. Repair using the modified two-flap technique (Madagascar patient). (a) Design of the modified two-flap technique. (b) Elevation of the two mucoperiosteal flaps. (c,d,e) In the nasal mucosa, a Z-plasty is performed. (f) A muscle sling is performed. (g) After the palatoplasty is finished, there is no raw surface on the anterior portion of the hard palate.
The occurrence of palatal fistula by cleft type according to the Veau classification was 0/14 patients with class I, 0/18 patients with class III (left 0/10, right 0/8), and 0/12 patients with class IV.

**Statistical analysis**

Since there were no cases of palatal fistula in the patients of this study, statistical analysis of each factor such as sex, age at time of surgery, cleft type, location of fistula and operating surgeon was judged to have no significance and was not carried out.

**Discussion**

The incidence of cleft lip and palate differs by race. The ancestral population of Madagascar is mainly Merina and Betsileo, who are Malay ethnic groups that came from Indonesia. Therefore, although they are located in Africa, the incidence of cleft lip and palate in the people of Madagascar is as high as in Asians. At present, the country has almost no palatoplasty specialists for cleft palate repair, therefore, medical collaboration with humanitarian organizations from developed countries is needed for cleft lip and palate surgery. Our group has been providing medical collaboration for cleft lip and palate repair at the Ave Maria Hospital in Madagascar’s
third largest city, Antsirabe. Fortunately, there is a Japanese nun working full-time there, and the system allows almost complete follow-up before and after surgery. Therefore, conditions are better than those described in a report published by a medical collaboration group that is working in a similar environment.

We investigated the incidence of palatal fistula, which is an early complication after primary palatoplasty, in 44 patients treated in Madagascar over a 7-year period, in which 100% follow-up during the acute phase was possible. Numerous papers have reported the incidence of palatal fistula after primary palatoplasty, with rates ranging from 0% to as high as 77.8% (1-10). In patients where a palatal fistula exists immediately after the primary palatoplasty, there is a strong tendency for the palatal fistula to still be present at the time of secondary bone grafting for alveolar cleft (15). Depending on the severity of the fistula, it may be necessary to plan a secondary palatal fistula closure to manage the problem. However, in secondary palatal fistula closure, because of the high risk of a fistula remaining after surgery, avoiding a postoperative fistula at the primary palatoplasty is an important objective (16, 17).

There are differences in the occurrence of palatal fistulas depending on the surgical technique used, but the main methods of palatoplasty are the pushback, Furlow, and two-flap techniques. Each method has advantages and disadvantages.

The pushback technique is a method reported by Veau in 1931 (4) and then by Wardill and Kilner in 1937, in which the cleft margins are sutured with mucoperiosteal flaps of the hard palate with the left and right greater palatine vessels as vascular pedicles, and pushed back (Fig. 5) (18-20). At the anterior region of the hard palate, since a raw surface is left on the side of the oral cavity by suturing only one layer of the nasal mucoperiosteum, it has the disadvantage that the raw surface heals by secondary granulation and scarring leads to suppression of maxillary growth.

The Furlow technique is a method reported by Furlow in 1986 as a double opposing Z-plasty, in which a Z is formed on the nasal side and the oral side to close the cleft (Fig. 6) (21, 22). Since Z-plasty is performed on the oral and the nasal sides of the soft palate, there is a risk that the tip of the mucosal flap will become necrotic due to blood flow disturbance.

The two-flap technique is a method reported by Bardach in 1967, in which two large mucoperiosteal flaps in the oral cavity are elevated, sutured in 2 layers to the hard palate and in 3 layers to the soft palate; it is a procedure that does not leave a palatal mucosal defect at the anterior region of the hard palate (23). Salyer et al (24) reported that the two-flap technique has shown good results over the course of 20 years. Currently it is the most selected surgical technique in America, and it is the most selected surgical technique in Asian countries, other than Japan. The flap has high blood circulation stability, and normal closure of the palatal muscular sling is possible.

Of 300 patients with complete unilateral cleft lip and palate who underwent primary palatoplasty in our facility in Japan during the 34 years from 1978 to 2012, there were no complications of hearing loss or delayed language development with surgery at 12-18 months of age. Of these patients, 234 (174, pushback technique; 38, Furlow technique; and 22, two-flap technique)
could be evaluated linguistically over time after surgery. According to survey results from these 234 patients, palatal fistula occurred in 81 patients (47%) with the pushback technique, 4 patients (11%) with the Furlow technique, and in no patients with the two-flap technique²⁵).

Many patients undergoing voluntary surgery in developing countries have poor nutritional status and poor oral hygiene due to poverty. For this reason, there are many patients undergoing such surgery where wound healing is generally poor, as the blood circulation to the wound is
bad and postoperative granulation does not proceed well. In the Furlow technique, this can lead to necrosis of the mucosal flap or poor filling of the dead space due to defective granulation, and a fistula is easily left after surgery. The mucosal flaps of both layers often become necrotic at the same time, and a fistula easily forms since granulation is poor (Fig. 7). In fact, in Nepal, where we participated in a medical assistance program before going to Madagascar, we encountered fistula development such as we had never experienced in Japan\(^9\).

In the pushback method performed in Nepal, the mucoperiosteal flap that is found after ‘push back’ is liable to leave a fistula due to failure to unite. Furthermore, in this method, since a raw surface remains after the operation, the vascular stump is exposed and not covered by granulation, and together with a fibrinolysis phenomenon in the exposed vascular stump, there may be repeated episodes of bleeding for about 1 week after surgery (Fig. 8). Also, the postoperative pain and irritation caused by food and drink is strong, making ingestion of meals difficult. Therefore, in such countries, the use of a flap with a high risk of poor blood circula-
tion, as with the Furlow technique, or surgical methods that leave a raw surface in the anterior region of the hard palate, as with the pushback technique, should be avoided. Selection of a simple two-flap technique which does not leave a raw surface strongly contributes to a marked reduction in the incidence of fistulas. In our work in Madagascar, we have been using a modified two-flap technique as the basic method of palatal closure since our first year working there in 2011. A modified two-flap technique enables a multiple-layer, tension-free, watertight closure which is important for minimizing the fistula rate^5. 

To confirm that the zero incidence of palatal fistula in this group was not due to patients having extremely narrow cleft widths, we measured 4 patients in 2016 and 6 patients in 2017 in whom a maxillary plaster model could be created. In the age groups at the time of surgery, there were 6 patients (4 males and 2 females) with deciduous dentition and 4 patients (2 males and 2 females) with permanent dentition. The method of measurement for deciduous dentition was to measure the cleft between the distal parts of the first primary molars, with reference to the measurement site defined by the Japanese Society of Pediatric Dentistry (Fig. 9)^26. For permanent dentition, the cleft between the distal parts of the first molars was measured with reference to the measurement site of Otsubo (Fig. 10)^27. Measurements were conducted using digital calipers that are accurate to 1/100 mm. The cleft width for deciduous dentition was 7.2–15.4 mm, averaging 11.9 mm, while the width for permanent dentition was 4.9–11.4 mm, averaging 9.1 mm. Furthermore, 69% of patients were categorized as Veau classifications III (18 patients, 41%) and IV (12 patients, 28%) which have a high fistula incidence, so the patient group did not consist only of cases of low difficulty.

The experience of the surgeon is also significant, and although three operations were performed by young residents with little experience, it was thought that their lack of experience had no effect on fistula development, since a supervising surgeon who guided and helped the residents was present as the first assistant during surgery from the beginning to the end.

**Perioperative Management**

Perioperative management immediately after surgery is also important as a part of after-treatment care. To reduce the chances of secondary infection, we instruct not only the patient but also the mother to wash and disinfect their hands, as they are highly likely to touch the palate area after the operation. Additionally, we advise them to increase their meals from partial rice porridge to whole rice porridge. We believe that these two instructions contribute to zero fistula incidence.

**Speech Results**

Surgery is desirable as early as possible to acquire normal speech function. Cleft palate is usually treated at our facility in Japan from the age of 12–18 months, as a rule. But in fact, the age of patients at the initial palate closure surgery was diverse in the study group from Madagascar.

If palatoplasty is performed and there are no other complications or mental delay, language
training is conducted with the aim of acquiring the same level of spoken language as normal children by the age of 4.

However, in developing countries there are no facilities that can provide specialized language guidance after palatoplasty, and there are no speech pathologists. Pastors or others provide guidance in actual practice, but linguistic evaluation cannot be done in developing countries, and with the differences in the linguistic system (pronunciation) between Japanese and other languages, the results of velopharyngeal sufficiency and articulation in Japan cannot be applied in these countries. According to Bardach, the creation of a muscle sling, rather than the length of the soft palate, is the important factor in language performance, and we are trying to implement this technique. In developing countries, we believe that the two-flap technique is advantageous for speech results as it can approximate the normal muscle course if it is executed reliably and forms a fistula-free palate.

**Maxillary development**

When the two-flap technique is selected for primary palatal closure, it is beneficial for maxillary development as it does not leave a raw surface at the anterior region of the hard palate. Furthermore, if there is no fistula in the primary palatoplasty, a secondary fistula closure can be avoided, and thus an increase in the number of surgeries which endanger maxilla development is also avoided.
Conclusion

In this study, we examined the incidence of fistulas as an early complication in 44 patients who underwent primary palatoplasty by the modified two-flap technique in Madagascar. The resulting incidence of palatal fistula was 0%. We considered the factors affecting the development of fistulas - in particular, factors contributing to the increased incidence of fistulas in medical collaboration programs conducted in developing countries, compared to the incidence of fistulas in our patients in Japan. The incidence of palatal fistula after primary palatoplasty in developing countries is generally higher than in developed countries, and a zero incidence has not been reported previously in developing countries.

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Conflict of interest disclosure

There are no conflicts of interest to declare concerning this study.

References


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