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Closure surgery using two-flap palatoplasty for oro-nasal fistula after removal of palatal pleomorphic adenoma

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An oro-nasal fistula is an abnormal passage formed between the maxillary sinus and the oral cavity. It is one of the complications that can occur after resection of a benign tumor in the palate, and it can occur despite successful surgery and proper postoperative management abiding by an appropriate protocol. When an oro-nasal fistula occurs, it can cause symptoms such as nasal discharge, reflux of water and food, and maxillary sinusitis. Oro-nasal fistulas that do not heal on their own must be reconstructed through a secondary surgery using palatoplasty. Pleomorphic adenoma is the most common benign tumor in the salivary glands, and when it occurs in the palate, it has an aggressive tendency. Because there is a possibility of recurrence and metastasis to malignancy, complete surgical resection of the tumor is required. In this study, we introduce two cases in which a secondary surgery using palatoplasty was performed to close a bone defect (oro-nasal fistula) that occurred after removal of a pleomorphic adenoma of the palate. The two cases were performed with the goal of: 1. Closing of the defect. 2. tension-free suturing to prevent recurrence. The two-flap palatoplasty technique was used in both patients and is reported because it has been performed successfully without recurrence to date.
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Key words: Pleomorphic adenoma, Oro-nasal fistula, Two-flap palatoplasty

INTRODUCTION

An intact hard and soft palate is important in everyday life for speaking, breathing, swallowing, and eating (chewing, drinking, and swallowing). Defects of the hard and soft palate can be caused by congenital cleft, benign or malignant tumor extraction, trauma, infection, disease, etc[1]. The procedure for closing palatal defects is one of the most difficult techniques in oral surgery, and small defects spanning the hard and soft palate can be successfully closed using a mucoperiosteal flap.

Palatoplasty can be applied to repair large or small congenital or acquired palate defects. Procedures such as von langenback palatoplasty, V-Y pushback palatoplasty, Bardach's two-flap paltoplasty, Fulow double opposing Z-plasty Schweckendiek two-stage repair, Alveolar extension palatoplasy, and Vomer flap have been modified, developed, and used for many years.

Two-flap palatoplasty is a modification of the von Langenbeck procedure and is relatively simple. It is one of the most commonly selected procedures of palatoplasty and has the advantage of being able to close the soft and hard palate in a relatively narrow deformity with a single procedure[2]. In this study, we report two cases of patients who underwent closure of an oronasal fistula that occurred after complete surgical resection of a pleomorphic adenoma, which occurred in the palate when using two-flap palatoplasty.

CASE REPORT

The two patients were an 89-year-old female patient (Case 1) and a 69-year-old male patient (Case 2), both of whom were elderly. There were periods of more than 10 years (Case 1) and over 1 year (Case 2) from the time the patient recognized symptoms until the patient visited the hospital. The size of the lesion from the posterior hard palate to the soft palate was 2 cm (Case 1) and 1.7 cm (Case 2), respectively, and biopsy results obtained through incisional biopsy before surgical resection were all pleomorphic adenomas.

All lesions were painless, grew slowly, had both induration and undulation, and showed no evidence of bone invasion radiologically. In Case 1, the lesion was larger, more elevated, and had clearer boundaries compared to Case 2 (Figure 1A-1, 1A-2). Both

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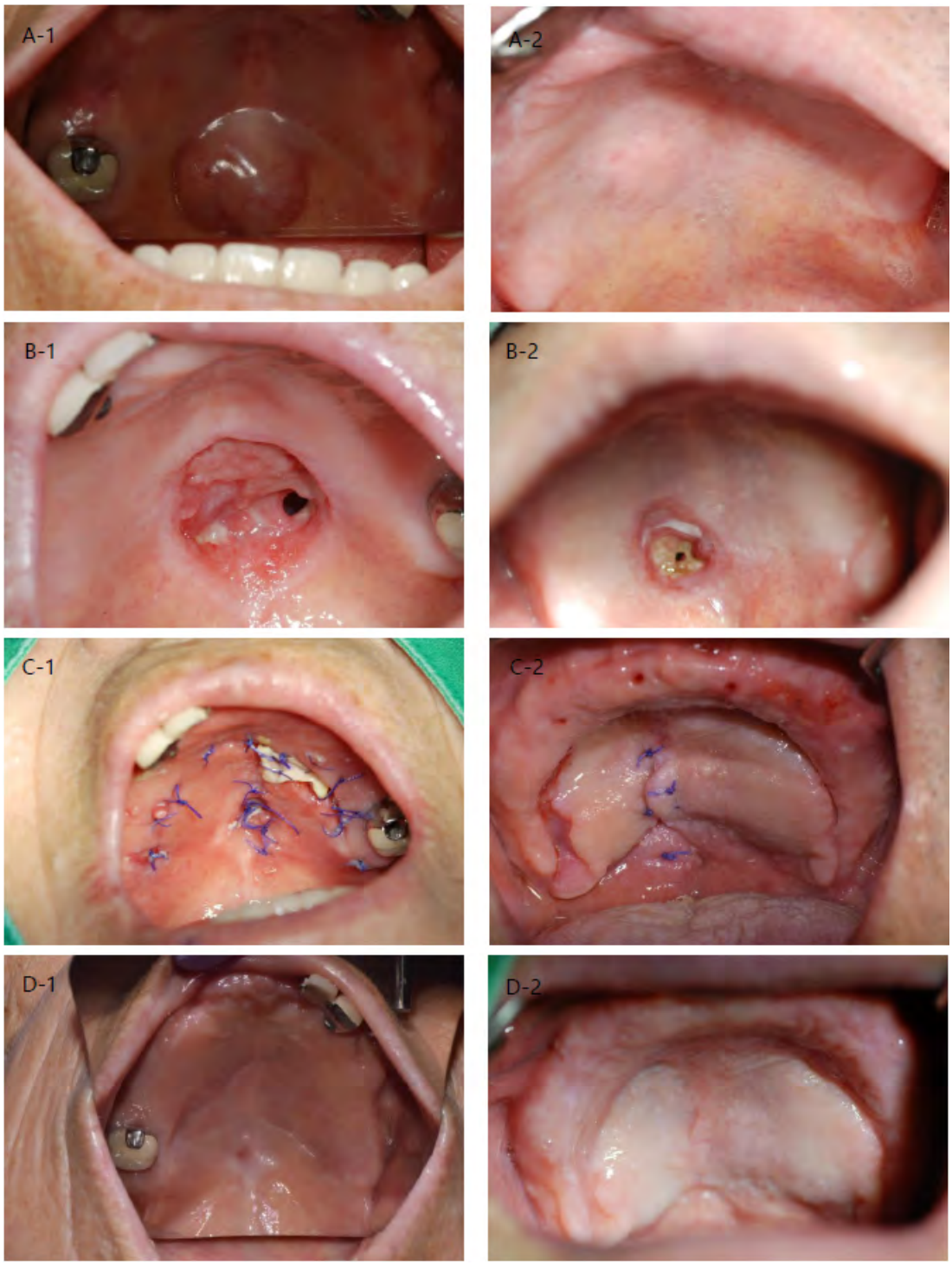

Figure 1. (A) The lesion of palatal on the first visit. (B) Oronasal fistula with a diameter of about 0.5 cm was created on the posterior part of the soft palate. (C) Healing state after two-flap palatoplasty. (D) One month post-operative clinical state (1: Case 1, 2: Case 2).

patients underwent surgical resection, including normal tissue surrounding the lesion, and the final biopsy results after surgery were all pleomorphic adenomas. Patients in Cases 1 and 2 were required to maintain a removable splint or used dentures at all times for 1 week post-operatively. They were discharged after removal. After 19 days (Case 1) and 10 days (Case 2), respectively, a 5 mm diameter oro-nasal lesion appeared in the perioperative area. It was confirmed that a fistula had formed (Figure 1B-1, 1B-2). Both

patients reported that food passed into their noses, and the patient in Case 2 also had nasal sounds. After observing the secondary healing and reduction of the surgical defect, two-flap palatoplasty was performed to close the oro-nasal fistula 6 weeks (Case 1) and 12 weeks (Case 2) postoperatively, respectively (Figure 1C-1, 1C-2). In the case of the patient in Case 2, the follow-up period for sufficient healing was longer due to excessive concerns about surgery.

The process of oro-nasal fistula closure using two-flap platoplasty was performed as follows. The surgical procedure of the Case 2 patient is attached in Figure 2.

1. Mapping & incision

Two flaps were mapped along the circumference of the oro-nasal fistula at intervals of approximately 3~5 mm and along the alveolar bone border. To prevent bleeding, a full-thickness incision was made on both sides towards the back of the alveolar crest after local anesthesia with 2% lidocaine (1/100,000 epinephrine) (Figure 2A).

2. Palatal flap elevation

The oral mucoperiosteal flap was elevated from the anterior to the posterior aspect of the oro-nasal fistula, and dissection was performed on the maxilla and palatal bone. The oral mucoperiosteal plate was lifted with a lifting motion using a periosteal device. At this time, the blood vessels and nerve bundles coming out of the foramen were carefully identified and preserved. The location of the greater palatine blood vessels and nerves was identified through preoperative cone beam CT, and a careful approach was employed to ensure complete dissection from the oral mucoperiosteal plate and palatal bone to maintain the circumference of the blood vessel and nerve bundles (Figure 2B).

3. Double layer closure

The fistula was first closed by suturing the nasal mucosa along the circumference of the oro-nasal fistula using absorbable 4-0 surgisobe suture. The two flaps (oral mucoperiosteal flaps) on both sides that had been dissected were pulled and repositioned to approach the midline and were sutured to form a double layer. The alveolar crest areas on both sides were left exposed without

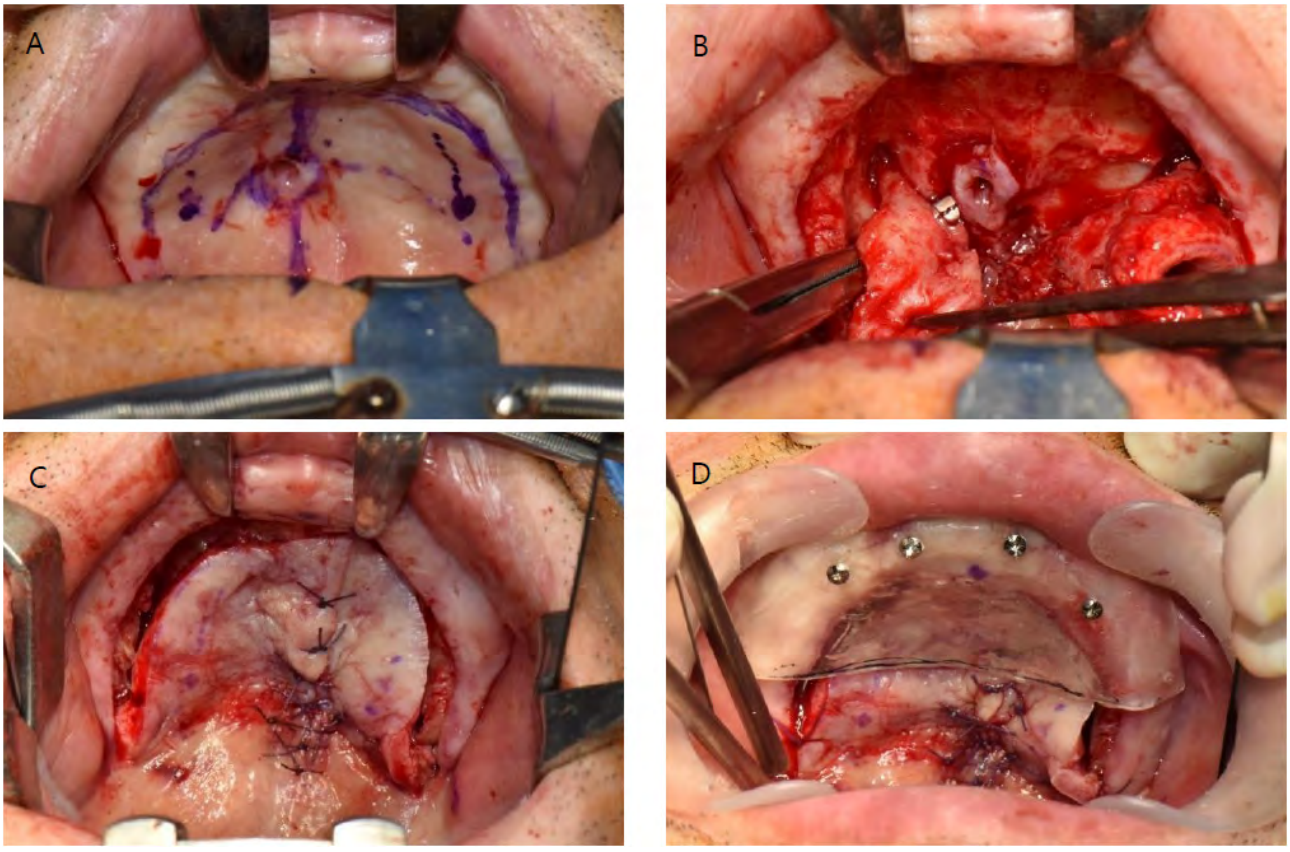

Figure 2. (A) Mapping & incision, (B) flap elevation, (C) double-layer closure, (D) splint fixation.

sutures to ensure a tension-free predicate, allowing secondary healing to wait (Figure 2C).

4. Operation site preservation

Before surgery, a pre-made splint device was fixed to the palatine bone using plate fixation screws to maintain the flap and protect the surgical area. As a result of the postoperative follow-up, both patients had no symptoms of nasal discharge or reflux of foreign substances, the wound area recovered well, and no evidence of recurrence was observed 12 weeks (Case 1) and 6 weeks (Case 2) after the secondary surgery (Figure 2D).

DISCUSSION

Pleomorphic adenoma accounts for 70% of minor salivary gland tumors and occurs in the oral cavity in this order: the palate, upper lip, buccal mucosa, and floor of the mouth[3]. Pleomorphic adenoma is classified as benign, but has a tendency to recur and has the potential to transform into malignancy, thus surgical excision is the basic treatment option. Although they tend to be well encapsulated, pleomorphic adenomas arising in the palate tend to be locally aggressive and often invade surrounding bone, so sufficient surgical resection of the lesion, including the vocal margin, is necessary[4]. Therefore, in the case of pleomorphic adenoma occurring in the palate, a large defect including the hard palate, soft palate, and bone may remain after surgery, and complications such as nasal sound, oro-nasal fistula, and speech abnormalities may occur, and treatment (secondary surgery) is required.

Soft tissue defects formed after complete resection can wait to heal on their own. An oro-nasal fistula is an abnormal passage formed between the nasal cavity (or maxillary sinus) and the oral cavity. When an oro-nasal fistula occurs, it can cause symptoms such as nasal discharge, reflux of water and food, and maxillary sinusitis[5]. If the defect does not close naturally after 4 weeks or if the defect is larger than 4~5 mm, secondary surgery is required for reconstruction[6]. In the two cases in this report, secondary surgery was performed through palatoplasty after 6 weeks (Case 1) and 12 weeks (Case 2) of postoperative observation to close the oronasal fistula that developed as a complication after removal of the pleomorphic adenoma.

The procedure for closure of oronasal fistula was two-flap palatoplasty, the most commonly used surgical method since cleft palate surgery was performed. Two-flap palatoplasty was first described by Bardach in 1976, and is used in various applications such as wardill V-Y palatoplasty, Bardach two-flap palatoplasty, and von langenbeck palatoplasty. The first Bardach two-flap palatoplasty could be used in relatively narrow defects because the oral mucoperiosteal plate was detached at the border of the cleft palate. Later, a modified method of more extensive dissection and additional incisions along the alveolar bone border was introduced, making tension-free suturing possible[7]. Wardrill V-Y palatoplasty (V-Y pushback palatoplasty) has the advantage of extending the length of the palate as the v-shaped flap is sutured into a y shape. However, in this case, extension of the length of the palate was not necessary, so the v-y palatoplasty design was not used.

The process of the two-flap palatoplasty procedure is summarized in Figure 3. The incision is made along the edge of the bone defect and around the alveolar crest, and the end of the valve is rounded, so the v-y pushback design is not used (Figure 3A). The palatal mucoperiosteal flap is completely detached from the maxilla and palatine bone. Since this flap is based on the greater palatine blood vessels, it is important to preserve the greater palatine blood vessels and nerves during the procedure (Figure 3B). Subperiosteal dissection is performed anteriorly and posteriorly to the hard palate such that the nasal mucosa can be sutured without tension (Figure 3C). At the midline, the palatal mucoperiosteal plate is repositioned and sutured, leaving the bilateral alveo-

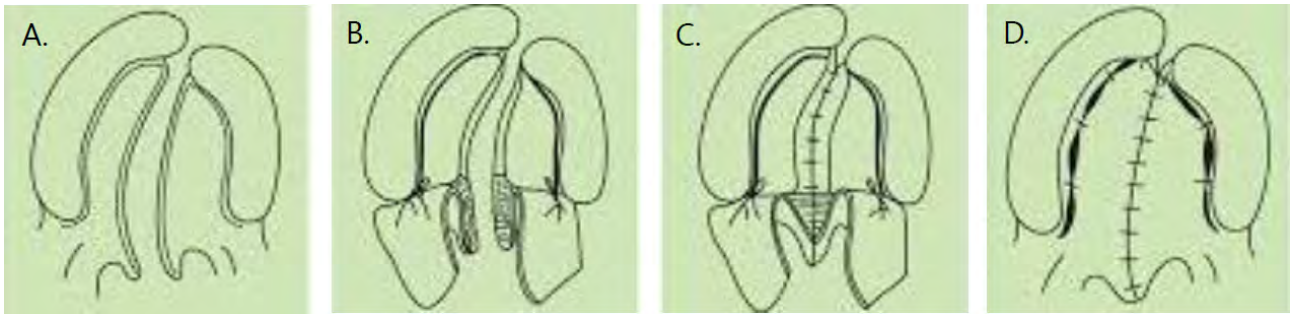

Figure 3. Bardach's two-flap palatoplasty.

lar bone areas exposed.

Emmanouel Koudoumnakis et al. reviewed two-flap palatoplasty performed on 257 cleft patients at the Cranio Facial Anomalies Center of Aghia Sophia Children's Hospital and reported a low recurrence rate of 5.4%. Two-flap palatoplasty has the advantage of effective healing and a low possibility of recurrence as it allows tension-free suturing while minimizing bone exposure through extensive dissection[8]. Therefore, in the case of this study, oronasal fistula closure was performed using two-flap palatoplasty as a procedure for the oronasal fistula that developed after surgical resection of a pleomorphic adenoma in the palatal. The fistula did not recur, and long-term follow-up will be conducted to check for recurrence of pleomorphic adenoma in the future.

CONCLUSION

Pleomorphic adenoma, the most common benign tumor in the palate, has the potential to become malignant, and thus local resection with sufficient margins including the tumor is essential. If natural healing does not occur for soft tissue or bone defects that occur after surgery, secondary surgery is required. Oronasal fistula cause great discomfort to the patient due to symptoms of nasal discharge and reflux of food, so secondary surgery for reconstruction must be performed appropriately. Two-flap palatoplasty can be selected as an effective approach for reconstruction of hard palate defects (oronasal fistula) as in this case due to its technical simplicity, effective healing, and low possibility of recurrence.

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