Closure of an Unusually Large Palatal Fistula in a Cleft Patient by Bony Transport and Corticotomy-Assisted Expansion

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The management of cleft lip and palate can vary among patients because the size of a cleft defect and its anatomy vary. Although treatments involving orthodontics and surgery have been developed that can help a large number of children with cleft lip and palate, there are always unusual clefts that defy conventional treatments. In such cases, with each gain made during treatment, there also is the possibility of introducing an additional complication. The secondary bone graft performed during the mixed dentition stage of dental development is an example of treatment gains that are balanced against additional complications. Ordinarily, the maxillary segments are expanded to attain a normal archform before the alveolar bone graft. If there is an anterior palatal fistula present before orthodontic expansion, the size of the fistula concomitantly increases as the expanders widen the anterior maxillary segments. There are, however, extreme cases that present with a combination of collapsed maxillary segment, large anterior palatal fistula, and large alveolar clefts. In such cases, orthodontic expansion could make both the alveolar clefts and anterior palatal fistula unmanageable. In the expanded position, there would be insufficient soft tissue to close the fistula or cover the alveolar bone graft. Consequently, dental prosthetics is often needed to cover the palatal fistula to support speech and eating.

In this report, we present the case of an 11-year-old girl who had a 20-mm anterior palatal fistula before orthodontic expansion. In the opinion of our surgeons, the fistula was too large to close before the segments were expanded and would prove more difficult to treat after expansion. To make the palatal fistula and cleft sites manageable, the treatment sequence and procedures were modified. Rather than expand the maxillary segments, the segments were compressed to make the cleft space narrower so that the surgeons could graft the alveolar cleft. To provide more bone support and donor soft tissue for a palatal flap, the palatal tooth and surrounding bone were distracted across the anterior palatal opening. A sequence of minor procedures allowed the cleft sites to be grafted, the unusually large palatal fistula to be closed, the lateral segments to be expanded, and the dental alignment to be improved.

Report of a Case

In 1999, a 9-year-old girl with bilateral cleft lip and palate and orofaciiodigital syndrome type I was seen by the craniofacial team at Childrens Hospital Los Angeles. She presented with collapsed maxillary segments, an anteriorly displaced premaxilla, and a large 20 × 10 mm anterior palatal fistula. Her speech was hypernasal and contained developmental and compensatory articulation errors. She had downward slant of her palpebral fissures, ankyloglossia, bifid tongue, and asymmetry to the left tongue. Her previous operations for the palate included an attempt to close the anterior
palatal fistula, palatoplasty, and a reduction in posterior cleft volume surgery. Initially, she was evaluated for palatal closure to improve her speech. The palatal defect was too large to close with soft tissue flaps (Fig 1A). Because orthodontic expansion would increase the size of the fistula and alveolar clefts, the treatment sequence and strategy were modified to narrow the arch form for the placement of an alveolar bone graft.

COLLAPSING THE SEGMENTS FOR AN ALVEOLAR BONE GRAFT

Our original objective was to narrow the palatal fistula and cleft space so that the maxillary segments could be grafted and the fistula closed. A quad helix expander was modified to compress cleft segments toward the center of the palate. Once the maxillary segments were in soft tissue contact, the bilateral cleft spaces were grafted with autogenous bone from the iliac crest. After the surgery, there remained a fistula that was still 15 mm long in an anteroposterior direction and 8 mm wide (Fig 1B). We had hoped that the fistula could be surgically closed by soft tissue flaps, but the palatal shelves were so narrow that they did not provide adequate soft tissue for rotating a flap over the palatal opening.

BONY TRANSPORT TO CLOSE THE PALATAL FISTULA

To produce additional bone and soft tissue to support a palatal flap and to narrow the palatal opening, a lateral incisor tooth and surrounding bone were transported across the palatal opening (Figs 1C, D). This patient had a permanent lateral incisor that had erupted into the palate. Based on our experience in closing large alveolar clefts by segmental transport,1 we used a similar approach to transport a small palatal transport disc consisting of the palatal tooth and the surrounding bone. A lingual arch was constructed to direct a palatal tooth and surrounding bone toward the center of the fistula under the tension of nickel titanium springs. As the patient was in mixed dentition and did not have fully erupted permanent first molars, 2 primary maxillary second molar bands were bonded to support the lingual arch. Two parallel wires were soldered across the lingual arch to provide a track for moving the palatal maxillary right lateral incisor (Fig 1D). Orthodontic brackets were bonded to the remaining maxillary teeth, including deciduous teeth, for a later stage of arch expansion. The lingual arch appliance was fitted but was not cemented until the day of surgery. Study models were taken to draw the osteotomy lines for surgery.

Under intravenous sedation, a full-thickness mucoperiosteal flap was used to expose the palatal and buccal cortical bone. Vertical cuts were made lateral to the root of the palatal tooth and were connected to a horizontal cut made above the apex. An osteotome was used to mobilize the segment of bone, which resembled a rectangular box holding the palatal tooth. After flap closure, the lingual arch was cemented into place and the transport disc firmly ligated with 0.012 ligature wire to the lingual arch to promote good callus formation. There was no mobility of the healing transport disc during a 1-week latency period. To tip the crown of the palatal tooth toward the center of the palate, ligation wires were removed and nickel titanium springs were used to pull the tooth toward the center (Figs 1C, D). The tooth was tipped rather than translated to prevent the apex of the root from protruding out of the thin maxillary floor into the nose. As the tooth and bone moved toward the center of the palate, new bone formed in the transport site.

After 5 weeks, most of the palatal opening was closed by the combination of segment compression and bony transport of the palatal tooth. The springs were removed, and the tooth was ligated into position for 2 months for bony consolidation.

CORTICOTOMY-ASSISTED TOOTH MOVEMENT

At the end of bony transport, most of the palatal fistula had closed but the arch form was distorted into a Y shape (Fig 2D). To restore the U shape to the archform, the buccally positioned teeth and bone would have to be expanded. However, the earlier alveolar bone graft would resist any segmental expansion and potentially force the roots of the buccal teeth out of the buccal plate if dental expansion were attempted. To accomplish the segmental expansion after bone grafting, we adapted a technique for corticotomy-assisted tooth movement and expansion that was originally described by Wilcko et al2 for the rapid correction of dental crowding in noncleft orthodontic patients. Under general anesthesia, full-thickness flaps were elevated. Vertical corticotomy lines were marked and then cut in the interproximal bone between the buccal teeth (Figs 2A, B). These vertical lines extended superiorly to the apical areas but stopped 5 to 7 mm below the gingival papilla. Completion of the vertical osteotomies was not necessary and was not performed. In the apical area, a horizontal corticotomy line was made so that each tooth had a U-shaped corticotomy outline surrounding it on the buccal side. The palatal side was left untouched because the goal was to widen the buccal alveolar ridge with orthodontic tooth movement. A corticotomy rather than an osteotomy was used to reduce the risk of injuring a dental root. The flaps were sutured. No archwire was placed in the orthodontic brackets during the latency period. After 1 week, a round elastic archwire was used to move the buccal teeth into an archform. The teeth moved outward with a flexible buccal plate of bone. During expansion, the alveolar ridge of bone

FIGURE 1. A, Palatal photograph showing palatal defect before surgery for palatal closure was attempted. B, Photograph of cast for making an appliance, taken after orthodontic compression of the lateral segments. A large 15-mm defect remained. C, Occlusal photograph of palate before activating bony transport. D, Occlusal photograph of palate during bony transport showing the lingual arch appliance.
was temporarily widened with outward tooth movement. Within 3 months, the U-shaped dental archform was attained by changing to larger, rectangular nickel titanium archwires.

During expansion, the palatal opening minimally changed as the palatal tooth was tipped back into an upright position. An important finding was a funnel-shaped wall of bone that narrowed to a 3 to 4 mm opening toward the nasal floor (Fig 2E). This new bone and soft tissue, formed by bony transport, provided a support and soft tissue source for the pedicle flap. It was now possible to undermine and elevate the mucosa from this vertical wall and close the palatal fistula with a small mucosal flap. The final flap procedure was performed after palatally erupting premolars were either brought into the dental arch or extracted (Fig 2F).

**Discussion**

This particular patient had 4 clinical problems that needed to be addressed: closure of an unusually large palatal fistula, 2 bone grafts for the bilateral cleft spaces, expansion of collapsed segments, and alignment of teeth into a dental arch. During the initial evaluation, several treatment plans were considered. The first plan was a conventional approach: expand the cleft segments, graft the cleft spaces, and then close the palatal fistula with a tongue flap. In this patient, expansion would have enlarged the cleft spaces and the palatal fistula. Based on this previous patient and others, our surgeons did not believe that they could completely close the palatal fistula unless it was reduced in size.

A second treatment option consisted of expanding the segments, and then distracting an osteotomized segment of bone and teeth across the cleft spaces for a secondary bone graft. In previous cases, we showed that a palatal fistula can close with bony transport. Unfortunately, in this case, the palatal fistula extended to the posterior palate such that the transport disc would not include transpalatal soft tissue; therefore, bony transport would have allowed the cleft spaces to be grafted but would have left a sizable palatal defect. Our segmental distraction experience is thus far limited to 12 distractions with the “Yen” orthodontic setup that is both tooth- and bone-borne and 10 cases using the KLS Martin (Jacksonville, FL) “Liou” distractor that is bone-borne. Both bony transport techniques have their advantages and disadvantages. In this case, bilateral bony transport with widely separated segments would have been technically difficult to control with orthodontic archwires. Alternatively, bone-borne devices have problems in vector control, loosening of load-bearing screws in
the proximal segment, and twisting and breakage of the connecting metal arms. In this case, the use of bony transport was excluded because the palatal fistula would not be adequately addressed.

A third plan was to leave the fistula open and obturate the fistula. However, we learned from previous patients that older patients were willing to go to great lengths to be rid of an obturator and their problem of oronasal fluid leakage. This plan was considered as a last resort because the patient’s chief complaint of a large palatal opening was not addressed.

A fourth treatment option consisted of expanding the segments, setting back the premaxilla, grafting the cleft spaces, and attempting a soft tissue closure of the palatal fistula. In this patient, the incisors were in normal overjet position. A premaxillary setback would produce a Class III malocclusion. The disadvantages to this approach are reduction of the maxillary length, the intrusion of incisor position to reduce the size of the palatal fistula, and the need for further orthognathic surgery. Padwa et al. reported that the growth impact on the midface would be minimized in older patients. However, at the time that we were developing the treatment plan, it was not known whether the patient would grow into a Class III malocclusion during her adolescence. However, if a premaxillary setback were attempted at the outset to reduce a 20-mm palatal fistula, the patient would have had an unesthetic Class III malocclusion during her adolescent years. This plan would necessitate a future orthognathic surgery for this patient. Instead, our team opted to replace more extensive surgeries such as orthognathic surgery and tongue flaps with a series of minor surgeries that would be better tolerated by our patient (Fig 3).

Although corticotomy-assisted palatal expansion and uncrowding of teeth have been described, this report concerns the first use of corticotomy-assisted tooth movement to correct a craniofacial anomaly after an alveolar bone graft. The orthodontic correction was much more dramatic than non-cleft palate applications because the distortion of the archform was so extreme. Corticotomy-assisted tooth movement has a long history and has been used by orthodontists and surgeons to align crowded teeth and to expand the palatal archform. A recent articulation of this technique has been popularized by Wilcko et al. Their technique was modified to expand dental archforms after bone grafting. According to Wilcko et al., this type of surgically assisted tooth movement differs from distraction osteogenesis. Their review of computed tomography scans and biopsies suggested that there is rapid resorption of bone resembling a local osteoporosis followed by gradual replacement of buccal bone. The phenomenon, also known as regional accelerated phenomenon, has been described as a transient response to mucoperiosteal flap surgery in the mandible and has also been reported in the orthopedic surgery literature. In this case, a second corticotomy was performed because the orthodontic expansion of a primary molar stopped when the primary tooth exfoliated (Fig 2E). The corticotomy-supported expansion was resumed after the replacement premolar erupted and could be banded. The second corticotomy occurred 8 months after the first corticotomy and provided an opportunity to evaluate the buccal bone. On the left side, the bone overlying the buccal dental roots was uncovered and photographed (Fig 2C). There was good visual confirmation of bone without dehisences.

Several lessons were learned during the management of the large palatal fistula and cleft spaces. First, segments could be collapsed, grafted, and later expanded. Second, palatal distraction could reduce a 20-mm fistula to a 3-mm opening and produce a fun-
nel of bone in the center of the palate. Third, the shape of the alveolar ridge could be redefined by orthodontic tooth movement assisted by buccal corticotomies. Buccal corticotomies with orthodontic tooth movement have since been used by our craniofacial team to correct problems in segment position and alveolar ridge width.

The treatment of this patient illustrated how different types of tissue-generating and molding techniques can be combined to solve a particular clinical problem. Every procedure has a set of benefits and complications. In this case, the additional surgery and orthodontic treatment were helpful in treating the large palatal fistula, the bilateral cleft maxillary segments, and the collapsed position of the segments.

References
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Life-Threatening Bleeding After Dental Extraction in a Hemophilia A Patient With Inhibitors to Factor VIII: A Case Report

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Hemophilia A is a X-chromosome–linked congenital bleeding disorder caused by a deficiency of the blood coagulation factor VIII (F.VIII).1 Typical manifestations of this disorder are soft tissue, muscle, and joint hemorrhage. Exogenous F.VIII substitution with either plasma-derived or recombinant products is the treatment of choice, being implemented during bleeding episodes or prophylactically, such as before surgery.

Currently, the most detrimental treatment complication is the development of inhibitors.2 Alloantibodies against foreign F.VIII develop in 25% to 30% of patients with severe hemophilia A who receive therapeutic infusions of F.VIII.3 Several investigators found a correlation between the type of genetic mutation and the development of inhibitors after exposure of hemophiliac patients to F.VIII substitution.4 Severe genetic mutations like intron 22- or nonsense mutations lead to dire molecular defects of F.VIII. Therefore, exogenously administered F.VIII during treatment periods is recognized as a foreign antigen, triggering the formation of alloantibodies. Less severe mutations result in a small amount of circulating F.VIII, eliciting an immune tolerance in most of the