FIGURE 2. (A) Intraoperative photograph of orbital dermoid cyst excised through a lid crease incision. (B) External photograph showing the gross specimen of the dermoid cyst. (C) Postoperative external photograph 3 weeks after excision demonstrating no recurrence and resolution of hypoglobus and ptosis.

causes progressive proptosis or diplopia or when acute rupture causes inflammation and compromised orbital function.[3,6]

Our case demonstrates a giant postseptal dermoid cyst that became symptomatic in infancy due to its growth into the preseptal space, which is not commonly seen. "Giant" has been used to describe an orbital dermoid cyst equal in size to or larger than the globe on the affected side.[7] There have been several reported cases of giant orbital dermoids presenting in adults.[6–8] To the best of our knowledge, there have been only 2 reported cases of giant postseptal orbital cysts presenting in infancy. Fasina and Ogun[9] describe a case of a neglected giant deep orbital dermoid cyst in a 3-month old infant presenting with proptosis and corneal exposure, and Leonardo et al[10] reported a recurrent giant orbital dermoid in a 15-day old infant. Imaging should be performed to rule out intracranial or intraorbital extension and assist with surgical planning.[3]

The management of dermoid cysts is dependent on the size and location of the mass and the presence of ocular symptoms. Small, asymptomatic orbital dermoid cysts may be monitored as they may stabilize or decrease in size over time. Larger cysts can be treated surgically with complete excision to prevent recurrence or abscess formation.[1–3] Postoperative edema can be significant particularly if there is cyst leakage at the time of excision. Left untreated, these cysts can lead to complications, including cyst rupture, intracranial extension, and inflammatory fibrosis.[2,6] Overall, early surgical intervention can achieve excellent results and is curative.[3]

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OPEN

Management of Tessier Type 3 Cleft With a Novel Reverse Palatal Expander

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Abstract: The purpose of this clinical report is to present the novel management of a type Tessier 3 cleft which was treated using a palatal expander in reverse fashion to reapproximate the craniofacial skeleton allowing for closure of the palate and soft tissue of the cleft. Reapproximation of the bony component of the cleft was achieved without osteotomies and allowed for easier and earlier realignment of the bony and soft tissue components of the cleft. To our knowledge, this is the first use of reverse palatal expansion in the treatment of type 3 Tessier cleft. Reverse palatal expansion made management of this cleft more straightforward and should be considered as a useful adjunct in the management of wide facial clefts.

In 1976, Paul Tessier introduced his comprehensive system for categorizing facial clefts. He numbered facial clefts based on their relationship to the sagittal midline and the orbit.[1] Craniofacial clefts are an uncommon congenital craniofacial anomaly occurring in

0.014 to 0.048 per 1000 live births.[2] Embryologically, type 3 Tessier clefts result from failure of fusion of the olfactory placode, the frontonasal process, and maxillary process along the naso-optic groove.[3]

Also known as oronaso-ocular clefts, type 3 Tessier clefts typically span from the philtrum of the upper lip and ipsilateral alar base to the medial canthus. Other soft tissue structures that may be involved include colobomas of lower eyelid, disruption of the lacrimal system, and inferior-lateral displacement of the globe. The bony cleft begins between the lateral incisor and canine and involves the palate, frontal process of the maxilla, and ends at the lacrimal groove. As such, there is typically direct communication between the oral, nasal, and orbital cavities on the affected side. Important to note is that type 3 Tessier clefts present on a wide clinical spectrum (incomplete, complete, unilateral, and bilateral) and can occur in combination with other Tessier clefts.[4]

Given their varied clinical presentation and involvement of both bony and soft tissue, preoperative planning and surgical treatment of type 3 Tessier clefts is complex at best. There is no widely accepted treatment algorithm and patients generally require several staged procedures to restore bony and soft tissue landmarks. As such, treatment for type 3 Tessier clefts is controversial and limited to case reports and series. We present here the successful management of a patient with a wide type 3 Tessier cleft using a palatal expander in reverse to help realign the bony and soft tissue components of the cleft in preparation for definitive repair.

CLINICAL REPORT

A newborn Caucasian male was referred to our center for evaluation of a unilateral type 3 Tessier cleft. He was found to be otherwise healthy and without genetic abnormalities.

On examination, his type 3 Tessier cleft involved the soft tissue from the left alar base to the punctum of his lower eyelid (Fig. 1A).

Computed tomography scan of the maxillofacial bones revealed partial absence of the left nasal spine and truncation of the left nasal bone in addition to absence of much of the left hemi palate with communication between the oral and nasal cavities. The maxillary palatal shelves were widely splayed with the interalveolar and palatal shelf distance measuring approximately 22 mm.

Lip taping was started after initial examination to approximate the lip and alveolar segments. At 3 weeks of age, after obtaining a palatal impression, a Rotterdam palatal expander was modified with acrylic caps over the alveolar maxillary segments (KLS Martin LP, Jacksonville, FL) (Fig. 1B). It was secured intraorally using six 11 mm self-tapping screws, (3 on each buccal aspect) placed through predrilled holes the acrylic alveolar caps (Fig. 1C). After a latency period of 3 days, the Rotterdam device was turned twice daily in reverse fashion to achieve a 0.6-mm of midline palatal shelf advancement/day. The activation period continued for an additional 7 days until the palatal shelves were approximated to approximately 10 mm apart. This was followed by a consolidation period of 12 weeks (Fig. 1D).

At 4 months of age the patient underwent a lip adhesion and exchange of the palatal approximator for a palatal prosthesis. At 8 months of age he underwent cleft lip repair with rotation advancement flaps, cleft rhinoplasty, cheek fasciocutaneous advancement flap to close the soft tissue over his maxilla and medial canthopexy (Fig. 2A). At 15 months of age, his soft palate was repaired via a Furlow palatoplasty with placement of a palatal prosthesis over the hard palate.

At 14 months follow up after the initial reverse palatal expansion the patient has had no complications and maintained stable maxillary position with no relapse. Aesthetically, he has significant improvement of facial appearance and functionally has achieved complete repositioning of the maxilla with good alignment of the palatal shelves and alveolar segments (Fig. 2B-D).

FIGURE 1. Preoperative and intraoperative photos. (A) Preoperatively the patient had a widely splayed unilateral type 3 Tessier cleft spanning soft tissue from the left alar base to the punctum of his lower eyelid. (B) A Rotterdam palatal expander with modified acrylic caps is pictured overlying the orthodontic stone impression. (C) The palatal expander in situ after initial insertion. (D) Frontal view of the patient after 12 weeks of consolidation with the modified palatal expander.
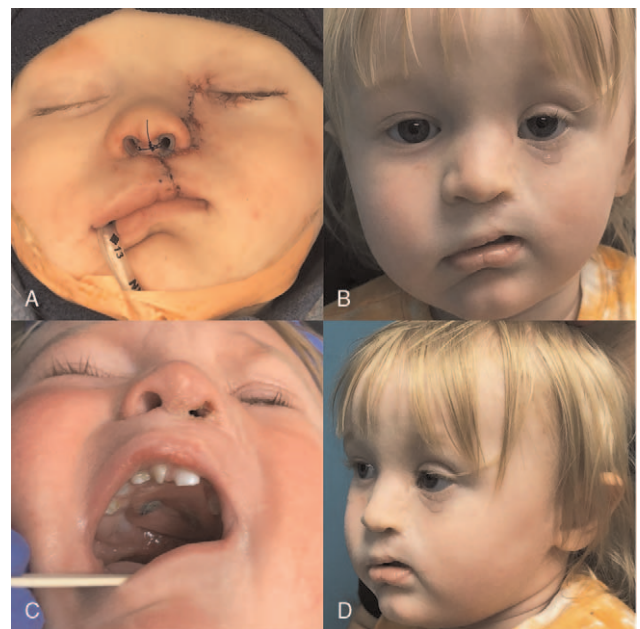

FIGURE 2. Postoperative photos. (A) Postoperative view after cleft lip repair with rotation advancement flaps, cleft rhinoplasty, cheek fasciocutaneous advancement flap. (B) Frontal view 14 months after initial reverse palatal expansion. (C) Intraoral view of alveolar arches 14 months after initial reverse palatal expansion. (D) Oblique view 14 months after initial reverse palatal expansion.

DISCUSSION

A variety of options have been reported for management of the soft tissue defects including rotation and advancement flaps,[5] the use of tissue expanders,[6] using a "split approach"[7] and various other techniques. Equally important, correction of the underlying bony deformity in craniofacial clefts serves as the foundation for soft tissue repair and still remains a critical piece of the puzzle whose treatment is not as clearly defined.

Distraction osteogenesis generates vascularized bone between cut ends of an osteotomy by gradually separating them from one another with a specialized distraction device. Because of its simplicity and decreased morbidity, distraction osteogenesis has become a mainstay in treating craniofacial differences where bony deficits exist. Distraction osteogenesis has wide applications including maxillary and mandibular advancement to expansion of the cranial vault in craniosynostosis. In patients with craniofacial excesses, contraction osteogenesis (CO) is a similar concept with movement of bone in the reverse direction without the need for osteotomies. CO is the application of an external compression force on bone with the concomitant induction of new bone formation/remodeling. The proposed mechanism of action of CO is that tension placed on the bone by a contractile force causes a mild decrease in vascular flow which induces remodeling and osteogenesis in growing bone.[8]

Castello et al[8] describe the use of an external maxillary distractor across the incisive foramen in growing rabbits to effect craniofacial shortening without the need for osteotomies. Similarly, nasoalveolar molding is a commonly used technique to presurgically mold the bony alveolus and soft tissue elements of the lip and columella to create better anatomic alignment make subsequent surgical repair more straightforward.[9]

Konofaos et al[10] describe the use of an external mandibular distractor device in reverse to reapproximate the bony shelves in a child with a type 2 to 12 Tessier cleft without the need for osteotomies. However, to our knowledge, there are no prior reports regarding the use of a palatal prosthesis in patients with a type 3 Tessier cleft to help restore normal anatomy.

In the presented case, the widely splayed maxillary segments created distortion in both the bony and soft tissue landmarks of the face. The goals in treatment were to restore normal facial landmarks and continuity of the palate and lip. By applying the palatal prosthesis early within the first few weeks of life, we were able to take advantage of the mobility of the facial bones and bring them into a more normal anatomic alignment without the need for osteotomies. Applying the force to the palatal expander in reverse fashion is a novel use that allowed us to narrow the cleft, recreate anatomic alignment early, and make the patients subsequent soft tissue repairs more predictable and straightforward. We acknowledge that long-term follow-up is necessary to fully assess quality of life and functional outcomes in a longitudinal manner. However, in short and medium-term follow-up, we found the use of a palatal expander in reverse fashion to be a useful and safe adjunct in the management of this patient with a wide type 3 Tessier cleft.

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Bioabsorbable System-Related Subcutaneous Swelling After Craniofacial Surgery

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Background: Bioabsorbable systems have been commonly used in pediatric patients for primary cranioplasty and other related surgeries. However, subcutaneous swelling, a unique complication related to bioabsorbable osteosynthesis, is a concern. Differences in the incidence of subcutaneous swelling, depending on the bioabsorbable material used to construct the plate, are still unknown.

Methods: The authors retrospectively reviewed all incidences of subcutaneous swelling related to resorbable systems used during primary cranioplasty for patients with craniosynostosis at their hospital between 2014 and 2018 during a 12-month follow-up period. Furthermore, the authors reviewed all published English-language articles (since 1995) on subcutaneous swelling in bioabsorbable systems used for craniosynostosis.

Results: The most common resorbable systems used in the literature were divided into 2 groups: mixtures of poly D-lactic acid and polyglycolic acid, and mixtures of poly D- and L-lactic acid. In patients for whom poly D-lactic acid and polyglycolic acid were used, the incidence of subcutaneous swelling during resorption was