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CASE REPORT

Serial Presurgical Orthopedics for Eye Repositioning and Optimization of Soft-Tissue Repair in an Infant With Tessier No. 4 Cleft

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Objective: **A male patient with Tessier No. 4 cleft (unilateral left) presented at 20 days of age. The cleft defect beginning between the cupid bow and oral commissure extended to the ipsilateral orbital floor, skirting the nose and lacrimal duct while passing through the cheek medial to the infraorbital nerve. With the lesser segment disposed 16 mm transversely, the wide gap included an absence of orbital floor and lower eyelid. A deficient midfacial platform caused a severe inferior globe dystopia, superiorly displaced left ala base, and severe vertical shortening between ala-canthus and ala-globe.**

Intervention: **A modified Latham device applied directional orthopedics to contract the cleft gap and with an eye part added to elevate the dystopic globe. Two different Latham devices used in succession were each applied for 4 weeks. Lastly, a removable plate further repositioned the eye. Each appliance was designed to differentially move the noncleft and cleft segments of the maxilla. Presurgical orthopedics began at 3 weeks lasted 14 weeks. Intraoperatively at 17 weeks, the inferior globe dystopia was effectively reduced, and the cleft gaps were nearly closed and aligned at the orbital floor, cheek, and the alveolus. Respecting the aesthetic units of face became possible with the soft-tissue repair yet were tight enough in the malar region to retract the lower lid.**

Conclusion: **The presurgical directional orthopedic and eye-globe mechanics were sufficient to enable medial canthal repositioning, sustainable correction of orbital distopia, and optimized primary soft-tissue repair. Early result suggests that surgery with presurgical orthopedics is superior to surgery alone.**

KEY WORDS: *eye repositioning, presurgical infant orthopedics, Tessier 4 cleft*

The incidence range of rare facial clefts reported by Kawamoto (1976) as 1.43 to 4.85 in 100,000 births is found from the literature as if they are exact ratios derived from birth records. These figures are actually estimates derived from reports on common cleft populations. The ratio of atypical to common clefts varies in the range of 0.75% to 5.4% (Van der Meulen, 1985; Kawamoto and Praveen-Kumar, 1998). If 1 over 1000 is accepted to be the occurrence rate of common cleft lip (palate), then we would expect the atypical clefts to be seen on the order of 1 per 100,000 live births.

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Tessier's (1976) easily retained, topographical numeric system is currently the universally accepted classification for these scarce disorders since it is simple yet efficient in specifying the deformities. In this classification, the clefts are numbered (left, counterclockwise; right, clockwise) around the orbit—the southbound (facial) clefts from the midline to the lateral and the northbound (cranial) clefts from lateral to the midline—from 0 to 14.

Among the rare clefts, the No. 4 facial cleft is so infrequent that there were only 8 of them in a series of 345 atypical clefts (Ortiz-Monasterio et al., 1987). Starting with the No. 4 cleft, the facial clefts move onto the cheek, sparing the median facial structures. Therefore, No. 4 cleft is sometimes called medial maxillary dysplasia (Van der Meulen, 1985) and described as melochisis. The cleft has been classified as an oro-ocular (American Association of Cleft Palate Rehabilitation) or oro-ocular type 1 (medial to the inferior orbital foramen) cleft (Boo-Chai, 1970). The ubiquitous term *oblique facial cleft* is also used for this malformation as well as for Nos. 3 and 5 clefts. When treating these clefts, generally complex surgeries such as dermatoplasty, transfer of skin flap, soft-tissue filling, craniofacial bone osteotomy, and reconstruction of bone morphology may be required according to the severity and characteristics of the case (Zhou et al., 2006).

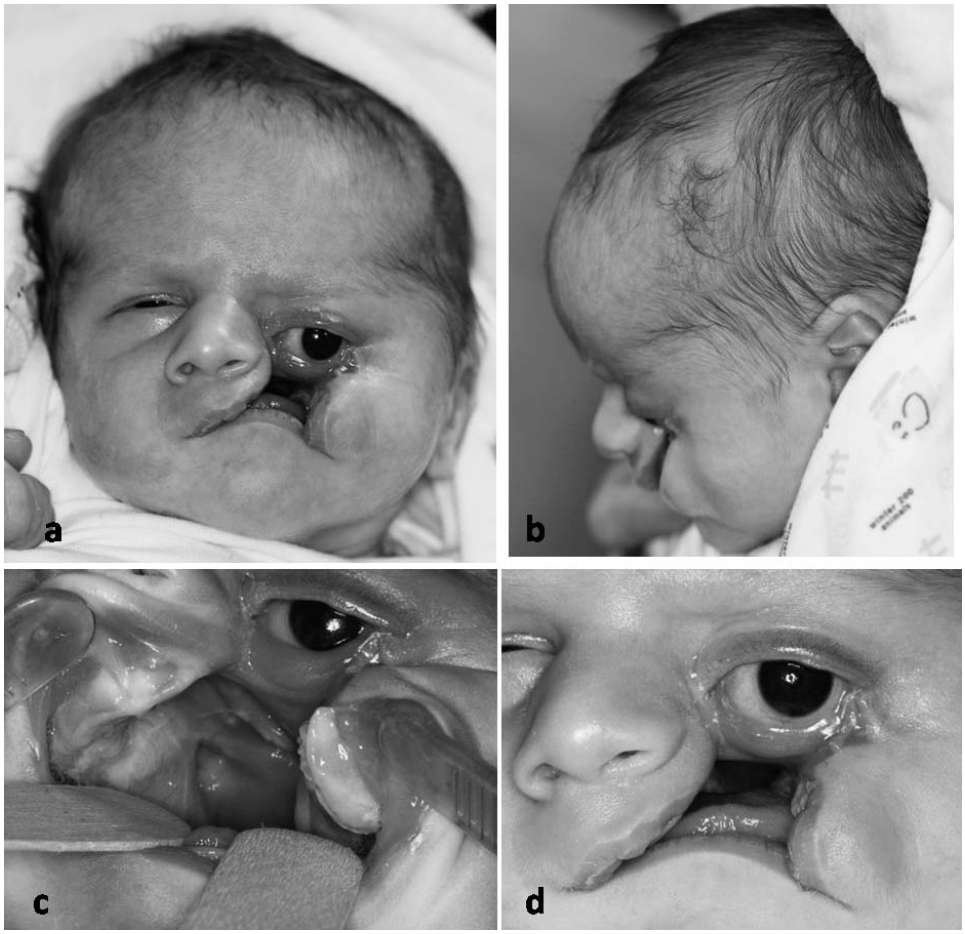

FIGURE 1 a–c: The initial presentation of the patient at the age of 3 weeks.

The long-term efficiency of presurgical infant orthopedics (PSIO) has been a major issue of controversy in the treatment of common cleft lip palate. With the introduction of nasoalveolar molding, evidence has been obtained for improved long-term nasolabial aesthetics in both unilateral and particularly bilateral cases (Rubin et al., 2015). There is also a growing agreement on the claimed benefit that when combined with gingivoperiosteoplasty, it may obviate the need for secondary bone grafting (Dec et al., 2013). Furthermore, either with passive obturator (plate molding therapy) and primary bone grafting (Hathaway et al., 1999) or with active Latham directional dentomaxillary advancement (DMA) therapy without primary alveolar surgery (Chan et al., 2003), both presurgical protocols achieved dental arch relationships in preadolescent-like children without presurgical intervention. Apart from the above concerns and the burdens of PSIO, there is a consensus that it reduces the severity of the initial cleft deformity and enables surgeons and ultimately patients to routinely enjoy the benefits associated with the repair of a minimal cleft deformity.

Herein, we present a Tessier No. 4 facial cleft with severe deficiency of bone and soft tissue, displacement of the maxillary segments in all planes, and prolapsed eye globe. At the initial presentation, a soft-tissue repair appeared unfeasible without local flaps traversing the aesthetic units of the face. Surgically repositioning the globe and maneuvers for its maintenance were judged to be not practicable. Therefore, a decision was made to pursue PSIO. Employment of PSIO in an oro-ocular facial cleft is rare. A literature search revealed a Tessier No. 3 cleft treatment with a molding plate to ease lip closure (Stellzig et al., 1997) and recently a pin-retained active PSIO for bilateral rare No. 4 cleft (Maeda et al., 2014).

Case Report

Diagnosis and Examination

A 20-day-old boy was referred to our clinic with a Tessier No. 4 facial cleft. The family and prenatal histories were unremarkable. The diagnosis had not been made prior to birth. His birth weight was 3600 g. No auditory or visual problems were detected. The cleft was located lateral to the cupid's bow and philtral crest, at approximately midway between the philtrum and left

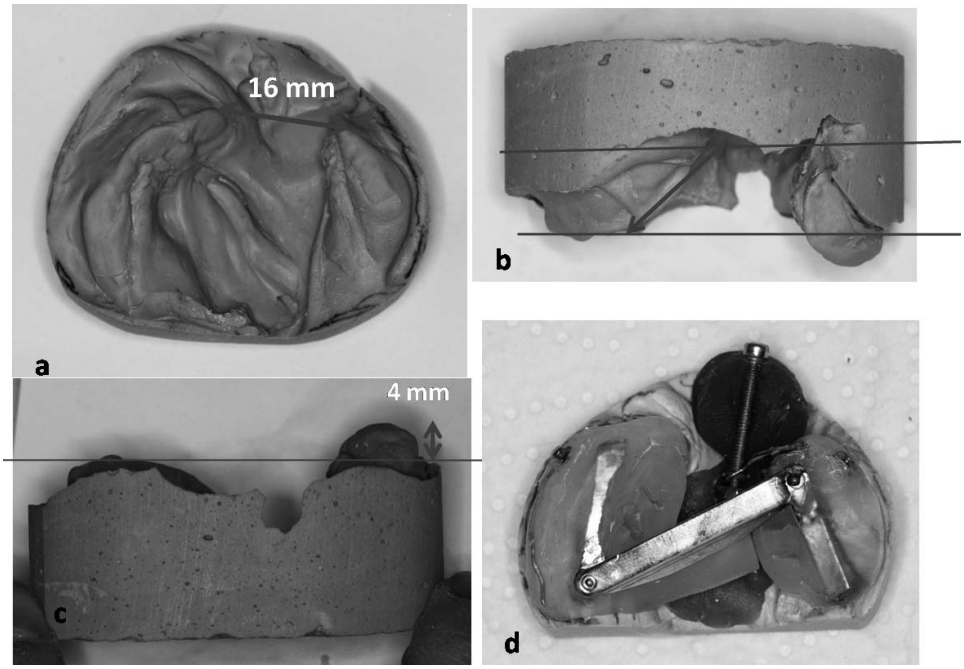

FIGURE 2 a: Alveolar cleft gap. b: Vertical premaxillary rotation. c: Vertical discrepancy between the alveolar segments. d: Modified framework of the first Latham device.

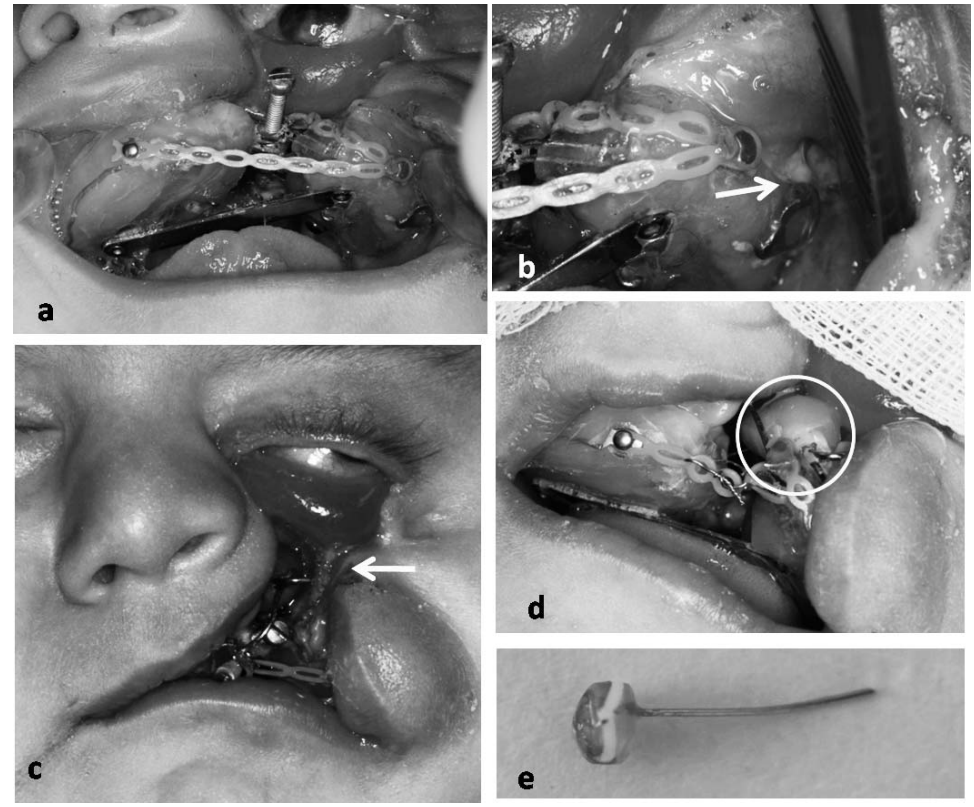

FIGURE 3 a: Latham device placed intraorally. b: Zygoma plate used for anchorage (yellow arrow). c: Prolene mesh base under the eye globe (yellow arrow). d: Eye part of the Latham device. e: Nasal septum bone pin.

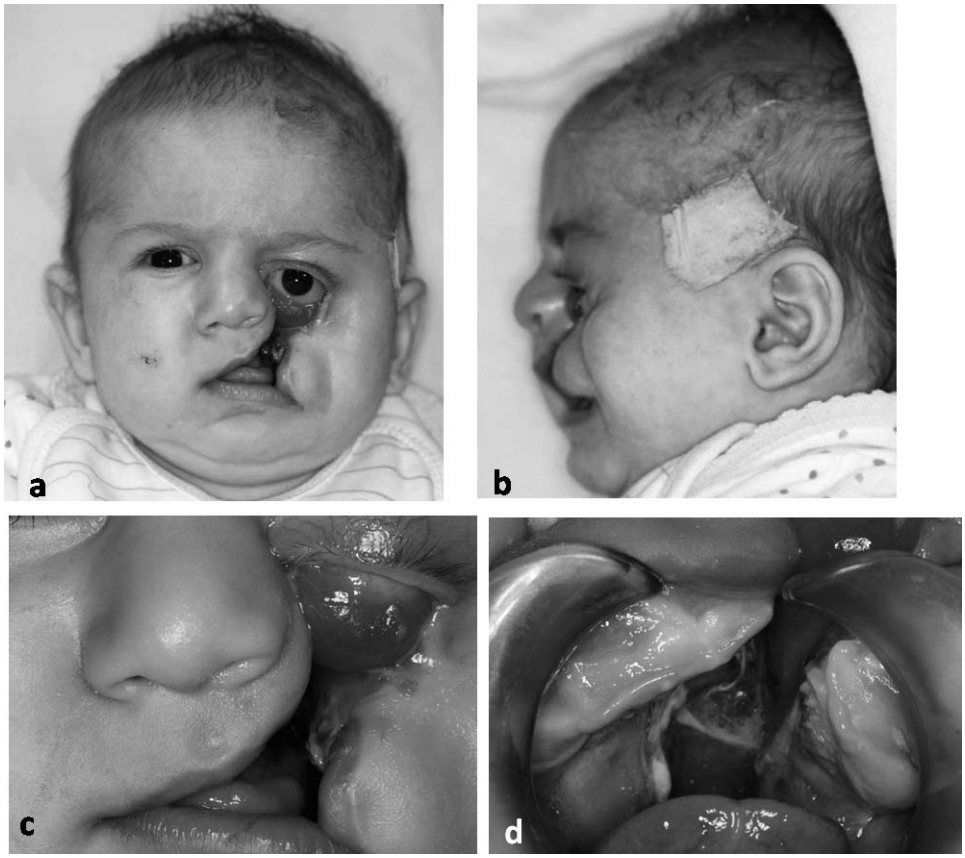

FIGURE 4 Facial and oral pictures after 8 weeks of Latham device.

corner of the mouth. The cleft continued onto the left cheek lateral to the nasal ala and ended in the lower lid. There was no sign of northbound continuation as No. 10 cleft in the upper lid, brow, forehead, and hairline. The contralateral side was completely normal. The size of the globe was comparable to the intact one. Although the nose looked anatomically normal, the left ala rotated upward. Excluding the tiny remnants at the medial and lateral corners, the lower lid was almost absent. The eye including the medial canthus was severely displaced inferiorly into the cleft. Associated with these findings, the midface was seriously diminished vertically on the cleft side (Fig. 1a and b). The cleft widely involved the alveolar process and extended posteriorly as a cleft of the secondary palate. The separation between the nasal cavity and the maxillary sinus remained intact. However, the orbital cavity, maxillary sinus, and oral cavity looked confluent (Fig. 1c and d). Ophthalmologic examination revealed sufficient upper lid closure and presence of cornea reflex. Premaxilla looked protruded and exhibited upward rotation into the defect. Assessment of the cast revealed a –6 mm alveolar cleft gap and a 4-mm vertical discrepancy between the greater and lesser segments (LSs; Fig. 2a–c).

Objectives and Performance of PSIO

The objectives of PSIO were defined as follows: (1) Narrow defect gaps for possible soft-tissue repair with minimal scar within the junctions of aesthetic units. (2) Reposition and align maxillary segments, which is also important to improve midfacial vertical symmetry. (3) Raise the eye for incremental correction and sustenance of the orbital dystopia in the novel position. To hasten treatment and attain the objectives, fixed directional orthopedic traction mechanics (Latham, 1980) known for effective skeletal platform correction in bone marker studies (Spolyar et al., 1992; Spolyar et al., 1993) were chosen. A silicone impression was taken to create the cast and to fabricate the device. Cast assessment indicated the LS was not posteriorly displaced contrary to expectation, while the greater segment (GS) was rotated to the noncleft side. Latham's DMA device was modified considering the patient's alveolar characteristics (Fig. 2d). By design, the LS component related to the zygoma to optimize device anchorage. The intersegmental anchorage (hinge cross-bar) was modified to a "Z" configuration instead of a "U" configuration. Hinge action at the cross-bar ends then occurred at the anterior of the LS and posterior tuberosity of the GS.

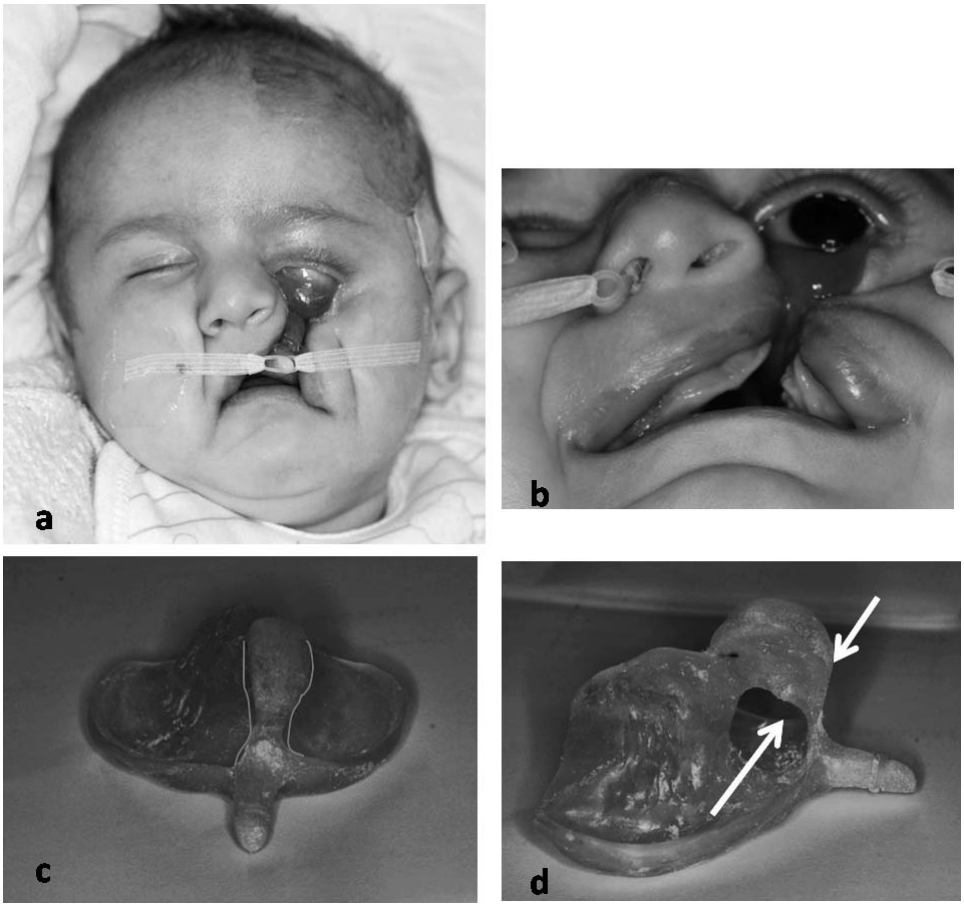

FIGURE 5 a, b: The molding plate for moving the eye upward. c, d: Grinding the molding plate transversally for closing the maxillary cleft gap.

Hence, the pivot center of the moment arm at the tuberosity became relatively posterior to increase the anterior arc of rotation for more medial translation of the LS. The Latham device was inserted under general anesthesia (Fig. 3a). The zygoma anchorage was performed by using a three-hole miniplate with one hole for plate attachment secured by light-cured material (Triad Gel, Dentsply, York, PA) and another screwed into the zygomatic bone (self-drilling orthodontic screw 1.6 × 10 mm; Fig. 3b). The device drive screw was activated a three-fourths turn daily. A nasal septal button-head pin (Spolyar, 2013) was inserted through the nasal septum and chain activated to the cleft side for septo-premaxillary molding. This pin was activated once a week by means of an elastic chain (Fig. 3e). Furthermore, an eye part added to DMA elevated the globe with two DMA devices applied successively and each maintained for 4 weeks. A second DMA was designed as previously described for greater distraction of LS to medial. Here, a zygoma anchorage was made onto the GS to improve the stimulated adjustment response of LS. Acrylic was added to the eye part of the device twice a week to elevate the globe gradually, and a Prolene mesh was attached under the globe to benefit from its tissue ingrowth effect (Bellón et al., 1994; Fig. 3c and d). Following 8 weeks of DMA treatment (Fig. 4a through d), a removable molding plate was applied and sequentially modified to reposition the eye. When the residual alveolar cleft gap (4–5 mm) was closed completely, closure of the malar cleft was achieved by grinding the acrylic plate transversally (Fig. 5a through d). Five weeks later, closure of the malar cleft was accompanied by formation of a bony septum under the elevated eye. Strip bands were applied across the cleft on the face to help cleft closure and as a means of soft-tissue relaxation. Another strip band was used also at the left lateral canthal region to move the soft tissue upward (Fig. 5a). The left eye was regularly checked for exposure disturbances, and no extra measure was required due to sufficient upper eyelid closure. The overall PSIO took 14 weeks, and primary surgical repair was performed at the age of 17 weeks (Fig. 6a through d).

Surgical Procedure

At the lateral side of the cleft, an incision was made at the skin-conjunctiva and skin-mucosa junctions starting

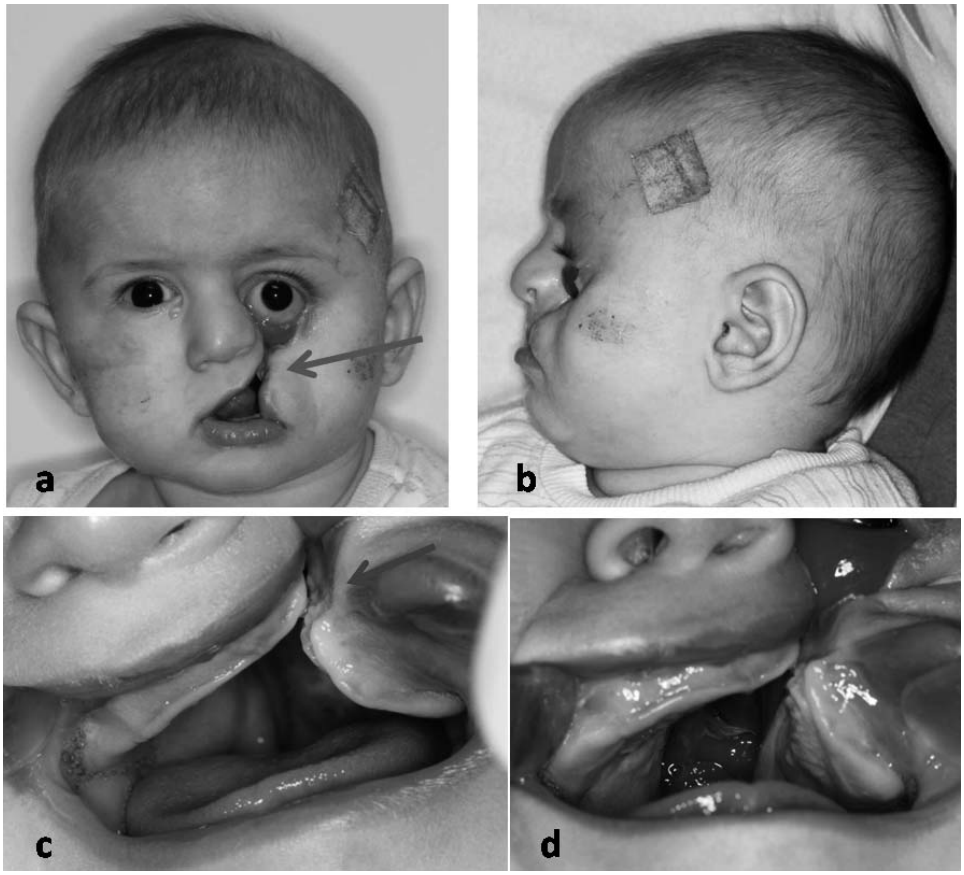

FIGURE 6 Facial and oral pictures at the end of presurgical orthopedics. Note cleft closure at both alveolar and maxillary regions and the markedly reduced vertical eye asymmetry.

from the lateral canthus to the point at which the white roll ended. As in the repair of unilateral cleft lip, A "V" flap was designed in the vermilion to adjust for the vermilion deficiency on the medial side. Lower lacrimal canaliculus was explored through 4.5× magnification but was not found. The dissection was deepened under the superficial muscular aponeurotic system by considering the facial nerve branches. A Tenzel semicircular rotation flap was added in the same submuscular plane by cutting the lower lateral canthal tendon. Despite this maneuver, the flap did not reach the medial canthus location projected according the normal side. Therefore, a paranasal vertical flap with a base (pivot) on the projected medial canthus was prepared. Some residues of medial canthal tendon were dissected and secured to the bone at its new location with a nonresorbable suture tied over a button on the contralateral side. Thus, the suture line was brought to within the nose-cheek junction. The alar wing was dissected from the piriform aperture, rotated inferiorly, and fixed in that position with a deep suture. The cleft of the lip was repaired by using the rotation advancement design as usual. Thus, the lip tissue between the cleft and the philtral column was discarded on the medial side Surgical procedures are demonstrated in Figure 7a and the closure at the end of the operation in 7b. Facial and oral pictures at 3.5 months postoperation are also seen in Figure 8a through c.

Discussion

By achieving all treatment objectives on this unusual Tessier No. 4 cleft, the vertical eye dystopia was effectively relieved, a bony septum sustaining this restored position was brought up, maxillary segments were anatomically aligned, the cleft size was far reduced, and soft-tissue closure within the nose-cheek border became feasible. Local flaps and remarkable z-plasties that would violate the borders between aesthetic facial units became unnecessary. Disfiguring scars, trap-door deformities, and tissue mismatches were avoided. Bone grafting was unnecessary. PSIO enabled an optimized primary surgical repair. Yet, in the early postoperative period, considerable tension was noted at the repair site. This *in situ* residual load became visibly manifest most prominently with lower lid retraction, reported as a frequent problem by Versnel et al. (2011). In succeeding surgeries, tension-relieving measures as with use of tissue expanders and those addressing the tarsal skeleton may alleviate this problem.

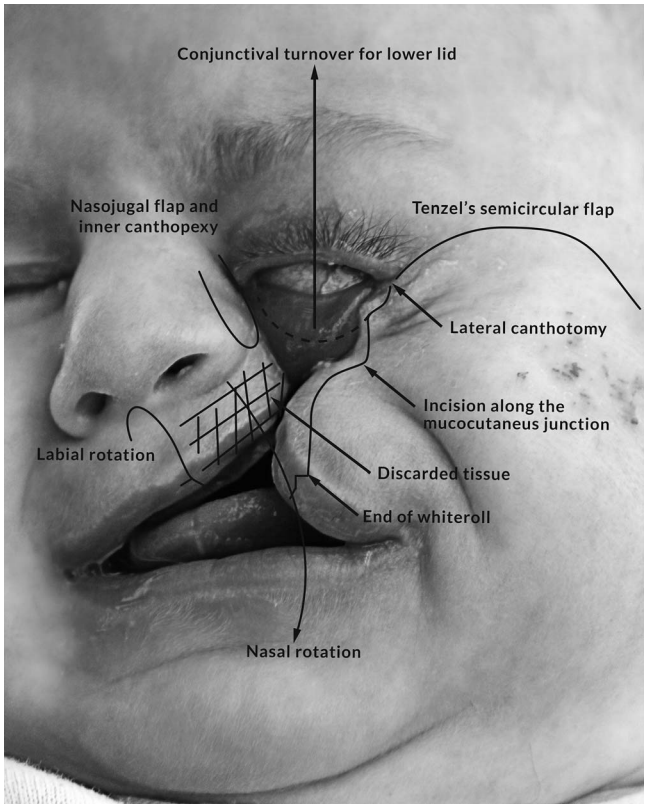

FIGURE 7 a: Diagrams of the surgical intervention. b: Final closure. Tarsoraphy was maintained for 5 days.

Achieving good facial growth, aesthetics, function, and speech are primary goals in cleft rehabilitation. Midface hypoplasia may be apparent at birth in some patients but becomes visible at around age 10 in others (Versnel et al., 2011). The hypoplasia is due to intrinsic growth retardation to some extent (Versnel et al., 2011). Also, the greater the number of interventions in the growing maxilla with resulting scar, the more growth retardation (Anastassov and Joos, 2001) should be expected. On the other hand, multiple surgeries are usually required for these anomalies from infancy to the adulthood (Versnel et al., 2011) for a satisfactory result. Severe three-dimensional underdevelopment was initially presented in our case. This was relieved to some extent by using PSIO. Long-term implications of this treatment cannot be foreseen (Ross, 1995; Henkel and Gunlack, 1997; Hathaway et al., 1999; Chan et al., 2003). Although different presurgical orthopedics devices have been developed to approximate the cleft segments, in the presented case, the Latham device provided efficient and predictable closed distraction for sufficient orthopedic correction by stimulated sutural adjustment response (Latham, 1974) during infantile rapid growth. During this infantile period, not only the alveolus but also the ascending maxilla and orbital floor were molded, and the eye globe was relocated. Once the eye was moved upward, a space arose below the eye, and this gap was closed by grinding transversally from the acrylic molding plate, permitting greater closure.

Reviewing the literature on treatment of oblique facial clefts revealed three challenging issues (Tessier, 1977; Mishra and Purwar, 2009; Laure et al., 2010). One was stated as skin shortage at the nasomalar area (Menard et al., 1999). Generally, Z flaps were used to increase the distance between the canthus and the lips. Tissue expanders placed under the cheek skin have been also used for tension-free closure (Menard et al., 1999). In addition, use of elastic bands presurgically has been suggested to reduce soft-tissue strain after surgery (Grayson and Shetye, 2009). It may have limited value, as seen in this case.

The second issue has been the asymmetric appearance of the eyes in unilateral cases, (Kara and Oçsel, 2001; Laure et al., 2010, Chen et al., 2012). A critical factor in determining the severity of orbital dystopia is the ability of the upper lid to cover the cornea (Mishra and Purwar, 2009). Moreover, reported failure of the normal upper eyelid to cover the cornea is a risk leading to exposure keratitis and blindness (Mishra and Purwar, 2009). Therefore, eye protection is most essential in these cases. In the present case, the upper eyelid showed sufficient closure of the eye, and cornea coverage was present. Thus, there was no need to cover the eye or for temporary tarsorrhaphy.

In previous reports, the reconstruction of the canthal area and the eyelids has been demonstrated in different ways (Sesenna et al, 2012). To define the canthus position in unilateral cases, the contralateral side can be used. Some authors used the transposition of a median forehead flap as an alternative approach (Van der Meulen and Vaandrager, 1989). Our presurgical orthopedics enabled the reduction of the soft-tissue strain causing the epicanthal fold after the canthoplasty procedure and also enabled a nearly symmetric eye position.

Third, although primary bone grafting was unnecessary in the case presented, its timing for the orbital floor and ascending maxillary region has been controversial. Tessier (1977) recommends one-stage surgery with different flaps, extensive dissections, and multiple osseous grafts. Some authors suggest that grafting should be performed at the same time as primary closure for preventing orbital dystopia (Tessier, 1977; Galante and Dado, 1991; Sari et al., 2003). They advocate early intervention in case exposure keratitis is imminent. Sesenna et al. (2012) stated that the indication for early bone surgery would be warranted depending on the width of the orbital floor defect, adequacy of eyelid closure, and whether the protection of the eye could be obtained simply with soft-tissue surgery alone. Finally, the advisability of harvesting bone grafts in infants is controversial (Resnick and Kawamoto, 1990). Bone grafts are used to fill the cleft or as an onlay graft along the orbital border to compensate for maxillary hypoplasia. However, it was reported that bone resorption was frequent and often required multiple graftings (Resnick and Kawamoto, 1990; Aköz et al.,

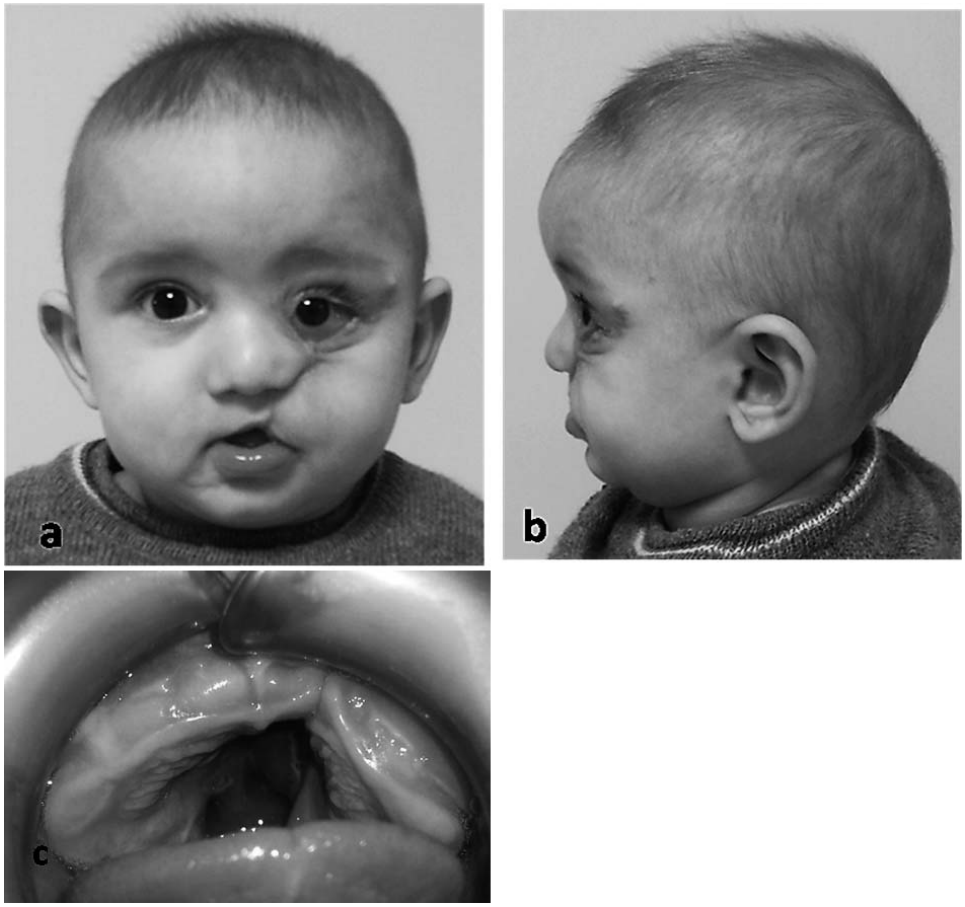

FIGURE 8 a–c: Facial and oral pictures at 3.5 months postoperative.

1996). Hence, bone-grafting procedures have been usually postponed to older ages (Tessier, 1977; Sano et al., 1983; Boo-Chai, 1990; Mishra and Purwar, 2009; da Silva Freitas et al., 2010). Midfacial deficiency has been reported in Tessier cleft 3, 4, or 5 cases with early maxillary bone grafts (Versnel et al., 2009; Versnel et al., 2011). Yet, unilateral maxillary hypoplasia has been accepted an intrinsic feature of unilateral oblique facial clefts. (Versnel et al., 2009). Versnel et al. (2009) stated that owing to deficient growth of all affected tissues in the cleft area, the subtle hypoplasia of one side of the midface at birth becomes more obvious over the years. Similarly, Van Der Meulen and Vaandrager (1989) treated nine patients with facial cleft by performing conventional procedures described in the literature, but the results, although initially acceptable, soon deteriorated. In this case, formation of a bony floor for the elevated position of the eye concomitantly with the approximation of maxillary segments was accomplished while adjusting the eye component of intraoral appliances. Maxillary realignment and sustainable eye repositioning with PSIO may reduce the conspicuousness of the deformity and may delay the use of bone grafts to postadolescent years with less psychosocial impairment in early childhood.

The rare facial clefts that present with severe tissue hypoplasia are real reconstructive challenges, with modest results even when approached in a multidisciplinary way. Therefore, discovery of the pathological mechanisms underlying these disorders may lead to potential advances in their treatment. The recently found mutations associated with oblique facial clefts point to a genetic basis for these disorders such as the common clefts and may have promising implications for the treatment of these disorders (Gfrerer et al., 2014).

Conclusion

A series of presurgical orthopedic procedures in a severe oblique facial cleft provided bony cleft closure at orbital floor, cheek, and alveolar process. This allowed a primary soft-tissue repair respecting aesthetic units of the face. Vertical eye asymmetry was considerably reduced. Lower lid retraction due to tight closure imposed secondary procedures for this region. Early results suggest that the number of operations would be significantly less than usual for these cases, that most of them could be postponed to the years when bone growth is completed, and that the

approach combining orthopedic and surgical procedures is superior to surgery alone.

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