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Review Article

Paediatric Facial Cleft Treatment - Lessons Learned

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Abstract

The title of this publication includes the words "Lessons Learned" in order to emphasise the knowledge and experience gained in more than 4 decades in treating 5000+ patients with facial-oral cleft anomalies. The choice, timing and optimization of the most appropriate paediatric surgical and paediatric orthodontic treatments are reflected in the extremely satisfactory treatment outcomes. This paper delineates hierarchically the conceptual framework of treatment for the most profound to the least severe cleft anomaly. Over the years multiple treatment techniques, options and protocols have been published internationally and yet the young professionals often use the procedures and timing of interventions they have learned from their 'masters' or seniors, irrespective of whether these have been chosen based on the best long-term treatment outcomes. This paper advocates specific treatment protocols and techniques, based on the results of research, on observation, clinical evaluation, judgement of outcome and the ideal treatment of a specific type of cleft anomaly, in order to optimize long-term outcomes. The initial short-term result, as well as the long-term facial growth, functional and aesthetic influence or their combined influence, have been recorded until the onset of the adolescence age. The application of technique(s) appropriate for treatment of a particular type of cleft anomaly are described and motivated for use, without mention the so-called original or first person who applied or described or used a particular technique or procedure for the first time. Conversely, those treatment technique(s) utilized during the very important developmental phases of the neonate, the infant and the child and which have had a profoundly negative effect on function and/or facial growth and/or facial aesthetic in the long-term, are also described, without mention the name of the person(s) who advocated their use. Paediatric patients who have been treated by means of these latter surgical techniques or interventions which may lead to undesirable or insufficiently positive outcomes, may require extensive drawn-out surgical restructuring and orthodontic treatment during their adolescent's years.

Keywords: Cleft Lip Palate; Paediatric Cleft Treatment; Cleft Ultimate Treatment; Cleft Comprehensive Surgical-Orthodontic Guide; Obturpaedics; Orthopaeddontics; Cleft Divisions

Abbreviations

C: Cleft; L: Lip; A: Alveolus; P: Palate; hP = Hard Palate; SP: Soft Palate; hPsP: Hard+Soft Palate; U: Unilateral; B: Bilateral; Oblique: Tessier type 0;4-14;30.

Introduction

The treatment protocol for paediatric cleft lip and palate or facial cleft anomaly, determines the conceptual framework designed to elucidate diverse cleft classifications, thereby optimizing treatment strategies for achieving optimal long-term outcomes. Notably, sections characterized by a higher incidence of specific cleft types also tend to exhibit a correspondingly elevated prevalence of severe and/or challenging soft and hard tissue defect. Whereas such correlation is clearly and distinctly reflected

in the clinical manifestations across various cleft types, however, excludes the group of extremely rare cleft facial anomalies as central-facial, oblique-facial, lateral-facial and mandibular clefts (Tessier type 0;4-14;30). Therefore, the conceptual presentation as delineation within the protocol consequently progresses hierarchically from the most profound to the comparatively milder categories of cleft anomalies. The combination of *'cleft lip - cleft alveolus - cleft hard palate - cleft soft palate'* type (CLAP = ±41%) is the most frequent type, representing the most severe anomaly, followed by the 'cleft soft palate' type (sP = ±18%) and the combination of '*cleft hard palate - cleft soft palate*' type (hPsP = ±17%). The next category of the *'cleft lip - cleft alveolus'* type (CLA = ±14%), is recorded more than twice than the isolated *'cleft lip'* anomaly (CL = ±6%) (Fig. 1). Both these types may also present as unilateral or bilateral deformities, similar to the CLAP.

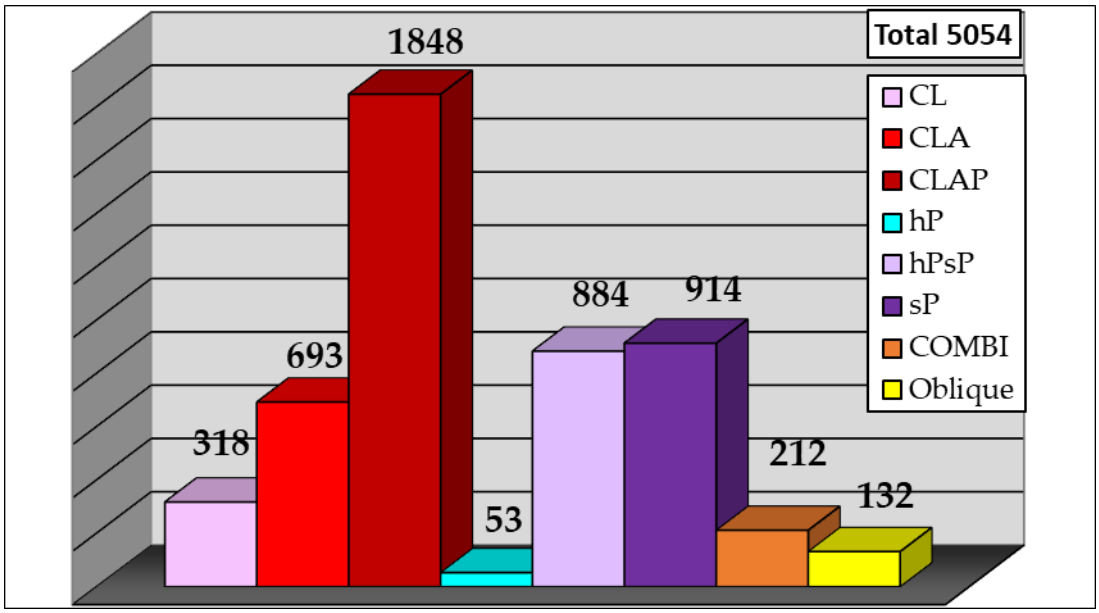

Figure 1: 5000+ facial-oral cleft anomalies.

Each of the aforementioned cleft types necessitates a particular treatment approach in order to optimise long-term surgical and orthodontic outcome. The significance of the early orthodontic treatment in paediatric cleft patients is very often undervalued despite its eventual very positive impact on the ultimate long-term result. Orthodontic treatment in these patients is essential, especially during the two pivotal phases; the infant presurgical period (obturpaedic treatment period) and in the crucial intermediate period, that coincides with the extreme active growth period of the child (orthopaeddontic treatment period). Subsequent to the completion of the obturpaedic and orthopaeddontic treatments, the orthodontic completion treatment becomes considerably less intricate. The final phase of orthodontic treatment culminates shortly before a possible potential last 'touch-up' surgery for lip and nose aesthetics at the onset of adolescence.

Concepts and Methods

1. CLAP (cleft - lip, alveolus, hard and soft palate)

This category of cleft anomalies is of paramount significance, not only due to its high prevalence (±41%), but also due to the extension of the deformity presented at birth, whether as a unilateral (±63%) or bilateral anomaly (±37%).

The Surgical Reconstruction

The cleft lip reconstruction or cheiloplasty for the CLAP closely resembles that of the isolated Cleft Lip (CL), serving as an aesthetic technique for both the primary cleft lip and primary cleft-deformity of the nose (cf. point 5 below).

An integral aspect in the cleft lip reconstruction process entails the strategic use of excessive lip red tissue, together with its adjacent cleft tissue for the formation of the anterior nasal floor [1,2]. Such a muco-subdermoid tissue flap for the anterior nasal floor reconstruction is therefore placed within the borders of the alveolar Cleft (CA), serving as a bridging structure in the sulcus, extending from the lip towards the hard palate cleft. This superior placement of the flap within the alveolar cleft, mitigates the

risk of the development of an anterior oro-nasal connection/fistula. Additionally, it establishes an anterior nasal floor to support the subsequent bone grafting, during the course of the secondary osteoplasty/osteofusion procedure [2-5]. The latter procedure is optimally undertaken between 8.5 to 9.5 years of age, corresponding to approximately ⅔ completion of facial growth.

The most optimal technique for closing the cleft hard palate in an infant/small child involves the use of a single-layered, cranial-based vomer-nasal-septum flap often referred to as one of the "vomerplasty"-types, avoiding at all costs a mucoperiosteal hard palate stripping [2,6-9]. The surgery of this special technique needs to be proceeded with caution. It is further enhanced with supplementation of a soluble gelatine plaque or a soluble lactose-acid plate (PdLAlLA) or very similar material, at the same horizontal level as the natural palatal vault [2,10]. This strategy aims to prevent the formation of a palatal groove.

A considerably less advisable alternative for the surgical closure of the hard palate, involves a delay of a procedure, preferably until the approximate age of 12 years (around ±5/6 completion of facial growth), in performing a muco-periosteal stripping technique of the hard palate [11-20]. Though there is some evidence that these techniques with delay can favour long-term midfacial growth [21,22]. However, poses a significant risk of leading to persistent speech impairment in a subset of patients [23-27]. It is of utmost importance to realize that any type of surgery that to a large extent or completely strips the muco-periosteum from the hard palate should never be used in the reconstruction of infants or small children. Further on, it is unequivocally discouraged to use any kind of variation of his technique. In the majority of cases, the stripping of the muco-periosteum from the palatal bone during infancy or early childhood, leads to severe midfacial growth disorders, manifesting in retrognathic or hypoplastic (retro-brevignathic) maxillae, as midfacial growth disturbances and subsequent dysgnathia, characterised by collapsed dento-alveolar arches and crossbites [21,22]. The correction of these effects, is often only partially feasible during the adolescence and typically demands extensive midfacial surgery and long-term orthodontics, demanding sometimes often surgical cranio-maxillo-facial distraction. Additionally, such corrective measures may result in residual permanent occlusal malalignment and velopharyngeal insufficiency.

The soft palate reconstruction in CLAP anomaly is the same as described for the isolated cleft Soft Palate (sP) (cf. point 2 below). It is imperative that the reconstruction extends slightly beyond the border of the hard palate bone, to prevent the development of an oro-nasal fistula at this particular anatomical transition zone.

The Orthodontic Treatment

To achieve most favourable immediate and lasting outcomes in patients with unilateral and bilateral CLAP, preoperative orthodontic management during infancy is necessary, where obturpaedics [28,29] is employed, a treatment possibility where several of an overall of seven available methods can be used for CLAP [29,30]. Furthermore, it is essential to rigorously use orthopaeddontic interventions during the important growth spurt period from 5 to 11 years of age [31]. It is imperative to avoid extraction of any than non-supernumerary primary and secondary teeth during this treatment intervention [28]. Moreover, it is highly significant to actively address any lateral and anterior crossbites in the mixed dentition phase [32] where the active midfacial growth is very active, therefore during the orthopaeddontic period. The primary deformities in CLAP cases are often involve the midface in various dimensions, including transversal (medio-lateral alveolar arch collapse), longitudinal (antero-posterior, such as retrognathia) and vertical (supero-inferior, as brevignathia/short-face with or without deep bite) [22]. These occluso-facial deformities must be corrected to establish an ideal initial jaw relationships before secondary osteoplasty/osteofusion procedures are undertaken.

2. sP (Cleft Soft Palate)

The reconstruction of the isolated (±18%) and other sP cleft type is crucial for a competent language production, however, is a notably intricate technical challenge. To a great extent, this complexity arises from the inherent shortage of soft tissue in this region, particularly in the antero-posterior dimension of the soft palate, being often exacerbated by an extreme cleft width and/or asymmetrical formation [33]. Furthermore, it is of utmost importance to avoid any infection by utilizing strict microbiological principles post-operatively and apply continuous testing for the first week. The risk of tissue compromise in the sP due to infections is a pressing concern [34,35]. The total cleft soft palate anomaly for sP, hPsP and CLAP is approximately 76%.

A crucial aspect represents the muscle reconstruction within the soft palate. In case of a submucosal to an extremely wide soft

palate cleft anomalies, the four pivotal soft palate muscles or muscle bundles should be dissected separately and anastomosed with the opposite counterpart. This intricate functional procedure is performed during an extended Intravelar Veloplasty (eIVV) [2,36-43]. The following muscles are reconstructed separately: 1) the tensor palatine muscle, for the activation of the pharyngo-tympanic (Eustachian) tube; 2) the levator palatine muscle, as the main elevation muscle sheet and 3) the combined palatoglossal-palatopharyngeal muscle bundle for the posterior velo-pharyngeal sphincter function [2,38-41]. To achieve partial lengthening, maintaining a normal aesthetical appearance, an uvuloplasty performing as a v-y-plasty of one of the two uvulas [42,43]. Superiorly to the uvula a temporary tonsillar pillar cross-over stitch that supports the healing process is used initially for a more V-shaped configuration of the tonsillar pillars [42].

In case of a quite narrow cleft this can be achieved through the application of double-opposing z-plasties, effecting a rotation and an overlapping of the "*cleft*" muscles [2,44].

In extremely short antero-posterior soft palate clefts, a substantial lengthening of the soft palate, involving the hard and soft palate anatomical transition zone, in using bilateral buccal fat pads [2,45] and to a lesser extent buccal-myo-mucosal flaps [46]. These additional tissue flaps are positioned in an overlapping manner between the hard and soft palate, employing a "Hammock hanger" technique alongside double-opposing z-plasties of the mucosa with muscles. This approach aims to achieve the considerable lengthening and velopharyngeal competence. Optimal timing for the sP reconstruction is advocated at the onset of the 'phonetic age' (±5-7 month of age) to facilitate enhanced speech development.

Speech Therapy Treatment

Initiation of active speech therapy is to established the correct placement of articulators with appropriate air flow [47]. It typically commences at approximately two years of age. In cases where velopharyngeal incompetence is confirmed, a velopharyngoplasty [2] may be considered, but only at the onset of the orthopaeddontic period. Early implementation of a velopharyngeal flap may potentially impede midfacial growth; however, any resultant dysgnathial influence can be mitigated through active orthopaeddontic intervention. Consequently, surgical placement of the velopharyngeal flap is not recommended during infancy or very early childhood. In instances of recurrent tonsilitis, a tonsillectomy, rather not than an adenectomy, may be contemplated under specific circumstances to maintain proficient speech.

3. hPsP (Cleft Hard Palate and Soft Palate)

To optimize the outcomes for individuals with hard palate and soft palate clefts (hPsP, approximately in 17%), while minimizing complications and very limiting dissection at the hard palate, the hPsP is addressed in two separate procedures, The initial stage involves reconstruction of the soft palate (eIVV), followed by a subsequent delay of 12 to 18 months, before addressing the hard palate cleft.

The Surgical Reconstruction

The basic reconstruction of the soft palate cleft is the same eIVV as described under point 2. The reconstruction of the hard palate, in combined Hard and Soft Palate cleft (hPsP) cases, involves a clearly different technique compared to the hard palate reconstruction in CLAP cases. In the majority of hPsP cleft cases are bilateral (in approximately 98%) and the hard palate cleft length is often significantly shorter (in approximately 82% in cases) than the length of the anatomical hard palate. The vomer-nasal septum is predominantly situated superiorly and separated from the bilateral cleft palatal shelves in this cleft, rendering it mostly inaccessible for palatal reconstruction.

The preceding closure of the sP cleft, followed by a 12 to 18 months delayed hard palate closure in infants or small children, allows for substantial medio-lateral growth of the hard palate. This time slot results in a substantial narrowing of the medio-lateral hard palate cleft size [48]. Therefore, the requirement for this approach is a tiny, partial incision (in width and length), resembling a micro to miniature unilateral or bilateral 'Von Langenbeck'-incision in the hard palate [2]. The width of this lateral minimized mucoperiosteal flap is determined by the width of the residual hP cleft deformity. Importantly, the hard palate in toto is certainly never or should be stripped for any types of major mucoperiosteal flaps [13-20]. This approach, based on an absolute tiny muco-periosteal flap due to the spontaneous narrowing of the hP and the absence of a Cleft Alveolus (CA), exerts minimal impact on midfacial growth development [2]. A notable key advantage of this approach is that breathing/ventilation

can still be maintained and be secured through the hP-cleft opening for a 12 to 18-month period after the sP reconstruction. This particularly proves most advantageous in infants born with a Pierre Robin sequence, subdivision Fairbairn-Robin triad [48], that occurs in approximately 32% of hPsP cleft cases.

An alternative strategy involves a long delayed surgical intervention for the hP or hPsP, at approximately 12 years of age, beyond the orthopaeddontic treatment period, however, with inherent drawbacks regarding a proficient speech development [25-28]. Once more, advocating the closure of the hPsP, as it is similar applicable in CLAP, using a comprehensive muco-periosteal stripping procedure of the total hard palate is unequivocally discouraged in infants and small children [13-20].

The Orthodontic Treatment

The obturpaedic interventions for the standard hPsP cleft may be modified with a slightly extended type of Obturator (OwE), with a small lengthening into the soft palate defect. However, the initial life-saving baton appliance is highly recommended for the infant Pierre Robin sequence in also avoiding mostly possible mandibular distraction surgery [28,29,50,51]. During the foetal period of the Pierre Robin sequence, microglossia predominantly occupies the cleft, exerting a negative influence on the mandibular growth. Consequently, treatment during the most active growth spurt phase, referred to as the orthopaeddontic phase [31], in children with congenital mandibular micrognathia or retrognathia, may stimulate mandibular growth actively, leading to a more favourable relationship between the upper and lower dental arches, as well as a more balanced aesthetic facial appearance [29].

4. CLA (Cleft Lip and Alveolus)

This group represents the fourth common cleft type (±14%). It may present as unilateral (±82%) or bilateral (±18%) anomaly.

The Surgical Reconstruction

In cases of a complete Lip and Alveolus Cleft (CLA), the surgical approach to lip reconstruction in CLA cases closely mirrors the one used for isolated Cleft Lip (CL). Consequently, an aesthetic surgical technique is employed to address primary lip and nose deformities (refer to point 5 below).

The reconstruction of an anterior nasal floor flap is executed, as previously described above (cf. point 1) in the superior part of complete alveolar clefts (CA). In CLA cases, where the alveolar cleft presents only as a notch or a partial mucosal or partial osseous cleft without involvement of the nasal floor, the required surgical intervention is limited to a cleft lip reconstruction alone.

The Orthodontic Treatment

Similar to the orthodontic treatment in CLAP, it involves obturpaedic treatment, primarily through cleft lip moulding and/or NAM1 (nasal-alveolar moulding) or NAM2 (nasal-alar moulding) [2,29,52]. However, a critical facet of the treatment during the orthopaeddontic phase, especially when addressing a potential anterior crossbite is to rectify this anterior maxillary part prior before the implementation of the secondary osteoplasty/osteofusion as outlined in point 1 above [31].

5. CL (Cleft Lip Anomaly)

Whereas this type of isolated cleft (approximately 6% of CL) appears to a remarkable 87% unilateral and only in about 13% bilateral. Compared to bilateral anomalies to CLAP and CLA, this ratio is consistently lower. The total cleft lip anomaly for CL, CLA and CLAP is approximately 61%.

The Surgical Reconstruction

The surgical reconstruction for the unilateral cleft lip and nasal deformity for CL, CLA and CLAP requires the attainment of a flawless aesthetic reconstructive result [53-63]. The optimal design during the lip-plasty or cheiloplasty for the perfect result should entail the following: a precise horizontal alignment of the mucosal red wet-dry lip line and the white-red lip roll with its Cupid's bow perfectly adjusted horizontally. Both these lines necessitate a tiny, for the white-red roll, aesthetically Z-triangular or v-y connection to prevent the formation of a vertical scar at these anatomically and aesthetically sensitive visible areas;

otherwise, visible scars or evidence or surgery will remain permanently at these regions after cleft repair.

The vertical cutaneous scar should be strategically placed to mimic a philtrum-edge or philtrum column, mirroring the natural non-cleft philtrum-edge [2,64]. The triangular shaped, rotated tissue, necessary for lip lengthening on the cleft side, is situated at the level of the nasal sill-nasal floor, positioned adjacent to the columella base. It is important, never to place such a triangular shaped piece of tissue for lip lengthening horizontally or obliquely into the area between the given white-red roll and the nasal floor. This would aesthetically compromise the vertical cutaneous dimension, thereby violating the created 'philtrum edge' or the scar mimicking the 'philtrum-edge'. The achievement of a central dimple effect involves the creation of a shallow natural concavity between the 'philtrum-edges', bridging the gap between the natural column and the lip scar [2,64].

The bilateral cleft lip reconstruction follows a principle similar to that of the unilateral CL, however, with some additional surgical considerations. The reconstructed prolabium with specially its red-lip mucosa, is typically smaller in size, must have a volume matching with that of the lateral red-lip mucosa [65]. The width, as a horizontal length, at the cutaneous-red lip mucosa area of the prolabium aims to emulate a Cupid's bow [66]. The bilateral cutaneous scars are strategically positioned to mimic 'curved or preferable less straight' philtrum edges/columns [2]. The appearance of the prolabium is significantly influenced by the width of the columella's foot plate. No horizontal incisions should be made in this important aesthetic area (refer to point 6 below). The final design may emulate the characteristics of one of the parents, creating a more natural 'inherited' appearance. Consequently, an evaluation of the parents' philtrum edges and lip length is important before commencing the bilateral lip surgery of an infant or small child.

To achieve a dimple effect, a shallow natural concavity is created between the 'philtrum edges', i.e. between the cleft lip scars. This effect is achieved by maintaining a separation of approximately 2 mm between the lateral orbicularis muscles parts during suturing and placement of a central horizontal pull-down suture intra-orally [2].

The Orthodontic Treatment

In the pre-primary surgical treatment phase, the obturpaedic treatment may include moulding the cleft lip width and defect by means of lip-taping and implementation of a NAM2 (nasal-alar moulding) [29].

6. Columella lengthening (for bilateral CL, CLA, CLAP)

The lengthening of the columella in a bilateral cleft lip (approximately 17%, of which approximately 87% present in a complete lip length deficiency) is typically undertaken at the age of 12 months to achieve a favorable long-term outcome through positive projection growth of the nasal dome, while minimizing lateral nasal skeletal growth stimulation. This prevents the widening of the nasal skeleton in the active growth phase. The surgical technique used for the primary bilateral cleft lip reconstruction, as well as the procedure for lengthening the columella should preserve the natural supero-inferiorly curved appearance at the central columella-lip-junction and narrowing the nasal sill area [2,67]. An important 'curved' natural aesthetic landmark, crucial for maintaining the natural appearance should never be compromised by any type of complete horizontal incision during the primary bilateral cleft lip reconstruction or during the columella lengthening procedure.

7. COMBI cleft (a subgroup of CLAP)

The combination cleft anomaly, described as COMBI (COMBI = ±4% of CLAP) may present as follows: CL+sP (with no CA and hP), CLA+sP (with no hP) or CL+hPsP (with no CA). Depending on their particular presentation, adequate variations of the previously described treatment approaches may be applied (cf. points 2 - 6).

Conclusion

The paediatric treatment outlined above for various facial clefts are grounded on more than four-decades of surgical and clinical experience. Drawing from extensive clinical observations, long-term monitoring and research, encompassing over 5000 reconstructed former facial cleft patients, across four distinct racial and cultural groups, the respective protocols have been continuously evaluated and where necessary adjusted with the aim of constantly improving post-operative optimal long-term result. The clinical findings are continuously and meticulously catalogued within a well-documented database.

Of paramount significance amongst these anomalies assessed with their long-term outcomes, over a minimum period of 14 years, are the CLAP and the sP anomalies. Accordingly, the continuing improvement of the ultimate long-term results for these two main anomalies in a cleft clinic or unit is a basis for the contingent upon the election of the optimal surgical techniques in infants, coupled with stringent adherence to obturpaedics, orthopaeddontics and speech-therapeutic interventions. The ultimate judgement of such a cleft clinic or unit will be rendered by medical and dental peers, allied health care providers, the parents and other family members of children with clefts and finally, by the affected children themselves.

Conflict of Interests

The authors have no conflict of interest to declare.

Language Evaluation

A.C. Wolmarans.

Confirmation of Patient's or Parent's Permission

Not applicable

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