The Impact of Alveolar Bone Grafting on Cleft Lip and Palate: A literature review

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ABSTRACT

Introduction: Alveolar bone grafting between the ages of nine years to eleven years is a routine procedure for children with a cleft involving the alveolus. It is believed to encourage dental development and subsequent treatment within the region of the cleft and to improve nasolabial aesthetics. The aims of this article are to review the literature as to its impact on dental development and subsequent treatment, nasolabial aesthetics and the nasal airway.

Methods: An electronic search was conducted using Medline and Embase, with no restriction as to date of publication, study design or language.

Results: The results suggest that secondary alveolar bone grafting when carried out at the appropriate time has significant benefits and for subsequent dental treatment, often allows space closure of adjacent teeth and eliminating the need for a prosthesis. Although it has an effect of nasolabial aesthetics it is equivocal as to whether this improves nasolabial aesthetics or merely improves the likelihood of aesthetic improvement of subsequent nasal surgery. Nasal obstruction is a significant issue in patients with cleft lip and palate with smaller nasal volume and mean cross-sectional area. It would appear that there is a reduction in the growth of the airway after an age that approximates to the timing for secondary alveolar grafting, although there are no studies that can refute or confirm its actual impact.

Conclusions: Alveolar bone grafting between the ages of 9 - 11 years appears to produce clear benefits in terms of dental development and subsequent dental treatment. Its impact on nasolabial aesthetics appear equivocal as although there are changes in some landmarks post-surgery it is unclear as to whether these changes produce a benefit in terms of aesthetics for the patient.

Gillgrass TJ, MacDonald JP, Mossey PA, Welbury RR. The Impact of Secondary Alveolar Bone Grafting on Cleft Lip and Palate: A literature review, South Eur J Orthod Dentofacial Res. 2014;1:15-22.

Submitted: November 11, 2013; Received in revised form: May 09,2014; Accepted: June 22, 2014; Published: September 12, 2014

INTRODUCTION

Cleft lip and palate is the most common craniofacial anomaly with a prevalence of 1 in 700 live births. The implications of having a cleft involving the lip, alveolus and palate are many but include: aesthetics and its psychological impact, speech, dental development and facial growth. The standard protocols for surgical management in the UK involve lip closure before the age of 6 months to improve facial aesthetics and encourage maternal bonding, and palate closure by the age of 1 year to encourage normal speech development. Although these operations essentially close the cleft

Corresponding Author: Toby J. Gillgrass Consultant Cleft Orthodontist, Department of Oral Orthopaedics, Glasgow Dental Hospital & School, 378 Sauchiehall Street, Glasgow, Scotland G2 3JZ e-mail: toby.gillgrass@nhs.net of the lip and palate the cleft involving the alveolus is usually left until a later age.

Attempts at early closure (within the first year of life) have some advocates but involve a course of pre-surgical orthopaedics to approximate the cleft segment allowing the periosteum of both segments to be joined (primary bone grafting). This early correction of the alveolar defect is appealing but appears to lead to unfavorable growth compared to later conventional grafting ¹ and although advocates suggest precludes the need for further grafting in many cases ² this depends upon whether orthodontic space opening is the primary objective in later orthodontic treatment.

Secondary grafting was first suggested by Boyne and Sands ^{3,4} is now considered the 'norm' in Europe rather than primary alveolar grafting. It involves local mucoperiosteal flaps being raised to fully define the cleft, the nasal floor is then closed and

the 'pocket' produced is filled with autogenous cancellous bone harvested from the anterior iliac crest. The mucoperiosteal flaps are then sutured to attempt a watertight seal with well vascularised attached gingival flaps that are essential for tooth eruption. ^{5,6}

The impact of secondary grafting procedure can best be described in terms of (i) dental development and subsequent treatment, (ii) nasolabial aesthetics and (iii) nasal aesthetics and function/patient attitude/psychology. This article intends to cover each in turn reviewing the available literature and determining where evidence exists as to its benefit.

METHODS

An electronic search was conducted using Medline and Embase, with no restriction as to date of publication, study design or language. Multiple searches using a combination of the following terms "cleft", "secondary", "alveolar", "dental", "implant", "autotransplant", "nasal", "airway" "outcome" were used. We then screened the searches by title and abstract and the references of the relevant full articles further searched.

Key details of the relevant articles are described in Tables 1-5.

The impact on dental development and subsequent treatment

The implications of a cleft involving the alveolus are well recorded. The impact is significant and includes a higher incidence of dental anomalies than in the unaffected population. The cleft area has been shown to be susceptible to disturbances in the dentition. ⁷ Common findings with previous studies include tooth agenesis within or peripheral to the cleft region, supernumerary teeth, impacted teeth, delayed dental development, and altered crown to root ratios. ⁸⁻¹¹ The timing of the grafting procedures dependent upon allowing tooth

eruption through the graft site and is most frequently performed between the ages of nine and eleven years. ⁵ The key tooth for eruption through the graft is cleft side canine tooth as the lateral incisor on that side is frequently absent or is diminutive. Where the lateral incisor does exists on the mesial side and is of good anatomical form then earlier grafting may be considered. ⁵

During normal canine development and eruption, the tooth moves more mesial and becomes more upright. This seems to be true also in cases where optimal alveolar bone grafting has been performed. ¹² However the rate of canine impaction is significantly greater ¹²⁻¹⁴ on both the cleft and non-cleft sides. ¹² Once erupted the canines maintain the alveolar bone within the grafted region as without this functional stimulation, the bone rapidly resorbs. ¹⁵ Once the tooth is erupted through the graft then its periodontal support is adequate to allow orthodontic intervention and tooth movement. ¹⁶

Prior to the advent of secondary alveolar bone grafting (ABG) the aims of orthodontic treatment was limited to expansion and alignment. The edentulous space across the cleft was restored with fixed or removable prosthesis. Orthodontic movement of teeth adjacent to the cleft was challenging due the close approximation of the roots to the cleft site and the risks of loss of vitality. As a result, the residual gap in the arch was rarely of an ideal size and often resulted in undesirable aesthetics and a deterioration of dental health often due to the fixed nature of the fixed prosthesis.¹⁷

Alveolar grafting has transformed the management of the cleft site, allowing complex post-graft orthodontic movement. Keeping the cleft related lateral incisor has been estimated to be possible in up to 30% of patients. ¹⁸ Where the lateral incisor is of poor quality or absent then orthodontic space closure should be completed to obviate the need for any form of prosthesis.

Primary Author	Participants & Assessment	Comment	
Turvey 1984 ¹⁹	- 24 randomly selected cleft patients post BG. (15 UCLP, 9 BCLP) mean age for BG 11.7 years (y) - clinical evaluation	- 12 of 24 (50%) had orthodontic space closure	
Bergland 1986 ⁵	- 41 BCLP, 82 BG sites, grafting age range 8-17y - evaluated post-ortho	- 41 of 43 (95%) closed if BG before canine eruption - 29 of 39 (74%) after canine eruption	
Bergland 1986 ⁶	- 340 patients with 389 grafted sites. UCLP and BCLP - age range 8-18y	- BG before canine eruption = 90% space closure - BG after canine eruption = 72% space closure	
Enermark 1987 ¹³	- 3 groups of BG; A) pre-canine eruption (94pts), B) post-canine 13y (72pts), C) post-canine 16 y (54pts). >4y post graft. - radiological & clinical assessment	- group A best marginal bone levels - orthodontic closure in 49% of UCLP/BCLP	
Dempf 2002 ²¹	- 91 patients 41 ULCP 49 BCLP. - secondary (mean 10.3y) vs. tertiary grafting mean age 21.3y)	 - 25/42 (60%) where orthodontic treatment finished achieved space closure - teeth within the graft, maintain graft 	
Schultze-Mosgau 2003 ²²	- 57pts (46 UCLP, 11 BCLP) with 68 secondary BG (mean age 8-11y) - 59 BG before canine eruption	- orthodontic space closure in 53 of 68 grafts (78%) - less bone resorbtion with gap closure	
Oosterkamp 2010 ²³	p 2010 ²³ - 27 BCLP's with 1 missing lateral - assess aesthetics, mandibular function. Clinical assessment - assess aesthetics assessment - 17/27 (63%) with closure - no difference for aesthetics - space closure group scored better for function		
Seike 2012 ²⁴	- 41 patients with 49 BG's - 15 UCLP, 10 BCLP, 14 ULA - early and late post-BG radiographs. Orthodontic closure or not	- 26 of 49 (53%) achieved orthodontic space closure. - early radiograph parameters were unable to predict successful closure	

Table 1. Review of studies that have evaluated the impact of alveolar bone grafting on orthodontic space closure across a grafted site

Primary Author	Participants/teeth	Comment	
Hillerup 1987 ²⁶	- 4 patients with UCLP - 4-20 months (mo) post-graft - observation time 1-4 years (y)	- all successful with signs clinical and radiological healing	
Hamamoto 1998 ²⁷	- 2 patients, 1 CLA & 1 UCLP - and histological specimens from BG site	- graft is still undergoing remodelling at 6mo & better to perform the transplant at this stage - the teeth can be moved orthodontically 3mo post-transplant	
Czochrowska 2002 ²⁸	- 5 consecutive pts. with two incisors on the cleft side missing - three had previously been BG - transplant age range 10-13y 4-26mo post graft	- 100% survival - gingival index and pocket depth as control teeth	
De Muynck 2004 ²⁹	- 1 patient, BG 8y 6mo and transplant 10y 5mo	- clinical and radiological success	
Tanimoto 2010 ³⁰	- 2 patients - pt. 1, BG 12y 10mo and transplant 5mo later - pt. 2, BG 12y 6mo and transplant 12y 1mo	- clinical and radiological success - orthodontic movement followed without complication	
Aizenbud 2013 ³¹	- 4 patients transplanted with maxillary second premolars	- 12-48mo follow up - orthodontic movement after 6mo	

Table 2. Review of studies looking at success of transplanting teeth into sites that recieved an alveolar bone graft

Table 1. summaries studies that have quoted the success rates of orthodontic space closure post-secondary grafting in patients with a cleft involving the alveolus. This may require challenging orthodontic mechanics in cases where the patients has a Class III skeletal growth tendency and an attempt is made to maintain the upper dental centreline. As can be seen in Table 1. success rates for total space closure in some studies have been as high as 90% and more ^{5,6,18} and in most others vary between 50-75% success. ¹⁹⁻²⁴

Bone grafting (BG) prior to canine eruption appears to increase the likelihood of successful space closure ^{5,6,13,18} because it allows the canine to erupt through the graft once it is in place. This also increases the likelihood of a parallel root position after the closure with greater stability. ¹⁹

Early radiological assessment following the graft allows assessment of bone reorganisation within the cleft site, but only weakly correlates with whether orthodontic space closure is possible or not in the future. ²⁴ Once space is closed, the tooth function appears to reduce likelihood of resorbtion of the graft ^{13,21,22} and improve mandibular function without compromising aesthetics compared to prosthetic replacement. ²³

Tooth replacement

Where space closure is not possible (and similar to the non-cleft patient) there are three possibilities for tooth replacement: (i) adhesive bridgework, (ii) tooth transplantation and (iii) implants.

Adhesive Bridgework

Modern restorative techniques have advanced significantly from the era of the fixed-fixed prosthesis requiring significant tooth reduction. Reduced caries rates in the cleft population and modern adhesive techniques have resulted in resinretained bridgework being the first choice for restoration in the adolescent. ²⁵ Despite multiple searches there appeared to be no studies suggesting the long term success of this type of bridgework compared to the non-cleft population.

Tooth auto-transplantation

Table 2. Summarises studies for tooth auto-transplantation into the secondary graft site in patients with a cleft involving the alveolus. The most common tooth for transplantation is the lower premolar and has been demonstrated in grafted sites in patients with cleft lip and palate. ²⁶⁻³⁰ The technique is operator technique-sensitive, but if successful the functional tooth will maintain the bone in the graft site.

The optimum time for auto-transplantation appears to be 6 - 12 months post-secondary alveolar graft when the graft is still remodeling ²⁷ although later tooth auto-transplants have resulted in radiological and clinical success. ²⁸⁻³⁰ Although there may be some advantage to performing the auto-transplant at the same time as the alveolar graft, this has been shown to lead to increased resorbtion in simulated alveolar clefts. ³¹ Orthodontic movement can commence usually after 3 months ²⁷ and is likely to be completed uneventfully.

Implants

Table 3. Illustrates studies where endosseous implants have been placed into secondary alveolar grafted sites in patients with a cleft involving the alveolus.

Recent studies suggest that the long-term success of these implants is good ³⁷⁻³⁹ and the implant acts as a functional stimulus to maintain the bone ²¹ but a significant number of the implants required further grafting (tertiary). In some studies all subjects received simultaneous grafting at the time of implant placement, ^{38,40,43} while some cases required a tertiary graft in 50% or less. ^{37,39,44,46} This extra graft may be performed simultaneously ^{37,38,41,47} or prior to implant placement. ^{42,48,49}

Primary Author	Participants/Implants	Comment	
Ronchi 1995 ³²	- 3 patients, (40y 26y 18y). Implants placed 6-8mo post graft	- 14-24mo later clinical and radiological success	
Kearns 1997 ⁴⁸	- prospective 14 patients (12-65y) - 20 implants	- 1-54mo follow up - 18/20 survived	
Takahashi 1997 ⁴⁴	- 19 patients (10-34y)	- 1-3y follow up	
	- 21 implants	- 5/21 required further graft at insertion - 20/21 survived	
Fukuda 1998 ⁴⁰	- 7 patients (9-27y) - 7 implants placed 0.6-10y later	- all received tertiary graft due to insufficient bone ht - all integrated at follow up 2-5y	
Jensen 1998 ⁴¹	- 16 patients (15-38y) - 20 implants	- all received tertiary graft at placement - 18/20 survived at follow up of 36-69mo	
		- follow up 43-92mo	
Lilja 1998 ³³	- 16 patients (16-53y) - 31 implants. 2 gps with (10) and without (6) additional graft	- 29/31 successes - both failures in additional BG gp	
		- 4-36mo follow up	
Härtel 1999 ⁴⁹	- 11 patients (14-27y) - 17 implants	- 16/17 successes.12 of 17 required additional BG	
	-/ mpano	- implant placement should be inserted <6-8 weeks post graft	
L	- 4 patients (ant max) (17-24y) - 5 implants	- tertiary grafting in all	
Jansma 1999 ⁴²	- secondary graft 9-11y	- implants placed 3mo post tertiary graft - 28-65mo follow up. 4/5 successes	
	- 14 patients	- 50% required further graft due to resorbtion of secondary gra (particularly vertically)	
Takahashi 1999 ³⁴	- 14 implants (same cohort as 1997)	- follow up 12-48 months	
		- 100% success	
		- 42-48 months follow up	
Fukuda 200047	- 2 patients (19y & 20y) - 2 implants	- grafting and placement at same time for one patient	
		- 100% success	
Dempf 2002 ²¹	- comparing secondary (60) & tertiary (25) BG's - 16 implants in tertiary group	- implants placed within the bone graft reduce its resorbtion	
		- tertiary graft required on 5/9	
C 200/45	- 9 consec. BG patients (18-31y) with 10 implants	- follow up 1.3-5.7y	
Cune 2004 ⁴⁵		- 10/10 functioned	
		- 7/10 aesthetics acceptable	
	- 45 non-syndrome patients (14.8-49.1y) - 75 implants	- all received a tertiary bone graft	
		- follow up 1.5-11.3y	
Kramer 2005 ⁴³		- 65/75 successes all failures lost in first year. Shorter implants had worse survival	
		- implants inserted the same time as graft have reduced survival	
Takahashi 2008 ³⁷	- 23 implants in 21 patients	- 19/21 successes	
	- implant surgery 13.9-33.6y	- 5/21 required tertiary graft at time of implant placement	
		- 39/71 received simultaneous tertiary grafting	
Matsui 2007 ⁴⁶	- 47 patients (14.6-54.6y) with 71 implants	- follow up 21-120mo	
		- 70/71 successes	
		- follow up 1-10y	
Lalo 200735	- 12 patients	- 100% successes	
De Barros Ferreira 2010 ³⁸	- 120 patients - 123 implants (15-40y at placement)	- 100% successes - mean follow up 34mo - all received extra graft at the time of placement	
Filho 2013 ³⁹	- 39 patients - 39 implants	 - 19/39 required further graft simultaneously with implant placement. Mean follow up was 42mo 	
	- characteristics of implants and smile assessed	 no relationship between timing of secondary BG and whether tertiary BG required 	

Table 3. Review of studies evaluating the success of implants placed within an alveolar bone grafted site

The extra grafting procedure is required to increase the vertical alveolar height ⁴⁰ and ultimately the final aesthetic result. However, Filho and de Almeida ³⁹ reported that despite deficiencies in interdental papilla and asymmetries between left and right crown width and height this does not appear to have a significant impact on patient satisfaction. It is of note however that most patients registered a low lip line which was likely to have masked the less than ideal result.

Table 3. Summary of articles where endosseous implants have been placed in previously secondary grafted sites in more than one patient with a cleft involving the alveolus.

In Summary of the dental impact of secondary alveolar grafting:

- Alveolar bone grafting offers significant benefits for successful restoration of the maxillary dental arch.
- The canine tooth associated with the cleft will erupt through the graft and this functioning tooth will maintain the graft and its periodontal health appears satisfactory to allow orthodontic movement.

- The edentulous area associated with the frequently absent cleft side lateral incisor may be closed by orthodontic tooth movement in the majority of cases.
- Osseointegrated (OI) implants or auto-transplantation may well be considered within the grafted site particularly where both incisors are absent and can be expected to have a good survival rate.
- Where OI implants are considered a further bone grafting procedure is often indicated in the adult patient.

The Impact on Nasolabial Aesthetics

Although clefts of the lip are closed within the first 6 months of life in the UK the impact on the underlying skeletal base and overlying soft tissues can be profound. The aesthetic impact is mainly localised around the nasal aperture on the cleft side, resulting in displaced nasal cartilages, and hypoplasia of the pyriform rim, that affects the nasolabial complex and results in significant nasal asymmetry. Residual facial deformity can

Table 4. Review of studies on the aesthetic impact of alveolar bone grafting and their evaluation methods

Author	Subjects	Assessment Method	Result	
Cho-Lee 201356	- retrospective - 90 patients (4-21y) - 3D on 19pts comp to cleft without and control - pre and 2mo post-op	- 3D using Computer tomography	- anterior movement Pronasal and subnasal distances compared to non-grafted	
Offert 201357	- retrospective - 29 children - early graft & rhinoplasty vs. non graft	- esthetic index 2D photos - lay and professional observers	- early graft produced better aesthetics than non-grafted patients	
Zhang 2014 ⁵³	- prospective - 26 patients (10-34y) - various operative procedures	- anthropometric - 3D CT scanning - patient questionnaire	 satisfaction not related to changes in soft tissue alar base depression not associated with pyriform rim classification 	
Kim 2012 ⁵⁸	- prospective - 18 UCLP (Mean age 9.9y)	- volume of bone	- significant alteration post-op for nostril elevation and lip length - no comment on volume of bone versus	
	- follow up 12-24mo	- 2D photos		
	- pre and post ABG	- pre and post op proportions compared	effect on points	
Sander 2011 ⁵⁹	- prospective - 39 patients (7-14y) - assessment <1mo pre and > 6mo post	- partial Facial Imps then scanned using - 3D (3dMd)	 symmetry improvement decreases with time no significant difference in symmetry scores post graft changes in inf. & lat. margins of alar base 	
Kau 2011 ⁵²	- prospective - 10 patients - pre-op and 6 weeks post-op	- 3D (3dMD)	- anterior positioning of ala base in grafted patients	
Li 2011 ⁶⁰	- prospective - 90 patients - ABG only, ABG + rhinoplasty, rhinoplasty only (age 8-11y)	- 2D pre- and post- surgery	- nasal base and lateral rim elevated by ABG but cleft side nostril further flattened	
Krimmel 2011 ⁶¹	- prospective - 22 patients (9-20y) - 18UCLP, 4 BCLP	- 3D 3dMD - 1 day pre and 6 weeks post ABG	- subalare & alare curvature had greatest anterior projection	
Wu 2010 ⁶²	- 38 (9-13y) pre- and 6mo post-op - 29 cases examined post-op	- 2D photos and Anthropometry	- cleft nasal profile flattened, nasal base with increased. Profile worsens post op	
Devlin 2007 ⁵⁴	- prospective - 18 patients - block graft post graft (11-27y)	- 3D (Di3D) - bone volume measurement	- landmark placement largest error - significant improvement in facial symmetry - bone volume not related to improvement	

have consequences for psychosocial child development and can influence interpersonal relationships as well as affect success at school. 50,51

The two-dimensional assessment of facial asymmetry has been performed in unilateral cleft lip and palate by direct anthropometry and by 2-dimensional photography. Both come with their own individual disadvantages. Anthropometric and 2-dimensional photography suffer from the inability to assess accurately the 3-dimensional impacts commonly associated with the residual deformity. Both require significant patient cooperation to complete and standardisation can be difficult. Three dimensional assessments have been accomplished in cleft lip and palate using laser scanning 52 computer tomography (CT) 53 and stereophotogrammetry. 54 Laser scanning has certain disadvantages in younger children due to the level of cooperation required for accurate facial capture. A significant level of cooperation is also required for computer tomography. The disadvantage of CT is a significant radiation dose. Stereo photogrammetry has the advantage of non-invasive short capture times and has been validated for use even in infants. ⁵⁵ Secondary alveolar bone grafting attempts to repair the pyriform rim and nasal floor. Studies evaluating the effect of grafting and their methods of assessment on nasal aesthetics are summarised in Table 4.

Summary of the Impact of secondary alveolar bone grafting on nasolabial aesthetics:

- The literature is equivocal as to whether secondary alveolar bone grafting has an impact on nasal form.
- The longer the interval between the ABG operation and assessment post-operatively the lesser the effect.
- The literature presents a wide range of surgical variations within the groups examined making direct comparisons difficult.
- Several studies examined patients within three months postoperatively and observed changes in aesthetics are likely to be associated with to post-operative swelling.
- Over filling the cleft site with bone is suggested by some authors but there is no evidence to support this approach.
- Reports that suggest an alteration of the form of the nose, particularly in relation to anterior positioning of the alar base may in fact flatten the nostril, thereby worsening aesthetics.
- Alterations in many clinical variables post-operatively may not be associated with an improved patient perception of aesthetics of the nose.

The Impact on Nasal Function

Various deformities of the nose can result from facial clefts with partial or total obstruction. Deformities can take the form of septal deviation, nasal stenosis or choenal atresia, which is recognised in CHARGE, De George and Velocardiofacial syndromes. A normally functioning nasal airway is considered critical for respiration, olfaction and growth of the face. Sleep apnea associated with partial/total obstruction has been associated with disruptive behaviour or and attention deficit disorder in children. ⁶³ Children with clefts of the lip and or palate have been found to be as much as five times more prone than age matched controls. ^{64,65} Patients with cleft have a greater incidence of nasal obstruction than age matched controls and when it exists it has a greater impact on daily life and physical activity with lower physical quality of life measures. ⁶⁶

Small number of longitudinal data exists in relation to nasal function in children with cleft lip and palate during the period of secondary bone grafting. The cross sectional study by Drake ⁶⁷ suggests that the nasal airway in children with or without a cleft grows at similar rates although children with a cleft have a 30% reduction in nasal volume compared to the non-cleft controls and the percentage of nasal breathers was considerably lower. The lowest increment of airway growth in the cleft group was found in the 12-15 year age. Drake suggested this may be due to the number of cosmetic procedures carried out within this age range but also suggested that it may be due to other surgeries. ⁶⁷ In non-cleft subjects the minimum cross sectional area and thus the area of highest resistance to airflow is located at the nasal valve. This area approximates to the region of the anterior head of inferior turbinate ⁶⁸ and is closely related to surgical area that receives the graft. Lino et al.⁶⁹ suggested partial inferior turbinectomy during secondary alveolar bone grafting to facilitate formation of a sufficient bone bridge although no comment is made as to its effect on the nasal airway.

CONCLUSIONS

Secondary alveolar bone grafting for repair of alveolar defects in cleft lip in palate is a reliable procedure associated with high success rates. The dental impacts in relation to the eventual restoration of the cleft area are significant allowing orthodontic tooth movement and space closure in the majority of patients. This guarantees the grafts longevity but also avoid the need for a dental prosthesis.

Poor nasolabial aesthetics in children with repaired cleft lip and palate has a significant psychosocial impact. Despite claims that secondary alveolar bone grafting has a positive effect on symmetry and overall aesthetics the literature is equivocal. Authors have suggested overfilling the defect as a method of improving aesthetic impact but this is unsubstantiated and it maybe that alteration in nasal aperture position may flatten the cleft side nostril worsening the symmetry.

Nasal obstruction had a significant negative impact in patients with cleft lip and palate. Growth in the airway in cleft children appears to plateau after the time of secondary alveolar grafting although little information exists as to its effect.

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