A RETROSPECTIVE STUDY OF CIRCUMPUBERTAL CLEFT LIP AND PALATE PATIENTS TREATED IN INFANCY WITH PRIMARY ALVEOLAR BONE GRAFTING

by

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INTRODUCTION

Clefting of the lip and palate represents a morphological deformity, which has emotional and treatment ramifications beyond the actual physical manifestations. Treatment of such individuals has long involved a multidisciplinary approach, and historically, this has resulted in a variety of treatment philosophies and surgical protocols. Despite ongoing controversy among treatment teams as to the best treatment protocols, the goal of all treatment centers remains to resolve the physical deformity without further accelerating growth attenuation inherent in cleft lip and palate.

Throughout the 19th and 20th century various treatment protocols became more or less standardized, and osseous grafting of the cleft deformity became commonplace. The real contention, which has persisted to this day, remained the timing and the source of the graft. Dr. Sheldon Rosenstein developed and advocated a specific surgical protocol, which incorporated early primary alveolar bone grafting and alveolar molding appliances.

The Riley Children's Hospital Craniofacial Anomalies Team rigorously follows Rosenstein's protocol for the treatment of cleft lip and palate patients. The Rosenstein protocol incorporates primary bone grafting and alveolar molding appliances for cleft lip and palate patients. While other cleft lip and palate treatment centers utilize alveolar molding appliances, there remains debate concerning primary bone grafting. The principal detraction of primary bone grafting involves the concern that such treatment affects maxillary and craniofacial growth and development. The purpose of this retrospective study was to analyze post-treatment lateral headplates and dental casts of cleft lip and palate circumpubertal patients treated in infancy at Riley Hospital by the Craniofacial Team following Rosenstein's protocol. The cephalometric values of the study group population were compared against a nongrafted group, an early primary grafted group, and the Bolton standard values (control group), all of which were cited in Rosenstein's 1982 study. This study may also lay the groundwork as a pilot study for a future prospective study of cleft lip and palate patients treated at Riley Children's Hospital.

REVIEW OF LITERATURE

Clefting of the lip and palate presents clinicians with multiple problems of a skeletal, dentoalveolar and soft tissue nature, each being one small piece of this treatment mosaic. The treatment of this anomaly has ramifications affecting not only the obvious physical and esthetic concerns but also the more subtle issues of social development and self-esteem. Early and sustained intervention is essential to resolve the anatomical problems and to provide the patient with positive feelings of self-worth which will reinforce good social development skills. Historically, the treatment of cleft lip and palate centered upon surgical resolution of the cleft sites, while the dental alveolar problems resulting from the collapse of the premaxilla were addressed with conventional dental prostheses and orthodontics.

Throughout the 19th century, plastic surgery techniques progressed rapidly, and more esthetic solutions for soft tissue revisions were possible; however, the glaring problem of how to adequately resolve hard tissue cleft sites remained. Pioneers such as von Eiselburg[1] and Lexer[2] in the early 1900s began to incorporate various bone grafting techniques in an attempt to bridge and resolve the bony cleft sites. Lexer[2] advocated the use of a free bone graft. Various sources of bone were utilized in the ensuing years with Drachter[3] reporting tibial bone and periosteum as a source in 1914. As surgical and orthodontic techniques continued to progress and become more refined, a variety of protocols were developed, utilized, rejected, and revised with the ultimate goal being the esthetic and functional resolution of hard and soft tissue clefted defects. Though soft tissue resolution continued to progress rapidly, the vexing crux of these treatment protocols remained the problem of achieving resolution of bony cleft sites. Given the technical challenges of resolving hard tissue defects, the advantages of utilizing bone grafts as the preferred means of treating this defect became an attractive option. The combination of clefting of the lip and palate is much more involved than just a physical defect of the involved tissues. Clefting inherently creates defects involving critical centers responsible for maxillofacial growth and development. These defects result in developmental deficiencies of bone and soft tissues eventually affecting growth and development of the midface.[4]

Alveolar grafting was viewed as a means in which the bony cleft site could be bridged and stabilized with the graft becoming incorporated and growing with the other maxillary structures in some semblance of normal facial growth and facial harmony.[4-7] The goals of alveolar grafting can be viewed in terms of functional and esthetic objectives. The advantages of alveolar bone grafting are: the stabilization of the premaxilla, prevention of segmental collapse, prevention of a oro-nasal fistula, provision of bony support to teeth adjacent to the cleft site, and the reestablishment of dentoalveolar bony contour while providing continuity to the maxilla.

The esthetic aims are described as restoring harmony via an intact dental arch and an improved appearance of the lip and alar base.[5-7] With the continued progression of plastic surgery and bone grafting techniques, there began research and debate as to the optimal age of children receiving this therapy. The timing of alveolar grafting reported in the literature is typically classified as primary, secondary, and delayed (Table I). The term primary grafting is applied to procedures performed before the eruption of the primary dentition or as late as one year of age. Secondary grafting is broadly defined as grafting following the eruption of a permanent dentition usually ranging from nine and 11 years of age. This broad classification is further divided into grafting procedures occurring from five to six years of age, which are categorized as early secondary. Secondary grafting is typically reported as procedures performed around nine to 11 years of age. Delayed grafting, sometimes reported as late secondary, is descriptive of procedures performed following the eruption of the permanent canines.

Helms et al.,[8] reporting on the timing and frequency of alveolar grafting, found primary and secondary alveolar grafting the most commonly performed procedure, followed by early secondary, with delayed grafting rarely reported. They[8] felt much of the conflict and disagreement surrounding alveolar grafting stemmed from three points of origin. The first related to the procedure reported and performed in terms of chronological rather than developmental age. The second discrepancy involves such intangible variables as particular definitions of success and socioeconomic ramifications driving the timing of grafting. Lastly, there is great difficulty in comparing research data produced from a variety of surgical centers, protocols, and differing philosophical approaches to treatment decisions. The advance of surgical and orthodontic technique led to the combination of both modalities during treatment, and a variety of protocols were developed that attempted to solve the goals of alveolar arch continuity, the prevention of a collapsed premaxilla, and the resolution of the clefted defect. This unified strategy, incorporating alveolar molding appliances, throughout the surgical treatment regimen, was introduced in the early 1950s in the hope that these appliances would stabilize and prevent the collapse of the greater and lesser segments of the premaxilla.[5-9]

The late 1950s and 1960s saw extensive use of alveolar molding appliances and various orthopedic appliances coupled with the use of autogenous bone grafting in cleft sites. During this era, bone grafts were placed in both infants and children (varying degrees of reported success) with the trend leading toward establishment of arch continuity and cleft resolution as early as possible in the treatment regimen. These early treatment protocols resulted in distinct opinions concerning the efficacy of primary bone grafting in infants.

Schruddle and Stellmach[10] published pioneering studies of primary alveolar grafting. Their technique involved conducting the lip adhesion and the alveolar graft simultaneously. Their efforts were also noteworthy in that they advocated restoring the "functional topology" of the alveolar segments through the use of infant orthopedic appliances. The concept of orthopedic alignment of the alveolar segments was advanced by the efforts of Skoog.[11]

Skoog advocated the approach that alveolar grafting be delayed until the maxillary segments were in a proper anatomical relationship following lip revision and orthopedic molding. Skoog[11] also incorporated Surgicel as a matrix to promote osseous bridging of the alveolar cleft site. Early results of this treatment regimen appeared beneficial; however, long term studies and follow-up were lacking to properly evaluate this premise. Though Skoog attempted early intervention in alveolar arch alignment and soft tissue adhesion, he thought delaying alveolar grafting until three to four months after soft tissue adhesion produced a more favorable outcome.

Pickrell et al.[4] published a study involving 25 patients who received primary alveolar bone grafts and were followed four years postoperatively. Their efforts were unique for that era in that his group performed primary alveolar rib grafts in infants aged two to six months with unilateral clefts of the lip and palate. They also incorporated passive and orthopedic appliances as soon as possible after birth and performed the lip adhesion and grafting procedure simultaneously. This effort laid the foundation for protocols that would eventually pursue primary alveolar grafting, because the concept of early orthopedic molding coupled with primary alveolar grafting and early lip adhesion was advanced, even though the results of Pickrell's group did not favor the efficacy of this regimen. They observed that there was no vertical growth of the graft that would maintain the height of the alveolar process; the graft alone would not prevent the collapse of the maxillary segment. Twenty-two of the twenty-five grafts were seen to incorporate with the medial and lateral segments; however, no growth in the size of the graft was evident. They concluded that alveolar grafts did not form a true alveolar process evidenced by: the presence of a permanent alveolar notch, non-migration or lack of spontaneous tooth migration and spontaneous eruption through the alveolar graft site, and a decrease in the orthopedic effect of the graft as the incorporation progresses.

Although Pickrell's group summarized that rib grafts failed to form an alveolar process and eliminate or minimize alar base deformity, Longacre et al.[12] reported a moderating effect of split rib grafts upon maxillofacial growth and stabilization of the alar base due to the formation of lamellar bone in the graft site. Friede and Johanson[13] contributed to the body of negative data surrounding primary alveolar grafting. Their efforts were consistent with other centers' management of primary alveolar grafts with the exception that they conducted extensive flap dissection with simultaneous lip revision and closure of the anterior palatal defect with a vomer flap. Friede and Johanson also advocated subsequent orthopedic arch expansion until the sixth month of life. Following arch expansion, they conducted a simultaneous lip adhesion and an alveolar graft using the tibia as the osseous graft source. Their results indicated abnormal maxillary development and a corresponding increased incidence of anterior and posterior crossbite. These adverse results led to the termination of primary alveolar bone grafting at their surgery center.

Robertson and Jolleys[14] reported similar negative findings in a comparison of maxillofacial development between a group of patients treated identically, that is, infant orthopedics, lip revision, and soft palate repair. The only exception being that the study group received primary alveolar bone grafts at 15 months while the control group were untreated. Robertson and Jolleys' data indicated a distinct deterioration of dental base relation compared with the nongrafted group. Results also indicated greater anterior posterior occlusion discrepancies among the grafted group when compared with the control group.

Despite negative results reported by many surgical centers and ambivalent data presented by others, Rosenstein et al.[5-7] developed and strongly advocated a specific protocol for primary alveolar bone grafting. Rosenstein's protocol advocates early and sustained orthopedic molding prior to lip repair. The exact sequence of Rosenstein's protocol is described by Dado[15]: the newborn infant has a dental impression made and a palatal appliance fitted before the lip is repaired. The cleft lip is repaired at 6 to 8 weeks of age with the functional cleft lip repair. This muscle repair then helps to mold the greater segment of the cleft alveolus as it grows to form a butt joint with the lesser segment. The palatal prosthesis is kept in place almost continually to prevent collapse of the alveolar arch after the lip is repaired. It is only when the alveolar segments are in perfect alignment without a large gap (i.e., the segments are just touching or 1 to 2 mm apart) that the bone graft is placed to stabilize the arch. This generally occurs at about four to five months of age.

At the time of operation, the palatal appliance is kept in place, and the infant is anesthetized with a Rae preformed endotracheal tube that sits on top of the lower lip and chin, so that interference with the site of surgery is avoided. The rib graft is taken first. A 2 to 3 cm incision is made over the sixth rib to avoid interference with the breast bud and to allow the resulting inconspicuous scar to fall in the breast fold as subsequent breast development occurs in females. The dissection of soft tissues over the rib is rapidly performed using a "spreading maneuver" with tenotomy scissors, taking care to keep the scissors over the center of the rib. Upon reaching the rib, an H-style incision is made in the periosteum near the costocartilage junction. The periosteum is elevated off of the rib on the anterior surface and around the superior and inferior borders, and the bony rib is then separated from the periosteum on the posterior surface. This can be accomplished with a dental scaler or periosteal elevator. This careful dissection, before lifting the rib, essentially eliminates the risk of entering the pleural cavity. Traction is then placed on the rib to lift it and disarticulate it at the costocartilage junction. The rib is cut approximately 1 to 2 cm away from the junction with a bone cutter. The wound is checked for any pleural tears by filling the small cavity with saline and asking the anesthesiologist to give several breaths to the infant with positive pressure. A small tear can easily be repaired with a suture around a small rubber catheter. The periosteum, muscle fascia, and subcutaneous tissues are then closed in layers. A running subcuticular suture in the skin allows easy removal 7 to 10 days after the operation.

The rib regenerates within several weeks, and indeed, if a bone graft is needed for a second operation (e.g., secondary orthognathic surgery), the same rib can be used and approached through the same scar. The rib is split with an osteotome and the posterior half cut into small chips of bone. The anterior is left intact as a larger strut of bone. Its natural curve follows the curve of the alveolus nicely. The cleft alveolus is then prepared for insertion of the rib graft. Small flaps of mucoperiosteum are incised on both sides of the alveolus at the cleft margin. Those flags are dissected only enough to turn back and create a posterior lining. This maneuver is best accomplished with a dental scaler. Another incision is made in the upper buccal sulcus just above the alveolar cleft. Dissection proceeds on the anterior and upper surfaces of the greater and lesser segments of the alveolus under the mucosa to strip the periosteum off of the maxilla for a distance of 4 to 5 mm on both sides. The undermining thus performed is minimal and is only extensive enough to create a small pocket to accommodate the small bone graft. Further soft tissue dissection is performed to free up a buccal flap of mucosa that can be brought down to close the anterior surface of the alveolar cleft. The mucoperiosteal flaps on either side of the cleft are turned back and sutured together to create a closed posterior lining with three simple stitches of 4-0 chromic suture material. The knots are tied on the palatal side of the flaps. The rib strut is then placed in the pocket that has been created on the anterior surface of the alveolar cleft, and the smaller bone chips are packed underneath it to increase the volume of bone. The buccal flap is brought downward as a V-shaped flap and sutured to the inferior edge of the posterior lining flaps and the anterior edges of mucosa on either side of the cleft with 4.0 chromic sutures. The alveolar cleft is thus completely closed. A notch remains at the inferior margin of the cleft but levels off later when tooth eruption occurs through the bone graft.

The palatal prosthesis is kept in place for 10 days before removing it for cleaning. It is then worn for an additional 6 to 8 weeks until the bone graft has become incorporated into the alveolar arch. As with all autogenous grafts, this is accomplished after vascularization of the graft allows some of the osteocytes to survive and maintain a calcified matrix, and also through oseoblastic activity, which forms new bone in the graft. Union occurs after osteoblasts from the alveolar segments act in conjunction with those of the graft to achieve consolidation. At this time the alveolar arch alignment is stable, and the baby wears the appliance only to facilitate feeding until the palate is closed at a separate, third operation. The contentious aspect of Rosenstein's treatment protocol is the timing of the alveolar bone graft. As cited earlier, the chief criticism of primary early bone grafting is that craniofacial development is retarded; however, Rosenstein and his proponents feel the effect upon craniofacial development is negligible while the ensuing arch development substantially minimizes the degree of future orthodontic treatment required.

Dado[15] summarizes Rosenstein's protocol as being quite different from the bone graft procedures described in the literature in 1950s and 1960s in that: it is not performed in conjunction with lip repair or palatoplasty; the extent of dissection is minimal and limited to the alveolar cleft margins, anterior maxilla, and does not involve the rest of the primary and secondary plates; the dissection does not cross the prevomerine suture; the elevation of large flaps is not required to cover the graft; the graft is not wedged into a large gap in the cleft alveolus and plate; the procedure is always performed in conjunction with neonatal orthopedics after correct arch alignment is achieved.

Surgical centers expressing an opposing viewpoint cite the primary alveolar graft and corresponding dissection as a source of disruption affecting the vomeropremaxillary suture and possibly the septopremaxillary ligament, thereby having a detrimental effect on maxillary growth and development.[1,13,15,16] Though most reported protocols incorporate some type of orthopedic appliance,[5-7,18] the timing and sequence in relation to lip revision and grafting varied greatly. Rosenstein feels his strict adherence to presurgical orthopedics, separation of lip adhesion and grafting procedure, and optimal presurgical approximation of the greater and lesser segment prior to grafting are responsible for his reported favorable outcomes. A follow-up study published by Rosenstein et al.[7] reviewed 16 unilateral cleft lip and palate patients treated at their surgical center 10 years postoperative. The patients demonstrated an improvement in maxillary segment position, dental alignment and occlusion.

Rosenstein et al.[19] also published a 25-year postoperative review of 36 primary alveolar bone-grafted patients treated by their service. The purpose of this retrospective study was to analyze maxillofacial development and determine the need for orthognathic surgery. Although six patients (22 percent) required maxillary advancement, Rosenstein felt their protocol did not contribute to any significant degree of maxillary hypoplasia. Trotman et al.[18] published results of a study which analyzed 43 patients treated by Rosenstein. Rosenstein's patients were matched by age, sex, and cleft type with a control group of nongrafted cleft patients. They[18] concluded that the grafted group exhibited less maxillary retrusion, lower anterior face height, and a depressed nasal base. Trotman's group postulated that the observed maxillary retrusion had produced a compensatory rotation of the mandible accounting for the observed decreased anterior face height.

Following review of 2200 cleft lip and palate patients treated in their surgical facility, Georgiade et al.[20] expressed disappointment in their case outcomes from a functional and esthetic viewpoint. The authors felt that a variety of highly variable intrinsic factors would necessitate a case by case treatment rationale. They expressed the view that bone grafting should be done prior to segmental collapse and that primary alveolar bone grafting held promise.

Ross[21] expressed the opinion that the conflicting data and viewpoints of primary alveolar bone grafting studies were correlated to the varying surgical protocols and not a result of the presence of the graft. He also concluded that primary alveolar bone grafting prevents skeletal collapse of the maxillary segments, and that such grafting should have no long-term effects on anteroposterior maxillary growth, because the growth site possesses no intrinsic growth potential. Ross[22] reported data from other studies that correlated maxillary growth attenuation in both primary alveolar bone grafting and in children treated between four and 11 years of age. He observed the resulting compensatory mandibular growth produced an increased lower face height and diminished facial esthetics. He expressed the view that vertical maxillary growth would be less affected if grafting was delayed until 15 years of age or later.

In a study involving palatal stripping and elevation of mucoperiosteum among a group of rhesus monkeys, Sarnat[23] observed no grossly evident growth arrest of the maxilla midface. Sarnat believed that maxillary growth attenuation observed in cleft lip and palate patients was not due to the primary alveolar bone graft, but the surgical dissection employed while placing the graft.

Robertson and Jolleys'[24] published results of an 11-year primary alveolar bone grafting study in which pairs of infants with similar cleft types were matched and treated with the same protocol, with the exception that one member of each group received an alveolar bone graft at a later date 12 to 15 months). The investigators found an increased incidence of crossbite, decreased palatal area, and an inhibition of anterior posterior maxillary growth. They concluded that no clinical advantages could be realized from primary alveolar bone grafting. Rosenstein[7,9] said any crossbite or other occlusal discrepancies could be readily addressed orthodontically throughout the mixed dentition, and that one did not have to delay treatment until after the circumpubertal growth spurt. Rosenstein remained adamant that the benefits of primary alveolar bone grafting were more efficacious than delayed grafting.

Forshall, Osborne, and Burston[25] published a study in which presurgical orthopedics were utilized without primary alveolar bone grafting. The authors reported that a significant number of their patients (66 percent) developed lateral crossbites. This large percentage was similar to the results of Klings[26] study (100 percent lateral crossbite) in which primary alveolar bone grafting was employed without presurgical orthopedics. Comparing these separate studies, Robertson and Jolleys[24] said that primary alveolar bone grafting did not prevent segmental collapse.

Caroll-Ann Trotman et al.[18] published a retrospective study comparing frontal craniofacial dimension in alveolar bone-grafted and nongrafted, complete unilateral cleft lip and palate patients, and in non-cleft subjects with normal occlusions and good facial balance. The control group of the non cleft subjects were taken from McNamara's[27,28] sample group of non-orthodontically treated European American ancestry, who were judged to have normal occlusion and possessed well-balanced facial aesthetics by a panel consisting of McNamara and three additional orthodontists all in unanimous agreement. The grafted and nongrafted patients were treated using a fixed protocol and a single surgeon permanently assigned to either the grafted or nongrafted case. The grafted group received secondary alveolar bone grafting timed in relation to two-thirds root development of the permanent cuspid nearest the cleft site. Although this study found no difference in maxillary width between the clefted groups, Molstead[29] found primary alveolar bone-grafted nonorthodontically treated patients presented with a greater degree of maxillary constriction than did patients treated nonorthodontically with secondary alveolar bone grafts. Trotman et al.[18,30] found that based on posterior anterior cephalometric radiographs, there were minimal effects upon craniofacial growth of alveolar bone, and that any significant findings were limited to the cleft site. Ross[31] published study results analyzing hard and soft palate repair within the first decade of life and found no influence in anterior posterior facial growth.

Wood[32] published research that contradicted many of the core issues raised by the detractors of primary alveolar bone grafting. Woods study reviewed 20 cases of unilateral complete cleft cases that were treated with a combination approach of presurgical alveolar molding appliances coupled with primary alveolar bone grafting. Wood used as his control group 24 nongrafted patients. A comparison was made between the groups four and one half years post-surgery. Wood[32,33] said the combination of presurgical orthopedics and primary grafting maintained the unity of the maxillary segments, prevented the collapse of the maxillary arch, and facilitated maxillary development as one unit.

Nylen et al.[34] published research that supported the findings of Wood. Nylen said presurgical orthodontics in conjunction with primary alveolar bone grafting results in stabilization and unity of the maxillary segments. Nylen et al. reported a low frequency of anterior crossbites and a matrix formation that failed to inhibit tooth eruption. These results were at odds with research published by Suzuki et al.[35] which found an increased incidence of anterior crossbite, inhibition of tooth eruption through the graft site, and the failure of grafts to integrate and keep pace with craniofacial development.

In research published by Pruzansky,[17] the author expressed the opinion that primary alveolar bone grafting and presurgical alveolar molding were a "needless and sometimes barbaric" protocol. Pruzansky said the infantile capsule or the dynamic relation of muscle forces paired with the oral orthopedic environment led to the development and anatomic relation of the dental arches and the growth of the midface. Pruzansky carried this concept to the point that he associated this muscular and connective tissue matrix as the driving force of the development of the cartilaginous nasal septum. His main argument against primary alveolar bone grafting was that the grafting dissection led to the disruption of the functional matrix of connective tissue and muscular forcers that drives midfacial development. The premise that primary alveolar bone grafting attenuates craniofacial development by disrupting potential growth centers, namely the vomeropremaxillary suture and the septopremaxillary ligament,[4,16,17] is not a new concept. This remains a highly contentious point in the literature and could be rationalized by the many variables among surgical centers and differing protocols.

Factors that contribute to the difficulty in comparing data among studies include the differences among surgical providers, the amount and nature of soft tissue dissection carried out during grafting and soft tissue revision, the type of graft preparation, and the extent or lack of presurgical orthopedics.

King and Schneiderman[36] experimentally addressed the issue of growth attenuation, as a consequence of growth site alteration, in animal research. The premise of their research was that cleft patients experienced craniofacial growth attenuation as an attribute of the developmental defect of the cleft, and that this problem is exacerbated from wound constricture following surgery,[14] which affects the developmental balance of muscular foraces acting on centers of maxillofacial development. King and Scheiderman[36] evaluated growth patterns of the palatine and maxillary segments in the hard palate of Rhesus monkeys. Their findings demonstrated that appositional bone growth in the transverse palatal suture promotes craniofacial development. Their conclusion was that surgical alterations in this area should be carefully performed with minimal disturbance of the potential growth sites.

Kremenak et al.[37,38] published literature that supported King and Schneiderman's assertion that wound contracture in growth areas could lead to attenuation of development. Kremenak et al. postulated that such contracture is the principal cause of maxillary growth attenuation. Latham et al.[39,40] published a study investigating the links between maxillary development and the vomer-nasal septum relationship in animal models. Lathams group resected the canine vomer while leaving the nasal septum unmolested. Although the subject group consisted of five subjects, retardation of maxillary growth in the anterior-posterior dimension was observed in all subjects.

Latham also speculated on the existence of a septopremaxillary ligament that was responsible for growth of the maxilla and midface. Latham et al. and Trotmann et al. [18,30] believed this ligament connected the maxillary hard palate with structures comprising the nasal capsule of the midface, and that disruption of this ligament as a consequence of surgical dissection would lead to growth attenuation of the midface. Latham et al. also theorized that in cases of unilateral cleft palate, the ligament is attached in the nonclefted area and is responsible for the deflection of the premaxilla from the cleft site.

Research published by Lynch and Peil[41] contradicted many of the observations of Latham et al. Lynch and Peil utilized beagle puppies as experimental animals. The animals were subjected to partial palatal stripping of the mucoperosteum, partial resection of the vomer and nasal septum, and given surgically induced palatal clefts. When the subject group was compared to the control animals, there were no marked different patterns of growth or development. In addition to craniofacial growth attenuation, clefting of the lip and palate dramatically affects development and symmetry of the maxillary alveolar segments. Ideal objectives following alveolar grafting are the restoration of arch symmetry, and the prevention of alveolar segment collapse. Maxillary arch stability has been attempted by a variety of techniques, including various regimens of presurgical alveolar molding through post-surgical orthodontics.

Derijcke et al.[42] addressed the issue of dental arch development by comparing untreated individuals possessing unilateral cleft lip and palate to unilateral cleft lip and alveolus. The comparison was made by analyzing dental casts from the respective groups. Derijcke et al.[42] concluded the arch forms of both groups were relatively symmetrical with the exception of the clefted area. The authors speculated that surgical intervention was responsible for aberrant development and not caused by the intrinsic nature of the cleft. Rosenstein said growth attenuation secondary to grafting is not a factor in his protocol due to the use of presurgical orthopedics and the minimal nature of soft-tissue dissection necessary to accomplish the primary grafting procedure. Rosenstein[5-7,9] said the use of presurgical alveolar molding in conjunction with early lip revision guides maxillary arch development and prevents segmental collapse, thus reducing the dimension of the cleft defect that must be grafted. By minimizing the cleft dimension to be grafted, the extent and surgical trauma incurred in alveolar grafting is also minimized.

Robertson and Fish[43] published a comparison of 50 patients presenting with complete unilateral cleft lip and palate to a group of 50 nonclefted patients. The cleft group was treated by a surgical protocol that included: presurgical orthopedic treatment from 0 to 3 months of life, lip and soft palate repair at 3 months, and hard palate repair at 11 months. This group was unique, because all treatment was conducted without bonegrafting. Robertson and Fish[43] were interested in the subsequent effects this treatment regimen had upon the development and continuity of the maxillary arch. The study found that at birth the cleft group possessed a broader maxillary arch in the canine and posterior alveolar arch than the nonclefted group. From birth to the third month of life, both groups were observed to have a decrease in the intercanine width; however, while the nonclefted subjects decreased in posterior width, the cleft group remained consistent in posterior alveolar width.

Using stereophotogrammetry, Berkowitz and Pruzansky[44] examined serial dental casts from a study group of three month old twins of opposite sex. The females in the group were noncleft patients, while their male siblings had unilateral complete cleft lip and palate. Their findings demonstrated similar values in anterior posterior and palatal widths. The authors concluded that vomeromaxillary separation did not cause significant retardation of maxillary growth.

Helms et al.[8] reported study results that correlated primary alveolar bone grafting and presurgical alveolar molding with greater maxillary arch symmetry, less disruption of tooth development, less incidence of cross bite, and no need for redrafting procedures. They analyzed grafting results that were divided into three study groups at three separate and distinct developmental stages. The primary group consisted of 20 rib grafts placed at less than one year of age; the secondary group consisted of 19 iliac crest grafts placed when the root of the permanent canine reached 25 to 50 percent formation, and a delayed group consisted of 18 iliac crest grafts placed following the eruption of the permanent canines. Records were taken at a minimum of five years post-surgically. The primary group consisted of patients treated by Drs. Rosenstein and Jacobson using Rosenstein's protocol for primary alveolar bone grafting. The secondary and delayed groups comprised patients treated by the authors at the University of Minnesota Cleft Palate Clinic. Growth inhibition is often cited by many detractors of primary alveolar grafting as a result of primary grafting. The authors speculated that the reported growth attenuation may not be significant as evidenced by the overall lack of crossbite observed in their study among the primary grafted groups, and by the lack of statistical significance among all groups in regard to anterior crossbites.

Athanasiou et al.[45] investigated dental dimensions that coincided with specific dentition phases in a population of repaired cleft lip and palate patients contrasted with a noncleft control group. This analysis was conducted using dental casts of the cleft group with landmarks defined in Moorree's[46] study (Figure 1). The developmental stages analyzed were at 3 to 4, 8 to 9, and 12 years of age. The maxillary arch widths were consistently smaller than the control group measurements; however, the measurements followed a harmonious increase with advancing age. Anterior-posterior dimensions were more affected than posterior width when compared with the control group. The researchers concluded that further evaluation was necessary to analyze growth attenuation in conjunction with specific surgical protocols.

Despite the voluminous research and efforts of many dedicated practitioners, the efficacy of primary alveolar bone grafting remains debated. A multitude of nonquantitative variables plague efforts to compare study results published by differing surgical groups and researchers. The clear agreements throughout the literature are about the desirability of establishing alveolar continuity, the elimination of the clefted defect, and the need for additional longitudinal studies.[47,48] The divisive issue regards the timing, modality, and surgical protocol that best serves the duality of correcting the physical defect without harming potential for craniofacial growth.

The purpose of this retrospective study was to evaluate (using exactly the same method of analysis published by Rosenstein in 1982) the craniofacial and dental arch development of circumpubertal unilateral cleft lip and palate patients treated in infancy at Riley Children's Hospital using Rosenstein's protocol for primary alveolar grafting and alveolar molding appliances. A secondary goal of this study was to stimulate interest in initiating a double blind longitudinal study of the cleft lip and palate patients treated at Riley Children's Hospital using Rosenstein's protocol and other treatment alternatives.

The authors hypothesis was that there would be no difference of statistical significance between the Riley experimental group and children who were treated using secondary alveolar bone grafting, thus, primary alveolar grafting via Rosenstein's protocol does not attenuate craniofacial growth to a more significant degree than non-primary alveolar grafting.

MATERIALS AND METHODS

The materials for this investigation consisted of lateral cephalometric films and dental study casts taken from an experimental group of 11 children. The experimental group consisted of 11 complete unilateral cleft lip and palate patients treated by the Craniofacial Team at Riley Children's Hospital using Rosenstein's protocol[6,15] for early primary alveolar bone grafting and were selected on a voluntary basis from patients reporting for follow-up visits at the Craniofacial Anomalies Clinic without regard to sex. The mean age of the experimental group at the time of record collection was 12 years 7 months and restricted to patients aged 13 years plus or minus 6 months. The experimental group consisted of Caucasian patients in an attempt to minimize variables impacting the uniformity of the study group. The small numbers of non-Caucasian cleft lip and palate patients referred for treatment at Riley Hospital make it difficult to get an adequate sample size for statistical analysis, given that ethnicity affects cephalometric standards.

METHODS

Lateral cephalometric films were taken on the 11 members of the experimental group, and dental impressions on seven members of the experimental group. The author was unable to acquire diagnostic impressions on the four remaining study group members due to restricted opening and severe gag reflexes. Orthodontic study casts were fabricated from the impressions, and the cephalometric films were manually traced with critical landmarks identified. The cephalometric films and orthodontic study models were then analyzed in an identical format used by Rosenstein[6],[15] (Figure 1,2,3). In order to eliminate as many variables as possible, the principal investigator was responsible for taking all radiographic films and impressions, and all measurements were made by three investigators in a blind format. The three investigators were the author, Dr. Jeffrey Dean, and Dr. Ronald Hathaway. Both Dr. Dean and Dr. Hathaway are orthodontists and members of the Craniofacial Anomalies Team at Riley Hospital.

CEPHALOMETRIC ANALYSIS

The lateral cephalometric films of the Riley group were exposed with the film cassette placed 12.5 mm from the patients midsagittal plane with the teeth in centric occlusion. The exposed films were traced on acetate paper with a white leaded orthodontic pencil. The tracing technique was consistent with current standard methods for tracing and identifying craniofacial structures from cephalometric films. All radiographic film placements and exposures were made by the author.

The following standard cephalometric landmarks[6] (Figure 2,3) were identified and traced manually by the three independent investigators.

A point (A): The deepest point on the curvature of the anterior maxilla between the anterior nasal spine and the crest of the alveolar process, the most forward point of the maxilla. When A point was obscured due to distortion in the anterior maxillary area, the midpoint between the anterior nasal spine and the alveolar crest was defined as A point.

Anterior Nasal Spine (ANS): The anterior-most point of the anterior nasal spine. Due to the anterior lateral displacement of the cleft segments, the anterior nasal spine is often deformed or obliterated. For the purpose of this investigation, the most anterior point of the maxillary basal bone was defined as anterior nasal spine.

Articulare (Ar): The point of intersection of the posterior margin of the ascending ramus and the external outline of the cranial base.

B point (B): The deepest point on the curvature of the mandibular alveolar process between the alveolar crest and pogonion; the most anterior point of the mandible in the median plane.

Basion (Ba): The most interior point on the occipital bone; representing the anterior margin of the foramen magnum.

Gonion (Go): A constructed point formed by the intersection of lines tangent to the posterior border of the ascending ramus and the mandibular planes.

Menton (M): The most inferior point on the mandibular symphysis.

Nasion (N): The most anterior point of the naso-frontal suture.

Pogonion (Pg): The most anterior point on the midsaggittal mandibular symphysis.

Sella turcica (S): The midpoint of the hypophyseal fossa.

The following linear and angular relationships were identified and traced in an identical fashion as the cephalometric landmarks:

SNA, SNB, ANB:	Established the anterior-posterior relation of the maxilla and mandible to each other and to the cranial base.
NA-Pg.	Established the hard tissue convexity of the face.
NS-MP:	Established the steepness of the mandibular plane angle.
N-ANS, ANS-Me:	Established the anterior vertical face height.

ANS-PNS, Go-Me: Established the maxillary and mandibular corpus length as percentage of linear relation.

The cephalometric landmarks listed above are identical to those used in Rosenstein's[6] study. The data derived from these angular and linear measurements of the Riley experimental group were compared with the same three groups that Rosenstein[6] used for comparison in his 1982 study.

The Riley group was compared with the three reference groups used and cited in Rosenstein's 1982 study.

Grafted: complete unilateral cleft lip and palate patients treated with early primary alveolar bone grafting and alveolar molding appliances. This group was Rosenstein's[6] experimental group used in his 1982 study (mean age 13 years 11 months).

Nongrafted: complete unilateral clefts treated without primary alveolar bone grafts and alveolar molding appliances (mean age 13 years 10 months). This data originated from The Cleft Lip and Palate Institute at Northwestern University Dental School and was used for comparison by Rosenstein.[6]

The control group (Bolton standard): consisting of averaged cephalometric values taken from nonclefted patients 12 to 16 years of age. This data originated from the Bolton-Brush Growth Study Center of Case Western Reserve University. This group was comprised of noncleft patients and compared with Rosenstein's[6] experimental group in Rosenstein's 1982 study.

ANALYSIS OF STUDY MODELS

Dental arch dimensions from the maxillary and mandibular study casts of the Riley experimental group were analyzed and compared against two groups (Figure 1, Table II) in a fashion identical to Rosenstein's 1982 study[6] and listed below:

Normal children (Moorrees, 1959)[46]: The values of dental arch dimensions of the normal patients (control group) consisted of the same data utilized by Moorrees in his study of the dentition of the growing child.

Nongrafted cleft patients (Athanasiou, 1988)[45]: The values of dental arch dimensions of the nongrafted cleft patients consisted of the same data utilized by Athanasiou in his study of the dentition of cleft patients treated surgically without bone grafting.

The maxillary and mandibular casts from the Riley experimental group were analyzed by using the incisal tip of the permanent cuspids, the lingual cusp tip of the second premolars, and the mesiolingual cusp tip of the first molars as reference points (Figure 1). The reference points were identified and marked with a standard number two pencil. If one of the reference teeth were missing or yet to erupt, a score corresponding to the mean value of the two neighboring teeth in the same segment was given. All measurements were made independently by the three investigators using the same sliding Miltex caliper.

RESULTS

CEPHALOMETRIC DATA

The three investigators in a blind format measured the cephalometric values, and examiner agreement reliability was assessed. The cephalometric values were analyzed for the mean value, the standard error of the means (SE), standard deviation (SD), intraclass correlation coefficients (ICC), and the range and number of values were established for the experimental group (Tables III, IV). The confidence levels for the individual intervals were established at the 95-percent level using 2-sample t-tests. The intraclass correlation coefficients (ICC) were calculated using the ANOVA technique. Intraclass correlation coefficients (Table IV) were used to measure the agreement between the three evaluators. There was good agreement among the investigators with regards to the NS-MP measurement.

The investigators moderately agreed on the SNB, ANB, and NAP values.

The investigators were in fair agreement in regard to the SNA, N-ANS, ME-ANS, and N-ANS / ME-ANS values.

Those values generating poor agreement were ANS-PNS, GO-ME, and ANS-PNS/ GO-ME.

The angular and linear measurements from the Riley experimental group were compared with the three groups cited by Rosenstein[6] (Table V) and summarized as follows (Table III):

SNA: Riley experimental group mean value was less than the Bolton standard but not different than the Grafted and Nongrafted groups.

SNB: Riley group mean value was not different than the Bolton standard, Grafted, and Nongrafted groups.

ANB: Riley group mean value was greater than the Bolton standard but not different than the Grafted and Nongrafted groups.

NAP: Riley group mean value was greater than the Bolton standard but not different than the Grafted and Nongrafted groups.

NS-MP: Riley group mean value was not different than the Bolton standard, Grafted, and Nongrafted groups.

N-ANS: Riley group mean value was greater than the Bolton standard, Grafted, and Nongrafted groups.

ME-ANS: Riley group mean value was greater than the Bolton standard, Grafted, and Nongrafted groups.

ANS-PNS / GO-ME: Riley group mean value was not different from the Bolton standard, Grafted, and Nongrafted groups.

ORTHODONTIC STUDY CAST DATA

The eight dental study casts measurements were analyzed (Figure 1, Table II) for the mean value, the standard error of the means (SE), standard deviation (SD), range of values, and intraclass correlation coefficients (ICC). Comparison of these dimensions for the dental cast data were made among the Riley group, the normal data used by Moorrees[46] and the nongrafted group of Athanasiou[45] (Table II). The confidence levels for the individual intervals were established at the 95-percent level using the Students t-test technique. All casts were measured independently in a blind format by the three investigators, and examiner reliability was assessed. UArL and Larl refer to maxillary and mandibular arch length.

The measurements were assessed (refer to Table IV for criteria) as follows: The 3+3, 5+5, UArL, 3-3, 5-5, and 6-6 all had ICCs greater than 0.90, which indicated excellent agreement among the examiners with the exception of one UArL measurement, which one examiner had scored lower than the remaining two examiners. The ICC for LArl was 0.63, indicating moderate agreement among all examiners. The ICC for 6+6 was 0.32, indicating poor agreement among the examiners.

All recorded dimensions were analyzed using the Student's t-test, and discrepancies were found to be statistically insignificant (p less than 0.05). Arch dimensions of the Riley experimental group were recorded to be smaller in all dimensions of length and width when compared with the normal group of Moorrees. The arch width dimensions of the Riley experimental group were not statistically different (p less than 0.05) from recorded measurements of Athanasiou's nongrafted group, except in the maxillary first permanent molar region (6+6). The length of the maxillary and mandibular study models arch dimensions (UArL and LArL) of the Riley experimental group were not statistically different from those of the nongrafted group of Athanasiou.

FIGURES AND TABLES

TABLE I

Primary, secondary, and delayed alveolar grafting

Primary	Typically performed before the eruption of the primary dentition or as late as one year of age.
Secondary	Grafting following the eruption of the permanent dentition usually ranging from nine to eleven years of age.
Early Secondary	Grafting procedures performed from five to six years of age.
Delayed (Late Secondary)	Grafting procedures following the eruption of the permanent canines.

TABLE II

Comparisons of dental arch length and width (Figure 1)

		N	Mean	S.D.	S.E.	Range
3 + 3	G	7	28.31	4.2	1.59	23.87-36.00
	NG	18	27.05	3.54	0.83	21.00-34.00
	N	*	33.11	2.06		
5 + 5	G	7	30.06	4.59	1.74	23.13-38.20
	NG	18	32.13	3.78	0.89	22.00-38.00
	N	*	35.84	2.9		
6 + 6	G	7	35.20	1.97	0.74	31.60-36.97
	NG	18	39.72	4.22	0.99	30.00-47.00
	N	*	40.41	2.9		
Uarl	G	7	25.35	3.6	1.36	20.23-29.97
	NG	18	27.44	2.7	0.63	23.00-35.00
	N	*	28.89	2.85		
3---3	G	7	24.78	3.02	1.14	20.90-28.10
	NG	18	24.94	2.13	0.5	21.00-30.00
	N	*	25.18	1.58		
5---5	G	7	28.36	3.39	1.28	23.57-32.77
	NG	18	29.44	2.81	0.66	26.00-34.00
	N	*	32.51	2.74		
6---6	G	7	31.75	2.57	0.97	38.57-35.73
	NG	18	34.38	3.5	0.82	29.00-42.00
	N	*	34.82	2.95		
Larl	G	7	24.63	1.01	0.38	22.90-25.93
	NG	18	24.86	2.18	0.51	20.00-28.50
	N	*	23.45	2.08		

G# = Riley Group.
NG# = Athanasiou Study (nongrafted).
N# = Moorrees Study (normal).
* = Patient number unknown.

TABLE III

Statistical analysis of the Riley experimental group[a]

	N	Mean	S.D.	S.E.	Minimum	Maximum	Bolton Standard	Grafted Group	Nongrafted Group
SNA	11	77.09	3.99	1.20	71.00	82.67	-	=	=
SNB	11	77.30	4.99	1.50	70.00	85.00	=	=	=
ANB	11	-0.21	2.47	0.74	-4.17	3.67	=	=	=
NAP	11	181.30	4.57	1.38	174.00	187.67	+	=	=
NS-MP	11	34.64	6.94	2.09	26.17	49.00	=	=	=
N-ANS	11	52.53	4.97	1.50	45.17	60.67	+	+	+
ME-ANS	11	70.05	3.94	1.19	65.50	78.00	+	+	+
N-ANS / ME-ANS	11	75.23	6.95	2.10	66.89	91.30	+	+	+
ANS-PNS	11	49.94	5.21	1.57	41.17	59.50	+	+	+
GO-ME	11	65.73	3.69	1.11	57.50	71.33	=	=	=
ANS-PNS / GO-ME	11	76.33	6.96	2.10	67.89	87.33	=	=	=

[a] = Denotes no statistical significance between the Riley group and the comparison groups.

+, - Riley group reported greater or lesser values than the comparison groups.

TABLE IV

Intraclass correlation coefficients[a] of the Riley experimental group

SNA	.59	fair agreement between evaluatiors
SNB	.78	moderate agreement between evaluators
ANB	.79	moderate agreement between evaluators
NAP	.74	moderate agreement between evaluators
NS-MP	.87	good agreement between evaluators
N-ANS	.52	fair agreement between evaluators
ME-ANS	.61	fair agreement between evaluators
N-ANS / ME-ANS	.46	fair agreement between evaluators
ANS-PNS	.09	poor agreement between evaluators
GO-ME	.32	poor agreement between evaluators
ANS-PNS / GO-ME	.28	poor agreement between evaluators

[a]Perfect agreement	=	1.0	
Good agreement	=	.8	-1.0
Moderate agreement	=	.7	-0.8
Fair agreement	=	.4	-0.6

TABLE V

Comparison of study results

Sample	SNA	SNB	ANB	NAP	NS-MP	N-ANS	ANS-Me	Maxillary / Mandibular
Grafted	74.5	72.9	1.60	178.8	40.9	43.5	56.6	71.5
(13 yrs., 11 mo.)	(.95)	(.64)	(.67)	(1.90)	(1.22)	(.52)	(.52)	(1.34)
n:16								
Nongrafted	74.9	73.5	1.40	180.0	42.9	44.1	55.9	70.9
(13 yrs., 10 mo.)	(.55)	(.72)	(.88)	(2.15	(1.29)	(.48)	(.49)	(1.20)
n:16								
Bolton standard	83.7	80.6	3.1	176.5	32.0	45.9	54.1	77.2
(14 yrs., 0 mo.)	(.26)	(.24)	(.10)	(.45)	(.32)	(.17)	(.17)	(.32)
n:16								
Riley study	77.1	77.3	-0.2	181.2	36.6	50.4	70.0	78.5
(13 yrs., +/- 6 mo.)	(1.22)	(1.5)	(.74)	(1.38)	(2.09)	(1.50)	(1.19)	(2.10)
n:11								

[a](xx) denotes standard error $p < 0.05$

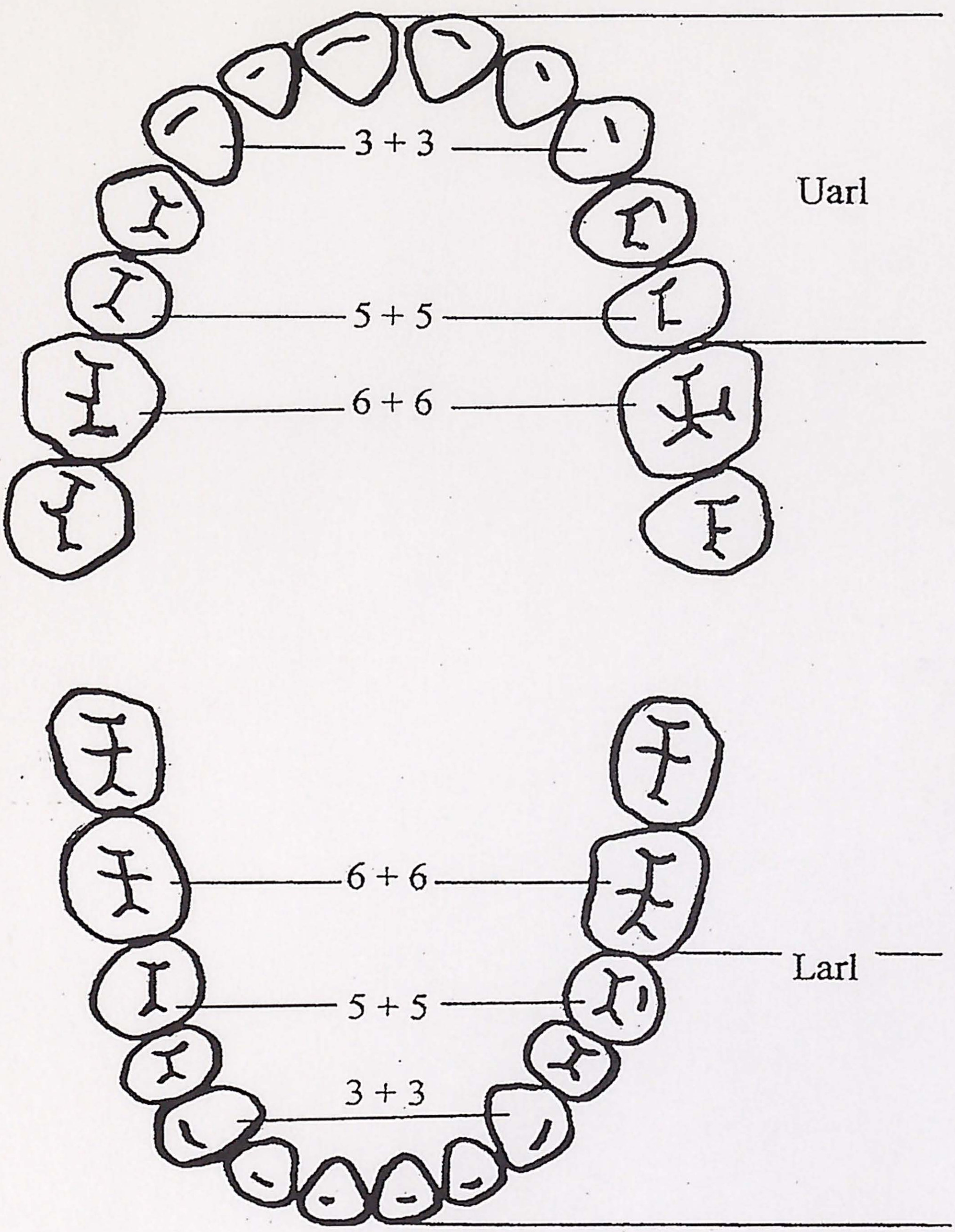

FIGURE 1. Dental arch dimensions and measurement parameters.

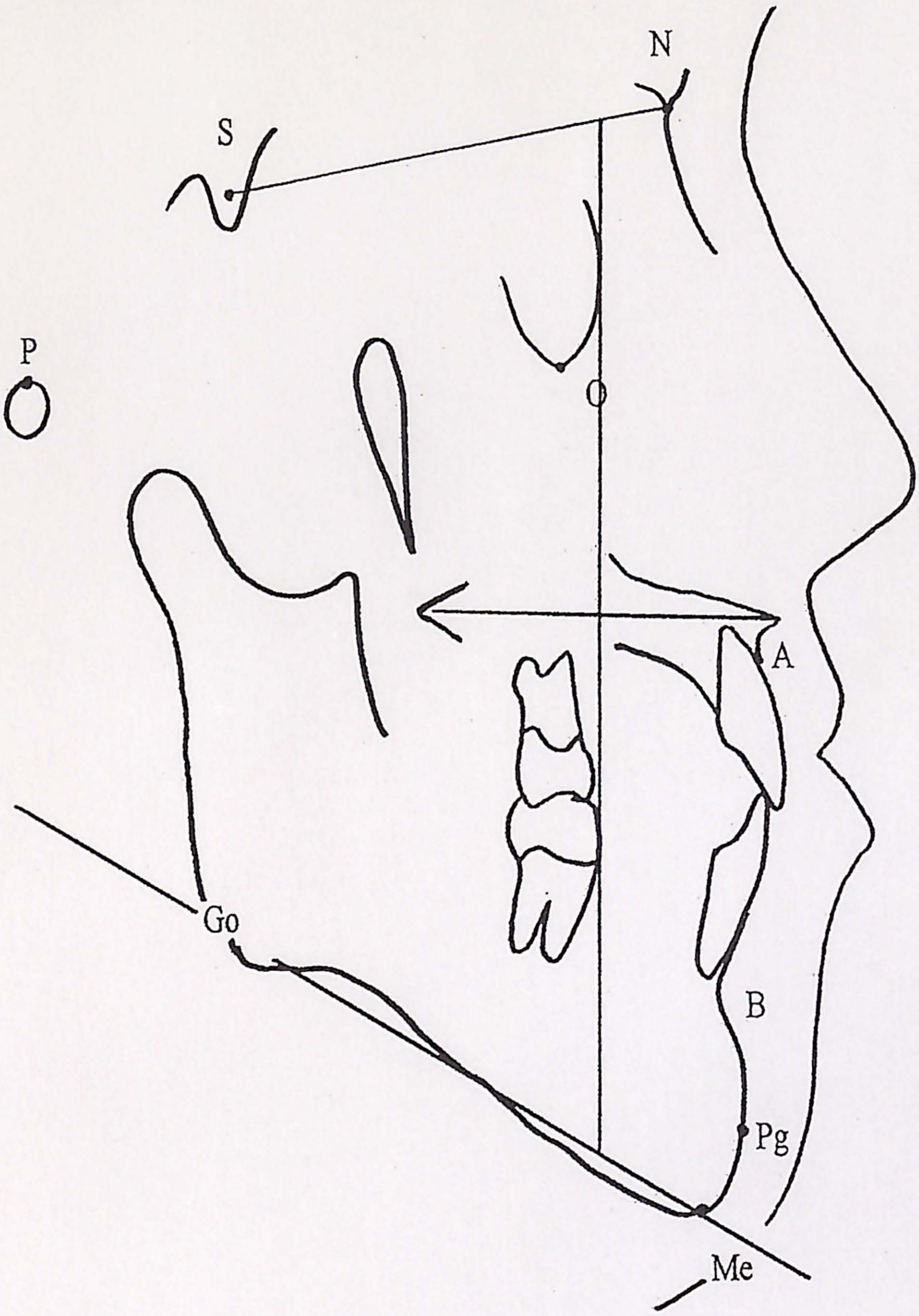

FIGURE 2. Cephalometric landmarks and linear measurements: N-ANS, ANS-Me, and ANS-PNS/Go-Me.

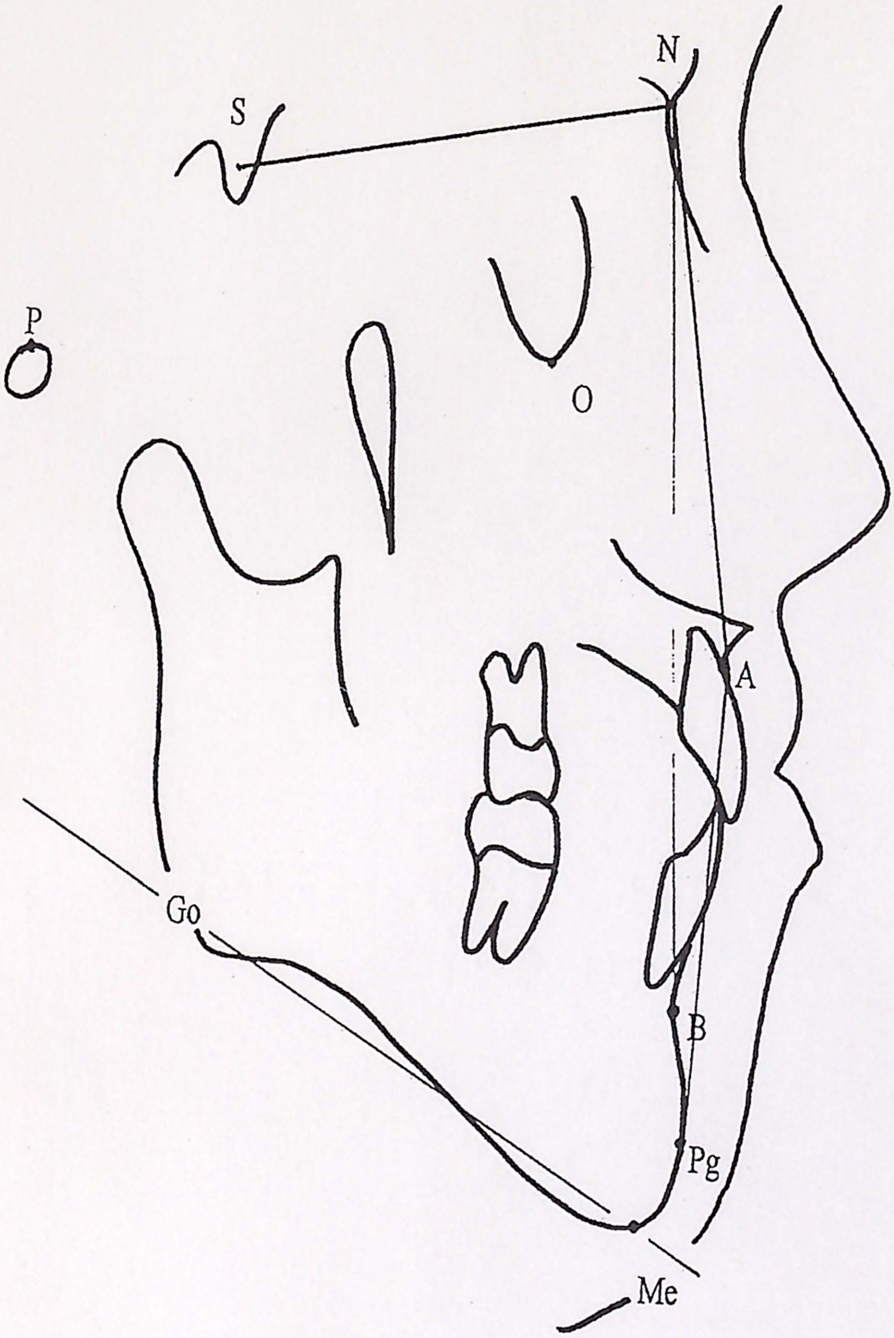

FIGURE 3. Cephalometric evaluation of angular relations: SNA, SNB, ANB, NAP, and NS-MP.

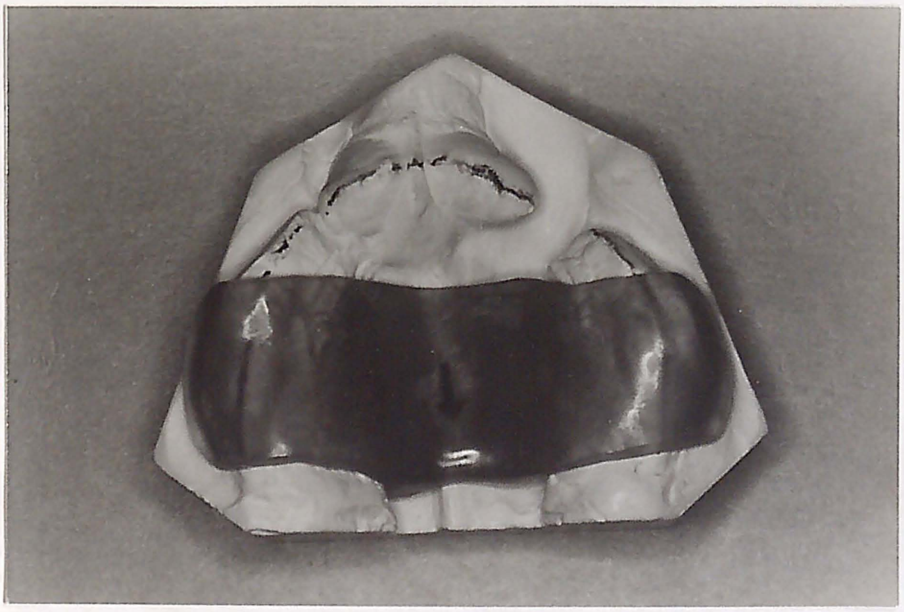

FIGURE 4. Alveolar molding appliance fabricated to Dr. Rosenstein's specifications.

DISCUSSION

Rosenstein[6] selected five angular and three linear craniofacial relations to compare reported maxillofacial growth attenuation as a consequence of primary alveolar bone grafting. Rosenstein compared his experimental group of primary alveolar grafted patients against a control group of nonclefted children and a group of nongrafted cleft lip and palate children. This investigation compared the Riley experimental group against Rosenstein's experimental group, a nongrafted group, and a control group of nonclefted children in a manner identical to Rosenstein's 1982 study.

The cephalometric values of SNA, SNB, and ANB establish the relation of the cranial base to the maxilla and mandible as well as the degree of harmony between the maxilla and mandible. Craniofacial growth attenuation among clefted individuals is expressed in significantly lower SNA and SNB values than those accorded to noncleft children. Though the SNA and SNB values of the Riley group were less than that of the control group (Bolton Standard), the Riley values closely approached the normal control values (Table IV) and were significantly better than those of Rosenstein's experimental group and the nongrafted group (Table IV). Although not ideal, the ANB results of the Riley group demonstrated a harmonious relationship of the maxilla and mandible. The lower than normal values for the Riley group (SNA, SNB, ANB) support the expected deficiency of the maxilla secondary to the cleft disruption of potential growth centers. The significance of these angular measurements lie in the improvement demonstrated in the Riley group when compared with the nongrafted group and Rosenstein's experimental group. This result indicates that Rosenstein's protocol, if not better than delayed grafting, is at least no worse than other treatment regimens. As with most radiographic films, which require manual location and measurement of landmarks, the possibility of error and poor correlation among different examiners can exist. The Riley group demonstrated fair to moderate agreement among the angular data (SNA, SNB, ANB). Factors that could have attributed to inaccuracy are: the quality of the radiographic film, difficulty in locating skeletal landmarks, and transposition of other anatomical structures and artifacts.

The hard tissue convexity of the face was evaluated by the angular values (Table IV) corresponding to the landmarks of N point, A point, and P point. These values provide the examiner with a snapshot of facial growth and harmony between the maxillary process and the mandible. The Riley group exhibited higher than control values (181.2 Vs 176.5) but remained close to the values of Rosenstein's experimental group and the nongrafted group. Higher values reflected in the NAP relationship could be attributed to the reported clockwise mandibular rotation in response to vertical growth attenuation subsequent to the cleft deformity.

The mandibular plane angle also provided a comparison of the mandible to the cranial base. The NS-MP values of the Riley group (Table V) were better than the reported values for the nongrafted group and Rosenstein's experimental group. While the Riley group was at least 4° closer to the control value than the nongrafted and Rosenstein's experimental group, the Riley group exhibited a steeper mandibular plane angle than that of the control group. This result appears to support previously mentioned reports of mandibular compensation (an increase in the mandibular angle) in response to a developmentally deficient maxilla.

The linear assessment of anterior face height (Table IV) between Rosenstein's group, the nongrafted group, and the Riley group demonstrated significantly greater values among the Riley group. The Riley measurements were also greater than the control group. These results indicate there was no great deficiency in face height resulting from primary alveolar bone grafting; however, this result could be skewed due to a compensatory mandibular rotation resulting from maxillary growth deficiency.

The linear comparison of maxillary corpus length (ANS-PNS / GO-ME) resulted in values for the Riley group, which were in close agreement to the control group (Table IV). The values expressed by the Riley group were significantly better than Rosenstein's experimental group and the nongrafted group. This ratio is significant in that a low numerical score would express any maxillary growth attenuation resulting from a cleft deformity or subsequent surgery. One point, which should be considered, is that of all measurement criteria in this study, the linear relationships exhibited the greatest variance in all categories of statistical measurement (Table IV). This variance could be attributed to the difficulty in accurately identifying the critical landmarks necessary for these measurements. Artifact and superimposed hard and soft tissue structures often mask these landmarks (ANS,PNS). The large picture one gathers from these linear and angular values is that the Riley results are not of such striking difference, when compared with the control group and the nongrafted group, that one could uniformly reject the surgical protocol (Rosenstein's protocol) used at Riley Hospital in treating cleft lip and palate patients.

The main argument against primary alveolar bone grafting is that the procedure is detrimental to and attenuates maxillofacial development. The clinical significance of the

Riley data is simply that no difference of statistical significance exists among the Riley group, Rosenstein's experimental group, the nongrafted group, and the control group. If primary alveolar bone grafting were of such detriment, one would expect to record cephalometric values much lower than the values cited in the nongrafted and control population.

The orthodontic study casts of seven members of the Riley experimental group were analyzed and compared with a nongrafted and control group (Table II). The Riley group exhibited smaller dimensions in width and length than those of the control group (nonclefted) and exhibited no difference of clinical significance from the nongrafted group. This limited analysis implies that children receiving primary alveolar bone grafting and alveolar molding appliances show no substantial improvement in arch dimension over the nongrafted group.

The agreement among examiners ranged from poor to excellent and could be attributed to the individual variability in choosing exact reference points on the designated landmarks. Distortion of impression material, due to patient compliance and restricted opening, could lead to inaccurate study models and measurement errors. The Riley study cast analysis shows no harm resulting from primary alveolar bone grafting; however, the sample size is too small for a definitive statement.

Although the Riley group demonstrated the expected lower values than the control group, the real issue is that there is no difference of statistical significance when compared with the nongrafted group. While the Riley data does not demonstrate significant benefit, the results contradict claims that primary alveolar bone grafting is a detrimental treatment regimen when compared with the nongrafted group.

SUMMARY AND CONCLUSIONS

Detractors of primary alveolar bone grafting contend that any esthetic or orthodontic advantage that could result from primary alveolar bone grafting is offset by the reported craniofacial growth attenuation. Though the data reported in this study is dimensionally less than that of nonclefted groups, the dimensional differences between the grafted and nongrafted groups is not of statistical significance and in some instances better. If primary alveolar bone grafting truly attenuated craniofacial growth, one would expect to see significant differences in craniofacial linear and angular values between the grafted and nongrafted groups.

The fact that both the grafted and nongrafted groups demonstrated smaller dimensions than the nonclefted group is not a surprise. That the physical deformity of cleft lip and palate adversely affected craniofacial growth centers and thus craniofacial growth has frequently been reported in the literature. The continued debate centers upon treatment regimens applied to resolve the physical defects. The data reported in this study showing no difference of statistical significance among patients treated with primary alveolar bone grafting and nongrafted groups lend weight to Rosenstein's contention that his surgical protocol is not a detriment to craniofacial growth.

This restrospective study was an effort to replicate the results reported in Sheldon Rosenstein's 1982 study of unilateral cleft lip and palate patients treated through early primary bone grafting and orthopedic alveolar molding appliances. The craniofacial Anomalies Team at Riley Children's Hospital has followed the Rosenstein protocol in treating cleft lip and palate patients. The age of the study group, drawn from this patient population, was at least 13 years postsurgery plus or minus six months.

The results of the cephalometric analysis show no difference of statistical significance between cleft patients treated with primary alveolar bone grafting and cleft patients not treated with primary alveolar bone grafting. Although the width between the maxillary molars was smaller in the grafted group, all other values of the dental study casts revealed differences of no statistical significance. The focal argument against primary alveolar bone grafting centers around literature reporting results of craniofacial growth attenuation. The majority of this reported data is complicated by a variety of intangible variables such as: a number of different surgical techniques and protocols, differing treatment facilities, extent of surgical dissection in the cleft areas, varying chronological ages of patients receiving treatment, the use or lack of presurgical orthopedics, and differing measures of parameters of success and failure. The magnitude of such variables makes relative comparison of the reported literature difficult. Rosenstein's protocol addresses these variables by applying a standardized and consistent treatment regimen covering surgical and orthopedic modalities.

The long-term data generated from Rosenstein et al. has consistently demonstrated cephalometric and model dimensions that are either statiscally better or at least of no significant statistical difference when compared with nongrafted groups. The study results reported from Riley Hospital also support Rosenstein's data. Although the Riley study group population was limited, it does accurately represent a cross-section of the patients treated for complete unilateral cleft lip and palate via Rosenstein's protocol. Our data also demonstrated either improved craniofacial dimensions or values that were not statistically significant from nongrafted patients, these data support similar findings published by Tanimura.[47]

Primary alveolar grafting allows early intervention and resolution of a physical deformity through a procedure with an extremely low morbidity and mortality rate. The resolution of this problem at such an early precognizant age has immeasurable value on such intangible issues as self-esteem, parental acceptance, and the integration of the patient and parents into specialty team care and follow-up. The physical and emotional effects of the surgical intervention are resolved early in life, and future treatment of dental and orthodontic problems are made much less complicated. The emotional and physical stigma the patient must endure as age and development progresses is much reduced, and the patient and parents have become informed participants in their treatment plan.

The next step in accurately assessing Rosenstein's protocol would be a controlled, randomized clinical trial in which one group of cleft lip and palate patients would be treated following Rosenstein's protocol and a separate group treated following a different surgical protocol such as secondary grafting. These groups could be standardized among each other in regards to age, sex, cleft type, attending surgeons, and comprehensive radiographic and concurrent alveolar arch models. This type of study when compared with the Bolton standard should resolve the continued debate concerning the efficacy of Rosenstein's protocol. It is the author's hope that this thesis will provide the motivation for such a prospective study.

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ABSTRACT

A RETROSPECTIVE STUDY OF CIRCUMPUBERTAL CLERT LIP AND PALATE PATIENTS TREATED IN INFANCY WITH PRIMARY ALVEOLAR BONE GRAFTING

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The Riley Children's Hospital Craniofacial Anomalies Team rigorously follows a treatment protocol developed by Dr. Sheldon Rosenstein for the treatment of cleft lip and palate patients. Rosenstein's protocol incorporates primary bone grafting and alveolar molding appliances for cleft lip and palate patients. While other cleft lip and palate treatment centers utilize alveolar molding appliances, there remains debate concerning the efficacy of primary bone grafting. The principal detraction of primary bone grafting is the concern that such early surgical treatment affects maxillary and craniofacial growth and development. The purpose of this retrospective study was to analyze post-treatment lateral headplates and dental casts of cleft lip and palate circumpubertal patients treated in

infancy at Riley Hospital in Indianapolis by the Craniofacial Team following Rosenstein's protocol. The hypothesis was that primary alveolar bone grafting in conjunction with the use of alveolar molding appliances contributes to the early stabilization of the alveolar segments, and produces no statistically significant difference in craniofacial development among primary bone grafted patients and nongrafted patients. The dental arch dimensions of the nongrafted patients (control group) consisted of the same data utilized by Moorrees in his study of the dentition of the growing child. The dental arch dimensions of nongrafted cleft patients consisted of the same data utilized by Athanasiou in his study of the dentition of cleft patients treated surgically without bone grafting.

Of the eight measurements made by the three examiners, six demonstrated excellent interexaminer agreement, one demonstrated moderate interexaminer agreement, and one demonstrated poor interexaminer agreement. The arch width and length for the grafted group was significantly smaller ($p < .05$, Student's t-test) than the normal group in all measures except for the mandibular canine width. The arch width and length for the grafted group was not significantly different ($p < .05$, Student's t-test) than the nongrafted group, except for the maxillary molar width where the grafted group was smaller than the nongrafted group.

The cephalometric values of the Riley group were compared against a nongrafted group, an early primary grafted group, and the Bolton standard values cited in Rosenstein's study. The Bolton standard values were used as the control group.

This study found the cephalometric values of the Riley experimental group (treated following Rosenstein's protocol) to be of no statistically significant difference (p < .05, Students t-test) when compared with cephalometric values of the nongrafted and primary alveolar grafted groups cited in Rosenstein's 1982 study. The cephalometric values of the Riley experimental group were less than the cephalometric values of the nonclefted patients (Bolton standard control group) cited in Rosenstein's 1982 study. Interexaminer agreement ranged from poor to good with the poorest agreement among the linear values of ANS/PNS and GO/ME. The intraclass correlation coefficient values for SNA,m ANB, and SNB ranged from fair to moderate.

The Riley cephalometric values were equal or slightly better than Rosenstein's grafted and nongrafted groups. Though smaller than the control group, the Riley cephalometric values were of no statistical significance (p < .05, Students t-test) when compared with the same parameters cited in Rosenstein's study. Although these findings infer that the patients treated following Rosenstein's protocol demonstrate some degree of craniofacial growth attenuation when compared with nonclefted patients (Bolton standard control group), the Riley patients showed no worse growth attenuation than similar patients treated without Rosenstein's protocol for primary alveolar grafting.

The hypothesis of this thesis was that Rosenstein's protocol was viable and non-detrimental when compared with other treatment regimens. The results of this study support the hypothesis that Rosenstein's surgical protocol is not a contributing factor in craniofacial growth attenuation among cleft lip and palate patients.

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