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AN EVALUATION OF DENTAL ARCH RELATIONSHIPS AMONG CHILDREN WITH UNILATERAL CLEFT LIP AND PALATE TREATED AT TWO DIFFERENT CENTERS

by

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Abstract

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The objectives of this study were to determine if differences exist in dental arch relationships (favorable vs. unfavorable) after primary surgery for children with unilateral cleft lip and palate between two craniofacial centers and to consider the reliability and validity of the Goslon Yardstick.

Non-syndromic unilateral cleft lip and palate children were treated. Several surgical protocols were used to repair a cleft lip and palate. The dental arch relationships were categorized using the Goslon Yardstick. Favorable inter-arch relationships were Groups 1, 2, and 3 and unfavorable arch relationships were Groups 4 or 5. Odds ratios were calculated to quantify the association between unfavorable dental arch

relationship and samples. Kruskal-Wallis statistics, Mann-Whitney statistics tested for differences in the dental arch relationship among the three samples.

The prevalence of unfavorable arch relationships was found for 22%, 11%, and 28% for Samples A, B, and C respectively. There was no statistically significant association between the samples and unfavorable outcome ($p > 0.05$). Sample A and B showed a statistically significant lower mean rank Goslon Group compared with Sample C ($p \leq 0.05$).

The Goslon Yardstick gives a general outcome assessment of the relationship of the dental arches. Even though a statistically significant higher prevalence of unfavorable outcomes was found in Sample C ($p < 0.05$), one cannot conclude that an unfavorable outcome will be more likely in any one sample.

DEDICATION

To my loving wife, Amanda, thank you for your support and understanding over the last three years and the sacrifices that you have endured.

To my mother and father, thank you for providing me with every opportunity to succeed and for your endless support and guidance.

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INTRODUCTION

There are many craniofacial anomalies and all exhibit variation in their effect on craniofacial morphology. One of the most common craniofacial birth defect is unilateral cleft lip and palate. The incidence of cleft lip and palate is one in 500 to 1000 live births (Moreno et al. 2004). Orthodontic treatment of the cleft lip and palate child is challenging. Many patients with this craniofacial anomaly experience a significant reduction in maxillary development. Therefore, it would be helpful to identify factors that influence future dental arch relationships.

Primary surgery to correct a cleft lip and palate contributes to maxillary deficiency (Ross 1987). The Eurocleft project surveyed 201 centers and identified 194 different protocols. Children underwent one to four operations following one of 17 different sequences. Lip repair was performed sometime between birth and twelve months of age. The primary and secondary palate was repaired sometime between birth and 13 years. Many

different surgical procedures were reported (Shaw et al. 2000). Factors contributing to arch discrepancies include tissue deficiency and displacement, abnormal nasal septum and abnormal muscle function. The orthodontist craniofacial team member focus on the dentition of the cleft patients, leads to their interest in differences in dental arch relationship between various craniofacial centers using different protocols.

Retrospective studies comparing two groups within a single center face the possibility of sampling bias and biases related to gradual changes in experience and/or technique when the data are collected over long time periods while inter-center studies can provide a sufficient number of patients to compare surgical outcomes such as speech, esthetics and growth.

Multi-center studies can allow direct comparison of components of the treatment protocol and comparison of the effects of primary surgeries on subsequent jaw growth (Shaw et al. 1992). It is difficult to collect data at multiple centers in a standardized unbiased format, due to different availability of potential explanatory factors, different levels of severity of cases, etc. Some centers may have higher percentage of more severe

patients. The Goslon (Great Ormond Street, London, and Oslo,Norway) Yardstick is a clinical tool used to evaluate the effects of primary surgery on the outcome of treatment. The Goslon Yardstick has been reported as a clinical tool to qualitatively assess dental arch relationships in three planes of space in patients with a cleft lip and palate (Mars et al. 1987). In a six-center international study in Europe, the Goslon Yardstick was used to assess favorable outcomes and demonstrated the importance, among other things, of having an organized central and uniform treatment protocol (Shaw et al. 1992).

The Goslon Yardstick is intended to measure the severity of the malocclusion and difficulty of correcting it, but does not consider generalized crowding and irregularity. Antero-posterior arch relationships, vertical labial segment relationships; and transverse relationships are considered. The yardstick was developed at the Hospital for Sick Children at Great Ormond Street in London. A favorable outcome has been defined as Groups 1, 2, and 3 because a good orthodontic result is expected without surgery. An unfavorable outcome has been identified as Group 4 would only need surgery if growth

was unfavorable and Group 5 would require orthognathic surgery (Mars et al. 1987; Morris et al. 2000; Shaw et al. 1992; Chan et al. 2003). Alternatively, Groups 1 and 2 were considered favorable and Groups 4 and 5 were considered unfavorable (Hathorn et al. 1996; Pigott et al. 2002).

Mars and coworkers stated "The Yardstick can be applied reliably only by those trained in its use with the original cases and who have undertaken the categorization of a calibration series" (Mars et al. 1987). The reliability of this measure has been reported to be consistent at different times. The results were found to be reproducible, repeatable, and consistent in a study of four orthodontists ranking the models twice, one week apart. Reliability was measured by Spearman's rank correlation coefficient and the Wilcoxon matched paired signed rank test. Intra-examiner and inter-examiner agreement was high (Mars et al. 1987).

REVIEW OF THE LITERATURE

Midface Growth

Fibrous Sutures

Little is known about the growth of the midface. Weinmann and Sicher (1947) theorized that the growth of the maxilla is thought to be due to four fibrous sutures: the frontomaxillary suture, zygomaticomaxillary suture, zygomaticotemporal suture, and the pterygopalatine suture. All four sutures grow in a parallel direction. The growth from these sutures "shifts" the maxilla downward and forward. Transverse growth occurs at the median palatine suture and the pterygoid processes. Growth also occurs in all the other sutures of the facial skeleton.

Chondrovomeral Joint

Scott studied the chondrovomeral joint and its connection between the vomer and the septal cartilage separated by loose connective tissue. The cartilage of the nasal septum is considered a mechanism of growth of the midface. There are three ossification centers:

basiocciptal, basisphenoid, and presphenoid center. The ethmoid and inferior conchae replace parts of the cartilage by endochrondral ossification. Scott stated that the cartilage and periosteum carry an intrinsic genetic message that guides their growth. The synchondroses and nasal septum are centers of facial growth. This explains the forward position of the nasal septum in patients with a bilateral cleft lip and palate (Scott 1953).

Cartilaginous Nasal Septum

Sarnet felt that the cartilaginous nasal septum is responsible for downward and forward growth of the mid-face. The hypothesis was tested by either dislocation or resection of the cartilaginous nasal septum. The dislocated septum deviated and showed no growth difference. In the studies where the septum was removed, the midface showed lack of forward growth (Sarnet 1991).

Hans et al. (1996) used anti-rat nasal septum cartilage antisera(RNS-IgG) to inject in rats. A decrease in snout length, premaxillary length, premaxillary displacement and bimaxillary width was found. The nasal septum was believed to be important for normal A-P development of the mid-face.

Anterior Septo-premaxillary Ligament

The anterior septo-premaxillary ligament is attached to the anterior border of the nasal septum and postero-inferiorly to blend with premaxillary periosteum and inter premaxillary suture which was found by Lathum to provide septum growth force in the embryonic period. Patients with septo-premaxillary deformity, such as a cleft lip and palate, do not get a septal growth force. Lack of the septo premaxillary ligament contributes to midface growth deficiencies (Lathum 1969).

Functional Maxtrix Theory

Behrents and Johnston (1984) wanted to test the hypothesis that the nervous system was responsible for growth of the midface. "Neurotrophism mechanisms" was suggested by Moss who presented the Functional Matrix Theory (Moss 1971). Behrents and Johnston found in animal studies that the trigeminal nerve, when severed, did not contribute to midface deficiencies.

Classification

Classification of cleft lip and palate is based on embryology of the defect rather than on clinical findings. In the 1920s and 1930s, Veau used a four category classification. In 1971, the striped Y symbolic

classification for cleft lip and palate by Kernahan is found to be simple and an easily recognizable classification system (Kernahan 1973). This system describes the different processes that do not come together to form the lip and palate.

Embryology

In the fifth week of pregnancy, the lateral swellings form the alae of nose. The medial nasal swellings form the middle portion of the upper lip, the nose, the maxilla and the entire portion of the primary palate. The maxillary swellings approach medial and lateral swellings and form grooves.

In 6-7 weeks, the upper lip is formed by two medial nasal swellings and the two maxillary swellings. The intermaxillary segment is also formed at a deeper level by the two medial swellings and the two maxillary swellings. This segment is continuous with the nasal septum which is formed by the frontal prominence. The intermaxillary segment is also the labial component or the philtrum of the lip, the maxillary four incisors and the palatal component.

The palatine shelves are located in vertical position on each side of the tongue at six and one-half

weeks. This is the secondary palate formed by two outgrowths from the maxillary swellings. The palatine shelves are horizontal and the tongue drops downward. The secondary palate is formed at seven and one-half weeks. The anterior shelves will fuse with the primary palate. The nasal septum also fuses with the secondary palatal shelves. At week ten, the palatine shelves fuse with each other and the nasal septum (Peterson 1993).

The incidence of cleft lip and palate is one in 500 to 1000 live births (Moreno et al. 2004). Cleft lip and palate is more common in boys than girls in a 3/2 ratio. Unilateral cleft is more common than bilateral cleft in a 3:1 ratio and the left side of the lip is more commonly than the right side. Cleft lip and palate is classified by the involvement in the primary and secondary palates (Ortiz-Posadas et al. 2001). The primary palate involves structures anterior to the incisive foramen, which are the lip and alveolus. The secondary palate involves structures posterior to the incisive foramen which includes the hard and soft palate.

Protocol

Pre-surgical Orthopedic Treatment

Pre-surgical orthopedic treatment (PSOT) has been suggested to be the first therapeutic intervention in the newborn with cleft lip and palate to improve arch form (Rosenstein et al. 2003; McNeil 1955; Lathum 1980). Pruzansky, in 1964, felt that this method hindered palatal reconstruction by preventing a narrowing of the cleft (Pruzansky 1964). Primary bone grafting can also be performed during the first year in conjunction with PSOT. Studies have shown this protocol produces positive growth results of the maxilla and have an additional benefit of improving the prognosis for the lateral incisor (Rosenstein 1963; 1969; Rothstein et al. 2003). Prahl et al. (1997) found no difference between those treated with PSOT and those without).

Pin-Retained Appliance

Lathum, in 1980, began using a pin-retained appliance in the newborn to align the lesser segment with the greater segment. The appliance was found to improve the transverse dimension and inhibit anterior-posterior growth. Lip repair was found to provide enough force to shape the maxillary arch (Kuijpers-Jagtman and Long 2000;

Chan et al. 2003). Kuijpers-Jagtman and Long found no long term benefits to justify use of the pin-retained appliance introduced by Latham. Pre-surgical orthopedics in the form of a pin retained appliance was used by a craniofacial clinic in Bristol, U.K. Mid-face deficiency was more prevalent in the Bristol group when compared to the protocol used by Oslo, Norway. Lateral cephalographs were used to compare the samples (Roberts et al. 1996).

Lip Repair

Lewin (1964) sent a questionnaire to practicing plastic surgeons in the United States and Canada. He determined that repair of the cleft lip should be done between 6 and 12 weeks after birth. A sample of 500 consecutive cleft lip repairs were studied over a 15 year period. The post-operative complication rate was five times higher if the following criteria were not met: Surgery delayed until the child weighed 10 pounds; hemoglobin at least 10 gm; and white blood cell count at least 10,000/mm^3(Wilhelmsen and Musgrave 1966; Fonseca et al. 2000).

Timing of Surgery

Lip surgery may be recommended in the first twenty-four hours of life. The earlier operation offered no

greater risk than repairing the lip several weeks later (Cannon 1967). In a study of one-hundred consecutive cleft lip repairs, half of the patients had lip repair within the first 3 weeks of life and the other half had surgery after 3 weeks. Long term follow up found no difference in the need for revisions between the two groups of patients (Weatherley et al.).

Types of Lip Repair

The Millard rotation-advancement lip repair, Tennison-Randall triangular flap lip repair, Davies Z-plasty, modified Z plasty, and Nakajima-Yoshimura straight-line lip repair were compared by Lazarus. The outcomes of all five methods were similar. The Millard lip repair is the most commonly used technique used today (Lazarus et al. 1998).

Lip repair in a unilateral cleft lip and alveolus patient will cause more remodeling in the anterior nasal spine compared with those, also with a unilateral cleft lip, that did not undergo surgery. The maxillary growth in patients with only a cleft palate who underwent surgery did not differ from those who did not undergo surgery for their cleft palate. However, those with both a cleft lip and palate who underwent surgery had

significant maxillary retrusion compared to those who did not have surgery for their cleft lip and palate (Normando et al. 1992).

Palatal Repair

The purpose of palatal surgery is to provide a competent velopharyngeal seal and prevent air release from the nasal cavity during speech. The production of normal speech is the most important objective in the repair of the soft palate. The Veau-Wardill-Kilner V-Y pushback procedure uses a mucoperiosteal flap. This is the pushback procedure in common use today. This procedure is used to lengthen the soft palate (Kilner 1937; Wardill 1937). In wide clefts, releasing the tensor palatine muscle toward the midline may be needed if the quantity of muscular tissue is inadequate. If the soft palate is short, articulation with the pharyngeal wall is impossible. When both sides of the cleft need to be brought together and lengthened, the W-Y push-back procedure (Wardill) and U shaped push back procedure (Dorrance and Brown) are used (Fonseca et al. 2000).

The Langenback procedure also has been shown to produce the same results in terms of speech. The von Langenback procedure utilizes a bipedicled flap and

minimal periosteal stripping to avoid maxillary growth inhibition. This procedure does not lengthen the soft palate. Both the W-Y push back and the von Langenback procedures are widely used today (Palmer et al. 1969; Weintraub 1981). The earlier the palate is repaired, the better the speech results (Dorf 1982). Kremenak found evidence to support maxillary deficiency in patients after palatal surgery (Kremenak et al. 1967). Ross (1987) reported evidence of scar tissue restricting growth of the maxilla, palatine bones, and the pterygoid plates of the sphenoid.

Multi-Center Studies

Goslon Yardstick

The original Goslon Yardstick was developed at the Hospital for Sick Children in London, England based on 22 of 30 models were ranked in the five categories. Forty-five percent of the patients were in Groups 1, 2 and 3 and 55% were in Groups of 4 and 5 (Mars et al. 1987). The patients had not received orthodontic treatment except for correction of an anterior crossbite in the early mixed dentition. Five categories were formed by 22 models. Eight models did not represent a definite group and were excluded from the sample (Mars et al. 1987). The

sagittal jaw relationships of each set of models were classified according to the examiner's clinical perception. First, the overjet is examined. A reverse overjet of 3-5 mm indicates this case would belong to Group 4. Group 4 represents a retrognathic maxilla and/or prognathic mandible. The criteria for Groups 1, 2, 3, and 5 sagittal jaw relationships were not discussed (Mars et al. 1987). After the model was categorized in groups from 1 to 5 for the saggital using the assessment, the vertical and transverse assessments placed the models in the next higher group that increase the group by one. The second consideration is the vertical assessment. The group, in which the model is placed, is not affected by a deep bite, but an open bite placed the models in the next higher group. The transverse assessment increased the group number by one if a posterior crossbite is present that can not be corrected by orthodontics alone (Mars et al. 1987).

A search of the literature found most cleft lip and palate studies using the Goslon Yardstick Index, used the weighted kappa statistical test to determine reliability and reproducibility (Chan et al. 2003; Noverraz et al. 1993; Hathorn et al. 1996; Morris et al. 2000; Pigott et

al. 2002). Reliability was also assessed using the Spearman Rank Correlation (Mars et al. 1987, Hathorn et al. 1996). Weighted kappa statistics take into account the extent of agreement between examiners whereas the unweighted kappa does not weight for closeness and only count an agreement if they agree. Both kappa statistics take into account the percent due to chance alone (Landis and Koch 1977).

The weighted kappa has been reported as a more useful statistical tool when the number of categories is small such as the 5 point ordinal scale of the Goslon Yardstick. Unweighted kappa values are always less than weighted kappa scores as found in this study (Table 3) (Landis and Koch 1977). In order to report a very good level of reliability, the previous publications reported the weighted kappa which considers the extent of agreement (see Table 3)(Chan et al. 2003; Noverraz et al., 1993; Hathorn et al. 1996; Morris et al. 2000; Pigott et al. 2002), rather than the unweighted kappa statistics that only counts complete agreement.

Examiners would score the models twice with one month between the first and second assessment (Noverraz et al. 1993). In other studies, examiners would score the

models two weeks between assessments (Pigott et al. 2002). The inter-examiner weighted kappa scores in this study ranged from 0.46-0.65. This range indicates a moderate to good agreement among the examiners (see Table 3). The intra-examiner scores ranged from 0.69-0.89 indicating good to very good agreement. Other studies have shown inter-examiner and intra-examiner weighted kappa scores from 0.78-0.85 (Morris et al. 2000). The European six centers study used five examiners. Weighted kappa scores ranged from 0.58-0.71. These scores were considered moderate to good agreement and similar to the agreement in this study. Other studies also used weighted kappa scores reporting inter-examiner and intra-examiner scores above 0.70 (Chan et al. 2003; Noverraz et al. 1993; Hathorn et al. 1996).

Another methodological approach we deem worthy of discussion is the statistical approach. A literature review of studies using the Goslon Yardstick found two studies used parametric analyses (Chan et al. 2003; Shaw et al. 1992). However, because the Goslon Yardstick records five discrete values as opposed to continuous, non-parametric tests were performed by Morris and coworkers (2003).

A study in Northern Ireland in 2004 did a comparison of one stage vs two stage palate repairs. The Northern Irish children with unilateral cleft lip and palate were compared to those treated with the Oslo protocol. The Northern Irish children were treated with the Skoog lip repair between 3-6 months, and the one stage Wardill-Kilner repair of the hard and soft palate between 12-18 months. The Veau-Wardill-Kilner procedure was found to allow satisfactory long-term mid-facial growth (Choudhary et al. 2003). The Oslo protocol used a Millard lip repair and vomer flap hard palate repair at 3-6 months followed by a von Langenbeck soft palate closure at 12-18 months. Dental arch relationships were assessed using the Goslon ranking system. The Northern Irish group had a greater number of Goslon scores that were 4 and 5, indicating unfavorable arch relationships (Johnston et al. 2004).

The Children's Memorial Hospital Cleft Palate Clinic in Chicago, Illinois performed primary alveolar bone grafting with definitive lip repair at age 4-6 months and one stage palatal repair by eighteen months. The treatment protocol for patients in Lancaster only had definitive triangular-flap lip repair at 3 months of age. The hard palate was repaired at twelve months with a

vomer flap. The soft palate was completed at eighteen months with intra-velar veloplasty and either simple midline closure with V-Y lengthening without primary bone grafting. Neither group used pre-orthopedics to align the upper arch. Primary bone grafting was thought to result in a more retrusive maxilla than a sample from the Lancaster Cleft Palate Clinic (Trotman et al. 1996). Primary bone grafting has been shown not to influence growth in patients treated before twelve months (Rosenstein et al. 2003).

Growth of the maxilla differs with different types of palatal surgeries. The modified V-Y closure, von Langenbeck and modified, medial von Langenbeck procedures were used in a study to evaluate maxillary growth. Minimizing periosteal undermining during hard palate closure using the modified, medial von Langenbeck procedure appeared to result in better maxillary growth. The mean dentoalveolar relationship Goslon Yardstick group in a sample of 32 unilateral cleft lip and palate patients was 3.59 (Pigott et al. 2002).

Dahl compared one layer vs two layer palato-vomerplasty. Danish patients were treated with Tennison lip repair and a Veau two-layer palato-vomerplasty of the

hard palate at two months. The soft palate repair is completed by 24 months using a Wardill push-back procedure. The Norwegian sample had the lip repair at five to six months of age by a Millard technique. The hard palate was closed by a one layer vomerplasty at the same time as the lip closure. The soft palate was closed at thiry-six months by a modified von Langerback procedure. The two layer palato-vomerplasty was found to be more traumatic causing less growth of the maxilla (Dahl et al. 1981).

Delaying repair of the hard palate until 12-14 yrs was proposed by Schweckendiek to improve growth of the maxilla. The soft palate was repaired at an early age to encourage proper speech function (Schweckendiek 1951; 1978). Bardach found poor speech results among Schweckendiek's patients (Bardach et al. 1984). Rohrich also found delay of the hard palate repair produced poor speech results and increase incidence of palatal fistulas (Rohrich et al. 1996).

The timing of palatal surgery was evaluated between the Bristol group and the Oslo group. The Bristol group soft palate surgery was repaired before 6 months which required more revisions. The Oslo group was repaired at

18 months. Maxillary growth was found to be poorer in the Bristol group. Transverse development of the maxilla was not considered in this study due to the limited information of the lateral cephalographs which was used for comparison (Roberts et al. 1996).

The Yorkshire sample had one-third of the cleft sample assigned to Goslon Yardstick Groups 1 and 2 (favorable results), another third to Group 3 (borderline results), and the remaining third to Group 4 and 5 (poor/very poor results) (Morris et al. 2000). Another study compared patients that used the Lathum appliance: 16% in Goslon Yardstick groups 1 and 2, 47% in Group 3 and 37% in groups 4 and 5; to patients that did not use the appliance: patients in Groups 1 and 2, less borderline Goslon Yardstick Groups of 3 and more patients in Groups 4 and 5 (Chan et al. 2003). The Goslon Yardstick was developed at the Hospital for Sick Children in London, England (Mars et al. 1987). Twenty-two models were ranked in the five categories. Forty-one percent of the patients were in Goslon Yardstick Groups 1, 2 or 3; while 15% of the models were in Goslon Yardstick Group 3 and 55% in Group of 4 and 5 (Mars et al. 1987). Hathorn

et al. (1996) studied a sample in Frenshey, England. Fifty-six percent of the patients were in Groups 4 and 5.

The Golson Yardstick gives a general outcome assessment of the relationship of the dental arches. Multi-center studies provide a means to compare the dental arch relationship differences among the different centers. This investigation will determine if differences exist in dental arch relationships (favorable vs. unfavorable) after primary surgery for children with unilateral cleft lip and palate between two craniofacial centers and to consider the reliability and validity of the Goslon Yardstick.

STATEMENT OF PURPOSE

The purpose of this study was to examine the reliability and validity of the Goslon Yardstick and to determine if differences exist in dental arch relationships (favorable vs. unfavorable) after primary surgery for children with unilateral cleft lip and palate between two craniofacial centers.

AN EVALUATION OF DENTAL ARCH RELATIONSHIPS AMONG CHILDREN WITH UNILATERAL CLEFT LIP AND PALATE TREATED AT TWO DIFFERENT CENTERS

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Abstract

Objective: To determine if differences exist in dental arch relationships (favorable vs. unfavorable) after primary surgery for children with unilateral cleft lip and palate between two craniofacial centers and to consider the reliability and validity of the Goslon Yardstick.

Design: Cross-sectional study

Setting: Two major cleft-craniofacial centers: Center 1 provided two separate samples; Sample A and Sample B; Center 2 provided one sample, Sample C.

Patients: Non-syndromic unilateral cleft lip and palate children (total n = 83).

Interventions: Several protocols were used to repair a cleft lip and palate.

Main outcome measure: The dental arch relationships were categorized using the Goslon Yardstick. Favorable inter-arch relationships were Groups 1, 2, and 3 and unfavorable arch relationships were Groups 4 or 5. Odds ratios were calculated to quantify the association between unfavorable dental arch relationship and samples. Kruskal-Wallis statistics, Mann-Whitney statistics tested for differences in the dental arch relationship among the three samples.

Results: The prevalence of unfavorable arch relationships was found for 22%, 11%, and 28% for Samples A, B, and C

respectively. There was no statistically significant association between the samples and unfavorable outcome ($p > 0.05$). Sample A and B showed a statistically significant lower mean rank Goslon Group compared with Sample C ($p \leq 0.05$).

Conclusions: The Goslon Yardstick gives a general outcome assessment of the relationship of the dental arches. Even though a statistically significant higher prevalence of unfavorable outcomes was found in Sample C ($p < 0.05$), we cannot conclude that an unfavorable outcome is more likely in any one sample.

KEY WORDS: Cleft lip and palate, multi-center research, treatment outcome, dental arch relationships, Goslon Yardstick.

Introduction

There are many craniofacial anomalies and all exhibit variation in their effect on craniofacial morphology. One of the most common craniofacial birth defect is unilateral cleft lip and palate. The incidence of cleft lip and palate is one in 500 to 1000 live births (Moreno et al., 2004). Orthodontic treatment of the cleft lip and palate child is challenging. Many patients with this craniofacial anomaly experience significant reductions in maxillary development. Therefore, it would be helpful to identify factors that influence future dental arch

relationships. Primary surgery to correct a cleft lip and palate contributes to maxillary deficiency (Ross, 1987). The Eurocleft project surveyed 201 centers and identified 194 different protocols. Children underwent one to four operations following one of 17 different sequences. Lip repair was performed sometime between birth and 12 months of age. The primary and secondary palate was repaired sometime between birth and 13 years. Many different surgical procedures were reported (Shaw et al., 2000). Factors contributing to arch discrepancies include tissue deficiency and displacement, abnormal nasal septum and abnormal muscle function. Retrospective studies comparing two groups within a center run the risk of sampling biases related to gradual changes in experience and/or technique when the data are collected over long time periods while inter-center studies can provide a sufficient number of patients to compare surgical outcomes such as speech, esthetics, and growth.

Multi-center studies allow for direct comparisons of components of the treatment protocol and of the effects of primary surgeries on subsequent jaw growth (Shaw et al., 1992). It is difficult to collect data at multiple centers in a standardized unbiased format, due to non-standardized data on potential explanatory factors, different levels of severity of cases, etc. Some centers may have a higher percentage of more severe patients. The Goslon (Great Ormond Street, London, and

Oslo, Norway) Yardstick is a clinical tool used to evaluate the effects of primary surgery on the outcome of treatment. The Goslon Yardstick qualitatively assesses dental arch relationships in three planes of space in patients with a cleft lip and palate (Mars et al., 1987). In a six-center international study in Europe, the Goslon Yardstick was used to assess favorable outcomes and demonstrated the importance, among other things, of having an organized, central and uniform treatment protocol (Shaw et al., 1992).

The Goslon Yardstick is intended to measure the severity of the malocclusion and difficulty of correcting it, but does not consider generalized crowding and irregularity. Antero-posterior arch relationships, vertical labial segment relationships; and transverse relationships are considered. The yardstick was developed at the Hospital for Sick Children at Great Ormond Street in London.

Mars and coworkers stated "The Yardstick can be applied reliably only by those trained in its use with the original cases and who have undertaken the categorization of a calibration series" (Mars et al., 1987).

The reliability of this measure has been reported to be consistent at different times (Chan et al., 2003; Shaw et al., 1992; Nollet et al., 2005; Leonard et al., 1998; Molsted et al., 2005; Noverraz et al., 1993). The results were found to be

reproducible, repeatable, and consistent in a study of four orthodontists ranking the models twice, one week apart. Reliability was measured by Spearman's rank correlation coefficient and the Wilcoxon matched paired signed rank test. Intra and inter-examiner agreement was high (Mars et al., 1987).

The purpose of this study was to evaluate the reliability and validity of the Goslon Yardstick and to determine if differences exist in dental arch relationships (favorable vs. unfavorable) after primary surgery for children with unilateral cleft lip and palate between two craniofacial centers.

Material and Methods

Study population

The Case Western Reserve University Institutional Review Board (IRB) approved the study. Sample A (n = 46) and Sample B (n = 19) were two independent samples provided by Center 1 that met the following criteria: 1. Non syndromic, complete unilateral cleft of the lip and palate; 2. All primary surgeries were performed at one center and by one surgeon; 3. Maxillary central incisors present; 4. No orthodontic, orthopedic or orthognathic intervention 5. Consecutively treated.

In Sample C (n = 18), which was provided by Center 2, identical criteria were used. However, patients were identified using lists of active and inactive from two orthodontic offices

and one university orthodontic clinic with all primary surgeries performed during 1985-1996.

Main Outcome

Three examiners were calibrated in the use of the Goslon Yardstick. Models from both centers were assigned a number by an investigator blinded to the identity of the patient and/or treatment centers. The relationship of the dental arches was categorized into 5 groups using the Goslon Yardstick. One investigator calibrated all three examiners to categorize each model into one of five groups: 1 - excellent; 2 - good; 3 - fair; 4 - poor; and 5 - very poor. Each examiner independently examined each set of models. The sagittal jaw relationship of each set of models was classified first. Group one represents prognathic maxilla and/or retrognathic mandible. Group 2 represents prognathic maxilla and/or retrognathic mandible with an anterior openbite or severe posterior crossbite.

Group 3 represents a prognathic maxilla with a posterior crossbite/ openbite or represents orthognathic sagittal jaw base relationships with incisors end to end or dentally compensated. Group 4 is represented by a retrognathic maxilla and/or prognathic mandible or may be represented by an orthognathic sagittal jaw base relationship with an anterior openbite or severe skeletal posterior crossbite. Group 5 had greater than

5mm of reverse overjet in which the maxilla was retrognathic and/or the mandible prognathic (Mars et al., 1987).

In addition to the assessment of the inter-arch relationship using Goslon Groups, the dental arch relationship was dichotomized as favorable or unfavorable outcome. A favorable dental arch relationship has been stated as Groups 1, 2, and 3 because a good orthodontic result is expected without surgery. An unfavorable dental arch relationship has been identified as Group 4 would only need surgery if growth was unfavorable and Group 5 would require orthognathic surgery (Mars et al, 1987; Morris et al., 2000; Nollet et al., 2005; Shaw et al., 1992; Chan et al., 2003; Molsted et al., 2005; Johnston et al., 2004; Choudhary et al., 2003; Pigott et al., 2002; Leonard et al., 1998).

Reliability

Intra and inter-examiner ratings were compared to check for reliability. Unweighted and weighted kappa statistics were used to assess intra and inter-examiner agreement. Intra and inter-examiner reliability tests were performed using Statistical Analysis Software (SAS 9.0). Weighted kappa scores over 0.80 were considered "almost perfect", 0.61 - 0.80 were "substantial", 0.40 - 0.60 "moderate" reliability, and 0.40 - 0.21 were considered to have "poor" agreement (see Table 1) (Landis and Koch, 1977).

Properties of an Ideal Index

The Goslon Yardstick was assessed using the following properties for an ideal index: ability to measure what it is intended to measure (validity); ability to consistently measure at different times (reliability); the criteria should be clear and unambiguous, with mutually exclusive categories; must be amenable to statistical analysis; and able to detect clinically-relevant but small shifts in the condition and acceptable to the patient (Burt and Eklund, 2005).

Statistical Analyses

The average Goslon Yardstick Group was calculated for each model based on two ratings assigned by each examiner. The Kruskal-Wallis statistics compared the mean rank of the three samples. The Mann-Whitney Statistic was used to test for differences between two samples (SPSS for Windows V6.1). Normality tests were done using Statistical Analysis Software (SAS version 9.01). Epi Info 6.04 was used to calculate the unadjusted odds ratio (OR) and 95% Confidence Interval to quantify the association between the samples and unfavorable outcome.

Results

The 46 patients from Sample A had a mean age of 8.7 years with 28 (61%) males and 18 (39%) females. The 19 patients from

Sample B had a mean age of 9.0 years with 11 (58%) male and 8 (42%) females. The 18 subjects from Sample C had a mean age of 9.0 years and included 13 (72%) male children and 5 (28%) female (see Table 2).

Intra-examiner reliability using weighted kappa statistics was considered substantial to almost perfect with scores ranging from 0.69 to 0.90 and unweighted kappa's 0.50 - 0.85. The inter-examiner reliability was moderate to substantial agreement between the examiners based on weighted kappa scores from 0.46 to 0.65 and poor to substantial unweighted agreement (see Table 3).

The distribution of the Goslon Yardstick Groups for each sample is shown in Table 4. Among the patients from Sample A 78% had favorable dental arch relationship and 22% had an unfavorable dental arch relationship. Among the patients from Sample B 89% had a favorable dental arch relationship and only 11% had an unfavorable dental arch relationship. In Sample C, 72% of the patients had a favorable dental arch relationship and 28% had an unfavorable dental arch relationship.

The Kruskal-Wallis Statistic found a statistically significant difference among the three samples ($p < 0.05$). The Mann-Whitney test found no difference in the mean Goslon Group rank between Sample A and B ($p > 0.05$). There was a significant

difference in the mean Goslon Group ranks between Sample A and C ($p<0.05$) and between Sample B and C ($p<0.05$).

In Table 5, the odds ratio and 95% CI for Sample A vs. Sample B was 2.36 (0.41 - 17.57). The lack of statistical significance can be explained by a lack of power due to a small sample size. With a prevalence of 11% unfavorable dental arch relationship in patients in Sample B, a sample size of approximately 100 from each sample would be needed to detect a statistically significant OR of 2.5 (Strom, 1994). The odds ratio and 95% CI for Sample C was 3.27 (0.44 to 29.45) more likely to have an unfavorable dental arch relationship. This was not statistically significant due to insufficient power caused by the limited sample size. It is estimated that a sample of 52 patients from each sample would be needed to detect an OR = 3.25 (Strom, 1994). Sample C vs. Sample A had an odds ratio and 95% CI of 1.38 (0.33 - 5.62) with 22% of the patients from Sample A having an unfavorable dental arch relationship, more than 950 patients would be needed from each sample to detect an OR = 1.50. Thus, there was no statistically significant association between the samples and an unfavorable dental arch relationship but clinically relevant trends may be identified.

Discussion

An outcome assessment program leads to improved clinical care and therefore benefits the patient. Although patient

treatment protocols varied between the three samples we can not conclude that any sample was more or less likely to have an unfavorable dental arch relationship. Sample A repaired the lip at 3 months with a triangular flap. The hard palate was repaired at 12 months with a vomer flap. The soft palate was completed at 18 months with intra-velar veloplasty and simple midline closure with V-Y lengthening. Sample B used a passive molding plate between 7 weeks and 14 months. Lip adhesion was preformed at 7 weeks and lip repair using the Millard technique was performed at 14 months. Soft and hard palate repair was completed by 14 months using the Wardill Pushback procedure. Sample C used a passive molding plate in half of the patients and lip repair at 3 months of age. The soft palate and hard palate repair was performed with either the Furlow Z plasty or Wardill Pushback procedure at a mean time of 18 months. Four surgeons were involved in the surgeries over a 10 year period.

Two potential explanations for the inability to find statistical significant association between the sample and an unfavorable dental arch relationship are: 1) insufficient power and small sample size, 2) the possibility of the Goslon Yardstick not meeting the criteria for an ideal index. Based on the original study that developed the Goslon Yardstick (Mars et al., 1987) there is an indication that this index is less than

ideal because of the lack of clarity, simplicity, objectivity, sensitivity and concerns about the validity and reliability.

The original Goslon Yardstick was developed at the Hospital for Sick Children in London, England based on ranking 22 of 30 models into one of five categories. 45% of the patients had a favorable dental arch relationship (Groups 1, 2, and 3) and 55% had an unfavorable dental arch relationship (Groups 4 and 5) (Mars et al., 1987). Eight models did not represent a definite Group and were excluded from the sample (Mars et al., 1987) indicating a concerns regarding the ideal requirements of an Index namely; clarity, simplicity, objectivity and sensitivity. The fact that 8 of 30 models (27%) were excluded may help explain our low intra and inter-examiner reliability. The eight models excluded in the original study may be similar to the models that the three examiners in this study could not agree upon. An example that exemplifies the limitations of the Index follows.

Models are examined to categorize the saggital relationships by assessment of the overjet. The models are categorized from 1 to 5. The vertical and transverse assessments could place the models into the next higher numbered group (that would indicate a poor dental arch relationship). The vertical assessment only takes into account the overbite. An openbite would place the models in the next higher group. The transverse

assessment increased the group number by one if a posterior crossbite is present that can not be corrected by orthodontics alone (Mars et al., 1987). Therefore, a Goslon Group of 4 could be a child with a prognathic mandible and retrusive maxilla or a patient with an orthognathic maxilla and mandible with a severe posterior crossbite that could not be treated with orthodontics alone. According to the criteria of the Goslon Yardstick, both patients' dental arch relationships would be considered unfavorable. However, the second patient could have a favorable dental arch relationship because the posterior crossbite could be corrected with a fixed rapid palatal expander. Similarly, an openbite would place a patient with an orthognathic maxilla and mandible into Group 4. This would be considered an unfavorable dental arch relationship. However, if the patient's maxillary incisors are not erupted fully, then with time the overbite will improve. By waiting several months this patient's dental arch relationship would be considered favorable. We therefore suggest careful evaluation of conclusions from studies employing the Goslon Yardstick.

We also had concerns about the reliability of the Goslon Yardstick. A search of the literature found most cleft lip and palate studies using the Goslon Yardstick index, used the weighted kappa statistical test to determine reliability and reproducibility (Chan et al., 2003; Noverraz et al., 1993;

Hathorn et al., 1996; Morris et al., 2000; Nollet et al., 2005; Pigott et al., 2002; Sandy et al., 1998; Molsted et al., 2005). Reliability was also assessed using the Spearman Rank Correlation (Mars et al., 1987, Hathorn et al., 1996; Nollet et al., 2005). Weighted kappa statistics take into account the extent of agreement between examiners whereas the unweighted kappa does not weight for closeness and only count an agreement if they agree. Both kappa statistics take into account the percent due to chance alone (Landis and Koch, 1977).

The weighted kappa has been reported as a more useful statistical tool when the number of categories is small such as the 5 point ordinal scale of the Goslon Yardstick (Landis and Koch, 1977). Unweighted kappa values are always less than weighted kappa scores as found in this study (Table 3) (Landis and Koch, 1977). We remain concerned that the weighting may mask the limitations of the less than ideal Goslon Yardstick index, so we have also reported the unweighted kappa (Table 3). Most previous publications have reported the weighted kappa which considers the extent of agreement (see Table 3) (Chan et al., 2003; Noverraz et al., 1993; Hathorn et al., 1996; Morris et al., 2000; Pigott et al., 2002; Molsted et al., 2005), which results in a higher than the true level of agreement.

Another methodological approach we deem worthy of discussion is the statistical approach. A literature review of

studies using the Goslon Yardstick found several studies used parametric analyses (Chan et al., 2003; Shaw et al., 1992; Nollet et al., 2005; Leonard et al., 1998; Molsted et al., 2005; Noverraz et al., 1993). However, because the Goslon Yardstick records five discrete values as opposed to continuous, we performed non-parametric as did two separate studies by Morris and Johnston (Morris et al., 2003; Morris et al., 2000; Johnston et al., 2004). Because of the frequent reporting of parametric analysis in studies using the Goslon Yardstick we re-ran our analysis using parametric tests and found that our conclusions remained the same.

Figure 1 depicts our three sample dental arch relationships and their distribution of Goslon Yardstick Group ratings compared with other craniofacial centers. The results of Sample C were similar with other studies whereas Samples A and B appear to have a different distribution. West Yorkshire, England had a similar distribution to Sample C in our study (Morris et al., 2000) with 70% having Goslon Yardstick Groups 1, 2 and 3 (favorable results) and 30% to Groups 4 and 5 (poor/very poor results) (Morris et al., 2000).

In another study, 37% of the patients treated with the Latham appliance were considered unfavorable while 48% who did not use the appliance were considered to have an unfavorable dental arch relationship (Chan et al., 2003). Another study in

Frenshey, England found 56% of the patients had an unfavorable dental arch relationship (Hathorn et al., 1996). Figure 2 shows the Goslon Yardstick Group distribution among the six international centers in Europe study (Shaw et al., 1992). Samples A and B in our study had similar dental arch relationships to Centers A, B, and E in the six center international study, while Sample C in our study had similar dental arch relationships to Centers F, C, and D in the six center international study.

Conclusion

While the Goslon Yardstick gives a general assessment of the relationship of the dental arches, to the best of our knowledge, data are lacking to support the contention that Groups 1-3 will be non-surgical and groups 4 and 5 will need surgery. Even though a statistically significant higher prevalence of unfavorable dental arch relationships was found in Sample C ($p<0.05$), one cannot conclude that an unfavorable dental arch relationship will be more likely in any one sample.

Further research is needed to investigate the validity of using the Goslon Groups to define favorable (non surgical) vs. unfavorable (need surgery) dental arch relationships and to further assess its limitations.

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Table 1. Interpretation of Kappa Statistics (Landis and Koch, 1977)

Value of Kappa	Strength of Agreement
<0.00	Poor
0.00 – 0.20	Slight
0.21 – 0.40	Fair
0.41 – 0.60	Moderate
0.61 – 0.80	Substantial
0.81 – 1.00	Almost Perfect

Table 2. Descriptive Summary

	n	Mean Age (yr)	Age Range (yr)	Male n (%)	Female n (%)
Sample A	46	8.7	8.0 - 10.0	28 (61%)	18 (39%)
Sample B	19	9.0	7.6 – 12.0	11 (58%)	8 (42%)
Sample C	18	9.0	6.3 – 14.0	13 (72%)	5 (28%)

Table 3. Intra-examiner and Inter-examiner Reliability

Examiners	Weighted Kappa Coefficient: Weighted Kappa	Standard Error	95% Confidence Limit	Unweighted Kappa Coefficient: Kappa	Standard Error	95% Confidence Limit
		Intra-examiner reliability			Intra-examiner reliability	
MP1 vs. MP2	0.73	0.05	0.64 - 0.83	0.57	0.07	0.43 - 0.71
JC1 vs. JC2	0.69	0.05	0.60 - 0.78	0.50	0.07	0.37 - 0.64
WF1 vs. WF2	0.90	0.03	0.83 - 0.96	0.85	0.05	0.75 - 0.94
		Inter-examiner reliability			Inter-examiner reliability	
MP1 vs. JC1	0.65	0.05	0.56 - 0.75	0.43	0.07	0.29 - 0.57
MP1 vs. WF1	0.60	0.06	0.50 - 0.71	0.39	0.07	0.25 - 0.54
JC1 vs. WF1	0.64	0.05	0.55 - 0.74	0.43	0.07	0.30 - 0.57
MP2 vs. JC2	0.46	0.06	0.35 - 0.58	0.23	0.07	0.10 - 0.36
MP2 vs. WF2	0.52	0.06	0.41 - 0.63	0.27	0.07	0.13 - 0.41
JC2 vs. WF2	0.55	0.06	0.43 - 0.66	0.36	0.07	0.22 - 0.50

Table 4. Goslon Group Distribution

	Goslon 1	Goslon 2	Goslon 3	Goslon 4	Goslon 5
Sample A	13(28.26%)	15(32.61%)	8(17.39%)	10(21.74%)	0(0.00%)
Sample B	2(10.53%)	8(42.11%)	7(36.84%)	2(10.52%)	0(0.00%)
Sample C	0(0.00%)	3(16.67%)	10(55.56%)	4(22.22%)	1(5.55%)

Table 5. Association of Samples with Unfavorable Outcome

	Unfavorable Outcome Goslon Group 4 and 5	Favorable Outcome Goslon Group 1 ,2, 3	Odds Ratio with C.I.	P value Fishers exact*
Sample A	10	36	2.36 (0.41 – 17.57)	P = 0.2893
Sample B	2	17		P = 0.4839
Sample C	5	13	3.27 (0.44 – 29.45)	P = 0.1805
Sample B	2	17		P = 0.2319
Sample C	5	13	1.38 (0.33 – 5.62)	P = 0.6081
Sample A	10	36		P = 0.7441

*Fishers exact test is used when sample sizes are less than 5.

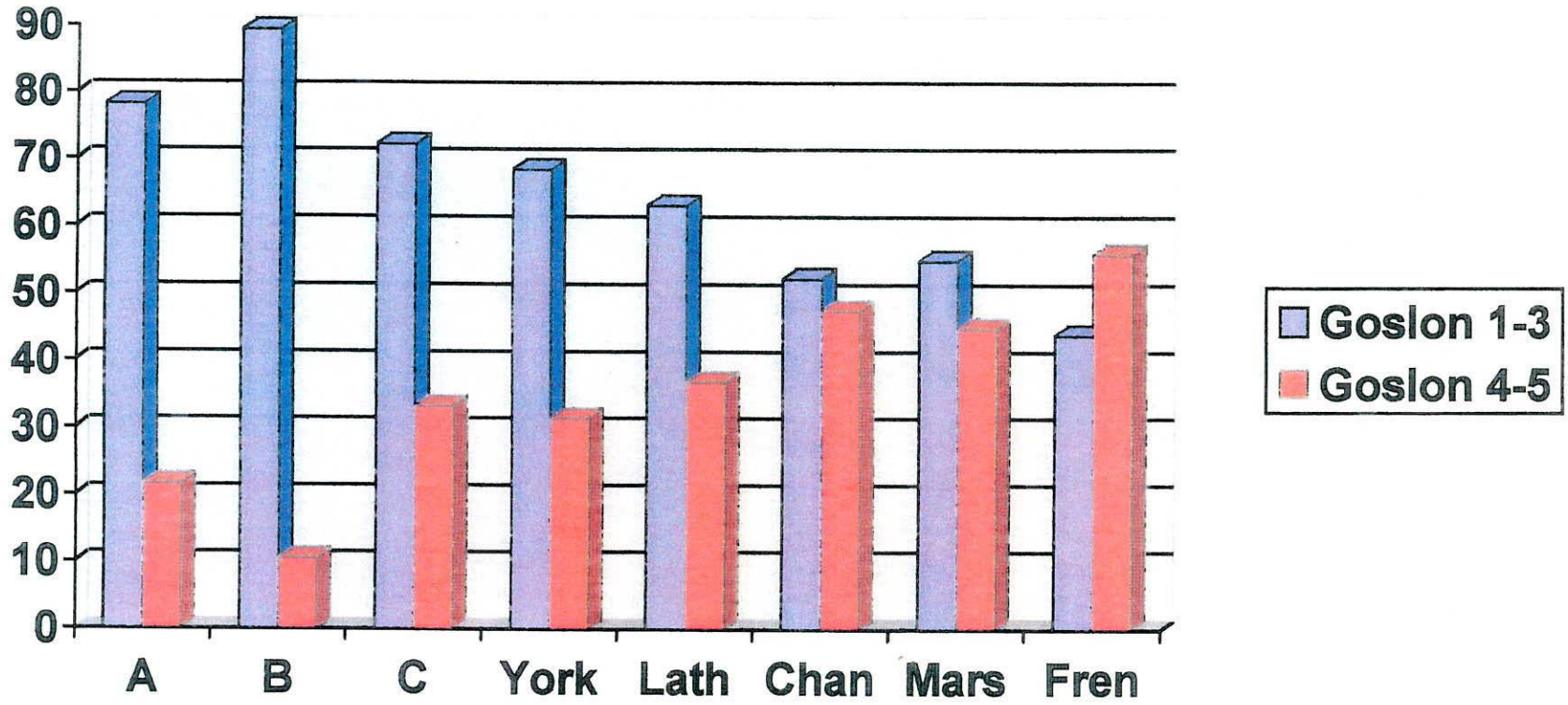


Fig. 1. Bar graph shows Goslon group distributions among our three samples (Sample A, B, and C) and five other samples in the literature. Morris studied patients in Yorkshire, England (York) (Morris et al., 2000); Chan studied a sample that was treated with the Lathum (Lath) appliance with a sample that was not treated with the Latham appliance (Chan) (Chan et al., 2003); Mars categorized a sample at Hospital for Sick Children in London, England (Mars) (Mars et al., 1987); Hathron studied children with an unilateral cleft lip and palate in Frenchey, England (Fren) (Hathron et al., 1996).

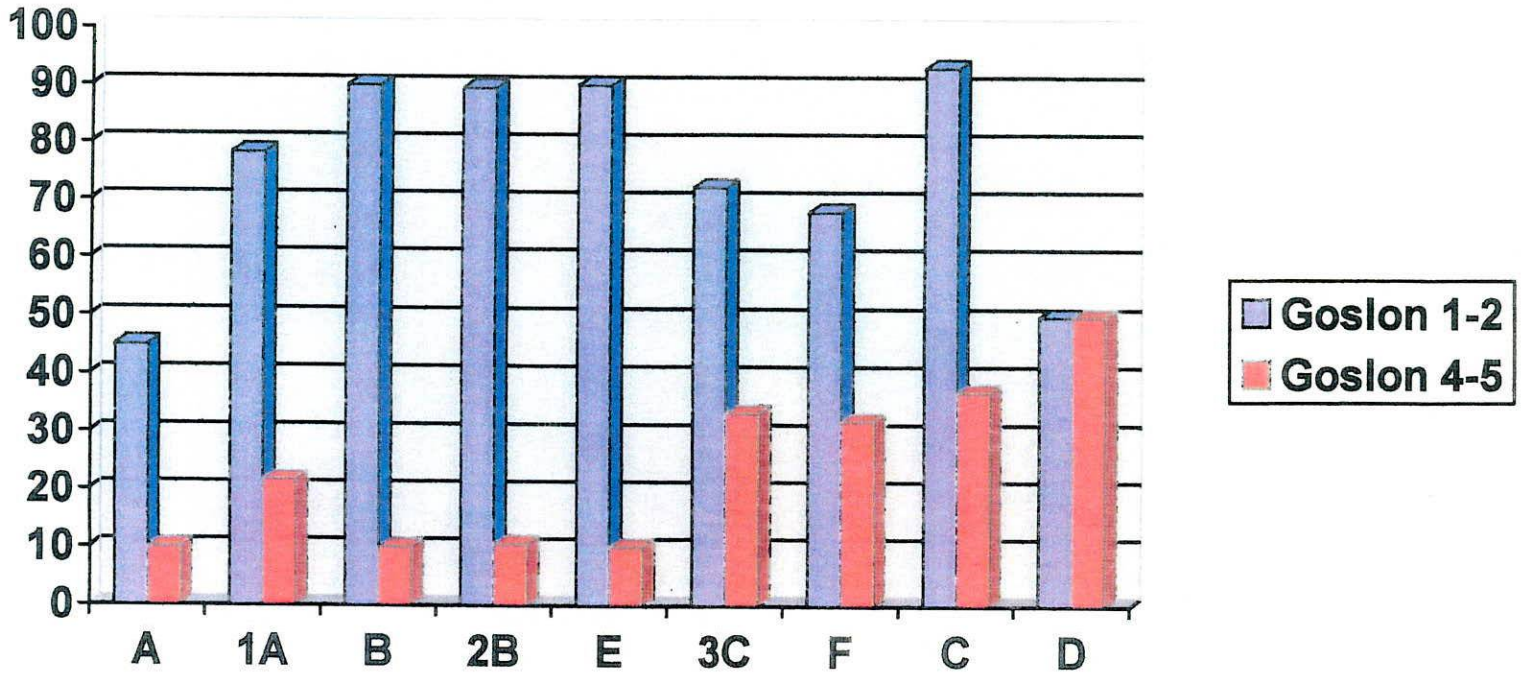


Figure 2. shows the Goslon Yardstick Group distribution among the six international centers in Europe study (Samples A, B, C, D, E, and F) (Shaw et al., 1992). Our samples are represented by Samples 1A, 2B, and 3C. 1A and 2B in our study had similar outcomes to Centers A, B, and E in the six center international study, while Sample 3C in our study had similar outcomes to Centers F, C, and D in the six center international study.

Raw Data

Center	MP	JC	WF	MP2	JC2	WF2	mean
1	1	1	1	1	1	1	1.00
1	1	1	2	1	2	1	1.33
1	1	1	2	1	1	2	1.33
1	1	1	1	1	2	1	1.17
1	2	2	2	1	3	2	2.00
1	4	4	4	4	4	4	4.00
1	3	4	4	3	4	4	3.67
1	2	2	2	1	2	2	1.83
1	2	3	3	2	3	3	2.67
1	2	1	2	1	3	2	1.83
1	3	4	4	4	4	4	3.83
1	1	1	1	1	1	1	1.00
1	1	1	1	1	1	2	1.17
1	1	1	2	1	2	2	1.50
1	3	3	4	3	4	4	3.50
1	1	2	1	1	1	2	1.33
1	2	3	3	2	3	2	2.50
1	1	3	2	2	3	2	2.17
1	1	2	3	2	3	3	2.33
1	1	1	1	1	1	2	1.17
1	1	1	1	1	1	2	1.17
1	1	1	2	1	2	2	1.50
1	2	2	2	1	2	2	1.83
1	1	3	2	2	3	2	2.17
1	3	3	2	2	3	3	2.67
1	3	2	3	2	3	3	2.67
1	4	4	3	3	5	3	3.67
1	4	4	4	4	4	4	4.00
1	4	5	4	4	4	4	4.17
1	3	4	4	3	4	4	3.67
1	1	1	2	1	3	2	1.67
1	2	1	2	1	2	2	1.67
1	3	4	4	4	4	4	3.83
1	3	4	3	3	4	3	3.33
1	1	1	1	1	1	1	1.00
1	4	5	4	4	5	4	4.30
1	2	1	2	1	2	2	1.67
1	2	1	1	1	2	1	1.33
1	3	3	3	3	3	3	3.00
1	1	1	2	1	1	2	1.33
1	2	2	1	2	2	2	1.83
1	1	2	2	1	2	2	1.67
1	2	2	2	1	3	2	2.00
1	1	1	1	1	2	1	1.17
1	2	3	2	1	3	2	2.17

Raw Data (continued)

Center	MP	JC	WF	MP2	JC2	WF2	mean
1	2	3	3	2	4	3	2.83
2	2	3	3	2	3	3	2.67
2	4	4	3	4	4	3	3.67
2	2	2	2	2	2	2	2.00
2	1	2	1	2	2	1	1.50
2	3	3	2	2	4	2	2.67
2	1	1	2	2	1	2	1.50
2	1	2	2	1	2	2	1.67
2	1	2	2	1	1	2	1.50
2	2	3	3	2	3	3	2.67
2	3	2	3	3	2	3	2.67
2	4	4	4	4	4	4	4.00
2	3	4	3	3	3	3	3.16
2	1	1	2	1	1	2	1.33
2	2	2	2	2	3	2	2.17
2	3	3	2	3	3	2	2.67
2	2	2	2	1	3	2	2.00
2	1	2	2	1	2	2	1.67
2	2	2	3	2	3	3	2.50
2	1	1	1	1	1	1	1.00
3	4	4	4	4	4	4	4.00
3	1	1	2	1	2	2	1.50
3	3	4	4	2	3	4	3.33
3	3	4	2	3	4	2	3.00
3	3	3	3	3	4	3	3.17
3	3	3	3	3	3	3	3.00
3	4	5	4	4	4	4	4.17
3	5	5	5	5	5	5	5.00
3	3	3	3	1	2	3	2.50
3	2	2	2	1	2	2	1.83
3	2	3	3	2	3	3	2.67
3	4	4	4	4	4	4	4.00
3	1	2	2	1	3	2	1.83
3	3	4	3	3	4	3	3.33
3	3	3	3	3	3	3	3.00
3	3	3	3	2	4	3	3.00
3	3	4	3	3	4	2	3.17
3	4	4	4	3	4	4	3.83

Kruskal-Wallis Test

Ranks

	Center	N	Mean Rank
Goslon	1	46	37.14
	2	19	40.58
	3	18	55.92
	Total	83	

Test Statistics(a,b)

	Goslon
Chi-Square	8.542
df	2
Asymp. Sig.	0.014

a Kruskal Wallis Test
b Grouping Variable: Center

Mann-Whitney Test

Center A and B

Ranks

	Center	N	Mean Rank	Sum of Ranks
Goslon	0	46	32.00	1472.00
	1	19	35.42	673.00
	Total	65		

Test Statistics(a)

	Goslon
Mann-Whitney U	391.000
Wilcoxon W	1472.000
Z	-.690
Asymp. Sig. (2-tailed)	0.490

a Grouping Variable: Center

Center A and C

Ranks

	Center A/C	N	Mean Rank	Sum of Ranks
Goslon	0	46	28.64	1317.50
	1	18	42.36	762.50
	Total	64		

Test Statistics(a)

	Goslon
Mann-Whitney U	236.500
Wilcoxon W	1317.500
Z	-2.738
Asymp. Sig. (2-tailed)	0.006

a Grouping Variable: Center A/C

Mann-Whitney Test
Center B and C

Ranks

	Center B/C	N	Mean Rank	Sum of Ranks
Goslon	0	19	15.16	288.00
	1	18	23.06	415.00
	Total	37		

Test Statistics(b)

	Goslon
Mann-Whitney U	98.000
Wilcoxon W	288.000
Z	-2.374
Asymp. Sig. (2-tailed)	0.018
Exact Sig. [2*(1-tailed Sig.)]	0.026(a)

a Not corrected for ties.
b Grouping Variable: Center B/C

ANOVA

Descriptives

	N	Mean	Std. Deviation	Std. Error	95% Confidence Interval for Mean		Minimum	Maximum
					Lower Bound	Upper Bound		
1	46	2.33	1.117	0.165	1.99	2.66	1	4
2	19	2.47	0.841	0.193	2.07	2.88	1	4
3	18	3.17	0.786	0.185	2.78	3.56	2	5
Total	83	2.54	1.039	0.114	2.32	2.77	1	5

Test of Homogeneity of Variances

Levene Statistic	df1	df2	Sig.
4.235	2	80	0.018

ANOVA

	Sum of Squares	df	Mean Square	F	Sig.
Between Groups	09.257	2	4.628	4.667	0.012
Within Groups	79.346	80	0.992		
Total	88.602	82			

Normality Test for Goslon Yardstick
21:22 Sunday, October 30, 2005

The UNIVARIATE Procedure

Moments

N	83	Sum Weights	83
Mean	2.44578313	Sum Observations	203
Std Deviation	1.00743834	Variance	1.014932
Skewness	0.39369949	Kurtosis	-0.9007848
Uncorrected SS	579.7184	Corrected SS	83.2244241
Coeff Variation	41.1908285	Std Error Mean	0.11058072

Basic Statistical Measures

Location		Variability	
Mean	2.445783	Std Deviation	1.00744
Median	2.330000	Variance	1.01493
Mode	2.670000	Range	4.00000
Interquartile Range	1.67000		

Tests for Location: Mu0=0

Test	-Statistic-		-----p Value------	
Student's t	t	22.11763	Pr > |t|	<.0001
Sign	M	41.5	Pr >= |M|	<.0001
Signed Rank	S	1743	Pr >= |S|	<.0001

Tests for Normality

Test	--Statistic---		-----p Value------	
Shapiro-Wilk	W	0.944603	Pr < W	0.0014
Kolmogorov-Smirnov	D	0.127069	Pr > D	<0.0100
Cramer-von Mises	W-Sq	0.221047	Pr > W-Sq	<0.0050
Anderson-Darling	A-Sq	1.436553	Pr > A-Sq	<0.0050

TABLE 6.--Sample Sizes for Cross Sectional Studies

	Odds Ratio to be detected								
Prevalence in unexposed group	1.5	2.0	2.5	3.0	3.5	4.0	5.0	7.5	10.0
0.10	2508	200	100	62	43	32	20	8	4
0.15	1566	121	59	36	24	17	10	-	-
0.20	1095	82	39	15	15	10	5	-	-
0.25	812	58	27	10	9	6	-	-	-

*Alpha = 0.05 (two-tailed), beta = 0.20 (power = 80%), control: exposed ratio = 1:1. The sample size listed is the number of subjects needed in the exposed group. An equivalent number would be included in the control group (Strom, 1994).

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From CWRU Institutional Review Board <cwru-irb@case.edu>
Sent Wednesday, December 15, 2004 3:32 pm
To mgh4@cwru.edu
Cc jsg@cwru.edu , mwp8@case.edu
Bcc
Subject Notice of Approval for 20041019

Case Western Reserve University
Institutional Review Board
NOTICE OF APPROVAL

Responsible Investigator: **Mark Haus**
Department: **Dental Clinic Ortho**
IRB Protocol #: **20041019**
Title: ***An Evaluation of Dental Arch Relationships among Children with Unilateral Cleft Lip and Palate Treated at Two Different Centers***
Co-Investigator: **Michael Paulus**

Approval Date: **December 13, 2004**
Continuing Review Deadline: **November 28, 2005**
Expiration Date: **December 12, 2005**
The Institutional Review Board (IRB) has APPROVED the above new protocol through the full review process.

It has been determined that this study involves minimal risk, and that the following vulnerable populations will be involved: minors.

As an investigator of human subjects, your responsibilities include the following (see full description of responsibilities at our website):

1. Report all adverse events and unanticipated problems involving human subjects to the IRB Office, located in the Office of Research Compliance (ORC), within three (3) business days of your knowledge of the occurrence.
2. Provide the IRB with a complete Continuing Review form (available at the CWRU IRB Web Pages, or from the ORC) by the continuing review deadline noted above, and when the study is terminated.
3. Discontinue all work pertaining to this protocol if a continuing review approval is not finalized by the expiration date noted above.
4. Submit all proposed changes to the protocol to the IRB, and receive approval from the IRB, before implementation of the change.
5. Keep all research data and original consent documents in your possession for at least three (3) years after the study is terminated.

Questions? Please visit our website: http://ora.ra.cwru.edu/orc_humansubjects_CWRU_IRB.asp

OR
contact our administrative office...

Isabel Sanchez, IRB Administrator
216.368.6993
Maureen Dore-Arshenovitz, IRB Assistant
216.368.6925
Fax: 216.368.3737

CASE Institutional Review Board
Office of Research Compliance
Sears Building 657

Lancaster Cleft Palate Clinic

November 18, 2004

CASE
School of Dental Medicine
Department of Orthodontics
Attention: Dr. Michael Paulas
10900 Euclid Ave.
Cleveland, Ohio 44106-4905

To Whom It May Concern:

I am writing to confirm the agreement for carrying out a joint clinical outcomes research project between the Lancaster Cleft Palate Clinic and the Orthodontic Program at CASE. Dr. Michael Paulas has submitted to me a description of his proposed research entitled, "An Evaluation of Dental Arch Relationships Among Children with Unilateral Cleft Lip and Palate Treated at Two Different Centers: Inter-Center Retrospective Study". As Director of the Lancaster Cleft Palate Clinic and the Head of the Orthodontic and Research Programs here, I approve of this research project and will make available to Dr. Paulas all necessary patient records and treatment information as necessary for successful completion of the project. In so doing, once the sample has been identified, the clinical records will be blinded with no patient identifiers available for use or display in the research. I will also provide for Dr. Paulas all necessary staff and administrative support possible in his attempt to gather these data and carry out the comparison described in his protocol.

Finally, it is understood that any presentation or publication of this material would similarly exclude any patient identifiers in order to protect the privacy of our patients here and acknowledge the Lancaster Cleft Palate Clinic and its staff when appropriate for their assistance in completion of this worthwhile venture.

I hope this information is of use to you. Please contact me if I can provide you with any additional confirmation.

Sincerely,

Ross E. Long, Jr., D.M.D., M.S., PhD.
Director, Orthodontist, Cleft-Craniofacial Program

REL/tjt



CASE

SCHOOL OF DENTAL MEDICINE

School of Dental Medicine

Case Western Reserve University
10900 Euclid Avenue
Cleveland, Ohio 44106-4905

Phone 216-368-3200
Fax 216-368-3204
www.case.edu

Letter of Cooperation

Date: November 18, 2004

Dr. Michael Paulus has described his proposed research to me, titled, **"An evaluation of dental arch relationships among children with unilateral cleft lip and palate treated at two different centers."**

As chairman of the orthodontic clinic at Case Western Reserve University in Cleveland, Ohio, I approve of this research to occur with our employees and administrative records.

Sincerely,

Mark G Hans, DDS, MS

Mark G. Hans, DDS, MSD

Letter Of Cooperation

Date: November 10, 2004

Dr. Michael Paulus has described his proposed research to me, titled, "**An Evaluation of Dental Arch Relationships among Children with Unilateral Cleft Lip and Palate Treated at Two Different Centers.**"

As owner of an orthodontic private practice in Berea, Ohio, I approve of this research to occur with our employees and administrative records.

Sincerely,

Mark G. Hans, DDS, MS

Board Certified
Specialist in Orthondontics

Associate Professor and Chairman
Department of Orthodontics
Case Western Reserve University
(216) 368-4649

347 Front Street
Berea, Ohio 44017
(440) 243-6724

Orthodontics
HOWARD E. KOSSOFF, D.D.S., M.S., INC.

Letter of Cooperation

November 23, 2004

Dr. Michael Paulus has described his proposed research to me, titled, "An Evaluation of Dental Arch Relationships among Children with Unilateral Cleft Lip and Palate Treated at Two Different Centers".

As owner of an Orthodontic private practice in Solon, Ohio, I approve of this research to occur with our employees and administrative records.

Sincerely,

Howard E. Kossoff, DDS, MS

Howard E. Kossoff, DDS MS

Member
American Association of Orthodontists

32901 Station Street • Solon, OH 44139 • Phone (440) 349-2454
9140 Lake Shore Blvd. • Mentor, OH 44060 • Phone (440) 257-0707

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