# International Society of Craniofacial Surgery
# VIIIth International Congress

*Taipei, Taiwan*
*October 31 ~ November 3, 1999*

The Elite Gathering
Song Emperor Hui Chung (1082-1135)
*National Palace Museum, Taipei, Taiwan, R.O.C.*

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INTERNATIONAL SOCIETY OF CRANIO FACIAL SURGERY
THE CRANIO FACIAL CHAPTER OF THE INTERNATIONAL CONFEDERATION
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Cleft Palate-Craniofacial Association

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## President's Welcome Message

On behalf of the Organizing Committee, and the Taiwanese people, I would like to warmly welcome you to the 8th International Congress of the International Society of Craniofacial Surgeons. On this special occasion, I hope you will be able to richly benefit from this experience in many ways.

First and foremost, we are fortunate to have attracted an abundance of excellent submissions to this congress, which has allowed the selection of a fine scientific and clinical program. I am therefore confident there will be innumerable opportunities for communication between people who care a great deal about craniofacial surgery. This is truly an international delegation, composed of 269 participants from 26 countries around the world. Merely bringing together such a varied and accomplished group of leaders in our field provides the most important ingredient for a provocative and successful exchange of ideas. It is my sincere hope that we will be able to use this forum as a springboard for furthering our understanding of the problems which challenge both our patients and us.

I am proud to have the privilege of hosting this meeting in Taiwan. Our country is blessed with a rich tradition, and filled with natural splendor. Within very short reach of our meeting venue, one can choose to view snow-capped peaks plunging into a blue Pacific, or to relax on a tropical beach. I hope you will take some time to sample these natural resources. Most of all, I am pleased that all of our guests will be able to experience our Chinese hospitality and culture, which is the heart of our society.

Yu-Ray Chen, MD
President and Chairman

# Abstract

The abstracts are numbered in the same manner

as they are on the program.

sagital suture also!

# THE DIFFERENTIAL EXPRESSION OF FGFR GENES IN HUMAN CRANIAL DEVELOPMENT AND HUMAN CRANIOSYNOSTOTIC SUTURES – TOWARDS A PATHOGENESIS OF SYNDROMIC CRANIOFACIAL DYSOSTOSIS

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**Introduction:**
Craniosynostosis is the premature ossification of the fibrous sutures of the cranial vault. Primary craniosynostosis presents most severely within a congenital craniofacial dysostosis syndrome, many of which share overlapping craniofacial and extracranial features, requiring multidisciplinary clinical care. Consistent mutations in the fibroblast growth factor receptor (FGFR) genes FGFR1, FGFR2 and FGFR3 are causally implicated in several craniosynostosis phenotypes, including those of Apert, Crouzon, and Pfeiffer.

**Materials & Methods:**
The expression of FGFR genes, selected ligands and Stat 1, a downstream target of FGFR signalling, has been accessed in cranial sections from Apert and normal human embryos; and a range of human craniosynostotic sutures, using the techniques of *in situ* hybridisation and immunohistochemistry.

**Results:**
FGFR1, 2 and 3, are differentially expressed in human craniosynostotic sutures, periosteum and osteoid bone, compared to the expression of *osteonectin* mRNA. Sutural cells, osteoblasts and osteocytes differentially express FGFR proteins and mRNAs in consistent sequence in "fusing" sagittal, coronal, and lambdoid sutures in the presence of FGF2, but not FGF4 or FGF7. Temporally and spatially specific FGFR gene expression also occurs in the human embryonic cranial skeleton and meninges, in domains overlapping those of FGF2 and FGF4. Stat 1 is activated in Apert skeletogenic tissues, compared to control.

**Conclusions:**
Clinical FGFR mutations are considered to act constitutively to promote proliferation and premature maturation of skeletogenic tissues. We therefore propose a model of mutant FGFR function in craniosynostosis, based upon our novel results indicating differential FGF and FGFR gene expression in the meninges and sutural primordia during human development, human postnatal craniosynostotic sutures, and the activation of the transcription factor Stat 1 in the Apert phenotype.

## TENSION-INDUCED RELEASE OF FGF-2 IN NEONATAL RAT CRANIAL SUTURES: A SELF-ORGANIZING MECHANISM IN CRANIAL DEVELOPMENT

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**Purpose:**
How does the neurocranium expand in perfect concert with the expanding brain? How does tension from a distractor cause increased osteogenesis? The objective of this study is to test components of the general hypothesis that tension across the cranial suture can cause cellular strain which in turn changes the permeability of the cell membrane. This leads to a local release of FGF-2 which set off the well-known down stream effects of FGF-2 including bone formation and thus reducing the local strain.

**Methods:**
10 Coronal sutures from I day-old Lewis rats were subjected to 5 minutes of 0.6 N tension in PBS. The Conditioned PBS was assayed for FGF-2 level by Slot Blot using monoclonal antibody against FGF-2. 30 sutures were also similarly strained in PBS containing fluorescein-conjugated dextran (MW 10 KD), normally unable to enter the cell due to the large molecular size. Laser confocal microscopy was employed to analyze the pattern of fluorescence and emission spectrophotometry was used for quantitative analysis.

**Results:**
5 minutes of 0.6 N tension caused significant FGF-2 release by slot blot ($p=0.003$, $N=10$ sutures). There was entry of large molecular weight tracers confirming transient disruption in plasma membrane integrity. The pattern of fluorescence suggest a Gaussian distribution in the orientation of cell-to-ECM adhesion.

**Conclusion:**
This series of experiments confirm that tension across cranial suture causes plasma membrane disruption and that FGF-2 is released. The pattern of cells undergoing maximum strain appears to follow the Gaussian distribution thus potentially leaving a group of unstrained cells to respond to the released FGF-2. These data support the above hypothesis.



## FGF4 OR FGF2 INJECTION NEAR THE DEVELOPING CORONAL SUTURE ENHANCES APOPTOSIS AND COLLAGEN TYPE I PRODUCTION SIMULTANEOUSLY

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**Introduction:**
Growth at the coronal suture takes place through proliferation of osteogenic stem cells, which express the fibroblast growth factor receptor (FGFR) 2. Normally, a balance is kept between rate of proliferation and bone differentiation of these cells. Differentiation involves, among others, downregulation of FGFR2, production of osteoid including collagen I, and mineralization of the bone matrix. A number of cells undergo apoptosis during these differentiation steps. This study aimed at elucidating the pathogenesis of FGFR-associated craniosynostosis, with particular reference to apoptosis and bone differentiation, in a mouse model mimicing the increased FGFR signaling resulting from the genetic mutations.

**Materials and Methods:**
Unilateral injection of 1.0 $\mu$-l fibroblast growth factor (FGF) 4 or 2 (10-100 $\mu$g/ml) near the coronal suture in mouse embryos (E14-E18) through ex utero surgery. Survival period from 7.5 to 48 hours, sectioning. Detection of apoptosis (Annexin V-biotin, and Tunel), differentiation (anti-collagen type 1), their double labeling (Annexin V-FITC / anti-collagen type I-TRITC), and mineralization (Alizarin Red, Von Kossa).

**Results:**
Simultaneous increase in apoptosis and bone differentiation within the suture, as soon as 7.5 hours postinjection, concerning two separate cell populations. The osteogenic fronts of the frontal and parietal bones were in closer approximity, with marked mineralization of both bones all the way to the periphery. Enhancement of apoptosis proceeded this accelerated mineralization. Within the control suture no apoptotic or collagen type I producing cells were detected, while mineralization was limited to the central part of the bone plates, leaving the peripheral rim unmineralized.

**Conclusions:**
FGF4 and 2 induce a simultaneous increase in apoptosis and differentiation within the developing coronal suture. These two cell populations could well be derived from one cell through so-called asymmetrical cell division. Apoptosis seems to be functionally involved in mineralization. In FGFR2-linked craniosynostosis, the upregulated receptor appears to force the FGFR2 expressing cells to undergo apoptosis and bone differentiation prematurely, at the expense of proliferation. This causes the loss of the stem cell population, and thus arrest of growth within the suture, with subsequent suture synostosis.

## DIFFERENTIAL DISTRIBUTIONS OF TRANSFORMING GROWTH FACTOR-BETA ISOFORMS BETA1 AND BETA3 AND RECEPTORS I AND II BETWEEN FETAL AND ADULT DERMAL FIBROBLASTS

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**Introduction:**
Transforming growth factor-beta (TGF-β) isoforms TGF-β1 and TGF-β3 have opposite effects during repair. TGF-β1 induces scar, yet TGF-β3 decreases scar formation. Both fetal and adult fibroblasts produce and respond to TGF-β. The purpose of this study is to determine the differences in production and distribution of TGF-β1, TGF-β3, transforming growth factor-beta receptor-I (TβR-I), and TβR-II between fetal and adult dermal fibroblasts.

**Methods:**
Fetal (14.5 and 18.5 day gestational age, term = 21 days), and adult Sprague-Dawley rat fibroblasts were established in primary cell culture. Third and fourth passage cells were immunostained with antibody to either TGF-β1, TGF-β3, TβR-I, TβR-II. Staining intensity was semiquantitated with the Image Pro Program®.

**Results:**
TGF-β1 and TGF-β3 were each present on both fetal and adult fibroblasts. However, the expression level of the isoforms was distinctly different. The fetal fibroblasts stained more intensely for TGF-β3 compared to the adult fibroblasts. The adult fibroblasts stained more intensely for TGF-β1 compared to fetal fibroblasts. No significant differences between fetal and adult fibroblast TβR-I and TβR-II expression level was observed. However, significant differences in the distribution of these receptors was found. In adult fibroblasts TβR-I had a granular pattern, which localized in the peri-nuclear area. TβR-II localized in a filamentous pattern, similar to the cytoskeletal structure. The fetal fibroblasts had no clear differences in TβR-I and TβR-II localization, with each showing a diffuse pattern on the cell.

**Conclusions:**
We conclude that 1) fetal fibroblasts produce relatively more TGF-β3 than TGF-β1; 2) adult fibroblasts produce relatively more TGF-β1 than TGF-β3; 3) fetal fibroblasts do not have discrete spatial localization of TβR-I and TβR-II; 4) adult fibroblasts have discrete spatial localization of TβR-I and TβR-II. We hypothesize that differences in TGF-β isoform and receptor production may contribute to differences in wound repair outcome between the fetus and adult.

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## GROWTH FACTORS IN IMMUNOHISTOCHEMISTRY OF CALVARIAL SUTURES

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The etiology of craniosynostosis remains unknown and continues to frustrate craniofadal surgeons. Several theories have been postulated including a possible chemical factor. Over the last decade growth factors have been an area of interest in wounds and other conditions. They have also shown to influence bone metabolism. Some of the growth factors have previously been investigated. The role of other growth factors needs to be investigated.

**PURPOSE:**
1. To determine the growth factor profiles in calvarial sutures.
2. To determine the difference in growth factor immunolocalization between fused and open sutures.

**METHOD:**
Biopsies of fused and open sutures were taken during surgery. These samples were then processed and immunohistochemistry performed on them. Some of the growth and other factors tested for were TGF $\beta_1$, TGF $\beta_2$, TGF $\beta_3$, FGF, IL-3, IGF, MMP1, MMP3 and anti-Annexin.

**RESULTS:**
The analysis of the growth factor profiles of the open and fused sutures will be presented.

**References:**
1. Immunolocalization of Transforming Growth Factor pi, P2, and pa and Insulin-Like Growth Factor I in Premature Cranial Suture Fusion. Douglas a Roth, M.D., Leslie I Gold, PhD., Victor K M Han, M.D., Joseph G McCarthy, M.D., Joanne J Sung, B.S., Jefferey H Wishoff, M.D., Michael T Longaker, M.D. Plast & Recon. Surg Vol 99 : 2, 1997
2. Bradley J P, Levine J P, Sung J J et al. The Biology of Cranial Suture Fusion : Growth Factors and Dural Cranial Interaction. Craniofadal Surgery : State of the Art Symposium. New York 1996
3. Roth DA, McCarthy JG, HanVKetal. Studies in cranial suture biology: Immunolocalization ofTGF β1, β2 and β3 and IGF-I in premature cranial suture fusion in humans. Plast Reconstr Surgery

## ROLE OF TGF-B SIGNALING IN THE REGULATION OF PROGRAMMED CRANIAL SUTURE FUSION

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**Introduction:**
Recent studies have implicated TGF-B growth factors in the regulation of pathologic sporadic cranial suture fusion. In addition, we have shown that these growth factors are highly expressed by the dura mater underlying fusing murine cranial sutures. The purpose of these experiments was to analyze the effects of disrupting TGF-B signaling during programmed mouse cranial suture fusion.

**Methods:**
Using recombinant DNA technology, a replication-deficient adenovirus encoding a defective TGF-B receptor (TB-R) was constructed. Infection with this virus completely blocked TGF-B signaling (all isoforms). Mouse posterior frontal sutures were harvested prior to the initiation of suture fusion (post-natal day 25), infected with vehicle, TB-R virus, or control virus (B-galactosidase; n=8 each), and placed in culture for 30 days. Sutures were harvested and analyzed histologically. A fusion score was calculated for each suture.

**Results:**
All sutures grew in size in culture. In all cases sutural tissue appeared intact without toxic effects from viral infection or organ culture. Analysis of uninfected and control infected sutures revealed suture fusion in 81.3 ±10%, and 74.5 ±9%, respectively (NS). In contrast, TB-R infected sutures exhibited suture fusion in only 38.1 ±14% (*$p<0.001$). In addition, infection with the TB-R virus resulted in a significant attenuation of anterior to posterior suture fusion with the majority of fused sections localized to anterior sections and only 20-30 % of the more posterior sections. In contrast, uninfected and control (B-Gal) infected sutures demonstrated between 65-100% suture fusion even as far as 2500 microns posteriorly.

**Discussion:**
These data strongly implicate TGF-B signaling in sutural fusion. In addition, this study demonstrates the utility of adenovirus-mediated gene transfer in preventing programmed sutural fusion. Future studies will assess the efficacy of this approach to the treatment of *in vivo* suture fusion.



## IMMUNOLOCALIZATION OF TGF-BETA ISOFORMS DURING THE REOSSIFICATION OF CALVARIAL DEFECTS

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Transforming growth factor-Betas (TGF-β) have been demonstrated to be upregulated during osteoblast function *in vitro.* Isofonns of TGF-β have also been demonstrated to be upregulated during cranial suture fusion *in vivo.* We hypothesized that spontaneous reossification of calvarial defects of less than critical size in the mature animal is also associated with upregulation of TGF-β ligands. The present study was designed 1) to evaluate the concept of a critical size defect (CSD) within the calvarium in an adult guinea pig model, and 2) to investigate the association between reossification ofcalvarial defects and TGF-β upregulation.

**Methods:**
Parietal defects 3, 5, 8, or 12 mm in diameter were made in 45 6-month-old skeletally mature guinea pigs. Animals were sacrificed after survival periods of 1, 4, 8, or 12 weeks. New bone ingrowth into the defects was measured histometrically, and ANOVA performed to test for statistical significance. Immunohistochemistry was performed to localize TGF-β1, β2 and β3.

**Results:**
After 12 weeks, all 3- and 5-mm defects were completely covered by a bone bridge, while defects remained within the 8- and 12-mm wounds. Within the 3-mm defects percent new bone formation steadily increased from a mean of 29% at I week to 93% at 12 weeks, and was significantly higher within 3-mm defects than in all larger defects at each time interval from 1 to 12 weeks ($p< .05$). There was no significant difference in percent new bone formation between defects 5mm or larger, all of which remained below a mean of 31% new bone formation by 12 weeks. Immunolocalization of TGF-β ligands demonstrated an osteogenic front in which the osteoblasts stained strongly for all isofonns of TGF-B located at the advancing bone edge of the defect as well as the endocranial side adjacent to the dura. TGF-β was upregulated in all size defects through the 8th week of survival, correlating with the period of maximum bone formation within the respective defects. There was a decrease in TGF-β activity by 12 weeks as bone formation waned.

**Conclusions:**
1) Histometric analysis provided a more accurate description of the biologic behavior of cranial defects than did the concept of the CSD.
2) Isoforms of TGF-β were upregulated during a limited "window" of time corresponding to the period of calvarial reossification, and were localized to osteoblasts within an osteogenic front at the periphery and dural surfaces of the defects.

## DEVELOPMENT OF AN *IN VITRO* RODENT MODEL OF FORCE-INDUCED CRANIOSYNOSTOSIS

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**Introduction:**
The etiology of craniosynostosis has remained elusive despice intensive scientific investigation. Recent reports at PSRC have advanced the role of soluble factors in promoting premature closure ofcalvarial sutures in a rodent model and dismissed the role of force as an etiologic factor in craniosynostosis. The purpose of this study is to develop an *in vitro* rodent model of force induced craniosynostosis and to further show that premature fusion of the sagittal suture can be induced by force alone.

**Methods:**
The sagittal sutures of post natal day 21 mouse calvaria (n=18) were harvested and cultured in 5 ml of serum free media. Utilizing a newly designed, built, and tested force delivery appliance capable of delivering controlled, cyclic mechanical loads, sagittal sutures (n=9) were subjected to 0.3 gm offeree, applied for 30 minutes each day. for a total of 14 days. A control group(n=9) were cultured and not subjected to load. Histologic specimens were then stained with hematoxylin and eosin.

**Results:**
The loading device is capable of delivering compressive loads to mouse calvaria within a tissue incubator. Sagittal sutures, a calvarial suture that does not normally fuse, showed histologic evidence of fusion in the majority of specimens analyzed. We have created fusion and/or increased osteoid production in the sagittal suture of the mouse establishing that a discrete range of compressive mechanical load is able to cause significant change in cranial suture morphogenesis.

**Conclusions:**
To our knowledge,this is the first report of the development of an in *vitro* rodent model of forced induced craniosynostosis. Premature fusion of the sagittal suture of the mouse was able to be induced by applying a compressive mechanical load. These results implicate abnormal forces in the pathophysiology of craniosynostosis. This unique in vitro mouse model should now enable us to determine the interplay between soluble factors and force transduction during suture morphogenesis.

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## A COMPREHENSIVE SCREEN FOR TWIST MUTATIONS IN SAETHRE-CHOTZEN SYNDROME IDENTIFIES A NEW MICRODELETION DISORDER OF CHROMOSOME 7.

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Saethre-Chotzen syndrome (SCS), first described in 1931, is one of the most common craniosynostosis syndromes, characteristically associated with 2/3 syndactyly and hallucal duplication. The recent discovery that heterozygous mutations in the *TWIST* gene on chromosome 7 are identified is some but not all patients with SCS, provides a useful confirmatory molecular diagnosis. These intragenic mutations lead to loss of function of the TWIST protein. This led us to investigate whether complete deletions of *TWIST* could contribute to this disorder by developing a comprehensive strategy to screen for both intragenic mutations and deletions.

We identified TWIST mutations in 8 of 10 patients with SCS and 2 of 43 craniosynostosis patients with no clear diagnosis. In addition to six intragenic mutations, our strategy revealed four complete TWIST deletions. These were characterised by a combination of analysis of polymorphic markers, fluorescence *in situ* hybridisation, Southern blotting and DNA sequencing.

Our results identify a new microdeletion disorder of chromosome 7 that contributes a significant proportion of SCS. This diagnostic strategy should achieve wide application in the molecular diagnosis of craniosynostosis. Learning difficulties were observed in the patients with large deletions suggesting that loss of genes neighbouring TWIST contributes to mental retardation.

## NORMAL INTRACRANIAL VOLUME

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Following a review of published normal intracranial volume measurements by Gault and colleagues in 1990, Lichtenberg's (1960) data have become a common reference for normal intracranial volume. However, we have found Lichtenberg's data inadequate. By measurement of the intracranial volume of 150 skeletally normal patients who had CT scans for medical indications, we have established the normal range of intracranial volume variation for a Caucasian population. Intracranial volumes were measured from the CT data using the Persona software package developed at the Australian Cranio-Facial Unit with volume determination based on measuring area in each CT slice.

Growth curves have been fitted to the data for determination of mean and variance as a function of age for each gender. Unlike Lichtenberg, who did not distinguish between males and females below the age of 2 years, we found that the male mean was significantly larger than the female mean, and that the difference is in accordance with standard head circumference measurements. In addition, our curves and standard deviations do not contain the anomalous variations of Lichtenberg that were presumably caused by small sample sizes.

The intracranial volumes of 66 patients with deformational plagiocephaly have also been measured and compared to our new CT normal. This group was of significance because the condition was at one time prone to misdiagnosis as craniosynostosis. We have found that their intracranial volume is in the normal range - if we had used Lichtenberg's normal their intracranial volume would have been found significantly larger than normal. Our finding is consistent with clinical experience. Our CT derived normal intracranial volume will be useful in assessing intracranial volume variation of patients with craniosynostosis. The dichotomy between measurement and clinical experience and practice has been a major source of concern, impeding rational thinking about growth and development of patients with craniosynostosis.



## TISSUE ENGINEERING CARTILAGE BY THE USE OF INJECTABLE POLYMER IN AUTOLOGOUS PORCINE

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**Objective:**
Tissue engineering cartilage has been successfully achieved in the nude mouse model. But in an immunocompetent animal model if the engineering cartilage can be formed is unknown. Investigation this question is important toward clinical application of engineering cartilage.

**Methods:**
Auricular cartilage was obtained from pigs. Chondrocytes were isolated and mixed with pluronic at 10, 20, 30, 40, 50, 60, $70x10^6$/ml concentrations. Chondrocyte – polymer constructs were injected as autologous implants subcutaneously into the pig's abdomen from which the cells had been isolated. Specimens were harvested and analyzed grossly and histologically after 1, 3, 6, 9, 15 weeks *in vivo*.

**Results:**
All implants had a good new cartilage formation and showed similar histological characteristics compared with original cartilage. The optimal chondrocyte density is $50x10^6$/ml, and the proper harvest time is the sixth weeks.

**Conclusion:**
The autologous chondrocyte-pluronics constructs can regenerate new cartilage in immunocompetent animal both grossly and histologically. The optimal chondrocyte density is $50x10^6$/ml and the proper harvest time is the sixth weeks. This suggest that tissue engineering cartilage can become a new method in the treatment of cartilage loss.

## COMPOSITES TISSUE ENGINEERED BONE AND CARTILAGE FOR MANDIBLE CONDYLAR PROCESS REPLACEMENT

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**Purpose:**
The purpose of this study was to engineer a composite bone and cartilage tissue in the shape of an adult human condyle by the transplantation of osteoblasts and chondrocytes using biodegradable polymers as cell delivery devices.

**Methods:**
Scaffolds in the shape of an adult human condyle was fabricated with a polyglycolic acid (PGA) mesh coated with a 1.5% solution of polylactic acid (PLA). Bovine osteoblasts and chondrocytes were isolated and cultured in vitro for three weeks. The chondrocytes were suspended in 30% (w/v) Pluronic F 127 gel and the osteoblasts in a 1.5% calcium alginate solution, both at a concentration of $5.0 \times 10^7$ cells/ml. Two milliliter aliquots of the osteoblast-calcium alginate mixture were then seeded onto each polymer fabricated in the shape of a human condyle. The articular surface of each polymer condyle was seeded with 0.3 ml of the chondrocyte-pluronic gel mixture. Each construct was implanted into a dorsal subcutaneous pocket of an athymic mouse. The study was divided into three groups. Group I consisted of animals with polymer matrices seeded with the osteoblast-calcium alginate mixture and the chondrocyte-pluronic gel mixture. Group II consisted of animals with polymer matrices seeded with osteoblasts and chondrocytes alone without calcium alginate and pluronic F127. Group III, as a control, was composed of animals with polymer matrices seeded with calcium alginate and pluronie-F127 without cells. All constructs were harvested at 12 week. Gross evaluation and histologic examination were performed.

**Results:**
In group I, all constructs exhibited a morphology nearly identical to that of the initial implant and mimicked the architecture of the original human condyle. In contrast, the specimens in group II demonstrated only rudimentary three-dimensional structure with decreased size and distorted shape Histology examination using H&E and Safranin-0 staining showed organized matrix of bone in the body ofcodyle and cartilage on the articular surface of the condylar head in groups I *&11.* With distinct transitional zone between bone and cartilage. In group III, no evidence of bone and cartilage formed grossly and histologically.

**Conclusion:**
This study demonstrated that we can engineer a bone/cartilage composite tissue in a shape of human condylar process and directed bone and cartilage tissue formation with selective placement of osteoblasts and chondrocytes onto a biodegradeable polymer scaffold bone. There is a strong and stable cartilaginous-osseous interface. The composite bone/cartilage joint constructs would be applied in oral and maxillofacial surgery in the near future.



## DEFINING THE PRESENT OBSTACLES FOR CLINICAL UTILITY OF "ENGINEERED" CARTILAGE

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**Introduction:**
The concept of tissue engineering involves harvesting a small number of cells (e.g., cartilage biopsy to provide chondrocytes), multiplying them in culture, and seeding them in a bioresorbable matrix which is then transplanted into the recipient for structural or functional needs. However, success in the laboratory has been achieved when large numbers of cells from immature animals have been seeded into polymers which are then implanted into immune incompetent animals.

**Purpose:**
This paper summarizes six years of experiments examining various aspects of cells implanted and matrices selected to produce "engineered" cartilage. The summation of these experiments is to define the fundamental obstacles preventing the "engineering" of cartilage from being clinically applicable at this time.

**Materials and Methods:**
A series of experiments using nude mice and genetically defined swine were performed to determine the effect of various parameters on the engineering of cartilage. Parameters included: cell age: number of cells implanted; multiplication of cells in tissue culture, and feasibility of calcium alginate, polyethylene oxide, and fibrin glue as injectable matrices. Cartilage formation was assessed by histomorphometric analysis, collagen typing, DNA analysis, and proteoglycan production. Appropriate statistical analyses were performed on the data.

**Results:**
Cells. Mature cells and even "aged" cells can be used to engineer cartilage. The density (or number) of cells implanted must be similar to that of mature cartilage. Cells multiplied in monolayer culture dedifferentiate but are capable of producing cartilage matrix proteins after encapsulation in polymer. Matrices. Existing matrices including calcium alginate, polyethylene oxide, and fibrin glue can be utilized for percutaneous injection. Calcium alginate degrades slowly, thereby limiting cell-to-cell interaction. Polyethylene oxide can be polymerized transdermally into a specific shape. Fibrin glue degrades rapidly from the outside in, thereby resulting in contour deformation and the need for over correction.

**Conclusions:**
"Engineered" cartilage requires large numbers of healthy cells to seed matrices whose degradation properties are well controlled. The present cell culturing techniques and available matrices are not sufficiently developed to allow clinical utility at this time. These areas should be the focus of future study.

## TISSUE ENGINEERING BONE USING BONE MARROW STROMAL CELLS-THE FIRST STEP TOWARD CRANIOFACIAL SKELETON REPAIR

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**Purpose:**
The core of the tissue engineering is to from a small biopsy, damaging as little as possible the donor area, obtain parenchyma cells, multiply them in vitro until sufficient cells are generated to seed them with a biodegradable polymer and generate the new tissue. This study was conducted to engineer bone using bone marrow stromal cells and polymers. The ability to form bony tissue may provide a significant surgical option for nonweight-bearing bone repair.

**Materials & Methods:**
The bone marrow stromal cells were harvested from bovine shoulder joints. After 4 weeks in vitro monolayered culture, the cells were mixed with 2% alginate. Subcutaneous injection on the back of nude mice, the cell-polymer constructs were taken at different time frames (every 4 weeks). Various cellular concentrations were tested. Histology was performed for Hematoxylin-Eosin, Von Kossa, Trichrome and Safranin-O stains. Biomechanic tests were done to each sample.

**Results:**
Grossly, new bone formation was noted with shape from at second week. Ball-like formation bony tissue with increased stiffness and more whitish in color. At the end of second week, chondrogenesis starts and shows in the Safranin-O stain. Mineralized bone formation starts at eighth week was highlighted by the black color in Von Kossa silver stain. Many areas show organized concentric lamellae. Till more than 4 months the quality of engineered bone was mature both by histology. There were many osteoblasts covered the entire surface of the trabecula and osteocytes trapped by surrounding themselves with bone matrix in their lacunae. The implants consisted of larger masses of lamella bone associated with small amounts of new woven bone. Implanted polymer pieces were mostly resorbed but their remnants were still seen in some areas. Osteoclastic resorption and bone remodeling were observed by this time. This indicates progressive deposition of matrix, suggesting the osteoblasts viability' and frinction. The increased modulus and decreased permeability were statistically significant in the biomechanic test.

**Conclusions:**
This study demonstrates that engineer a bony tissue by the construct formed by the cell and biodegradable polymer. The unique ultra-pure formula of this alginate will enable future study in the autologous animal models. Current study attempts to engineer bone for nonweight-bearing skeletal defect. This is the first step toward craniofacial bony reconstruction.



## THE BIOLOGY OF DISTRACTION OSTEOGENESIS: HISTOLOGIC ANALYSIS OF MANDIBULAR REGENERATE BONE IN HUMANS

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**Introduction:**
While distraction osteogenesis (D.O.) has become a widely accepted method of reconstruction for mandibular hypoplasia, the basic biology of D.O. in membranous bone remains largely unknown. The aim of this study was to characterize the histology of human regenerate mandibular bone produced by D.O.

**Methods:**
Five patients with mandibular hypoplasia (2 unilateral and 3 bilateral) underwent mandibular distraction. An intraoral expansion device was placed following a mandibular osteotomy and secured with two screws in each side of the osteotomy. The devices were actuated at a rate of 1mm per day (range of distraction: 8-17mm) using a Microtract internal fixation device. The devices were left in situ allowing the bony regenerate to consolidate and removed at a second procedure: excisional biospies were taken of bone adjacent to and within the regenerate. Hematoxylin-eosin stained sections were analyzed with light micros copy, for the presence of woven and lamellar bone, osteoid, fibroplasia, fibrotic scar, and osteoblasts.

**Results:**
Samples harvested adjacent to areas of consolidation in three patients were composed almost entirely of hypocellular tissue with minimal osteoblast proliferation and fibroplasia, consistent with mature lamellar bone. In these patients, examination of regenerate areas revealed the presence of fibroplasia and osteoblastic proliferation; in one patient, irregular cement lines were observed, suggesting ongoing destruction and remodeling. Interestingly, in one patient, primarily immature woven bone was observed in both the area immediately adjacent to and within the consolidated bone. In another patient, intraoperative findings of soft, cartilaginous like consolidated tissue was confirmed histologically with fibrocollagenous tissue fibroplasia resembling scar.

**Conclusions:**
These preliminary studies demonstrate that the regenerate often consists of a greater amount of immature bone, which may allow for molding in the consolidation period. Future studies may confirm these findings and further characterize the mechanism of regenerate bone formation.

## A MATHEMATICAL ANALYSIS OF TENSILE FORCES INVOLVED IN MANDIBULAR DISTRACTION OSTEOGENESIS

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**Background:**
Distraction osteogenesis of the mandible remains still in its infancy since the mechanobiological processes involved are not fully understood. In this preliminary study, we analyze the tensile forces applied to the mandible and the changes in this force throughout the distraction procedure via a specially designed distractor coupled to a force transducer device.

**Materials and Methods:**
An external distractor was specially designed with an Entran (Fairfield, N.J.) ELH-401 load cell force transducer and tensile forces were recorded digitally using an Entran MM45 sensor monitor. The device was placed externally on five beagles (3-5 months old). After 3 days of external fixation, each mandible was distracted at a rate of I mm/day for 10 days. Force measurements were made for two hours following each distraction. All animals were sacrificed within 2 days after completion of distraction and the mandibles underwent routine histology. The force values were then plotted as a function of time and analyzed based on our uniaxial elastoplastic model of mandibular distraction proposed earlier.

**Results and Discussion:**
During the post-operative latent period of 3 days, the vector force on the mandible experienced an average gradual increase of 0.541bs/day, most likely a reflection of the healing of soft tissues, periosteum, and marrow. Thereafter, each mm of distraction resulted in a successively higher spike in tensile force, ranging from 0.75 to 7 Ibs. Furthermore, the returning baseline levels of force steadily increased throughout the distraction process, from 2.15 to 6.1 lbs. Our previous uniaxial model of mandibular distraction states that with an optimal rate of distraction $(dd_{opt}/dt)$, the growth of bone along the distraction axis ($g$) should not exceed the distracted distance $(d)$. It can further be inferred that for $g$ to equal $d$, the returning baseline force $(f)$, as well as the force required to distract 1mm, must remain relatively constant (i.e. $df/dt=0$) after each distraction. We conclude that despite the adequate evidence of bone formation at the periphery of the radiolucent zone using a rate of I mm/day, the steady increase in returning baseline force, as well as the increase in the required force to distract each successive millimeter suggest that the mandible is capable of withstanding higher rates of distraction since $g$ seems to have exceeded $d$. In our proposed model, slightly higher distraction rates would shift the balance between distracted distance $(d)$ and bone growth $(g)$ so that at $dd_{opt}/dt$, distraction would precisely follow the growth rate of the mandible. We are currently testing higher distraction rates.

**Conclusions:**
Thus far, mandibular distraction has been studied empirically. We believe that an analytical model is invaluable in evaluating such techniques. An optimal distraction rate would cause less patient morbidity, require less time with the mounted device, and minimize the risk of infection, damage, and socio-phsychological problems while maintaining adequate bone quality.



## DISTRACTION OSTEOGENESIS AFTER MEMBRANOUS BONE ONLAY GRAFT IN DOG MODEL

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The purpose of this project is to study the possibility of distraction osteogenesis in the membranous bone onlay graft on the mandible and to clarify the histology of bone repair during distraction osteogenesis in the membranous onlay bone in the dog model. Four canine dogs, 5 months of age at the begining of the experiment, were used for this study. The zygomatic arch was exposed in the subperiosteal plane and the full-thickness zygomatic arch was harvested to 3 centimeters in length. The lateral surface of the mandibular body was exposed in the subperiosteal plane and the membranous bone onlay graft was done with firm contact using screw. The osteotomy on the membranous bone onlay graft and underlying mandibular body was carried down at week I in dog I, week 2 in dog 2, week 3 in dog 3, and week 4 in dog 4 after membranous bone onlay graft. The external distraction device was applied to the mandibular body. Mandibular distraction was started 7 days after the operation at a rate of I mm per day for a total of 10 mm distraction for 10 days. After completion of distraction, the distraction device was left in place for 6 weeks to allow for bony consolidation of the distracted area. Radiographs were carried out 2 weeks, 4 weeks and 6 weeks of the post-distraction. Dogs were sacrificed 6 weeks after distraction. The new bone between native underlying mandibular segments was generated in the distracted zone in all dogs. The new bone between segments of the membranous bone onlay graft was not generated in dog 1, but generated in dog 2, dog 3 and dog 4. However, in the dog 2 and 3, the new bone between segments of the distracted membranous bone onlay graft presented less firmness with fibrous tissue than that of native underlying mandibular segment. Histologically, the distracted gap between segments of the membranous bone onlay graft was composed of much fibrous tissue in the central zone and activated osteoblastic cells formed new bone in the margins of the distracted gap in dog 2 and dog 3. In dog 4, there were much activated osteoblastic activities in the distracted gap and the new bone appeared nearly normal cortical bone. In conclusion, these findings suggested that distraction osteogenesis is possible in the membranous bone onlay graft.

## MANDIBULAR RIB GRAFT DISTRACTION OSTEOGENESIS: TECHNICAL PEARLS AND PITFALLS

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**Introduction:**
Costochondral grafting of the Pruzansky type III mandible has given variable results. Patients in whom growth of the neomandible is inadequate, lengthening of the rib via distraction had been advocated. This retrospective study reviews our series of patients with costochondral neomandibles who underwent distraction osteogenesis of the graft.

**Methods:**
A retrospective review identified two groups of patients. Group I consisted of individuals (N=9) who underwent costochondral rib grafting of the mandible followed by distraction osteogenesis at rate of I mm/day. Group 2 contained patients who had distraction osteogenesis without prior rib grafting (N=9). Biomechanical parameters, orthodontic treatments, and complications were examined vs. age and the quality of the rib graft.

**Results:**
Distraction osteogenesis was successfully performed on 78% of the rib graft patients (group 1) and in 100% of the group 2 individuals. All the regenerates were classified by CT as either "standard or external", with the exception of the 2 fibrous non-unions in group 1 which were classified as either "agenic" or "pillar". In group 1, the average device was expanded 23 mm (range 20-30 mm) resulting in a 5.1 mm mean increase in ramal height (Cd-Go) (range 6.2-11.4 mm): translating to a 35% distraction efficiency. Group 2 patients had an average device expansion of 22.4 mm (range 16-30 mm) which resulted in an 8.5 mm increase in ramal height (range 3.2-13.4 mm) and a calculated efficiency of 38%. The length of consolidation averaged 12.6 weeks in group 1 compared to 8.5 weeks in the standard mandibular distraction patients. The dental midline mean shift to the contralateral side was 2.5 mm in group 1 vs. 4 mm in group 2. Complex mutliplanar and transport distractions were successfully performed on grafts of adequate bone stock. All 4 patients in group 1 with tracheostomies were successfully decannulated post consolidation. Rib graft distraction complications included 2 pin tract infections, 1 hardware failure with premature pin pull out, and 2 fibrous non-unions in young patients with single, thin rib grafts. In group 2 there were no distraction failures.

**Conclusions:**
Distraction osteogenesis can be successfully performed on costochondral rib grafts of the mandible; however, the complication rate is higher than in non-rib graft patients. This rate can be reduced by performing the technique on older individuals. In addition, placement of a double nb graft or an iliac graft of sufficient volume to create a neo-mandible with greater bone stock should decrease the risk of fibrous non-union and provide a bone base of adequate size for munipulation of the distraction regenerate.



## DISTRACTION OF THE PATIENT WITH MANDIBULAR DEFICIENCY: A CLINICAL TREATMENT ALGORITHM

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In an over ten year experience with mandibular distraction, the authors have developed a comprehensive treatment program for mandibular distraction based on a clinical algorithm. Multiple variables must be considered: 1) diagnosis; 2) severity of skeletal pathology; 3) associated non-skeletal pathology; 4) age of patient; 5) functional vs. aesthetic requirements; 6) occlusal status

There are several treatment age groups:
1) The neonate - birth to six months
2) The young child - eighteen to forty eight months
3) The pediatric patient - five to twelve years
4) The adolescent - thirteen to seventeen years
5) The adult patient

The algorithm also takes into consideration alternative surgical and non-surgical treatment techniques, such as advancement osteotomies, bone grafts, and orthodontic therapy. Special attention is paid to the relative role of each and the increasing role of distraction osteogenesis in the reconstruction of the hypoplastic mandible.

## PREOPERATIVE CONTOURING OF DISTRACTION DEVICE USING MEDICAL MODELS

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Distraction osteogenesis has been employed with increased frequency over the past four years. Contouring of the plates used in the distraction can be time consuming. We have developed a protocol which utilizes medical models for the preoperative planning and contouring of plates.

Patients scheduled for distraction osteogenesis undergo a preoperative CT scan by protocol. The protocol includes a spiral acquisition axial CT, 1.0mm thickness, with a soft tissue algorithm. The CT data are converted to a readable format and the bone window segmentation performed (threshold=1250). The two dimensional views are edited appropriately and a three dimensional representation created. A model is then created from the CT data ( MIMICS, Materialise, Inc.). We have utilized a three dimensional inkjet printing device (Z410, Zcorp, Inc.) and the models are treated with a resin (Z 10, Zcorp, Inc.). The plate component of the distraction device is contoured and cut to shape before proceeding with surgery and then the plates are sterilized in the usual fashion for an implant (Modular Internal Distraction System, Stryker Leibinger, Inc.).

All patients that underwent surgery were found to have anatomy which correlated with the model. The precontoured plates fit exactly and there was no need for adjustment. There was an estimated time saving of about one hour for this portion of the procedure.

Distraction osteogenesis is undergoing continued evolution as the techniques develop. Since most pateints are young, any measures to decrease operative time are welcome. There have been great advances in three dimensional imaging over the past ten years which has improved three dimensional imaging techniques. At the same time solid model sciences have improved. The new techniques of creating solid models are faster, cheaper, and just as accurate at the resolution required. We will present our protocol for utilizing solid models for preoperative planning and plate contouring while saving operative time.



## CRANIOFACIAL DISTRACTION WITH INTERNAL DEVICES

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**Purpose:**
Since 1993, a modular system for craniofacial distraction has evolved. The advantages of the system are internal placement and the ability for intraoperative customization to permit distraction in any region of the craniofacial skeleton.

**Methods:**
A flexible cable sheaved in Teflon permits access to the distraction frame which drives titanium rigid fixation mesh that can be contoured to virtually all anatomic variants. In September 1997, the modular internal distraction system (MIDS, Leibinger) was approved by the FDA, and first tier clinical trials were initiated.

**Results:**
We have now gained sufficient experience to report our first 18 completed cases, utilizing 34 distraction devices for mandibular distraction (n=7); LeFort I (n=3); LeFort III (n=3) and monobloc with or without facial bipartition (n=5). The operative time necessary for placement of the devices ranged from 100 min. (mandibular distraction) to 337 min. (first monobloc distraction). Hospital stay ranged from 2-12 days, dependent upon the anatomic site of distraction. Patients who were admitted to the ICU stayed a mean of 2.5 days. Patients undergoing mandibular and LeFort I distraction were sent to the floor. The distraction lengths varied from 1 5-30 mm. Consolidation periods ranged from 2-3 months. Complications occurred in one patient who developed a late cable tract infection. There were no device failures. During distraction in one patient, one of the distraction cables became stiff but, ultimately, distraction was able to be carried out. The modular internal distraction system has been utilized for virtually all craniofacial applications.

**Conclusions:**
To date, we have been pleased with the easy adaptability of the plates to virtually any anatomic situation, allowing predictable and safe utilization of the distraction technique. A second-generation system is planned to permit easier explantation, bidirectional movement, mandibular widening, and removable cables of varying length.

## CRANIOFACIAL DISTRACTTON - A NEW SYSTEM

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There continues to be a need for effective, reliable calvarial expansion in syndromic craniosynostoses and in some plagiocephalies.Over the past 18 months we have been using a technique for cranial expansion by distraction combined with traditional surgery. The distraction force is applied to removable wires. Twelve patients are presented with radiographic, neurologic and photographic studies of at least six months' follow-up. Only one patient was prematurely discontinued due to pin tract infection. Results in eleven of twelve patients reveal an increase in cranial volume versus traditional technique, and a possible improvement in craniofacial shape. This new technique may permit in the future the full potential of distraction osteogenesis: [1] minimally invasive surgery, [2] measurable, sufficient force, [3] accurate and simultaneous vectors of force, and [3] the potential for simple, future repetition of distraction.



## SIMULTANEOUS MIDFACE AND FOREHEAD DISTRACTION USING MULTIPLE INTERNAL DEVICES

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We have experienced the simultaneous midface and forehead distraction using multiple internal devices in 3 cases. The cases were two Crouzon patients and a brachycephaly patient with exophthalmos.

We developed two types of the internal distraction device, one for the midface distraction and the other for the universal craniofacial distraction such as mandible, zygoma or cranium.

The osteotomies preceding both the midface and the forehead distraction were complete osteotomies. The frontal bone was kept attached to the underlying dura during the whole course of the surgery.

In the two Crouzon patients, the midface and the forehead were separately osteotomized and distracted in the different direction. The forehead was distracted forward using two universal type internal devices, and the midface was distracted in the antero-inferior direction using two midface type devices.

In the brachycephaly patient, the midface and the forehead were osteotomized in monoblock and distracted forward (the same direction) using four universal type internal devices.

## SPRING ASSISTED CRANIOFACIAL SURGERY. AN EARLY REPORT

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The clinical utilization of implantable springs in craniofacial surgery was First performed at this unit in October 1997 and is now a routinely used clinical reality whenever suitable conditions prevail. It was the intention of this study to sum up and analyze results, peri- and postoperative morbidity and dices the indications for this type of procedures. 10 surgeries in 9 patients were studied. Five of these had craniosynostosis. 4 patients had severe craniofacial dysostosis. There was no death and no postoperative infection. Operative time was lower than what would have been the case if another technique for the same diagnosis would have been used. Dissection of the dura mater from the brain could be avoided in the craniosynostosis patients and among these patients blood loss was significantly reduced as these basically underwent strip craniectomies alone with the addition of metal springs. Pre- and postoperative CT and cephalometry was used for assessment of outcome. The results in all patients were interesting enough to make spring assistance now a routinely considered adjunct in a variety of craniofacial reconstructive surgery at this unite. It was also concluded that the use of springs in craniofacial surgery not only has been virtually free of complications but also has enhanced results with less surgical trauma.



## IMPLANTABLE SPRINGS IN RECONSTRUCTIVE SURGERY FOR CRANIOFACIAL DYSOSTOSIS

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In reconstructive surgery the utilization of various distraction procedures is well established and widely accepted . In order to explore the possibilities of achieving continues distraction over long time metal springs have been used at this center since October 1997. Among these patients 5 surgeries have been performed in 4 patients with severe craniofacial syndromes. These procedures included 2 monobloc facial advancement for airway improvement (1 Apert and 1 Pfeiffer), 2 cranioplasties in stages for a severe skull deformity (Crouzon) and 1 spectacles plasly (Pfeiffer). There was no complication. The results were encouraging even dramatic. The patients will be presented and future implications will be discussed based on these findings.

## A NEW GENERATION OF SUBMERGED DISTRACTION DEVICES : USE OF BIDIRECTIONAL INTRAORAL MANDIBULAR DISTRACTION IN CHILDREN

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**Purpose:**
This new generation of Intraoral Submerged Device is designed to provide the surgeon greater control in guiding deficient or misshapen mandibles. Its goal is to manipulate the callus, to obtain simultaneously the best morphological and occlusal results.

**Materials-Methods:**
After feasibility animal study achieved succesfully, clinical human use started in 1998. We report the results in 7 patients. 4 suffered from craniofacial microsomia. 2 from developmental micrognathia. One from mandibuiar hypoplasia after the TMJ ankylosis. The mean age was 11.6 years. The prototype used was manufactured by Leibinger according to our needs and ideas. The distraction length is 22 mm, or 18 mm, the angulation is 30°. The device is fixed by self drilling pins. The distraction and angulation carried out independtiy by turning one of the two rods. For the three last cases a more miniaturized prototype has been used. All the devices were placed intraorally under the mucosa. One single osteotomy was performed. The angulation was proximal and started in the last days of lengthening

**Results:**
None of the distractors led to premature removal for infections and mechanical problems. The action was to retroposition the ramus and angulate the callus. The authors show its interest for fighting against the shift of the midline, for increasing the vertical vector of distraction, and for providing more control in cases of bilateral mandibular augmentation. Real effects of bidirectional Intraoral Devices are demonstrated. Improvements regarding angulation, miniaturization, activation, remain necessary.



## MIDFACE DISTRACTION WITH INTERNAL DEVICE AND TRANSFACIAL PIN

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Maxillary (midface) distraction has been carried out with external or internal devices to correct midface hypoplasia, but in most instances, it is difficult to implement them in the youngest patients. Recently, it has been suggested to combine an external distraction device with a transfacial pin (Pellerin. 1998).

We have performed an upper midface distraction in Five patients (mean age of 4 years) with a combination of an internal swivelling-axis device (Microfrance) and a transfacial zygomatic pin. The procedure was carried out immediately after Lefort III osteotomies had been performed. The implantation of the internal device in the temporal fossa was performed bilaterally. An horizontal, transfacial 2.5 mm K-wire was inserted from one zygoma to the other, and each of its end was connected to the internal distraction device.

The distraction was initiated seven days after the procedure, at a rate of 1 mm per day (range 5 - 10 days). The period of distraction was 10 to 20 days according to the length of advancement required. Correction of the mid-face was achieved in all cases and there was no infection in these small series. No dislocation of the face occured during the distraction process. The advancement was symetrically planned in 4 out of 5 cases, but in one patient it was not possible to correct an existing preoperative asymmetry despite an asymetrical rate of distraction. The devices were removed at least 3 months after implantation, with minimal incisions,

We believe this procedure can be performed early in life to correct midface hypoplasia in faciocraniosynostosis with less morbidity.

## LENGTHENING OF HYPOPLASTIC MAXILLA IN CLEFT PATIENTS USING INTERDENTAL DISTRACTION OSTEOGENESIS AND RAPID ORTHODONTIC TOOTH MOVEMENT

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In this paper, we proposed the interdental distraction osteogenesis (IDO) to lengthen the hypoplastic maxilla between teeth and create a segment of new alveolar bone and attached gingiva for relieve dental crowding by rapid orthodontic tooth movement soon after IDO in cleft patients. Interdental and maxillary osteotomics were performed on both sides of the dental arches by the cleft. After a latency period of 3 days, the osteotomized distal segments of the dental arches were then distracted anteriorly by using tooth-born intraoral distraction devices on both sides. The dental arches were lengthened anterio-posterorly and the alveoli and gingivae were created on both sides of the dental arches. The hypoplastic maxilla, midface retrusion, and anterior cross crossbite were corrected after IDO. Teeth were orthodontically moved into the newly formed alveolar bone to relieve dental crowding one week after IDO. The orthodontic tooth movement was rapidly completed in 3 months and the edentulous space was eliminated. The IDO is an effective method in lengthening hypoplastic maxilla and at the same time creating new alveolar bone and gingiva for the rapid orthodontic tooth movement in cleft patients.



## APPROXIMATE ALVEOLAR CLEFT OR ORONASAL FISTULA USING INTERDENTAL DISTRACTION OSTEOGENESIS AND RAPID ORTHODONTIC TOOTH MOVEMENT

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The closure of a wide alveolar cleft and fistula in cleft patients and reconstruction of a maxillary dentoalveolar defect in traumatic patients always are big challenges for both orthodontists and surgeons. Not only does the difficulty in complete closure by using local attached gingiva but also the great volume of bone graft. In this paper, we proposed the interdental distraction osteogenesis (IDO) to create a segment of new alveolar bone and attached gingiva for complete approximation of a wide alveolar cleft/fistula and reconstruction of a maxillary dentoalveolar defect. Interdental and maxillary osteotomies were performed on one of the dental arches by the cleft or defect. After a latency period of 3 days, the osteotomized distal segment of the dental arch was then distracted and transported toward the cleft or defect by using a tooth-borne intraoral distraction device. The alveoli and gingivae on both ends of the cleft or defect were approximated after IDO. The need of extensive alveolar bone grafting was eliminated. Teeth were orthodontically moved into the newly formed alveolar bone to relieve dental crowding one week after IDO. The orthodontic tooth movement was rapidly completed in 3 months and the edentulous space was eliminated. The IDO minimizes alveolar cleft/fistula and reconstructs maxillary dentoalveolar defect by approximating the native alveoli and gingivae, and creates new alveolar bone and gingiva for the rapid orthodontic tooth movement.

## IMMEDIATE VERSUS DELAYED MIDFACE DISTRACTION IN A PRIMATE MODEL USING A NEW INTRAORAL INTERNAL DEVICE

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**Introduction:**
The theoretic advantage of distraction osteogenesis of the craniofacial skeleton, especially in cases of severe midface retrusion and in the presence of maxillary scarring, is prevention of relapse following significant advancements. The purpose of this study is to demonstrate the ease and utility of a new low profile, intraoral internal device for midface distraction at the conventional Le Fort I level. In addition, the present study compares the efficacy of *immediate* versus *delayed* distraction on subsequent maxillary relapse.

**Methods:**
Four adult Rhesus *Macaca mulatto* monkeys were divided into two groups. Group 1 consisted of 2 monkeys that underwent *immediate* midface distraction; Group 2 consisted of 2 monkeys that underwent *delayed* distraction. All 4 monkeys underwent a conventional Le Fort I osteotomy through an upper buccal sulcus incision and bilateral application of the intraoral midface distraction devices. No other osteotomies or incisions were necessary. Immediate distraction, performed in Group 1, entailed intraoperative activation of the devices and distraction of 10 mm followed by a 5-day lag period prior to postoperative activation and istraction of an additional 10 mm at the rate of 1 mm/day. Delayed distraction, performed in Group 2, entailed a 5-day postoperative lag period prior to device activation and distraction of 20 mm at the rate of 1 mm/day. Both groups thus underwent 20 mm of midface distraction. All devices were removed 6 weeks following completion of distraction.

**Results:**
All monkeys tolerated the distraction devices and daily distraction uneventfully. A semi-solid diet had been provided postoperatively and was readily tolerated. Preliminary data following distraction demonstrate no evidence of relapse in either the immediate or delayed distraction groups. Serial cephalograms and dental models obtained throughout the 6-month postoperative period, and bone and soft tissue histology following sacrifice, will be presented.

**Conclusion:**
Significant midface advancement is feasible using a new low profile, inttaoral distraction device placed through an upper buccal sulcus incision, and a conventional Le Fort I osteotomy. This distraction device presents several advantages over other internal devices that require coronal incisions and additional osteotomies to achieve midface advancement.



## MOLECULAR DIFFERENCES BETWEEN DISTRACTION OSTEOGENESIS AND ACUTE MANDIBULAR BONE LENGTHENING

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**Introduction:**
Although the histological and ultrastructural changes associated with mandibular distraction osteogenesis (DO) have been well described, the molecular mechanisms governing these changes remain unknown. We have recently described a rat model of DO in which gradual distraction at a rate of 0.25mm twice daily for 6 days (12% increase in length) results in bone formation while acute lengthening results in Fibrous union. Here, for the first time, we report dramatic differences between DO and acute lengthening in the expression of extracellular matrix molecules.

**Methods:**
The mandibles of 40 adult male rats were osteotomized between the $2^{nd}$ and $3^{rd}$ molars and a miniaturized distraction device was applied. Animals were divided into a gradually distracted group (0.25mm BID for 6 days; 3mm total; n=20) or acutely lengthened 3mm immediately postoperatively (n=20). Animals from both groups were sacrificed at various times postoperatively and mRNA expression of collagen I, osteocalcin, TIMP-I, and VEGF was analyzed using northern blot analysis.

**Results:**
Marked differences in the expression of bone extracellular matrix molecules (ECM) collagen I and osteocalcin were noted when distracted and acutely lengthened mandibles were compared. In gradually distracted mandibles, collagen I and osteocalcin expression was markedly upregulated (3.5 fold and 5 fold, respectively) starting 10 days postoperatively (at the end of distraction period) and remained increased even 4 weeks after. TIMP-I, a modulator of ECM. turnover, was similarly upregulated (nearly 4 fold) in distracted specimens. Interestingly, no significant differences in VEGF mRNA expression were noted.

**Discussion:**
We have demonstrated that gradual DO has significant effects on cellular gene expression and that these effects result in the upregulation of bone specific ECM products. In addition, these data suggest that diminished bone formation associated with acute lengthening is due to a decrease in the production of bone scaffold (collagen 1) and its mineralization (osteocalcin). These studies may provide a molecular "blue-print" by which interventions designed to accelerate bone healing may be identified.

## DYNAMIC GRADUAL SKULL EXPANSION FOR THE TREATMENT OF CRANIOSYNOSTOSIS

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We have been performing Dynamic Gradual Skull Expansion (DGSE) for the treatment of craniosynostosis since 1996. Here we present the follow-up studies for cases with I year or longer follow-up.

**Materials and Methods:**
DGSE was performed on 4 patients with Crouzon syndrome, 1 patient with Apert syndrome, 2 patients with bicoronal synostosis, and 5 patients with sagittal synostosis. Patient ages ranged from 5 months to 11 years. Expansion was started on the fourth postoperative day. After obtaining the proper expansion, the devices were retained for an additional 4 to 6 weeks and then removed. We evaluated patients clinically with cephalograms, CT, and 3DCT.

**Results:**
There were no major complications. Excellent results were obtained in the case of clinocephaly and Crouzon syndrome. Expansion treatment on patients with Apert syndrome and scaphocephaly resuited in fair evaluations. There was no relapse of the moved bone flap after surgery. Slight growth of the advanced flap was observed in some cases.

**Discussion:**
Indications of suitability for DGSE at the present time would be summarized as follows. 1) Patients under 5 years old who may experience brain growth. 2) Patients with clino-cephaly, bicoronal synostosis, unicoronal synostosis, and Crouzon syndrome. 3) Patients who refuse blood transfusion. 4) Patients who have a risk of sinus opening.



## CRANIAL DISTRACTION: A NEW APPROACH TO COMPLEX PROBLEMS

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Complex recurrent cranial deformities are a challenge to the craniofacial surgeon. Usually the scalp is scarred and rigid which resists cranial bone repositioning. Distraction osteogenesis can be an helpful adjunct for reconstruction.

Four patients with cranial deformities due to secondary synostosis presented with signs of increased intracranial pressure. Three of the four had undergone two previous cranial reconstructions which initially improved their symptoms, but recurred as the child grew. Each of these patients underwent revision of their cranial reconstructions with the assistence of a partially buried modular distraction device. Distraction preceded without difficulty and was monitored clinically for improvement of symptoms. Plain x-rays were helpful in assessing progress and 3-D volumetric CTs quantitated the expansion.

Each of the patients had improvement in their symptoms, which has been stable for a longer period of time than the patient previously experienced. Volume analysis revealed an average increase of 8.2 ( 5-11) percent. This was greater than the amount previously achieved in the prior surgeries, despite the scarred nature of the tissue.

Cranial expansion is a useful adjunct to the treatment of recurrent, complex cranial deformities. As new distractors are developed it is expected that this technique will be more frequently applied in craniofacial surgery.

## ORBITAL EXPANSION BY DISTRACTION OSTEOGENESIS: A CASE REPORT OF THREE-VECTOR DISTRACTION

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**Introduction:**
The microphthalmic and anophthalmic orbit have continued to be resistant to satisfactory enlargement by a variety of techniques. One that has not been reported is distraction osteogenesis. A case report of a 4 year old patient witn right craniofacial microsomia and anophthalmia will be presented.

**Materials and Methods:**
Right mandibular deficiency was previously corrected in this patient utilizing distraction osteogenesis. Two previous surgical attempts at right orbital enlargement had been performed and were unsuccessful over time. The method of distraction osteogenesis using two external distraction devices to produce three vectors of enlargement was used. Eight millimeters of vertical and 8 mm of lateral displacement of the malar complex as measured at the malar eminence was obtained. Simultaneously, a second distractor was used to advance the malar complex 5 mm anteriorly as measured at the malar eminence. Simultaneous use of the two distractors produced a true three-dimensional volume increase. Final photographs and three-dimensional CT scans are presented for review.

**Discussion:**
This technique brings all of the advantages of distraction osteogenesis to orbital enlargement. Because pins are placed in the orbital rim it can be used effectively for either anophthalmia or microphthalmia. Bone defects of complex configuration are obturated by matrix ossification and soft tissue expansion occurs just as in the mandible in response to progressive distraction. Bone grafts which resorb totally are avoided.

**Conclusion:**
This case presentation illustrates the advantages of distraction osteogenesis to yet another complex congenital deformity correction.



## DOG IN PIERRE ROBIN SEQUENCE AND RELATED RESPIRATORY PROBLEMS IN CHILDREN

*Ortiz Monasterio, F., Molina, F.*
*Hospital General "Manuel Gea Gonzalez", Mexico City*

Several congenital anomalies such as bilateral transverse clefts, alcoholic fetus and Pierre Robin sequence present mandibular hypoplasia associated with respiratory distress leading to tracheotomy in the severe cases and all the problems inherent to this procedure.

**Material and Methods:**
A series of 27 patients were treated, 16 were new born infants with acute respiratory distress. Another 5 patients (ages 2 to 5 years) had been treated conservatively but presented sleep apnea. One patient had previous tongue fixation and 5 more (ages 3 to 6 years) had a permanent tracheostomy.

A bilateral mandibular corticotomy was done and intraosseous pins were inserted in front and behind. Distraction was initiated 3 to 5 days later at a rate of 0.5 millimeters every 12 hours. Range of distraction was 12 to 16 millimeters.

In 3 infants one premature and 2 in poor general condition, tongue fixation and gastrostomy were performed as a preliminary procedure.

Respiratory distress was corrected in all the infants. Tracheostomies were closed without problems. Sleep apnea was corrected in all the 5 patients.

## CT ANALYSIS OF THE MEDIAL PTERYGOID MUSCLE AFTER DISIRACTION OSTEOGENESIS

*Richard Mackool, Eric Stelnicki, Larry Hollier, Barry Grayson, Court Cutting, and Joseph G McCarthy*
*Institute of Reconstructive Plastic Surgery, NYU Medical Center*

**Introduction:**
Distraction osteogenesis increases rnandibular length through the formation of regenerate bone. However, the changes in rnandibular musculature with distraction have not been quanticated. The purpose of this study was to determine the volumetric changes of the medial pterygoid muscle following distraction.

**Methods:**
A retrospective volumetric analysis of the distracted hemimandibles of human subjects was performed. All subjects underwent unilateral mandibular distraction. Four sets of CT scans were utilized in the study, and only CT scans of equal quality and similar parameters were included in the study. Analysis of the medial pterygoid was performed by calculating the areas of the medial pterygoid muscle on 2D CT slices, and summing the areas to obtain the total volume for the muscle pre and post discraction.

**Results:**
The average percent increase in volume of the medial pterygoid was 27% on the distracted side of the mandible and 1% on the non-distracted side of the mandible (See table below).

**% Increase in Volume**

| Subject | Non-distracted | Distracted |
|---|---|---|
| #1 | 7.2 | 30.3 |
| #2 | 4.1 | 23.3 |
| #3 | 22.4 | 34.8 |
| #4 | -29.3 | 18.7 |

**Conclusions:**
The medial pterygoid muscle increases in volume with distraction. Furthermore, the relative volumetric increase of the medial pterygoid is far greater on the distracted side of the mandible when compared to the non-distracted side of the mandible. This study suggests that muscle, and in turn the soft tissue envelope, is significantly enhanced through distraction osteogenesis.



## DISTRACTION OSTEOGENESIS - THE VARIABLES THAT INFLUENCE EFFICIENCY OF SKELETAL CHANGE IN RAMAL HEIGHT

*WY Lin, DDS, BH Grayson, DDS, Deirdre Maull, DDS, MS, Pedro Santiago,DMD, CB Cutting, MD, JG McCarthy, MD*
*New York Medical Center, USA*

**Purpose:**
The purpose of this study was to describe the variables that affect efficiency of change in ramal height during distraction osteogenesis. The influence of distraction device activation on ramal lengthening was recorded and analyzed in a three dimensional cephalometric model.

**Methods:**
Thirteen patients who underwent unilateral extraoral mandibular distraction were studied. Radiographs (including panoramic, posteroanterior and lateral cephalograms) and CT scans were obtained: T=1 predistraction, T=2 end of distraction, T=3 end of consolidation. The distraction device orientation or vector of placement, the amount of device activation and the changes in ramal height were recorded at each of the three time intervals. The three dimensional cephalometric method was used for measurements of change in ramal height.

**Results:**
The ratio of change in ramal height to device activation (efficiency of distraction, ranged from 33% - 71%) was responsive to the position or vector of device placement. Devices that were vertical demonstrated the most favorable distraction efficiency. An average increase in bony length of 2.8mm occurred during the consolidation period, along the axis of distraction, which may be the result of unloading the residual stress contained in the distraction pins following a period of activation.

**Conclusion:**
The efficiency of ramal lengthing is linked to the vector of distraction. The post distraction unloading of residual pin stress may account for small amounts of ramal lengthening that occurred during the consolidation period.

## 4-YEAR FOLLOW-UP IN TREATMENT OF CRANIOSYNOSTOSIS BY GRADUAL BONE DISTRACTION

*CM Raposo do Amaral, LA Athayde Cardoso, Celso Luiz Buzzo, Thomaz Rinco, Gino Di Domizio, Valdenize Tiziani, Leda Ap. Abib Turchiari, Nelson Bolzani*
*Institute of Craniofacial Plastic Surgery - SOBRAPAR*
*State University of Campinas - UNICAMP*

Seven patients with craniosynostosis (mean age 8 years. Apert Syndrome, n=4, Crouzon Disease, n=3) underwent lengthening of the skull by gradual bone distraction. Three patients (Group A) were treated by coronal craniectomy reaching the orbital fissure and gradual bone distraction. The other four(Group B) underwent monoblock craniofacial disjunction and gradual bone distraction. The patients' follow-up was monitored clinically as well as by radiographs and photographs during four years. The results showed that craniofacial disjunction followed by gradual bone distraction produced complete correction of exophthalmos and an improvement in the functional and aesthetic aspects of the middle third of the face without the need for bone grafts.

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## RIGID EXTERNAL DISTRACTION, LONG TERM FOLLOW-UP

*John W Polley, MD, Alvaro A Figueroa, DDS, MS and W Ko, DDS, MS**
*University of Illinois at Chicago, USA, ChangGung Memorial Hospital, Taipei**

Two years ago in Santa Fe, NM, we reported our early experience with maxillary distraction utilizing an external device. This experience was remarkable for it's ability to perform large sagittal maxillary advancements, consistently, predictably, and at any age in childhood. We now report the one year follow-up on 18 consecutive orofacial cleft patients who underwent reconstruction of their maxillary hypoplasia with rigid external distraction. The average age at the time of distraction was 9 years. Pre-distraction, post-distraction, and long term (mean 15 months post-distraction) clinical and cephalometric data is presented.

Dental and skeletal cephalometric data has confirmed our clinical impression of the stability of the maxilla obtained with this method of distraction. Sagittal measurements of maxilla in these patients revealed modest reshaping of the anterior surface of the maxilla ( pt, ANS), but no posterior repositioning or relapse of the body of the maxilla. These finding were true regardless of the patient age.

External distraction now allows us to advance the maxilla in the sagittal plane without limits. Following retention of the maxilla, there is no clinically detectable maxillary relapse. The advantages of the technique are numerous, the greatest being the ability to successfully treat children with maxillary hypoplasia at any age.

## MIDFACE DISTRACTION OSTEOGENESIS IN CLEFT PATIENTS

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*Department of Plastic Surgery, Singapore General Hospital and*
*Craniotacial Center, Chang Gung Memorial Hospital, Taipei**

Orthognathic surgical procedures for the correction of the hypoplastic maxilla and associated Class III malocclusion are presently the mainstay of treatment for impaired craniofacial growth in cleft patients. However, due to the soft tissue deficiency, scarring and the underlying pathology there is a high rate of relapse giving rise to suboptimal results in the long term.

Surgical advancement of the midface by distraction osteogenesis presents an exciting and alternative treatment option in the management of severe midface hypoplasia in cleft patients. Surgically induced bone growth concomitant to maxillary advancement. provides a stable and adequate dentofacial profile without the need for bone grafts, rigid fixation or interaxillary fixation.

We present 10 consecutive cases of midface distraction in cleft patients. There were no complications and follow up was at one year post distraction. All patients had improved facial aesthetics and dental occlusion. Speech was unaffected in nine patients.

Distraction osteogenesis of the midfacial skeleton in cleft patients offers the possibility to remodel not only the underlying bony skeleton but also all the soft tissues of the face and palate. Results to date have shown excellent functional and esthetic outcomes with potentially tar reaching consequences.



## SOFT TISSUE PROFILE CHANGES AFTER MAXILLARY RIGID EXTERNAL DISTRACTION: A ONE-YEAR FOLLOW UP

*Ellen W Ko, DDS, MS*. Alvaro A Figuaroa, DDS, MS and John W Polley, MD*
*Chang Gung Memorial Hospital, Taipei*, University of Illinois at Chicago, USA*

Distraction osteogenesis with a rigid external distraction device has been used to correct severe midface hypoplasia with quite stable results in patients with secondary cleft lip and palate deformities or other craniofacial anomalies. This study was conducted to evaluate the soft tissue profile changes after maxillary advancement with distraction osteogenesis.

Twenty-two subjects, ages 5.2 to 25.7 years with various forms of facial clefts were included. Pre-and post-treatment lateral cephalograms were compared to evaluate the changes in soft tissue profile.

After maxillary advancement with rigid external distraction, the facial profile was improved by reducing the facial concavity, increasing nasal projection, normalizing the nasolabial angle, forward movement of the upper lip, balancing lip posture and exposing more upper incisor teeth. The ratio for soft tissue to hard tissue movement was 0.5:1 for nasal tip and ANS. This ratio was negatively correlated with the age of the patient. The ratio for soft tissue A point to skeletal A point was 0.9:1; and 0.8:1 for the incisal edge to vermilion border of upper lip. The treatment results remained stable one year after maxillary distraction.

## ALTERATION OF THE VELOPHARYNGEAL FUNCTIONS AFTER RIGID MIDFACE DISTRACTION

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*Craniofacial Center, Chang Gung Memorial Hospital, Taipei*

**Purpose:**
To study the effects of midface distraction on the velopharyngeal function

**Material and methods:**
This group of the cases contain 30 cleft lip and palate patients associated with severe midface retrusion at the age 9 to 14. Le Fort I osteotomy was done and solid dental appliance fixed to the upper maxillary arch for a rigid fixation to the distal segment of the maxilla. Halo external distractor was used as the driving force. Activation of the distraction at the third day after the surgery. Daily advancement of the maxilla at the rate of 1 mm per day. The movement of the maxilla was closely monitored through cephalometric and dental relationship. All the patients received speech evaluation before the surgery and at regular 3 month intervals after the surgery. The longest follow up was 2 years after distraction. The study methods contained clinical assessment, nasopharyngoscopy and videofluoroscopy of the velopharyngeal space. The categorization of the patients at three different levels: adequate, marginal and inadequate.

**Results:**
Change of the velopharyngeal function was noted immediately after the distraction procedure started. It worsens at 3 months post surgery and gradually recovered one to two year after the surgery. However, increase of nasality and air leakage was observed. 30 % of the patients the speech results remain the same as before. 60 % of the patients had mild deterioration of the nasality and resonance after the surgery. 10% of the patients got improvement.

**Conclusion:**
The distraction movement of the maxillary segment at the slow pace by using the external distractor does alteration of the velopharyngeal function on our cleft patients.



## COMPLICATIONS OF MAXILLARY (MIDFACE) DISTRACTION IN CHILDREN

*Daniel Marchac, Eric Arnaud, Dominique Renier*
*Hopilal Necker Enfants Malades, Paris, France*

We report the results in 15 patients with faciocraniosynostosis who underwent maxillary distraction. The mean age was 4.4 years. Three types of distractors were used : zygomato-malar single axis (in 7 patients) ; temporo-malar single axis (in 6 patients); and ternporo-rnalar swivelling axis (in 2 patients) . The associated surgery was a Lefort III advancement in 11 cases (1 with bipartition), and frontal advancement in 4 cases. Pterygoido-maxillary disjunction was performed in all patients except 2. Both of whom were less than 6 months old.

The complications rate was high (8/15). However, exorbitism was corrected in 14/15 patients. Two dislocations of the system, and two infections occurred with the anterior distraction, leading to early withdrawal of the distractor. A transfixion of the malar bone threatening the skin occurred in two patients. A bilateral medial dislocation occurred in two infants (one of them leading to an unexpected bipartition). When distraction was completed, an open-bite was constant, even when the swivelling-axis distractor was used. Permanent tracheostomy could be withdrawn in one out of three children, the other two being temporarily improved.

Our third generation distractor seems to be more efficient, but follow-up is still short. We believe maxillary distractions in infancy have a greatly promising future in craniofacial surgery, but need improvement.

## LEFORT III OSTEOTOMY WITH DISTRACTION: A LONG TERM FOLLOW-UP OF NASAL, ORBITAL AND MIDFACE POSITION

*Bryant A. Toth, M.D. and Michael Cedars, M.D.*
*Childrens Hospital of Northern California, Oakland CA, USA*

LeFort III with distraction has emerged as an exciting advance in improving midface position in the child with midface hypoplasia. Our experience with an entirely implantable device was presented to this forum two years ago in Santa Fe. Our early results show satisfactory midface position.[1]

Fifteen patients with midface hypoplasia unden/vent LeFort ill osteotomy with distraction between August 1994 and July 1997. In this presentation we review the long-term results with midface position comparing standard results with our distraction results. A critical look has been made to evaluate both the nose and the orbit regarding bony and soft tissue position. In several patients, nasal lengthening and nasal shifting has resulted secondary to midface malposition. There are also long-term implications regarding ocular adnexae when advancement is in excess of 15 mm. Morbidity to examine the nose and orbit will be reviewed. Important technical recommendations will be made in an effort to minimize nasal and periorbital morbidity.

[1] Toth, BA, Kirn, JW, Chin, M and Cedars, M. Distraction osteogenesis and its application to the midface and bony orbit in craniosynostosis syndromes. J Craniofacial Surg. 9:2, 1-13, 1998



## TREACHER COLLINS SYNDROME: TREATMENT: MALAR BONE EXPANSION AND MANDIBULAR LENGTHENING BY GRADUAL BONE DISTRACTION

*Romulo Guerrero, MD, Adriana Salazar DDS*
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The Treacher Collins syndrome is a congenital craniofacial malformation. It is an autosomal dominant disease of variable penetrance and phenotypic expresivity. This syndrome or mandibulofacial dysostosis involves the middle and lower parts of the craniofacial structure, affecting bone, soft tissues and upper airway.

The patients with Treacher Collins syndrome have a characteristic appearance secondary to abnormal zygomatic-malar complex, dysmorphic mandible. These anomalous charecteristics resulting in a facial structure rotation addressed to low and back. It is resulting in a parrot face.

The surgical correction should always be directed toward correction of bony deficiencies also lead to the reorientation of craniofacial growth and development.

The surgical management will require supero-anterior bone expansion of zigomatic-malar complex. The procedure should be directed toward reconstruction of the orbits and zygomatic arches. This reconstruction is accomplised with bone grafts. The goal is orbital gap correction and rotation of the orbital inclination.

In the same surgical stage the internal mandibular distractors are placed in the mandibular ramus. These device will permit a ramus mandibular lengthening in vertical direction. It change the mandible. The dimension of the mandibular ramus is significantly increased and there is an anterior rotation of the mandibular body.

Treacher Collins syndrome is a relatively rare deformity affecting both bony structures and soft tissues. Satisfactory reconstruction depends on a clear understanding to etiololgy of skeletal problems.

## CT SCAN EVALUATION OF LE FORT III ADVANCEMENT BY DISTRACTION

*MG Cedars, B A Toth, and M Chin*
*Children's Hospital of Oakland*

14 patients with craniofacial dysostosis underwent LeFort III osteotomy and advancement using distraction techniques. "3-D" computed tomography was performed on 4 patients at least I year post-surgery.

Scans show bone formation at the osteotomy sites of the pterygoid regions, lateral orbital rims, and, to a lesser extent, the zygomatic arches.

This confirms the presence of solid bony healing, supporting the stability previously suggested by lateral cephalograms and clinical findings. Scan images will be presented. The amount of advancement obtained evidently did not exceed the critical distraction distance.



## DISTRACTION OSTEOGENESIS vs. PRIMARY OSTEOTOMY: INDICATIONS AND CONTRAINDICATONS: A CLINICAL PERSPECTIVE FROM A BUSY CRANIOFACIAL SERVICE

*Kenneth E. Salyer, M.D.; Eric H. Hubli, M.D.; David G. Genecov, M.D.*
*International Craniofacial Institute, 7777 Forest Lane, Suite C-717, Dallas, TX 75230*

Distraction osteogenesis quickly has become a mainstay in craniofacial surgery. Little has been reported on the indications, contraindications, advantages/disadvantages of this technique. This study was undertaken to look at a 6 year experience at our Institute by reviewing the operative log for patients having distraction osteogenesis from January 1993 to January 1999 and to compare this with the total number of patients, during the same period of time, having classic osteotomy reconstruction. Records were reviewed for all maxillary, intracranial and mandibular osteotomies including all distraction procedures.

**Materals and Metods:**
There were 740 operations performed from January 1993 to January 1999. 364 were intracranial operations performed on patients with craniosynostosis (syndromal and simple), hypertelorism, encephalocele, fibrous dysplasia and other miscellaneous diagnosis. There were 387 mandibular and maxillary osteotomies performed during the same period for syndromal craniosynostosis, cleft lip and palate, hemifacial and craniofacial microsomia, Treacher Collins, amniotic band syndrome and Rhomberg's hemifacial atrophy, traumatic deformities and other miscellaneous diagnosis. From this group there were 53 distraction procedures on 61 hemimandibles, 3 midfaces and 1 monobloc.

**Results:**
All cases were followed up from 3 months to 5.5 years. The average distance is 21.3 mm. Ten patients had previous costochondral rib grafts placed and subsequently had successful distraction. The average midfacial distraction distance was 16.0 mm. Complications of all distraction cases include pin site infections (5), hardware failure (7), nerve injury (3) and fibrous malunion (1). All classic osteotomy complications will be presented. The indications for classic asteotomy vs. distraction osteogenesis regarding age, procedure, timing, contraindications, advantages, disadvantages of these techniques will be presented based on this 5 year experience.

**Conclusion:**
In our experience, classic osteotomy remains the primary treatment modality in craniofacial surgery today. There are specific indications where distraction osteogenesis is superior and the preferred method of treatment.

## THE LEFORT III OSTEOTOMY: TO DISTRACT, OR NOT TO DISTRACT?

*Jeffrey A. Fearon, MD*
*Dallas Craniofacial Center*

Although the Lefort III midfacial advancement is critically important for the correction of both the phenotypic stigmata and respiratory compromise associated with the craniofacial dysostoses (C.F.D.), it remains a relatively uncommonly performed procedure at most craniofacial centers. When performed in small children, this procedure can be especially challenging. Recently, osteodistraction techniques have been applied to midfacial advancements, however, aside from isolated case reports there has been no evaluation of how osteodistraction compares with the standard Lefort III.

**Purpose:**
This retrospective clinical outcome study was designed to compare the results of osteodistraction with those achieved with a standard Lefort III, to help evaluate what potential role this relatively new technology might have in the future.

**Methods:**
The charts of twenty growing children who had undergone Lefort III advancements were reviewed. Ten children (mean 6.6 years) with C.F.D. underwent distraction (8 halodistraction/2 bilateral subcutaneous distraction) over the past 14 months, and were compared with a cohort of age-matched controls (mean 6.9 years) who underwent a traditional Lefort III.

**Results:**
Pre and postoperative cephalograms were available on 8 distraction and 7 traditional Lefort III patients. The mean measured midfacial dvancement was 19mm in the halodistraction group vs. 6mm in the traditional group. SNA increased by 23 degrees in the distraction group vs. 5 degrees in the traditional group. Length of hospitalization and complication rates were comparable among the two groups. Improvements in respiratory comprise could not be demonstrated in the traditional Lefort III patients but did improve in the distraction group as measured by sleep respiratory disturbance index. Somewhat surprisingly, patient satisfaction was higher with halodistraction; perhaps secondary to the very high proportion of patients in the traditional group which required intermaxillary fixation.

**Conclusions:**
This retrospective outcome analysis of distraction vs. traditional Lefort III osteotomies showed that distraction achieved significantly better results in children with C.F.D. as measured by midfacial advancement and respiratory symptoms, without significant differences in hospitalization or complications. In this surgeon's experience, halodistraction was found to be superior to bilateral buried distraction, and aesthetic differences are discussed. It is hoped that through the use of distraction, significant advancements will be sufficient to eliminate the standard repeat Lefort III required for these patients in teenage years.



## HEMIFACIAL MICROSOMIA - AN ARGUMENT AGAINST BONE DISTRACTION

*DJ David*
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From a database of 128 patients with Hemifacial Microsomia treated at the Australian Craniofacial Unit over the last 25 years, 25 consecutive patients who had completed their treatment protocol, were selected for this study. Their clinical results are presented in terms of achievement of the treatment goals. A retrospective analysis is made of the episodes of intervention over the growing period. The interventions (surgical and orthodontic) are seen as "growth enhancing maneouvres" during development; and definitive interventions at the completion of skeletal growth. Each treatment episode is reported in terms of the clinical advantage, pain and inconvenience to the patient, time, and cost.

The potential for replacement of episodes of traditional surgical/orthodontic treatment with bone distraction are made in each case to address the following questions.

1. Arc the results likely to be better?
2. Is there less patient inconvenience using the bone distraction device?
3. Are other interventions made redundant?
4. Are the end results achieved in a shorter time?
5. Is it cheaper?
6. Does bone distraction during the growing phase obviate end of growth surgery?

When analysing the introduction of this new technology to a well known disease process with well established protocols, using the modern criteria of outcome measurements against the established protocol steps, it is hard to justify the use of this technology in the clinical management of Hemifacial Microsomia at this time.

## IONALLY GRADED SURFACE LAYERS ON TITANIUM NTS

*, T Yuen, R Smart+, M Henneberg*, W Skinner+, CM Leigh*, DJ David*
*alian Crania-Facial Unit, +Ian Wark Research Institute, University of South*
**Department of Anatomy, University of Adelaide*

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ne last 10 years manufacturers of titanium implants have introduced mplant surface treatments in the quest for improvements in the olant interface. For example, there has been the application to the surface of acid etching, grit blasting and hydroxyapatite (calcium e ceramic) layers. The expectation is then that these surface tions will enhance bone growth around the implant thereby leading to xture stability. Other techniques which have been used to accelerate position on dental implant surfaces include the use of electrical on, bone grafting and the use of growth factors. The development of ethod using low temperature plasma reactions to produce silicate s in the thin oxide layers on titanium surfaces has led to the present tions; to determine if there are differences in bone apposition around reated and untreated surfaces.

**ology:**

implants were plasma sprayed to develop functionally graded surface the order titanium/titanium oxide/silicate structures/silica. These were examined using TEM and XPS spectra. Along with the y investigations an animal implant trial was commenced. Internally slowly rotating burs were used to prepare sites in the sheep mandible ion of sterilised implants. The implant protocol involved the nt of the surface modified implant into the left side of the mandible contralateral side as a control. In the pilot study six sheep were d and subsequently sacrificed at 1, 2 and 3 month intervals. Histology ron optical analyses were used to study differences in bone n.

ysis of the Si parameter in the XPS spectra resolved profiles through ce layer revealing a gradient from oxide, orthosilicate, pyrosilicate, cates through to bulk silicate. TEM evidence from the reacted ayer shows grain detachment and interpenetration of oxide and silicate Testing has also shown that the plasma-induced silica layer is not ntly altered by insertion and removal from bone under stress and load is similar to implantation. The initial Findings of the animal study hat modification of implant surface layers may be important in g bone growth.



## ABSORBABLE PLATING SYSTEM IN FIXATION OF THE CRANIOFACIAL SKELETON

*Mutaz B. Habal, MD, FRCSC*
*The Tampa Bay Craniofacial Center*

To elucidate further on our previous report and present new applications in the last three years were we have utilized the absorbable plating system in fixation of the craniofacial skeleton in 168 patients with congenital and acquired deformities after correction of their abnormalities. We found this system to be most useful and the utilities most appropriate for these procedures. The absorbable plating system is composed of copolymers of polylactic and polyglycolic acids in a ratio of 82/18. The patients did well and they progressed to have a satisfactory outcome.

**Clinical Material:**
186 patients with congenital and acquired deformities were evaluated. We have utilized 2467 different components of the absorbable material. The patients with congenital deformities were over 65%, and over 60% were children . The fixation of the craniofacial skeleton we followed the same principles for the standard and traditional techniques used in all the procedures. All the patients progressed to a satisfactory end result. We did not have any unfavorable outcome due to the utilization of this system. There was one extrusion intraoral of the screw without unfavorable outcome in one patient.

**Conclusion:**
The absorbable plating system represent the state of the art in Fixation of the craniofacial skeleton particularly in children. The utility in the latter group is to obviate the passive migration and to avoid any growth disturbances. We found the system is useful in patients that will need radiation for oncological and malignant disease for the absorbable plates will obviate the scatter effect and the precipitation of unwarranted disturbances on the dosimetry as well as the abnormal absorption of the radiation and side effect on the surrounding soft and bony tissues.

Other new copolymers are in the works which will primarily composed of different forms of polylactic acid polymers as L or D variant with and without collimation.

## RESORBABLE FIXATION IN CRANIOFACIAL SURGERY: LONG TERM RESULTS

*Robert J Havlik, Barry Eppley, A Michael Sadove, Thomas Luerssen, Joel Boaz, John Kalsbeck*

The use of resorbable fixation has become widespread in pediatric craniofacial surgery. Over a period of 5 years, 273 patients between 3 and 24 months of age underwent cranio-orbital, occipital, or total cranial vault reconstruction using resorbable plates fabricated of a copolymer of polylactic and polyglycolid acids. From 1993-95 plates were fixed using metallic microscrews, and from 1995-98, plates were fixed using resorbable screws. A total of 181 frontal reconstructions, 26 total cranial vault reconstructions, and 65 occipital reconstructions were performed. Resorbable fixation was used in predominantly stress-loaded situations (e.g. frontonasal and temporal regions) whereas resorbable sutures were used for stabilizatyion of bone fragments in other non-loaded locations.

240 patients are now greater than one year post-operative, which exceeds the known resorption time for this polymer. Over this time frame and clinical experience complications have occurred in twelve patients. In 6 patiente in whom the resorbable plates were fixed with metallic microscrews, the plates resorbed and the microscrews became palpable and required removal. In two patients with ventriculoperitoneal shunts present, fracture of the plate fixation occurred which required re-operation and repeat fixation. In one patient, dislocation of the resorbable plate from the nasofrontal junction occurred and required re-operation. In two patients, intra-operative fracture of the plate occurred and these plates were readily replaced. In one patient post-operative ; exposure occurred which required plate removal.

In this large experience, resorbable plates have been used in a myriad of reconstructive situations throughout the range of pediatric craniofacial surgery. They have proven to be extremely reliable and safe in the vast majority of applications. The presentation will review some limitations in their use.



## HISTOLOGIC, STRUCTURAL AND CEPHALOMETRIC ANALYSIS OF HYDROXYAPATITE CEMENT RECONSTRUCTION IN THE GROWING CRANIOFACIAL SKELETON- A PIG MODEL

*Robert R. Lorenz, Francis Papay, Brook Seeley, Steven Barthel*
*The Cleveland Clinic Foundation, Departments of Plastic Surgery and Otolaryngology*

**Introduction:**
Hydroxyapatite cement (HAC) is a relatively new alloplastic implant which can be applied as a paste and contoured intraoperatively to reconstruct defects of the craniofacial skeleton. Usage in adult patients has demonstrated osseointegration and vascularization of the HAC with excellent contouring results. The long-term effects of HAC upon the growing craniofacial skeleton when used in pediatric reconstruction have yet to be conclusively studied. The objective of this pilot study is to assess the effects of HAC on the developing porcine craniofacial skeleton which has a similar growth curve as that seen in humans.

**Methods and Results:**
Unilateral frontoparietal craniotomies were performed on 10 Yorkshire piglets at 4 weeks of age. The control animals (3 piglets) underwent craniotomy and subsequent reconstruction with orthotopic bone flap replacement (group 1). The remaining animals (7 piglets, group 2) underwent craniotomy and reconstruction using hydroxyapatite cement (BoneSource® Howmedica Leibinger Inc., Carrollton, Tex). After achieving skeletal maturity at 6 months of age, animals were sacrificed and the heads skeletonized. In addition, 2 non-operated pigs were sacrificed, one at 4 weeks of age and one at 6 months. These two specimens were used to calculate the growth rate of the porcine skull in comparison to the rate in humans. Comparisons were conducted between the two experimental groups in measurements of brain weight and cranial vault volumes. In addition, five standardized cranial measurements were obtained by sliding caliper to determine inter-group difference in skull morphologic development. Areas of reconstruction were then excised and biomechanically tested in 3 point bending mode to determine mechanical stability. Histologic evaluation of HAC determined the amount of new bone formation and bone type (compact vs. trabecular). The final results of this project will be available in the spring of 1999.

**Conclusion:**
It will be determined if HAC has restrictive or deforming effects upon the growing craniofacial skeleton in terms of overall size and/or morphologic structure in the porcine animal model. These results will be extrapolated for HAC usage in the human pediatric population.

## ROLE OF REVASCULARIZATION IN THE LONG TERM SURVIVAL OF CALVARIAL BONE GRAFTS

*CR Forrest, RA Hopper, JR Zhang, I Morovna-Protzner, K Protzner, V Fournasier, CY Pang*
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The role ofrevascularization in volume maintenance and resorption of autogenous bone grafts in the craniofacial skeleton is unclear. The purpose of this study was to investigate the contribution of the periosteum and dura to the revascularization and volume maintenance of autogenous calvarial bone grafts in the rabbit.

Adult rabbits randomized into 4 groups (n=10/group) underwent creation of a critical size cranial defect (13 mm diam.) and reconstruction with autogenous calvarial bone graft. Silicone sheeting (0.5 mm) was used to isolate dura (Group II), periosteum (Group III), or dura and periosteum (Group IV), Group I remained as a control group with no silicone. Calvaria were harvested 10 weeks post-operatively for assessment of volume, histology, and revascularization.

Inhibition ofrevascularization from dura and periosteum resulted in less interface healing and significantly ($p<0.05$) higher % graft necrosis, lower % mature bone formation, and less % surface osteoblasts in Group III compared to control (see table). Volumetric analysis and immuno-histochemistry for revascularization are pending.

| Group | Necrosis (%) | Mature Bone (%) | Osteoblasts (% surface) | Interface Healing |
|---|---|---|---|---|
| I | 11±9 | 73±21 | 67±12 | 10/10 |
| II | 11±15 | 69±21 | 47±14 | 8/10 |
| III | 12±9 | 65±12 | 45±10 | 7/10 |
| IV | 42±27* | 42±21* | 22±12* | 5/10 |

(mean ±SD, * Significantly different from control ($p<0.05$). One way ANOVA with bonferroni correction for multiple comparisons).

Preliminary results suggest that inhibition of vascular ingrowth from dura and periosteum does produece significant histological alterations of autogenous calvarial inlay bone grafts in rabbits. Correlation of these findings with bone graft volume and revascularization is currently underway and will be available for presentation.



## RANDOM-EFFECTS MODELS OF MULTI-CENTER CLINICAL TRIALS IN CRANIOFACIAL AND MAXILLOFACIAL SURGERY

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**Purpose:**
The Multi-Center Clinical Trial (MCCT) is an important tool in the clinical researcher's armamentarium. Multiple sites using a common treatment protocol to assess treatment safety and efficacy characterize the MCCT. If the disease of interest is relatively rare, a MCCT can enroll the desired number of sample patients in a short period of time. Even though centers follow the same clinical protocol, the set of clinical centers may be a significant source of variability. When analyzing the data, assumptions must be made regarding the behavior of the clinical centers set. Specifically, does the set of centers behave as a Fixed or Random effect? If set of centers is regarded as random and the set number of centers is small the effect on assessing treatment results may be seriously affected. The purpose of this study was to demonstrate how the variability of the set of clinical center can affect the interpretation of the effect of treatment.

**Methods:**
To address the study purpose we used simulation data to model the effects of center, treatment and center* treatment interactions using Analysis of Variance (ANOVA). Simulation studies were conducted using SAS statistical software and varying the: 1) number of centers, 2) mean difference between treatment group response, and 3) variance between the treatment group response.

**Results:**
In the first model, we varied the number of centers but held the mean and variance of the treatment effect constant. If five centers were used, treatment effect was statistically significant regardless of whether the set of centers was treated as a random or fixed effect. When the number of centers was decreased from five to two, treatment was statistically significant in the fixed effects model, but non-significant in the random effects model. In the second simulation, we varied the mean difference between treatment group response in each medical center, and treatment was significant in the fixed effects model. The treatment was non-significant in the random effects model. In the third simulation, we varied the variance of treatment group response in each medical center. In the fixed effects model, treatment was significant. In the random effects model, treatment was non-significant.

**Conclusion:**
Simulation studies revealed significant differences in treatment effect depending on whether the set of centers was treated as having fixed and random effects. Interpreting the results requires an understanding and justification of the assumptions guiding the data analyses. This research is supported in part by Dentist Scientist Award NIH/NIDCR K16 DE00275 for Dr. Chuang and Massachusetts General Hospital Oral and Maxillofacial Surgery Research Fund for Dr. Dodson.

## PNEUMATIC ORTHOTIC CRANIAL MOLDING HELMET

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The management of positional plagiocephaly range from conservative nonsurgical approaches to surgical cranioplasty. Orthotic helmeting and/or positioning of patient's head while monitoring craniofacial growth has been successful in those patients with mild to moderate positional plagiocephaly identified in the neonatal period. Orthotic helmets have been developed over the decades and range from an inner stacked lined helmet to he dynamic orthotic cranioplasty helmet. Both helmets assist in directing cranial growth by applying resistant forces to cranial regions of excessive bossing or protuberance. A new helmet has been developed which allows monitoring of those resistance forces through pneumatic pressures. The pneumatic orthotic cranial molding helmet consists of a custom made clear aerated helmet with clear pneumatic bladders which contain one-way valves. These valves allow pneumatic inflation and pressure monitoring of the clear bladders so as to measure the exact forces to the scalp and its resultant effect in cranial growth.

**Methods and Results:**
Over the past two years 63 patients have been treated with the pneumatic orthotic cranial molding helmet. Fifty nine patients were treated with positional plagiocephaly with ages ranging from 2.5 months to 10 months. Four patients had sagittal craniosynostosis and were post craniectomy and cranioplasty with ages ranging from 4 months to 9 months of age. The average period of orthotic helmeting was 3.& months. During the orthotic management there was no signs of scalp skin ischemia or inflammation. In four cases, temporary alopecia in the region of the pneumatic bladders were reported but resolved after discontinuation of helmet therapy. Cephalometric analysis confirmed changes and cranial shape improved during the helmeting period. Occipital pressure was measured in a series of 20 patients at various postnatal ages, height and weight to determine the average pressure which may induce restrictive growth in patients with posterior positional plagiocephaly. The pneumatic pressure measured ranged from 23 mm to 30 mm Hg and was not related to patient height and weight in the neonatal period

**Conclusion:**
Restrictive cranial growth in the postnatal environment maintains abnormal growth vectors of the cranial vault and may be a consequence of chronic positioning. Pressures ranging from 23 mm to 30 mm Hg have been measured in the occipital region in the supine child. Pneumatic orthotic cranial molding helmets assist in directing cranial growth by applying similar forces in the postnatal period. To date there have been no significant sequelae or complications caused by the use of the pneumatic helmets.



## CRANIAL GROWTH UNRESTRICTED DURING DOC TREATMENT OF PLAGIOCEPHALY

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**Objectives:**
The DOC Band™ is a cranial orthosis used to provide nonsurgical treatment of defonnational plagiocephaly. The ability of this device to re-direct growth and improve craniofacial asymmetry has raised concerns regarding the potential restriction of cranial growth. The purpose of this study was to evaluate the growth of the head during correction of plagiocephaly.

**Methods:**
The study sample consisted of 190 children: 81 females (42.6%) and 109 males (57.4%) All patients had been diagnosed with non-synostotic plagiocephaly, did not have other contributing medical conditions, were compliant with DOC protocol, and had complete anthropometric measurements at entrance and exit from treatment. Growth of the head was evaluated using head circumference, maximum cranial width, and maximum cranial length. Correction of plagiocephaly was evaluated by documenting the reduction of craniofacial asymmetry of the cranial vault, skull base. and face. Paired t-tests were used to assess the significance of changes in these anthropometric measurements. Differences were considered significant if $p < 0.05$.

**Results:**
Average entrance age was 6.5 months with a mean treatment time of 4.1 months. Statistical analysis demonstrated highly significant reductions in asymmetry in all three regions ($p<0.001$). More importantly, these corrections were achieved with synchronous growth of the skull as demonstrated by highly significant increases ($p <0.001$) in head circumference, maximum cranial width and maximum cranial length.

**Conclusions:**
These findings document statistically significant increases in cranial growth in association with concomitant reductions of the cranial asymmetries associated with deformational plagiocephaly.

## CRANIOFACIAL MIDSAGITTAL PLANE VISUALIZATION AND QUANTIFICATION: AN ASSESSMENT TOOL FOR UNICORONAL SYNOSTOSIS

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**Purpose:**
Three dimensional dysmorphology characterizes a number of craniofacial anomalies, especially those with right/left asymmetry such as unicoronal synostosis (UCS). A means for documenting and quantifying the anatomic craniofacial midsagittal plane (MSP) would be useful to document the natural history of such anomalies and to assess the effect of therapeutic intervention both perioperatively and with long-term growth of children.

**Method:**
3D volumes were reconstructed, using ANALYZ™ software, from archived thin-slice high-resolution CT scan digital data of 11 individuals with untreated UCS, ranging in age from 1-21 years, 1 infant for whom there was CT data both preoperative and one year post-craniorbital reconstruction in infancy, and one unaffected 3 month old normal. The volumes were aligned to a standard coordinate system based on the Frankfurt horizontal plane. Anatomic landmarks (typically 150) in the MSP were selected in orthogonal sections through the volume including points along the outline as well as within the cranium, the cranial base, the brain and the maxilla. Using these points, a continuous surface representing the MSP was created using Matlab™ software. The surface was color coded according to deviation in mm from an ideal flat plane, or according to local bending of the surface.

**Results:**
Thp MSP in untreated UCS deviates from the theoretical midsagittal line of symmetry in a consistent fashion resembling a sigmoid curve: the anterior cranium, nose and midface are deviated toward the side of the synostosis; the superoposterior cranium is deviated away from the synostosis with the posterior cranial base minimally perturbed. There is diminution of the MSP deformation one year following calvarial surgery in infancy.

**Conclusion:**
This new MSP visualization tool is capable of capturing and communicating essential properties of craniofacial morphology to better define anomalies and quantitatively assess the effects of surgery and growth upon outcome in infants treated for UCS.



## THE NATURAL HISTORY OF SAGITTAL CRANIOSYNOSTOSIS

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**Introduction:**
Controversy exists as to whether the skull deformity seen in unoperated sagittal craniosynostosis is a progressive one. Very few quantitatively descriptive studies looking at skull dimensions in these patients exist, and those that do compare only pre and postoperative dimensions. The purpose of this study was to examine patients with sagittal craniosynostosis to determine how their skull morphologies change with growth prior to surgical correction.

**Material & Methods:**
Nine consecutive infants with sagittal craniosynostosis were included in the study. Standard CT scans images of their skulls were obtained at initial presentation, and again just prior to their surgical procedures. Craniometric measurements of maximum length and width were obtained from the images, and a length to width ratio, reflecting "roundness" of the skull, was also derived. Statistical analysis for significance was performed.

**Results:**
The study group consisted of 3 girls and 6 boys whose average age at presentation was 65.7 days (2.2 mos). The average age just prior to surgery for the group was 148 days (5 mos). Craniometric measurements when compared to age-matched controls showed that the average maximum cranial length increased from 101.9% to 107.1% ($p=.05$) whereas maximum cranial width decreased from 94.8% to 93% ($p=.5$) The roundness ratio in controls decreased from 1.34 to 1.25, whereas in the study group it increased overtime from 1.08 to 1.15.

**Conclusions:**
The characteristic deformity seen in sagittal craniosynostosis is a skull disproportionately long compared to its width. This study indicates that the disproportion, expressed as a ratio of length over width, increases over time in the unoperated skull. Continued growth in length coupled with less growth in width resulted in the progressive nature of the deformity. Clinical implications of this finding will be discussed.

## METOPIC SYNOSTOSIS: DEFINING PARAMETERS THAT PERMIT DIAGNOSIS BASED ON CT IMAGES

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**Introduction:**
Chopra, in 1957, demonstrated that the normal metopic suture fuses before the second year of life. Therefore, demonstration of earlier fusion has typically been interpreted as evidence of craniosynostosis. The exact temporal sequence of metopic suture closure, however, remains undefined. The purpose of this study was to radiographically determine the timing of normal metopic suture closure, and assess normal metopic suture morphology and craniofacial measurements, thus providing normative data to which those obtained from patients with metopic synostosis could be compared.

**Methods:**
Axial CT scans of 76 male trauma patients ranging in age from 10 days to 18 months were reviewed. The endocranial and ectocranial morphology and patency of the metopic suture were evaluated. Direct measurements of the frontoparietal, frontozygomatic, frontobasal, and orbitozygomatic regions were obtained. Thirty patients surgically treated for metopic synostosis who had preoperative CT scans were similarly evaluated.

**Results:**
Control patients demonstrated evidence of metopic suture fusion as early as 3 months of age. By 8 months of age complete suture fusion was seen in all patients. Anterior interorbital, lateral orbital, and bitemporal distances were consistently decreased in the synostotic patients, whereas biparietal distances and cephalic lengths were consistently increased. An endocranial notch at the site of the suture was seen in all patients with metopic synostosis in contrast with the typical bony ridge seen in patients following normal fusion.

**Conclusion:**
This study establishes that metopic fusion is normally completed between six and eight months of age. Fusion of the metopic suture is a progressive process initiated at the nasion and completed at the fontanelle during this time interval. Therefore, CT scan findings of suture closure after six to eight months of age cannot be interpreted as evidence of craniosynostosis. However, the craniofacial regional indices derived from direct measurements, and endocranial morphology, do provide parameters that permit diagnosis of metopic synostosis based on CT images.



## UNILATERAL CORONAL SYNOSTOSIS: ANTHROPOMETRIC EVALUATION OF FRONTO-NASO-ORBITAL SYMMETRY AFTER CORRECTION WITH AND WITHOUT CLOSING WEDGE NASAL OSTEOTOMY

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**Introduction:**
Conventional surgical correction of unilateral coronal synostosis (UCS) focuses on asymmetry of the forehead and orbit. There is discussion as to whether angulation of the nasal dorsum should be addressed during fronto-orbital advancement.

**Methods:**
Pre- and post-operative anthropometry documented nasal anguiation (na), nasion to endocanthion (n-en), nasion to exocanthion (n-ex), exocanthion to tragus (ex-trag), corneal apex to orbitale superius (ac-os) and transcranial distances in 8 patients with unilateral coronal synostosis. All patients had bilateral parallelogrammic fronto-orbital advancement. Closing wedge nasal osteotomy was performed in group I (n=6), and was not performed in group II (n=2).

**Results:**
No significant anthropometric differences were found between the two groups with the exception of nasal angulation as correlated with n-en and n-ex distances. Group I had an average pre-operative nasal anguiation of 8.9 degrees, which improved to 3.5 degrees, whereas the nasal anguiation was unchanged in group II. Post-operative fronto-orbital-nasal symmetry was better in group I as evidenced by a reduction in n-en and n-ex differences between the ipsilateral and contralateral sides.

**Conclusions:**
This study indicates that: 1) nasal root angulation does not predictably self-correct after standard bilateral fronto-orbital correction of UCS, and 2) closing wedge nasal osteotomy improves nasal angulation and naso-orbital symmetry.

## AN ULTRASTRUCTURE, HISTOLOGIC, AND BIOMECHANICAL STUDY OF CORONAL SYNOSTOSIS

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**Purpose:**
Mooney and Losken (1994) established a breed of New Zealand white rabbits with non-syndromic coronal synostosis. This study proposes to perform a qualitative and quantitative ultra-structural comparison of delayed onset synostotic (DOS) and early onset synostotic (EOS) coronal sutures on non-synostotic coronal and sagittal sutures.

**Methods:**
Cranial sutures from 19 six-week old New Zealand white rabbits were examined. We studied 6 non-synostotic coronal sutures, 18 DOS coronal sutures, 14 EOS coronal sutures, and 19 sagittal sutures. All specimens were analyzed using micro-computed tomography (MCT), scanning electron microscopy (SEM), and histomorphometry which enabled detailed qualitative and quantitative characterization of these suture's trabecular micro-architecture.

**Results:**
MCT algorithms determined bone volume fraction (BVF), bone surface to bone volume (BS/BV), trabecular number (Tb.N), and anisotropy for all specimens. We found statistically significant differences ($p<0.05$) in BVF, BS/BT, and anisotropy when non-synostotic coronal sutures were compared to EOS and DOS coronal sutures. In addition, there was also statistically significant differences ($p<0.05$) in BVF, BS/BT, and Tb.N when non-synostotic, DOS, and EOS coronal sutures were compared to their matched sagittal sutures. SEM and histologic results showed distinct qualitative differences between the trabecular pattern of non-synostotic, DOS, and EOS coronal and sagittal sutures.

| *Suture Type* | *BVF* | *BS/BV* | *Tb.N* | *Anisotropy* |
|---|---|---|---|---|
| EOS | 68.95±5.18 | 5.63±0.91 | 1.82±0.29 | 1.92±0.14 |
| DOS | 75.54±8.10 | 4.98±1.09 | 1.73±0.22 | 2.21±0.39 |
| Normal | 78.25±3.30 | 4.63±0.56 | 1.71±0.19 | 2.04±0.11 |
| Sagittal | 80.70±4.64 | 4.08±0.91 | 1.55±0.27 | 2.02±0.12 |

**Conclusions:**
We found a significant difference in the bone micro-architecture of the DOS and EOS coronal sutures when compared to non-synostotic coronal and sagittal sutures. The quantitative and qualitative results of MCT and SEM suggest that biomechanical forces are transmitted to sutures and are manifested through changes in the bony ultra-structure of synostosed sutures. This study has helped expand our knowledge of craniosynostosis at the micro-architectural level.



## PROGRESSIVE POSTNATAL CRANIOSYNOSTOSIS AND INCREASED INTRACRANIAL PRESSURE

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Craniosynostosis is thought to occur as an early event in embryological development in the prenatal period. The correct diagnosis is usually made in the immediate post natal period, based on characteristic clinical findings confirmed by modern radiological techniques. From 1991 to 1998 we have treated 14 infants and children with progressive craniosynostosis occurring in the post natal period. All patients had a clinically normal skull shape at birth and no radiological evidence of skull suture fusion in association with other features suggestive of syndromic involvement of Crozon's in 11, Saethre Chotzen in 1, and Aperts in 2. One child with Aperts had a classic Crouzon's skull. All patients had clinical, radiological or opthalmological evidence of increased intracranial pressure and required skull expansion. Two children with Crouzon's, aged 4 and 8, developed total , and 2 others, partial visual loss. An important clinical feature in most cases was the progressive bulging in the region of the previously closed anterior fontinelle. This group of patients illustrates the importance of careful follow-up of all syndromic patients. The etiology of post natal craniosynostosis remains uncertain.

## PROMINENT BASILAR EMISSARY VEINS IN SYNDROMIC CRANIOSYNOSTOSIS

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**Purpose:**
To describe the prevalence of enlarged basal emissary veins in children with mutations in fibroblast growth factor receptor (FGFR) 2:Crouzon syndrome and FGFR 3:Crouzonoid-acanthosis nigricans.

**Patients and Methods:**
Twelve children (8 males, 4 females) with FGFR 2 and 3 mutations, aged 6 months to 14 years, were imaged with thin section, non-enhanced axial CT. Additionally, six of the patients had MR and 2 had cerebral angiography. Images were assessed for size and location ofoccipitomastoid emissary veins, size of jugular foramina, and patency of internal jugular veins. Although no standards in the literature exist for the size of emissary foramina (EF), a size of 2mm was chosen as the upper limits of normal based on comparison with CT scans in non-synostotic patients.

**Results:**
EF greater than or equal to 2mm in diameter were identified on axial CT in 10 patients. In 8 of 10 the right occipitomastoid EF were enlarged, measuring up to 10 mm in diameter. Ten patients had enlarged left occipitomastoid EF measuring up to 5 mm in diameter. Seven patients had enlarged central occipital EF traversing the internal occipital protruberance measuring up to 7 mm diameter. There were variable degrees of jugular foraminal stenosis, and only 2 of 12 patients had normal jugular foramina and lacked enlarged EF (both had FGFR 2 Cys342Tyr mutation). Two of 10 patients had angiographically demonstrated atresia of the right transverse sinus and internal jugular vein (both had FGFR 3 Ala391Glu mutation).

**Conclusion:**
1) Enlarged basal emissary veins are common in syndromic craniosynostosis, probably associated with jugular venous/foraminal stenosis or atresia. 2) Degree of jugular stenosis may correlate with location of the FGFR mutation. 3) Preoperative recognition of enlarged EF is important as disruption of emissary veins can produce massive hemorrhage.



## THE INCREASED INCIDENCE OF WORMIAN BONES IN PLAGIOCEPHALY WITHOUT SYNOSTOSIS

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It had been observed in tbe Unit that Wormian bones appeared to be common in patients who presented for assessment of Plagiocephaly without Synostosis (PWS). The question- "If all children sleep supine, why don't all children develop flat heads"-had also been raised?

We investigated the exact incidence of Wonnian Bones in PWS and propose the hypothesis that occipital flattening may result from a combination of nursing in the "Back to Sleep"' position and the presence of a potentially more malleable skull than usual.

The records of all patients who had presented to the Oxford Craniofacial Unit for assessment of posterior skull deformity (n = 201) *were* reviewed. Patients with insufficient clinical and radiological data, as well as cases with definitive pathological diagnoses were excluded. Radiological investigations were assessed and compared to an age-matched control group. Clinical photography and notes were reviewed to grade the site and severity of deformation, which was then correlated with the clinical history and radiological number and site of Wormian Bones.

A statistically significant difference in the incidence of Wormian Bones was documented between the study and control groups ($p < 0.001$; $X^2$ test) indicating a direct correlation between PWS and the presence of Wormian Bones. Results of relationships between other correlates will be presented. The hypothesis of potentially increased malleability will be discussed in the light of available literature. Other non-pathological factors, which may also contribute to increased malleability (such as prematurity), are subject to ongoing investigation.

## CRANIAL VAULT EXPANSION IN THE MANAGEMENT OF POST-SHUNT CRANIOSYNOSTOSIS AND SLIT VENTRICLE SYNDROME

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**Introduction:**
*Slit ventricle syndrome* occurs as a complication in 3% of patients after shunting procedures for hydrocephalus during infancy. Excessive intracranial decompression may result in ventricular collapse and the formation of slit-like ventricles and cranial vault collapse with secondary craniosynostosis. As the brain continues to grow within the now-restricted cranium, increased intracranial pressure [ICP] recurs. The present study reviewed a group of such patients treated with cranial vault expansion.

**Methods:**
A retrospective review of 12 patients who had undergone cranial vault expansion for management of post-shunt craniosynostosis and slit ventricle syndrome refractory to other treatment modalities was performed. All patients had initially been shunted for congenital hydrocephalus and subsequently underwent a mean of 4.9 shunt revisions prior to cranial vault expansion. The most frequent symptom of shunt malfunction seen in all patients was chronic headaches. Seizures, papiUedema, bradycardia, and cortical blindness occurred less commonly.

**Results:**
All patients demonstrated decreased ventricular volume with noncompliant slit-like ventricles. Five patients demonstrated pancraniosynostosis. Multiple sutures were involved in 3 cases; the sagittal suture alone in 2 cases. Two patients appeared to have functional synostoses with narrowed sutures. Eleven patients underwent bilateral fronto-orbital advancement with fronto-temporo-parietal expansion; 1 patient underwent only posterior vault expansion. Only 3/12 patients required subsequent vault procedures after the initial expansion; in one of these cases, 9 years after the original procedure. Seven of the 12 patients required a mean of 2.3 shunt revisions after cranial vault expansion; 5 patients did not require subsequent revisons. Improved cranial vault shape was achieved in all 12 patients as well as improvement of neurologic symptoms.

**Conclusion:**
Cranial vault expansion is a useful approach in the management of the restricted cranium associated with slit ventricle syndrome. It results in a decreased need for shunt revisions with improvement of neurologic symptoms. Pre-expansion and post-expansion intracranial vault and intraventricular volumes are currently being determined.

## THE IMPORTANCE OF SUPRA-ORBITAL RIDGE RECONSTRUCTION IN THE CONNECTION OF UNILATERAL AND BILATERAL CORONAL SYNOSTOSIS

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**Purpose:**
Patients with unilateral and bilateral coronal synostosis manifest a characteristic craniofacial deformity which has been extensively descnbed previously. A critical part of the deformity which has received little attention is the anatomical absence of the normal supra-orbital ridge on the affected side, or both sides, in the bilateral deformity. The purpose of this study was to evaluate the role of primary onlay bone graft reconstruction of the supra-orbital ridge at the time of the definitive craniofacial surgical correction.

**Methods:**
From 1991 to 1998, 57 patients with unicoronal and 49 patients with bicoronal synostosis have undergone surgical correction with a fronto-orbital advancement and reshaping combined with onlay bone graft reconstruction of the absent supra-orbital ridge or ridges. All patients have been followed for six months to seven years.

**Results:**
This simple adjunctive technique has given dramatic improvement in results. The common flattening present in the supra-orbital region has been replaced by a natural contour and projection. There has been no clinical evidence of resorption of bone grafts.

**Conclusion:**
In our experience, conventional reshaping and contouring of the frontal and supra-orbital region, no matter how extensive, cannot restore the normal contour of the supra-orbital ridge adequately. These patients almost always manifest flattening in this area following correction. Primary bone graft reconstruction of the absent supra-orbital ridge provides a simple, definitive correction.

## MIDFACIAL CORRECTION IN CRANIOFACIAL DYSOSTOSIS UTILIZING SEGMENTAL OSTEOTOMIES AND DIFFERENTIAL BONY SHIFTS

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Correction of the forehead, orbital and midface deformity in the craniofacial dysostoses can involve a monoblock frontofacial advancement, two-stage fronto-orbital advancement, followed by an extracranial LeFort III, or an extracranial LeFort III, combined with augmentation of the frontal region. The usual midface deformity is characterized by an anatomically abnormal shaped skeleton with the deformity worse centrally than peripherally. Standard midface osteotomies or midface distraction techniques move this anatomically abnormal bony skeleton into a new position and usually fails to correct the underlying problem. Our experience over the past 20 years with segmental fracture repair and rigid internal fixation, has been applied to the correction of the midface in the craniofacial dysostosis patient. The abnormal anatomical configuration of the bony skeleton is completely changed with segmental osteotomies and differential bony shifts utilizing rigid fixation combined with onlay bone grafting and nasal correction using open rhinoplasty techniques and forehead augmentation. From 1991 to 1998, these techniques have been utilized in 19 patients with dramatic improvement in results. These include 7 Aperts, 8 Crouzons, 2 Pfeiffers, and 2 patients with Saethre Chotzen syndrome. Surgical details and long-term results will be demonstrated in detail.



## EVALUATION OF AESTHETIC OUTCOME, PSYCHOSOCIAL DEVELOPMENT AND ASSESSMENT OF THE DEFORMITY AND SURGICAL RESULTS BASED ON 3D CT SCANS IN METOPIC SYNOSTOSIS

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During the period 1993 – 1997, 57 patients with metopic synostosis were evaluated in the Australian Cranio-Facial Unit (41 males and 16 females). These patients were followed from the age of 2 months to 22 years. Operations were performed on 34 of these patients. The patients age at the first operation ranged from 2 months to 29 months:

- 25 patients were operated on before 6 months
- 5 patients 6 to 12 months
- 3 patients from 13 – 24 months
- 1 patient at 29 months
- 1 case was redone due to increased intra-cranial pressure.

Retrospective studies of non-syndromal and syndromal metopic synostosis based on cranio-facial data and CT scan measurements, for pre and post fronto-orbital advancement and remodelling showed that in non syndromal cases the aesthetic outcome was generally very pleasing with minor or no frontal deformity and minor or no psychosocial problems. The follow up CT's showed improvement of the full features of trigonocephaly including midline ridging, bifrontal recession, hypotelorism, shortened anterior cranial fossa, deficient projection of lateral orbital and bitemporal narrowing.

## LEFORT III MIDFACIAL ADVANCEMENT: THE CLASSIC OPERATION REMAINS A SAFE AND EFFECTIVE PROCEDURE

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Recent developments in craniofacial surgery provide for the advancement of the midface in patients with syndromal anomalies using distraction techniques. The classic osteotomies, however, remain the gold standards for bone movement in craniofacial surgery. hroughout the last 20 years the authors have performed over 120 Lefort III midfacial advancement procedures in patients with syndromal anomalies. From January 1987 to January 1998, the surgeons at the International Craniofacial Institute have performed craniofacial procedures with the same anesthesia, operative and post operative care teams. With the current discussion comparing distraction to standard osteotomies, we felt that it would be beneficial to review our positive results associated with the classic Lefort III osteotomy and midfacial advancement.

**Materals and Methods:**
Seventy-six Lefort III advancements were performed as primary and secondary advancements over the 11-year period from January 1988 to January 1999. Diagnoses included Crouzon's syndrome (37), Apert's syndrome (25), Pfeiffer's syndrome (5), clefting anomalies (5), and others (4). Charts and operative records were reviewed retrospectively to evaluate intra-operative complications, length of operative procedures, blood loss, transfusion requirements and postoperative complications. In addition, length of stay was evaluated on all patients. Anesthetic technique used involved controlled hypotension in all patients and hemodilution in patients over 25 kilograms.

**Results:**
Patient age ranged from 4.1 to 20.3 years of age with 48 patients in the age group less than eight years, and 28 patients in the age group greater than eight years. Secondary Lefort III was performed in 12 patients. Additional procedures were performed in all 76 cases, most commonly medially and/or lateral canthopexies, cranial bone grafts in 57 patients and demineralized bone grafts in 15, and iliac bone grafting in 4 patients. Associated cranial vault remodeling was performed in 11% of cases. Infra operative blood loss ranged from 300 cc to 3,400 cc with an average of 580 cc. Transfusions were required in 14 patients (18.4%). Average transfusional requirements were 230 cc (excluding the single case of a 15 unit pack cell transfusion.) Transfusions were given in non-hemodiluted patients at a ratio of 6:1. Complications include wound infection/surgical site infection in 8 patients, pneumonia in three patients, CSF leak in two patients (both required lumbar drains) and airway compromise in one patient requiring tracheotomy. There were no deaths, and length of stay averaged 6.3 days. Average advancement of the procedures was 14.5 millimeters with a range of 11-19 millimeters.

**Conclusion:**
In our experience, the classic Lefort III osteotomy with advancement is a safe procedure with acceptable risks, and when used in combination with controlled hypotension and hemodilutional anesthesia, transfusion requirements are less than 20% and occur mostly commonly in patients under the age of eight years. With the present day desire to use distraction in midfacial advancement, we feel that this new modality must offer equal or better results when compared to the gold standard of a classic Lefort III advancement.

## THE DENTAL OCCLUSION FOLLOWING LE FORT III CRANIOFACIAL OSTEOTOMY

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The aim of this study is to look at the dental occlusion before and after Le Fort III craniofacial osteotomy. Nine patients underwent Le Fort III craniofacial osteotomy at Hospital University Sains Malaysia had their dental occlusion thoroughly assessed before and after the procedure. Dental clinical examinations, dental study models, bite registrations and dental orthopantomograms were done before the operation and on further follow up one year post operatively, patients were also asked if they have any complaints regarding their dental occlusion postoperatively. Clinical assessment with the aid of study models and x-ray showed that all patients had Class III malocclusion with overjet of between 8 mm and 22 mm, and an open bite of between 2 mm and 6 mm, Class III dental occlusion on a Class III skeletal base relationship. There is no facial asymmetry in all the nine cases selected for this study. All patients underwent pre and post orthodontic management. Bone allografts were used to sandwich the osteotomy sites.

Results showed that patients are nor worried about the dental malocclusion but they are more concerned about their facial aesthetic in which all of them had a depressed midface. A Class I dental base relationship was achieved in all cases post operatively but a Class I dental occlusion is only achieved with respect to incisal relationship in six out of nine cases. At one year post operative, a relapse of dental occlusion was observed with a slight open bite. Further assessment showed a slight posterior tilt of the whole midface. Dental occlusal relapse occur following Le Fort III craniofacial osteotomy with maxillary advancement, and the condition takes place by backward rotation of the anterior mid face producing a slight anterior incisal open bite.

## LONG TERM FOLLOW-UP OF THE DYNAMIC CRANIOPLASTY FOR BRACHYCEPHALV

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A follow up study was made of brachycephaly patients uniformly treated with the dynamic cramoplasty presented by Lauritzen et al. in 1996. With this technique the growing brain is directed in an anteroposterior direction at the same time as wire mediated restraint is exerted on the transverse and vertical skull expansion. The operative technique is reviewed together with analyses of the surgical results and complications in 30 patients. 15 of these have a follow-up period of 5 years or more. The mean operative time was 188.8 minutes. The mean post-operative anesthesia duration was 33.8 minutes. Total blood transfusion (intra operative + post operative) averaged 158.1% of estimated red cell mass. Mean intensive care unit stay was 1.7 days. The surgical results were analyzed by means of cephalometric radiography and tracings plus medical photography. There was no mortality and few complications. To the authors' knowledge, no other series on the treatment of brachycephaly with equal or better results documented with strict scientific criteria is available for comparison in the literature.



## FUNCTIONAL OUTCOME AFTER SURGERY FOR BRACHYCEPHALY

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Bicoronal craniosynostosis is a severe condition which can result in intracranial hypertension when non operated on, leading to mental delay associated to dysmorphy.

A group of 99 non syndromic operated patients was prospectively investigated for functional outcome. Were excluded all syndromic related cases (Crouzon, Apert, Pfeiffer, ....). All patients were operated on with the same standardized technique (forehead advancement). Mean age at surgery was 10 months (range 1 month - 5 years). Eighty one percent of the patients were operated before one year of age, and 19% after one year. The mean follow-up was 5 years, and all intellectual quotient (1. Q.) was carried out by the same psychological team.

The mean postoperative IQ evaluation was 97 (range 55 - 134). Seventy percent of the patients had an IQ evaluation greater or equal to 90 (normal). When considering the age at surgery, the mean 1. Q. of the patients operated before I year of age was 99, while it was 89 in the patients operated after I year of age. This difference was statistically significant ($p<0.05$). Other factors of prognosis were analyzed: existence of mutation (P250R), familial cases, associated malformations...

These data confirmed our strategy that bicoronal craniosynostosis in non syndromic patients should be operated before one year of age to get the best functional outcome.

## ANTHROPOMETRIC ANALYSIS OF HOMEOTOPIC VERSUS HETEROTOPIC POSITIONING OF FRONTAL ELEMENTS IN CORRECTION OF UNILATERAL CORONAL SYNOSTOSIS

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**Introduction:**
The correction of unilateral coronal synoslosis (UCS) has evolved from unilateral coronal suturectomy. to unilateral fronto-orbilal advancement, to bilateral fronto-orbital modeling and positioning. Reconstruction of the forehead can be *heturotopic* (i.e..exchange and rotation of the frontal segments) or *homeotopic* (i.e.. contouring and replacing the frontal elements). Our null hypothesis is there would be no significant difference in forehead symmetry using the two techniques.

**Methods:**
Forehead symmetry was compared in 2 groups of children (average age 4vrs 9mos) who had either *homeotopic* (n=6) or *heterotopic* (n=6) placement of the frontal elements during correction of UCS in infancy (average age 7.5inos). A forehead iiionlage was taken on all 12 children for anthropomeiric analysis. We developed an objective anthropometric measure of forehead asymmetry based on asymmetries along three projected arcs. Each of the arcs adhered to the contour of the forehead, measured from lhe left lo right fronto-temporale (ft'), and intersecting one of three midpoints equidistant placed along a vertical line connecting the glabella (g) and trichion (tr). Each arc was measured three times and reproducibility was assessed. A global asymmetry score was computed for each forehead using a mathematical model incorporating these 3 arcs. that served as the dcpeiidein variable, operationally defining the measure of surgical success.

**Results:**
The postoperative forehead asymmetry score was 45 in the *heterotopic* technique group compared to 21 in the *homeotopic* group ($p<0.01$). With liltle practice, reproducibility of the measurements was high.

**Conclusions:**
Analysis of forehead synimetry showed superior postoperative results with *homeotopic* reconstruction as compared to *heterotopic* reconstruction. There are also technical advantages for the *homeotopic* forehead technique, including better adaptation of the frontal elements to the supraorbital rim, fewer bone segments smaller calvarial gaps, and diminished operative time for this portion of the operation.



## LONG TERM NEUROPSYCHOLOGICAL EFFECTS OF SINGLE-SUTURE CRANIOSYNOSTOSIS ON CHILD DEVELOPMENT

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**Purpose:**
The purpose of this study was to examine the long term effects of single-suture craniosynostosis on the neurologic development of children, by going beyond gross measures of mental function (IQ), in an attempt to assess the incidence of more subtle neuropsychological sequelae.

**Methods and Results:**
Retrospective analysis of Yale Department of Neurosurgery records between 1980 and 1990 was used to identify potential study subjects born with single suture craniosynostosis, and currently between six and sixteen years of age. There were 16 born with sagittal synostosis (n = 16). These patients were treated with simple linear craniectomy procedures. Of the patients with sagittal craniosynostosis, 50% were identified as having a learning disability by a battery of psychologic and neurodevelopment tests (Wechsler Intelligence Sclae for Children, Wide Range Advancement Test, Conner Continuous Performance Test, Wisconsin Card Sorting Test, Developmental Test of Visual-Motor Integration and Vineland Adaptive Behavior Scale).

**Conclusions:**
This study indicates that although subjects fall within the normal range of intelligence, there is a significantly higher incidence of learning disabilities in this group compared with the general population. The implications for mode and timing of treatment will be discussed.

## EFFECTS OF A CRANIOFACIAL ANOMALY ON THE STABILITY OF PARENTAL RELATIONSHIPS

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**Introduction:**
Parents of children born with major craniofacial abnonnalities are confronted with many social and psychologic challenges. It has been suggested that asymmetric anomalies or major malformations with a greater degree of anatomic involvement cause more psychosocial problems. The purpose of this paper is to determine rates of divorce in parents of children with asymmetric vs. symmetric, and major vs. minor anomalies.

**Materials and Methods:**
A 29-question retrospective survey was sent to the parents of all children treated in the craniofacial program between 1992 and 1997. Using deformational posterior plagiocephaly as a control group, rates of divorce vs. non-divorce were compared to all craniofacial anomalies. Deformities were separated for analysis into *asymmetric* (hemifacial microsomia. unilateral coronal synostosis. cleft lip. cleft lip/palate) and *symmetric* (syndromic-crailiosynostosis. orbital hypertelorism. Treacher Collins syndrome). Comparison was also made using the anatomic classification of *major* (uailateral coronal synostosis. hemifacial microsomia. cramosynostosis. orbital hypertelorism. Treacher Collins) vs. *minor* (cleft lip. cleft lip/palate) anomalies.

**Results:**
Surveys were sent to both parents of 412 families. 403 surveys were returned, and results were evaluated in 275 families (67%). Frequency analysis demonslrated an overall divorce rate of 6.8% at the time of questioning, with an additional 4.9% reponing separation. Anomalies associated with the highest rates of divorce in order of frequency were hemifacial microsomia (24.0%). syndroinic-craniosynostosis (12.2%). and cleft lip/palate (6.8). Using a two-sided Fisher exact test. comparison of control vs. all other anomalies showed significance at 030 for rates of divorce. Separation of anomalies into asymmetric vs. symmetric demonstrated no significance in divorce rate ($p > .05$). Similarly there was no statistical difference when anomalies were categorized as major vs. minor.

**Conclusion:**
Compared to control, divorce rates are higher in those families coping with a craniofacial anomaly, and the frequency of divorce is higher with specific abnormalities. Asymmetry and anatomic severity do not seem to be important variables. However, other possible determinant factors need to be analyzed such as number of operations required to correct each anomaly, birth rank of the child, length of time married prior to the birth, degree of spousal involvement following birth, and support structure for the parents.



## SAGITTAL CRANIECTOMY: DIFFERENT SURGICAL TECHNIQUES EVALUATED IN 161 CASES OF SAGITTAL SYNOSTOSIS

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Determination of the appropriate management of either isolated nonsyndromal or syndromal sagittal synostosis, in association with craniofacial syndrome, is at the base of a full patient recovery. A critical review of a craniectomy, performed on 161 patients, managed by the Australian Craniofacial Unit, is shown in this study. In about 17% of these cases, the result is unsatisfactory, requiring re-operation. All cranial measurements, from the pre-operative stage through to followup at full recovery, have been considered. We can conclude that sagittal craniectomy is not always adequate, and as a result, cranial vault reshaping is required.

## MORTALITY IN 1500 OPERATIVE PROCEDURES FOR CRANIOFACIAL MALFORMATION

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**Purpose:**
Surgery of severe skull and facial deformities carries major risks of complications and mortality. These patients may also have an early death for reasons related to their medical problems but unrelated to surgery. With focus on patients with malformation this survey reports the overall mortality in patients examined for surgery by the craniofacial unit in Goteborg during a 20 year period.

**Methods:**
The medical records of all patients seen by the craniofacial unit were retrospectively reviewed. A total of 1260 patients have been seen at the institution; the medical disorder was related to either tumour, trauma, or malformation in 126, 152, and 982 patients respectively. In 1016 patients 1859 operations were done; 1500 in patients with malformation.

**Results:**
Of patients with craniofacial malformation 17 patients (at age 2 months to 28 years, six with Aperts and three with Crouzons syndromes) were found to be dead, most commonly from infectious problems. Death was unrelated to suicide but drug abuse was the cause in two patients. Two patients operated on at our hospital for craniofacial malformation, and two patients operated on at another hospital undergoing orthopaedic surgery and tonsillectomy died less than 10 days after surgery, either at hospital or at home. Three of these patients had Aperts syndrome. The mortality rate within one months of surgery thus comes to 0.13% at our institution. In two cases, both Apert, death was a direct consequence of postoperative complications. There was no intraoperative death.

**Conclusion:**
Patients with craniofacial disorders requiring surgery represent a major challenge for both surgeons and anaesthesiologists. The concentration of these otherwise rare patients to one unit and by involving *a* limited amount of surgeons and anaesthesiologists mortality has been very low.



## HYPONATREMIA IN THE POSTOPERATIVE PEDIATRIC CRANIOFACIAL POPULATION: A CONNECTION TO CEREBRAL SALT WASTING SYNDROME AS AN ETIOLOGY AND MANAGEMENT OF THE DISORDER

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**Purpose:**
We have noted that some pediatric patients who have undergone cranial vault remodeling (CVR) have developed hyponatremia. Traditionally, these patients have been diagnosed with syndrome of inappropriate antidiuretic hormone (SIADH) however, in our patients the treatment has been resuscitation with normal saline (NS) as opposed to fluid restriction (normal treatment of SIADH) placing the diagnosis of SIADH in question. We therefore, developed a prospective study to evaluate the etiology of the sodium imbalance.

**Methods:**
Three consecutive pediatric patients were operated on at NYU for treatment of various craniosynostotic disorders. All were monitored postoperatively in the pediatric intensive care unit including hemodynamic, respiratory and fluid management. Each patient had daily (including intraoperative) measurements of standard serum electrolytes, random urine electrolytes, osmolarity, ADH (antidiuretic hormone), aldosterone and ANF (atrial natriuretic hormone). These patients were also treated with NS in the peri-operative period.

**Results:**
All three patients had normal to low serum sodium concentration throughout their hospital stay, as low as 128 (normal 137-145), even with NS resuscitation. All had elevated urine output in postoperative days 2-4 (>1 cc/hr/kg). All had an elevation of ANF, as high as 277pg/ml in one patient (normal 25-77), in the early postoperative period. ADH levels were low or normal in two patients but the third had a marked elevation along with his ANF at the same time point. Aldosterone levels were variable in all patients, Two patients showed clear evidence of cerebral salt wasting (CSW) including increased urine output, normal or elevated urine sodium, low serum sodium and elevated ANF levels. The third patient had similar findings but also had an elevation in ADH likely giving a mixed clinical picture of SIADH and CSW.

**Conclusions:**
CSW is associated with increased urine output and urine sodium concentration. The association of CSW to CVR has previously been ignored making us reconsider SIADH as the etiology of all hyponatremic episodes in these patients. An etiologic role has been given to ANF and as yet undiscovered CNS factors. Only 1 of 3 patients showed signs of SIADH, the other 2 had clear evidence of CSW syndrome. The third patient had a mixture of CSW and SIADH. All of these patients required NS resuscitation, contrary to the normal management of SIADH. These are important findings, which have changed our postoperative management of these patients. We now prophylactically treat our intracranial cases with NS instead of a more hypotonic solution and are more aware of sodium balance problems in these patients.

## PREVENTION AND MANAGEMENT OF COMPLICATIONS IN CRANIOFACIAL SURGERY INVOVING THE ANTERIOR CRANIAL BASE: A RETROSPECTIVE REVIEW OF 118 PATIENTS

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An uninvited complication arising in craniofacial surgery can have a tremendous impact on the final outcome. This is reflected not only on the significant morbidity, but that it can lead to surgical mortality.

The author has made a retrospective review of 118 patients undergone craniofacial surgery of the anterior cranial base. The series included 53 congenital anomalies (49 craniosynostosis, 4 encephaloceles), 31 benign craniofacial tumors, 16 malignancies, and 18 secondary posttraumatic reconstructions.

The most frequent complication was infection, totaling 9 patients. Two were craniofacial synostosis patients and the remaining 7 were traumatic cases developing infection after failed primary care. Prolonged CSF leakage was encountered in 4 patients. Postoperative hydrocephalus developed in one patient. Infecious complications have been categorized into 3 groups; those with prolonged CSF leakage, localized infections leading to abscess formation, and extensively progressive infections. The most frequent intraoperative antecedent cause was nasofrontal communication.

Tissel or galeopericranial flaps proved to be most effective in minor defects of the anterior cranial base. For moderate sized defects, pericranial and galeofrontalis flaps were sufficient. For significant defects or coexisting preoperative infections, a well vascularized tissue transfer such as the reverse temporalis flap or free vascularized tissue transfer have been most useful.



## ALLOGRAFT BONE USED IN EXTENSIVE CRANIO-ORBITAL RECONSTRUCTION FOR FIBROUS DYSPLASIA

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Banked allograft bone grafts are described for reconstruction of endochondral bones. We report its use in reconstruction of membranous bone. A 37 year old woman, with a 22 year history of progressive craniofacial fibrous dysplasia for which she had undergone multiple operations, complained of chronic pain in the right orbit. CT scans revealed extensive disease with cystic changes in the right orbital roof.

Psychiatric counselling and etidronate both failed to alleviate her pain. Further surgery was indicated, but the paucity of remaining donor sites led to a plan for cadaveric allograft reconstruction from the Victorian Bone Bank. Following animal studies on irradiated fresh frozen allografts in the rabbit, a human protocol was developed including screening for infectious diseases. When a suitable donor was available the entire fronto-orbital region and right zygoma was harvested, reconstructing the donor with a similar section from a plastic skull.

At surgery the anterior half of the cranial vault, the right greater wing of sphenoid and lateral orbital wall was removed. The reconstruction was performed with the cadaveric allograft bone in two pieces: the fronto-orbital-zygomatic component, and the neo-forehead. Autogenous split cranial bone grafts reconstructed the floor of the anterior cranial fossa. Fixation was with titanium microplates and screws. The anatomical "fit" was very close, requiring burring only to achieve an excellent contour. There were no postoperative complications.

At 3 years of follow up, she has minimal occasional pain, completely relieved by panadol, and a good contour. We would recommend cadaveric allograft bone reconstruction for extensive craniofacial reconstructions when there is a paucity of autogenous donor sites.

## THE EFFICACY OF OPTIC NERVE DECOMPRESSION IN FIBROUS DYSPLASIA

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There exists controversy in the strategy in management optic nerve decompression for cases of fibrous dvsplasia especial for prophylactic purpose. To clarify this point, we review eighteen patients with craniomaxillofacial fibroup dysplasia who under went 21 optic nerve decompression procedures in Chang Gung Medical Center between 1982 to 1998. There were thirteen procedures for prophylactic and eight for therapheutic reseaon.

One patient gained a useful improvement in vision in two decompression, and another patient had significant improvement with visual acuity recovered from 0.4 to 1.0.

No patient suffered permanent damage of vision in the procedure of decompression no mater what therapeutic or prophylactic. One patient got extraocular muscle dysfunction but which was corrected in subsequent surgery. In summary: The efficacy and safety of optic nerve decompression in case of fibrous dysplasia can be secured. In patient with a gradual deterioration of vision who are not yet totally blind and patients with a recent sudden onset of visual loss secondary to fibrous dysplasia may be benefit from optic nerve decompression.



## DESMOPLASTIC FIBROMA OF THE ZYGOMA

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Desmoplastic fibroma is an exceedingly rare primary bone bumor that is the osseous equivalent of the extra-abdominal desmoid tumor. Although previously described as occurring in several different bones of the facial skeleton, there are no reports of desmoplastic fibromas involving the zygoma. This case, in a 5-year-old boy, is the first report of a desmoplastic fibroma occuring in the zygoma. Surgical management utilizing a craniofacial approach with preservation of the facial nerve, enblock resection of the total zygoma and reconstruction with full thickness oranial bone grafts is described. The result at two years with no evidence of recurrence is shown with successful preservation of facial nerve function and maintenance of normal appearance.

## RADICAL RESECTION OF ANTERIOR SKULL BASE TUMORS USING THE SIX LEVEL CLASSIFICATION SCHEME OF TRANSFACIAL APPROACHES

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Between 1990 and 1998, 63 patients (39 males and 24 females) underwent a transfacial approach for resection of midline skull base lesions utilizing the six level classification scheme previously described in the literature. A total of 83 transfacial procedures were performed on 68 patients using a single transfacial level and 9 patients combining two levels. In 6 cases an upper transfacial approach was combined with either a pterional craniotomy or orbitozygomatic craniotomy. In total, thirty-five different pathological processes were addressed with chordoma (7), angiofibroma (7), and pituitary macroadenoma (5) being the most prevalent. One patient (1.2%) died in the post-operative period. There were 95 major and minor complications. Thirty-one patients had no complications. The complications were classified as follows: neurologic (27), systemic (21), CSF leak (15), wound healing (10), central venous line (5), hardware (3), epiphora (3), malocclusion (3), speech (3), bleeding (3), ocular (2). The six level classification scheme and surgical techniques will be briefly reviewed with an emphasis on clinical results including long-term outcome, complications and their treatment and avoidance.



## FACIAL TRANSLOCATION FOR SKULL BASE TUMORS

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The complexity of cranial base surgery is a refection of skull base anatomy as well as technical demands for maximum exposure to facilitate adequate tumor resection and/or reconstruction. Facial translocation, with temporary removal of facial bone and reinsertion at the end of the procedure, has been developed as one of the best surgical approach to anterior and middle cranial base. Versatility of this approach also permits expansion into neighboring cranial fossae.

From July 1993 to June 1998, sixty patients with tumors of skull base and paranasal sinuses underwent facial translocation approach for resection. The patients ranged in age from 3 years to 102 years and were followed up to 5 years post surgery.

There were 19 benign and 41 malignant lesions. 43 (71.7%) patients survived with a mean survival of 26.8 months. There was no peri-operative motality. The perioperative morbidity was 30% and included bone graft necrosis (NBG) (12), postoperative bleeding (4), CSF leak (2) and blindness (1). Analysis of the patients with postoperative NBG revealed that NBG was increased by postoperative irradiation (P=0.04) and was decreased by soft tissue reconstruction of the surgical defect (P=0.006).

The facial translocation approach offers favorable exposure of the critical zones of the anterior and middle cranial base to facilitate extensive resection and reconstruction. With this approach, 43 (71.7%) patients survived with a mean survival of 26.8 minths. The mobidity of skull base surgery remains high (30%). To further lower down NBG rate, we suggested the translocated bone graft should be attached to the cheek soft tissue or the surgical defect should be reconstructed with vaseularized flap.

## CRANIOFACIAL RECONSTRUCTION WITH FREE TISSUE TRANSFER

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Craniofacial region is a complex anatomical structure to be important in aesthetical and functional viewpoint. Craniofacial defect can be developed by tumor ablation and trauma. Local and regional flap reconstruction is suitable for many smaller defects in the craniofacial region. However, as the surgical defect become more medial and more extensive, free tissue transfer assumes greater role in the reconstructive procedure.

We have been experienced 19 cases of craniofacial reconstruction using free tissue transfer. Male were 13 cases and female 6 in the sex distribution. The range of age was from 19 to 72 years old. The cause of defect was benign tumor (2 cases), malignant tumor (12 cases) and trauma (5 cases). The defects were reconstructed by using latissimus dorsi myocutaneous flap (14 cases), radial forearm flap (2 cases), omental flap (1 case), groin flap (I case), and dorsalis pedis flap (I case).

In our experiences with the success rate of a hundred percent, free tissue transfer was quite reliable for craniofacial reconstruction. Appropriate flap selection and experience with flap design, elevation, and inset have proved to be very important in the success of these procedures.



## HYDROXYAPATITE BIO-COMPOSITE FREE FLAP FOR TOTAL MAXILLECTOMY DEFECT

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This paper presents a new method of maxillary reconstruction using prelamination technique to render vascularized three-dimensional bone reconstruction. Two pieces of artificial porous hydroxyapatite (HA) blocks were fabricated based on preoperative CT scan data; one for orbital floor and the other for the anterior wall of the maxilla. These blocks were implanted into a subcutaneous pocket of radial forearm flap or latissimus dorsi myocutaneous flap. 3 weeks later, the maxillectomy was performed and the prelaminated flap including the HA blocks was harvested. The two pieces of HA blocks were assemled together; in this way, the skin paddle of the flap ended up with providing the lining for the paranasal cavity. Two patients undergone total maxillectomy including the orbital floor. Their defects were successfully repaired with HA prelaminated forearm flap or latissimus dorsi flap without postoperative infection. Their aesthetic outcomes were satisfactory. The prelaminated flap with HA blocks decreased a risk of infection related to HA blocks. The presented method provided us with more precise three-dimensional reconstruction for total maxillectomy defect.

## CRANIOFACIAL RESECTION FOR ADVANCED MALIGNANT TUMORS IN ORAL AND MAXILLOFACIAL REGION: A RETROSPECTIVE STUDY OF 46 CONSECUTIVE CASES

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**Objective:**
This study was aimed to investigate the clinical significance of craniofacial resection for advanced malignant tumors in oral and maxillofacial regions.

**Materials and Methods:**
Forty-six patients who underwent craniofacial resection for malignancies involving the anterior and middle cranial fossae over a 20-year period between June 1978 and December 1997 in our Department were evaluated. Twenty patients received radiotherapy as an adjunctive therapy after operation (4500-6000cGray, with a mean of 5230cGray). Eleven patients received chemotherapy of various regimes as a postoperative adjunctive therapy. The dural defects were repaired with temporal fascial flap in 10 cases and galeal-pericranial flap in 4 cases. The defects of cranial base and extracranial tissues were immediately reconstructed with free pectoralis major myocutaneous flap combined with free radial forearm flap (5 cases), latissimus dorsi myocutaneous flap (2 cases), frontal flap (13 cases) and temporal muscular flap (7 cases). All the patients were followed up for 3 to 15 years. The survival rates were calculated using Kaplan-Meier Method.

**Results:**
The overall incidence of complications was 41.3%. 9 cases had major complications associated with surgery. Postoperative complications included cerebrospinal fluid leakage (4 cases), meningitis (3 cases), cerebral edema (2 cases). One patient died of acute cerebral edema postoperatively. The 3, 5, 10-year survival rate was 48.8%, 35.1% and 20%, respectively.

**Conclusions:**
The results showed a relatively fine prospect of craniofacial resection for patients with advanced malignancies in oral and maxillofacial regions. With the use of composite free flaps, the quality of reconstruction after ablative surgery improved significantly. We suggest that the indications for craniofacial resection might be extended to some selected T3 or T4 patients.



## LARGE ARTERIOVENOUS MALFORMATIONS OF THE FACE: AESTHETIC RESULTS WITH RECURRENCE CONTROL

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**Purpose:**
The management of targe and often recurrent facial arteriovenous malformations (AVMs) has undergone a rapid evolution - The purpose of this study was to look critically at the treatment of facial AVMs from different anatomic locations (labial, preauricular, eyelid, cheek, chin and occipitoparietal) including patient presentation, angiographic procedures, surgical planning and technique and postoperative follow-up care.

**Methods:**
From a series of 300 facial AVMs, 85 patients were treated with embolization and excision. A multidisciplinary approach was used which included interventional radiologists, reconstructive plastic surgeons, oral surgeons, otolaryngologists, ophthomologists, and neurosurgeons. As a first step the goals of the therapy were determined including cure or palliation of ischemic pain, bleeding, or functional disabilities, such as vision loss or feeding problems. Then the territory of involvement was defined by CT scan, MRI and most importantly superselective angiography. Preoperative devascularization by intra-arterial or direct lesional embolization was performed using variable stifiness microcatheters, flow control techniques and a variety of embolization materials. Crucial to the approach was the understanding of functional vascular anatomy and the hemodynamics of each lesion. Within 4 days after embolization, judicial resection (sparing vital structures) and reconstruction with local and expanded tissue flaps were performed. Several weeks after the procedure residual fistulization not directly apparent was identified by follow-up arteriogram and controlled by embolization.

**Results:**
Cures were possible only in some patients. In the majority of these large facial AVM cases aesthetic results were obtained but recurrences were common. However, long term palliation of these minor problems was achieved with embolic control. Preoperative devascularization with new embolization techniques, intraoperative control of blood pressure, use of tumescent injection, sparing of vital structures and staged reconstructions led to safer procedures. Early control of postoperative fistulas enhanced the stability of the treatment for a better long-term outcome.

**Conclusion:**
Long-term control of large facial AVMs with acceptable aesthetic results can be achieved.

## SURGICAL EXCISION OF MAXILLARY ANTRAL HEMANGIOMA FOLLOWING ARTERIAL EMBOLIZATION

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We report a rare, large maxillary antrum hemangioma in a 30 year-old male who presented with profuse epistaxis, blocked nose and facial pain. Preoperative MRI with gadolinium- DTPA contrast revealed an intensely enhanced 3.5x3.4x3.2cm right maxillary antral tumor with expansion of the medial wall. Digital Subtraction Aangiography of the right carotid was scheduled a day before operation in an attempt to embolize the tumor prior to surgical excision. Tumor staining was seen during arterial and persisted through to the venous phase. The main feeders were from branches of right internal maxillary artery namely the infraorbital, greater palatine and superior alveolar artery. Successful embolization was achieved with two liquid coils to the right internal maxillary artery proximal to the origin of right infraorbital artery and distal to middle meningeal artery. Right lateral rhinotomy and antrostomy was performed instead of rnaxillectomy as an approach to surgical excision following embolization. Intraoperative blood loss was minimal (approximately 200mls) and complete excision achieved. Subsequent pathology confirmed the tumor to be a benign hemangioma. With planned preoperative arterial embolization prior to surgical excision of a large hemangioma in a difficult location such as the central maxilla, we were able to avoid a major surgery with threat of exsanguination and resultant facial disfigurement to a relatively simple and almost bloodless procedure.



## CRANIOFACIAL INFILTRATING LIPOMATOSIS

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**Purpose:**
Craniofacial infiltrating lipomatosis (IL) is a rare overgrowth disorder in which mature lipocytes invade adjacent tissue accompanied by an increased number of blood vessels and nerves. Other features are skeletal hypertrophy, premature dental eruption, and regional macrodontia. Regrowth after surgical resection is common. Natural history, etiology and management are unknown.

**Methods:**
We documented clinical features, radiographic findings, response to resection, histopathology and analyzed for the RET mutation in 10 patients with craniofacial IL.

**Results:**
Diagnosis was made in infancy (6 males, 4 females) with involvement of the cheek (n=9) and chin (n=l). All patients had lateral facial hypertrophy and 6 had an ipsilateral capillary blush of the overlying skin; 5 patients had ipsilateral macroglossia; 5 had mucosal lesions and a histopathologic diagnosis of "mucosal neuroma." Overgrowth of the underlying bone and early eruption of ipsilateral deciduous and permanent teeth was observed in 8/10 patients. MRI showed excess soft tissue of fatty density and CT confirmed skeletal overgrowth. Repeated resections were done on 4 patients (mean = 2; range 0 - 6) and regrowth occurred in all 4 cases.

**Conclusions:**
Given the likelihood for post resectional regrowth, future management requires understanding of pathogenesis. We hypothesize a somatic mutation leading to a localized increase in growth factor(s) and studies are being done to test this theory.

## LONG-TERM FOLLOW UP OF VASCULARIZED BONE GRAFTS IN CHILDREN WITH TREACHER COLLINS SYNDROME

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**Introduction:**
Since 1982 the use of vascularized calvarial bone grafts is our first choice in reconstructing the bone defects in the malar region in Treacher Collins syndrome in children.

**Material and methods:**
Between 1982 and 1995 eleven children with Treacher Collins syndrome underwent vascularized calvarial bone reconstruction. Of eight children, mean age at operation 7.4 years, (range 2.4-15.5 years) post operative CT-scans were available. Total number of vascularized bone grafts 15. In 7 patients bilateral and in 1 patient unilateral malar reconstruction was performed.ln 3 patients the composite bone flap was taken from the frontal bone and pedicled on the temporal muscle and deep temporal artery. In the remaining 5 patients the graft was taken from thfi parietal bone pedicied on the galea and superficial temporal artery.To determine bone resorption or bone growth, measurements of the bone thickness of the bone graft and of the donorside of the skull were performed on CT-scan slices. The maximal thickness was measured in a plane rectangular to its longitudinal axis. To compare the bone thickness of the graft with the bone thickness of the donorsite a ratio was defined. Ratio < 1 indicates growth less then the calvarial donorsite suggesting resorption. Ratio > 1 indicates that thickness of the bone graft exceeds the bone thickness of the donorsite which means bone (over) growth.

**Results:**
None of the bone grafts had a ratio < 1, no resorption compared to the donor area. 10 out of 14 bone grafts had a ratio > 1 (1.2-1.8). This means that the growth of the bone graft exceeds the normal bone growth.

**Conclusion:**
In this retrospective study we demonstrated that vascularized calvarial bone grafts for malar reconstruction do not undergo resorption but show in majority growth and even overgrowth on the long-term. Also remodelling of the graft can be seen, adapting to its localisation in the orbital region.



## HEMIFACIAL MICROSOMIA: A LONG FACE SYNDROME

*CB Gordon, MD, XP Reyna Rodriguez, DDS, F Ortiz Monasterio, MD*

Hemifacial Microsomia (HFM) has been tradifionally thought of as a unilateral facial hypoplasa which affects both bone and soft tissue. Reconstruclion has focused on the mancKbular defomlily: maBollary involvement is less well understood. Clinical observations gained over 350 cases seem to contradict the concept. Most patients had stigmata not of urotatenal vertical deficiency, but of bilateral vertical maxillary excess. To test this hypothesis, 25 skeletally immature and 19 adult preoperative patients with HFM were paired with age-matched controls. They underwent cephalometric analysis with tiadifional and unoithodox measurements. These data were analyzed using multivariate analysis.

Immature A/P analysis showed nasal height (a measure of superior maxillary height) to be significantly increased by 29% ($p<0.001$). Mean ocdusal plane distance and midtacial heights were significantly reduced from controls on the affected side, and not signfficantly different from controls on the nonaffected side. Since these reflect dentoatveolar compensations, lower maxillary height, angular maxitery height and upper maxillary height were evaluated in the lateral ceph. These do not reflect dental compensations, and were all significantly increased compared with controls ($p<0.001$). Mean angular maxillary height, lower maxalafy height, and upper maxillary heights showed 13%. 18%, and 13% increases, respectively.

Adult A/P analysis was similar nasal height was increased by 6% ($p<0.001$). OPD on the affected side was equivalent to controls where it was shorter in the immature group, and the nonaffected side was augmented by 16% ($p<0.001$ ). Midfacial heights were 9% increased on the affected side, and 27% increased on the nonaffected side. Lateral ceph data revealed angular maxillary height increase of 14%, lower maxillary height increase of 24%, and upper maxillary height increase *of* 14% ($p<0.001$ for all groups).

Therefore, immature patients start with mildly long faces with severe dental compensations; maxillary growth leadis to very long facies by adolescence. Many measurements are increased by 5-6 SD. Dentoalveolar compensations did not account for much of increased maxillary height. The centrofacial triangle did not significantly vary in size from controls to experimentals. The lower maxilla seems to rotate and grow downward to seek contact. The greatest height increases were in grades I and II, but in many measurements, the "affected" side was longer than controls in all grades. This may indicate that the pathologic process affects eariy maxillary growth, but does not limit its growth potential. The lower and mid-maxilla appear to grow vertically until occlusion is achieved, regardless of grade of deformity. This phenomenon is being harnessed in the pediatric group with bite blocks and functional appliances to achieve occlusion. Knowledge of this propensity for maxillary overgrowth may have implications for current surgical strategies.

## MICROTIA RECONSTNICTION USING A POLYETHYLENE IMPLANT: AN EIGHT YEAR SURGICAL EXPERIENCE

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In the hands of a relatively few very experienced surgeons, microtia reconstruction using rib cartilage can give consistently excellent results. However, the results obtained by many surgeons can be variable and often disappointing. We became interested in performing alloplastic ear reconstructions and began using polyethylene frameworks for microtia patients in 1991. We present our experience with this technique in a large group of pediatric patients.

All patients having microtia reconstruction with a polyethylene implant by this author were reviewed. The surgical technique and implant design used were noted. Outcomes with respect to final appearance, implant exposure, infections, implant fracture, and length of hospitalizations were tabulated.

A total of 116 patients were reconstructed during an eight year period. The first stage was done on patients with an average age of 4.7 years and required an average hospital stay of 1.2 days. All subsequent surgeries were done on an outpatient basis. Because the implant and surgical technique was modified with the gain of experience during the study period, we compared two subgroups of 25 consecutive patients within our series. Each group had a minimum of at least two years post surgical follow-up. Between 1992 and 1994, patients were reconstructed using our earlier implant design and technique. There was a 44% incidence of implant exposure and a 25% incidence of frame fracture in this group within two years of reconstruction. These patients were compared to a second group of 25 consecutive patients treated from 1995 to 1996 using our modified technique. In this second group, there were no exposures and no frame fractures. The appearance and projection of the reconstructed ears were consistently better in the later group.

This study shows that our current technique of microtia reconstruction using a porous polyethylene framework gives improved aesthetic outcomes and significantly reduces postoperative complication rates compared to our *older* technique. The advantages of this method over rib cartilage reconstruction are numerous and include: 1) a shorter hospital stay, 2) fewer reconstructive stages (two), 3) no chest donor site morbidity, 4) the ability to reconstruct children at a younger age, and 5) a shorter learning curve for the surgeon. Although longer follow-up will be needed, the porous polyethylene framework may be an excellent reconstructive option for patients requiring total ear reconstruction.



## MANDIBULAR DISTRACTION-OSTEOGENESIS IN TREACHER-COLLINS-SYNDROME. PRO AND CONTRA

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Among others Treacher-Collins-Syndrome is characterised by a steep mandibular base, an anti SPEE'S curve, an open bite and mandibular retrognathia, caused by a more or minor pronounced hypoplasia of the ascending mandibular ramus. Mandibular advancement and simultanious anterior rotation is requested for functional and occlusal rehabilitation. In moderate cases mandibular-rotation-advancement procedures of the mandibular body (i.e. Peninsula mand. body osteotomy) lead to satisfactory results with excellent long term stability in adults.

In early childhood and more severe cases bone-expansion-techniques are requested to avoid severe funktionel disorders during growth. While distraction-osteogenesis for lengthening the mandibular body leads to satisfactory results and long term stability, the expansion of the hypoplastic ascending ramus in vertical direction does include remarcable risks of relaps, caused by permanent acting forces of the masticatory muscle sling, a phenomen, which is very well known from sagittal-split procedures simultaneously with mandibular anterior rotation in orthognatic surgery. Clinical examples additionally will explain these principles.

## CLINICAL EXPERIENCE OF NASAL RECONSTRUCTIONS IN SYNDROMATIC PATIENTS

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Nasal reconstruction in syndromatic patients is one of the difficult surgical procedures. The severity of the deformities vary from minimal degree such as tip dimple, bifid nose to severe degree, underdeveloped nasal framework. The reconstruction will have three components, mucosa, nasal framework and soft tissue coverage.

We have reconstructed some of the noses in syndromatic nasal deformities such as Binder syndrome, facial clefts, frontonasal dysplasia etc. Using the autologous tissue is preferred but still has some disadvantage. One of them had nasal framework collapse due to bone graft resorption and required re-operation.



## TISSUE EXPANSION IN THE RECONSTRUCTION OF TESSIER CRANIOFACIAL CLEFTS - A SERIES OF SEVENTEEN PATIENTS

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**Purpose:**
Atypical craniofacial clefts, as classified by Tessier in 1973, manifest the most extreme examples and variety of craniofacial dysmorphology. These clefts are characterised by hypoplasia of soft tissue and skeletal elements throughout the three dimensional extent of the cleft. Whereas bone grafting and craniofacial osteotomies have been successful towards correcting the underlying skeletal abnormalities, the ultimate success of these reconstructions has been limited by the deficiency of skin and soft tissue. Craniofacial tissue expansion has been utilized to reconstruct these facial clefts with like quality tissue, allowing for tension free reconstruction following osteotomy and bone grafting.

**Methods:**
We have incorporated craniofacial soft tissue expansion in the management of seventeen patients with Tessier craniofacial clefts ( five patients with bilateral Tessier no. 4 clefts, two patients with unilateral Tessier no. 4 clefts, four patients with Tessier no. 0-14 clefts, and one patient each with Tessier no. 2-12, 3-11, 4-10, 6-9,3-9-12, and 12-13-14 clefts). Tissue expansion has been used in the primary correction of facial clefts in eight patients, with nine patients undergoing expansion prior to secondary revisionary surgery. The mean follow-up time for the patients is five years (range one to ten years). Representative cases have been included with the abstract.

**Summary and Conclusions:**
Reconstruction of Tessier clefts involves the restoration of skeletal, soft tissue, and skin elements to an area of craniofacial hypoplasia. Previously reported techniques have highlighted the osteotomies and bone grafting necessary to correct the underlying skeletal abnormality and have attempted to address the soft tissue deficiency by means of local transposition flaps; these often prove to be insufficient in moderate to severe facial clefting. In our series, tissue expansion has evolved as an important element in overcoming the skin and soft tissue deficiency associated with these clefts, allowing for tension free closure and improved aesthetic results in these surgically challenging patients.

## APLASIA CUTIS CONGENITA: A RARE ANOMALY

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Two male patients were diagnosed with Craniofacial Aplasia Cutis Congenita from 815 registered Craniofacial patients since 1985.

The first patient was seen on day one in 1989 after a terminal forceps delivery with a scalp and skull defect measuring 11 x 11 cm and severe convulsions due to trauma at birth. Split thickness skin grafts were applied to the dura on the same day. By 3 months the mile stones were normal and the convulsions have ceased. At one year it was evident that new skull bone has partly formed under the transplanted skin.

In 1996 he started in a normal school and in December of that year, a Latissimus Dorsi free flap was done followed by split rib grafts in 1997.

Two Croissant tissue expanders were inserted in 1998 to advance hair bearing scalp with an excellent result.

Patient two presented at age 24 in 1997 with a deviated nose, aplasia of forehead skin, alopecia and a depression in the external table of the frontal bone. The patient was successfully treated with croissant tissue expantion and scalp advancement.



## QUADRIPARTITE FACIAL OSTEOTOMIES: SYNCHRONOUS CORRECTION OF ASYMMETRIC HYPERTELORISM AND MALOCCLUSION SECONDARY TO MAJOR FACIAL CLEFTS

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**Purpose:**
Standard craniofacial operations maintain the continuity of orbit and maxilla-zygoma as either a full face ("monobloc" or LF III) or hemiface ("facial bipartition") osseous unit. While this is adequate for most patients with craniofacial anomalies, occasionally there is enough discrepancy between the orbits and the occlusion that neither can be optimized in continuity. Independent mobilization of each orbit and hemimaxilla allows for such optimization in a single operation. Two cases are reported having been managed with such a novel approach. Patients: Case I (USA) - 9-year-old boy with a partially repaired left Tessier #3-l 1. His residual dysmorphology included: frontal recession, asymmetric hypertelorism, maxillary retrusion, maxillary arch collapse and anterior open bite. Case 2 (Taiwan) - 10-year-old girl with frontal encephalocele and bilateral asymmetric facial clefts (Tessier # 2-12 and 3). Her residual dysmorphology included: frontal calvarial defect, asymmetric hypertelorism, partially unrepaired facial clefts, maxillary hypoplasia, and "V" maxillary arch deformity with anterior open-bite.

**Methods:**
High resolution thin slice CT data was obtained for each patient to define the aberrant anatomy, simulate surgical options and provide a definitive intraoperative surgical "blueprint". After graphics workstation surgical simulation demonstrated the inability of facial bipartition to optimize both orbital position and occlusion for Case 1, a novel quadripartite partition was designed, simulated and then executed clinically for one-stage total reconstruction of frontal calvarial, orbital, nasal, and maxillary deformities. Having had the experience of Case 1, when Case 2 presented five years later it was planned and executed similarly, half a world away.

**Results:**
In both cases, the surgery was performed uneventfully according to the simulator "blueprint". In the second case, postoperative infection occurred which was successfully controlled with debridement of part of the calvarial and nasal bone grafts and antibiotic treatment. For both patients, the follow-up CT scan documented satisfactory frontal, orbital, and maxillary reconstruction. Ocular globe translation did not correlate well with osseous orbital movement.

**Conclusions:**
Quadripartite facial osteotomies allow optimization of orbital and occlusal deformities associated with facial clefts and asymmetric hypertelorism.

## ANALYSIS OF 361 CASES OF CRANIOFACIAL CLEFT IN CHINESE CHILDREN

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**Objective:**
This study is aim to indicate the character of craniofacial cleft in Chinese patients.

**Material and Method:**
There were 182 cases in this series. According to Tessier's classification, from No. 0 to No. 14 craniofacial cleft were used to describes the deformities around orbit which including cranial, orbital, and maxillofacial regions.

**Result:**
No. 0-14 cleft and No. 1-13 cleft associated with hypertelorism were most common in this series. Then No. 3 and No. 4 cleft were seen in our clinic. No. 8 and No. 9 were rare seen in this study. We found a few of craniofacial cleft cases combined with cleft lip or cleft plate.

**Conclusion:**
There were a little bit difference between Chinese patients and Western patients in the craniofacial cleft.



## SONOGRAPHIC DIAGNOSIS OF MAJOR CRANIOFACIAL ANOMALIES IN THE FETUS

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Five percent of newboms have a birth defect; the incidence is even higher by examination of aborted embryos and ultiasonography offetuses eliminated through natural selection. We reviewed the literature and our experience with prenatal ultrasonographic examination of the 4 maJor categories of craniofacial anomalies. Three issues were addressed: 1) age at which diagnosis is possible, 2) detection of associated anomalies, and 3) the possibility of molecular diagnosis.

***Craniosynostoses***
Cranial sutures can be imaged by 3D uitrasonography as early as 13 weeks gestation, thus isolated and eponymous (syndromic) craniosynostosis can be diagnosed from 16 weeks. Earlier detection of a syndromic fetus is by targeted (level 2) examination, presence of associated anomalies (such as limb deformities), and a positive family history. For the known mutations, the diagnosis is confirmed by chorionic villus sampling (CVS) or amniocentesis.

***Syndromes with Hypertelorism***
Both orbital and interorbital hypertelorism can be differentiated by standardized measurements and subcategorized as frontonasal malformation, craniofrontonasal dysplasia (CFND), paramedian facial clefts, encephalocele, and miscellaneous.

***Pharyngeal arch anomalies***
Auricular shape can be graded from 16 weeks gestation to term, permitting prenatal diagnosis of microtia and anotia. Micrognathia is measured in lateral view and compared to normal length. Hemifacial microsomia is confirmed by orbital, auricular, and mandibular anomalies. Associated CNS, skeletal, cardiac, and genitourinary anomalies (expanded spectrum) must be evaluated. Treacher Collins syndrome can be diagnosed by sonographic detection of downturned palpebral fissures, bilateral microtia, micrognathia, cleft palate, and molecular identification of the "Treacle" mutation (chromosome 5q).

***Oromandibular anomalies***
The two most common causes of syndromic Robin sequence can be determined by CVS and amniocentesis: Stickler syndrome (type 2 and type 11 collagen mutation) and velocardiofacial syndrome (VCFS, chromosome 22q11 deletion), Stickler syndrome has no extracranial prenatal sonographic findings whereas VCFS fetuses typically exhibit microcephaly, micrognathia, and characteristic cardiac defects.

***Conclusions***
1) Craniofacial anomalies can be prenatally diagnosed by second trimester level 1 sonography (72 % detection) but are more accurately detected by level 2 targeted examination.
2) Presence of associated extracranial deformities can increase detection of major craniofacial anomalies to 100%.
3) Fetal imaging is compromised by maternal obesity, fetal position, and miscellaneous conditions such as oligohydramnios.
4) First trimester diagnosis of known genetic mutations is possible by CVS or amniocentesis.

## THE FETAL CLEFT PALATE: MIDFACIAL GROWTH AND BONY PALATAL CHANGES FOLLOWING *IN UTERO* REPAIR OF A CONGENITAL MODEL

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**Introduction:**
Recently, we characterized a congenital model of cleft palate in the goat. Induction of clefting in this caprine model was achieved in 97% of fetuses following gavage with anabasine, a teratogenic compound extracted from *Nicotiana glauca.* Subsequently, we demonstrated scarless palatal healing and the development of a dynamic bilaminar palate following *in utero* cleft repair. The purpose of this study is to demonstrate the influence of *in utero* palatoplasty on midfacial growth and bony palatal changes in this congenital model.

**Methods:**
At 85 days gestation 8 clefted fetuses underwent *in utero* cleft palate repair using a modified von Langenbeck technique; 8 fetuses remained as unrepaired controls. All goats were sacrificed at 6 months of age and dry skull measurements were performed. Eleven direct measurements and 6 derived indices were determined for each skull. Six 6-month-old imciefted normal goats served as controls.

**Results:**
Repaired clefts demonstrated a reduction in bony cleft width of 48% anteriorly and 60% posteriorly compared with unrepaired clefts. Three of the eight repaired clefts demonstrated partially fused bony clefts. Cephalic length was decreased and cranial base width was increased in the clefted goats compared with controls; however, no significant difference in these measurements was observed between repaired and unrepaired clefted goats. Maxillary width of repaired clefted goats was decreased compared with both unrepaired clefted and control goats. Maxillary length was similar in all groups

**Conclusion:**
*In utero* cleft palate repair did not produce significant changes in facial growth compared with unrepaired clefts or controls. Partial palatal fusion in 37% of repaired clefts and bony cleft narrowing in all repaired clefts may account for the decreased maxillary width in this group. Scarless palatal healing may account for the unimpaired maxillary growth observed following cleft repair. A decrease in cephalic length and increase in cranial base width in both clefted groups, in the presence of unimpaired maxillary growth, suggest an intrinsic growth disturbance involving the cranial base.



## AN OPTIMIZING CT-BASED CRANIOFACIAL SURGICAL SIMULATOR FOR THE WINDOWS™ PERSONAL COMPUTER

*Court Cutting, Deljou Khoramabadi, Barry Grayson, and Joseph G McCarthy*
*New York University Medical Center*

**Introduction:**
For the past ten years our unit has used a CT-based simulator to plan surgery for our craniofacial patients. This program numerically optimizes the position of each bone fragment to best fit an age, race, and sex matched average normal skull. These plans can be performed by hand or output to a module which allows virtual reality techniques to be used in the operating room for execution with greater precision. Until recently this software required an expensive, high performance graphics workstation which was economically and technically out of reach for many craniofacial teams.

**Materials and Methods:**
A standard Microsoft Windows™ personal compute with a Pentium processor and 16 megabytes of RAM was used. Although any graphics card can be used, the new generation of OpenGL© accelerator cards are much preferred. The software was rewritten from its original UNIX base to conform to the familiar Microsoft Windows™ user interface using a Visual C++ 6.0 compiler. All code was carefully re-examined and rewritten if performance improvements were possible.

**Results:**
The advent of the OpenGL three dimensional graphics accelerator card permits "realtime" movements of the skull and bone fragments at an exceptionally low cost. The PC-based simulator described outperforms the original software run on compute that cost ten times as much.

**Discussion:**
The mass market for three dimensional compute games has resulted in a dramatic drop in the price of powerful three dimensional compute graphics. Numerically optimized CT-based craniofacial surgical planning can now be performed on an inexpensive PC using the familiar Windows™ interface.

## MANDIBLILAR RECONSTRUCTION USING A NEW COMPUTERIZED 3D SURGICAL PLANNING SYSTEM

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Mandibular reconstruction presents a challenge to the surgeon because of the 3-dimensional shape of the jaw. Free vascularized fibula transfers have become the method of choice for the majority of mandible defects. Precise shaping of the straight fibula gives the best-quality esthetic results. Usually four osteotomies are necessary in order to turn the straight fibula into a three dimensional shape that has the same dimensions as the missing mandible. Care needs to be taken so that the periosteal blood supply to the mandible is preserved. After the osteotomies are made, a significant amount of time is spent to burr the bone edges for additional angle precision.

We have developed a 3D surgical planning system that enables the surgeon to create blueprints for the fibular osteotomies. Preoperative CT scans of the face are reconstructed into high quality surface meshes and images are then visualized in a virtual environment. The mandible is disarticulated from the remaining skull and virtual surgical tools are used to make osteotomies and "unfold" the real mandible into a linear structure. The angles and length of straight bony segments are measured. Using a generic model of a 3D reconstructed adult fibula, these angles and linear segments are mapped and virtual osteotomies are automatically performed. Diamond-shaped transparency printouts function as actual blueprints and are taken to the operating room. These shapes are positioned over the actual fibula where marks are made for the proposed osteotomies.

Extremely precise osteotomies are possible using this method, resulting in optimal esthetic results for the patient. A number of clinical photographs of actual patients will be presented to illustrate the technique.



## THE INTERNET AND PRIVACY

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The Internet has permitted an exponential increase in data sharing. Files containing patient data can be easily shared and transferred from investigator to investigator. The Internet is, however, far from secure. The open systems make it easy for eavesdroppers to glance at the information sent through this modality. Examples of misused craniofacial images and patient data already exist on World Wide Web sites.

I will discuses the security flaws in the Internet and methods to insure security including a recommendation to members of this organization.

## THREE DIMENSIONAL DIGITAL PHOTOGRAPHY: A POTENTIAL NEW TECHNIQUE IN FACIAL ANALYSIS

*Greg M. Galdino, M.D., Eric Chang, M.D., & Craig A. Vander Kolk, M.D.*
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A new camera has recently been developed by Genex Technologies, Inc. that provides real time three dimensional digital images for analysis of shape, surface characteristics and volume. This camera was evaluated for use in facial analysis in craniofacial surgery.

The Rainbow 3-D camera is an eye safe, no laser camera which uses visible light to capture two-dimensional color and three dimensional information simultaneously and in real time (>30 frames/second). It has no moving parts and is relatively easy to use because of the speed in image acquisition. Software for rendering 3-D images is supplied with the camera and can run on a desktop computer. Images are input directly into the computer form the camera and rendered in less than 2 minutes.

Initial trials with the camera revealed it to be useful in the evaluation of the breast for reconstruction, augmentation and reduction. Comparisons with CT and MRI surface analysis and volume analysis are being performed.

Three dimensional digital photography and analysis with a camera like the one being presented hold the key to further understanding the results of craniofacial reconstruction.



## ENDOSCOPIC ASSISTED FOREHEAD RECONSTNICTION USING HYDROXYAPATITE CEMENT

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*Primary Children's Hospital, Salt Lake City, Utah*

The ability to reconstruct craniofacial defects utilizing hydroxyapatite cement has been shown to provide excellent results. Coronal incisions have been the surgical approach of choice, but now the ability to use the endoscope has provided a new approach to reconstructing defects of the forehead, temporal areas and supraorbit.

A total of 21 patients underwent an endoscopic assisted reconstnrclion of the forehead during a 17 month period (July 1997 lo Dee 1998). The average age at the time of operation was 9.5 years with a range of 5 yrs to 22 yrs. There were 13 males and 8 females. Average length of follow-up was 7 months with a range of 2 months to 17 months. The typical defect encountered was a temporal depression and some recession along the lateral supraorbit. The most common group was patients who underwent craniosynostosis correction during the first year of life with progressive recession along the temporal regions. Other diagnoses include posttranmalic deformities of the forehead and Rombergs Disease ("coup de sabre") deformity. There was one patient who presented with a significant deformity of the lateral forehead and supraorbital ridge from a previous excision of a dermoid cyst during early childhood. The average amount of Bone Source utilized per patient varied from 5 grams to as much as 25 grams depending on the nature and extent of the deformity. The typical time to perform the procedure was from 45 minutes to one hour. Maintenance of the contour and stability was excellent in all patients. Delivery of the Bone Source has been improved by increasing the ratio of solvent to the Bone Source and by providing a better mechanism for delivery. Thus far, we have not seen any resorption or shifting of the material, although it is still eariy to adequately assess the potential resorption and maintenance of the reconstruction. There have been no infections and only one seroma. This seroma was resolved with immediate drainage and compression dressing with no obvious loss of contour. There were 2 patients who were slightly under-corrected in portions of the original defect but who have not required further augmentation. These were early in the experience of the authors and were in areas that required further augmentation along the inferior temporal region. There were only 2 patients who demonstrated some minor amount of over-correction and only one required removal of the excess under local anesthesia in the office. There were also some irregularities that became more evident postoperatively and this was felt to be secondary to the rapid setting of the material before adequate contouring by finger pressure could be performed.

## ENDOSCOPICALLY ASSISTED REPAIR OF ORBITAL FLOOR FRACTURES

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Traditionally the orbital floor fractures were repaired with standard transcutaneous or transconjunctival approaches. Complications such as external scarring, eyelid edema, ectropion, entropion and granuloma formation have been associated with these traditionally open lid techniques. The endoscope has been used to assist in orbital floor exploration, reduce entrapped orbital tissue and identify the posterior shelf for implant placement; however, an open lid incision was inevitable to be used for implant placement when necessary. We present our experiences in repairing the orbital floor fractures using transsantral endoscopy without open lid incision.

A 0-degree, 4-mm endoscope was placed through a 2 x 1.5 cm2 maxillary antrostomy to dissect sinus roof. The endoscope was used for reduction of floor fracture and prolapsed orbital tissue into orbital cavity if present. The orbital floor defect was reconstructed with titanium mesh through the antrostomy under the endoscopic control. The mesh was fixed with microsrews to avoid dropping from the sinus roof. This technique was applied to 8 patients who had orbital floor fractures. One of the eight patients had zygomatico-orbital fractures, and the others had isolated orbital floor fractures. One patient received reduction only and the remaining 7 patients had titanium mesh reconstruction.

The patients were followed up for 4 to 12 months. The enophthalmos was corrected in all patients except one had mild enophthalmos due to uncorrected orbital medial wall fractures. Preoperatively, three patients had diplopia which resolved in two patients and improved in one postoperatively. There have been no complications apart from a transient anesthesia in the dennatome of the infraorbital nerve. The technique successfully reconstructed the orbital floor defects, minimized ocular globe manipulation and eliminated the need for lower eyelid incision.



## ENDOSCOPIC MONOBLOCK OSTEOTOMIES USING A NEW HIGH SPEED MICRO MALLEABLE DRILL

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**Introduction:**
Recent years have seen many innovative approaches to craniofacial surgery. These techniques have included distraction osleogenesis, endoscopic approaches and resorbable fixation devices. Much of credit goes to the manufacturers and their engineers working in concert with the surgeons. The evolution of craniofacial surgery by minimal exposure has been hindered by traditional power instruments. A prototype high speed micro malleable drill has been developed for this use. Ten cadavers and two clinical cases are discussed.

**Methods:**
Ten fresh cadavers heads were collected at the University of Brno. (CZ), Three 2.5cm access incisions were made in the hair bearing scalp. A subperiosteal dissection was performed of the forehead and facial skeleton. Burr holes were performed at the locus minoris resistentiae of Bishat. through the lateral incisions and parasagittally through the central incision. Dural elevation was completed in standard fashion. A protected bit on the malleable drill was used to create the frontal osteotomy. With the dura protected by a malleable retractor and the orbital contents protected by a malleable retractor placed through the upper eyelid, the osleotomy across the orbital roof was performed using side cutting bit on the malleable drill. This osteolomy joined the previous one laterally. Extracranially the drill was used to make the osteolomy from the pterionai region, lateral to the lateral orbital rim, to the pterygomaxillary junction. Again the orbital contents were protected. The orbital floor and medial wall were cut through a transconjunctival approach using the Midas Rex TCA tool. The frontoethmoid osteolomy was done while intracranial contents were protected and observed with the endoscope. The zygomalic arch was osteotomized through the temporal incision. Rowe forceps were used to perform lhe dysjunction and advancement. After each procedure, the dura was carefully inspected. The dura in each case was seen to be completely intact. With this experience completed. 2 clinical cases were then done. The first case is a five year old boy with recurrent plagiocephaly. In this case, a coronal incision was used. The second case was a 3 year old boy with trigonocephaly performed through the previous described incisions.

**Results:**
No complication resulted from this technique, the new malleable drill proved to be well designed for such procedure.

## WHAT LATERAL CANTHOPEXY CAN AND CANNOT DO

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Recently with increasing use in cosmetic procedures, and in more complex craniofacial procedures being done through the lower eyelid, lateral canthopexy has received increased attention. There are benefits from lateral canthopexies which haven't been emphasized, as well as short comings which need more emphasis. Based on an experience with more than 3,000 lateral canthopexies over the past 25 years, what can and cannot be expected from a lateral canthopexy has become clarified.

The four things that the procedure will accomplish in decreasing order of effectiveness are as follows: 1) alter position of the palpebral fissure; 2) change the shape of the palpebral fissure; 3) tighten the eyelid margins on the eye; 4) decrease scleral shown.

What lateral canthopexy cannot effectively do is to selectively tighten the lower lid margin on the eye. It is also not as effective as a v-shaped wedge or tarsal strip for lower lid laxity or ectropion. It is more effective than those two procedures in exophthalmos or exorbitism. Finally, for canthopexy to have a real effect, the canthus must be detached and reset. Otherwise, the procedure is extremely short lived, and of little or no effectiveness. Benefits, limitations and possible problems with canthopexy will be discussed.



## AESTHETICAL LE FORT IV MINUS LE FORT I DISTRACTIVE ADVANCEMENT

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Recent advances of distractive surgery for craniofacial bones are creating new Line of osteotomy as well as the new possibility of indications for patients who are seeking aesthetical improvement for the craniofacial region.

In this case, without taking out the cranial bone, a line of osteotomy was carried out on the cranium and zygomatic arch and fall range of orbital wall osteotomy in which osteotomy of the orbital roof was carried out through a creating bar hole which located just superior to the front zygomatic suture and the frontal sinasis. After the serious of osteotomy, four distraction devise were applied to on each side, two on each side a block of the cone is distracted anteriorly.

In this presentation, technical detail and the result will be presented.

## ONE STAGE CORRECTION OF RADIATED ORBIT AND CONTRACTED EYE SOCKET USING ORBITAL OSTEOTOMY AND PEDICAL FLAP ROTATION BOTH POSTAURICULAR SKIN FLAP AND TEMPORAL FASCIAL FLAP

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**Objective:**
Patients suffered from retinoblastoma or other tumor in the eyeball in infant usually were underwent previous surgery and radiotherapy. It develops secondary deformities in the middle face and orbit such as contracted eye socket and orbitozygomatic hypoplasia.

**Methods:**
We employed a one-stage surgical reconstruction using orbital osteotomy and pedical flap rotation including both postauricular skin flap and temporal fascial flap. Orbital osteotomy was made along the lateral third of orbital rim (C-shape) and repositioning with 5 mm advancement. Postauricular skin flap and temporal fascial flap were elevated one by one, than both of them were rotated as eye socket reconstruction and cover of repositioning orbital rim.

**Results:**
In our 6-case experience the results were satisfying and encouraging without any complications, which are presented.



## FACIAL FORM ANALYSIS OF THE LOWER AND MIDDLE FACE IN YOUNG ORIENTAL WOMEN COMPARED WITH YOUNG CAUCASIAN WOMEN

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Nowadays facial skeletal contouring, which have developed in craniofacial surgery, acquired popularity in aesthetic surgery. On planning aesthetic surgery, patient's desire must be considered first. But it seems to be essential to have an objective standard of facial form. Anthropometric analysis, cepahalometric analysis, and photogrammetri analysis were developed for this purpose but with a limitation in three dimensional analysis. Bamett and Whitaker (1986) developed a simple three dimensional analytic system of middle and lower face. We performed the same analysis for young Oriental women (n=50) and age matched young Caucasian women (n=20).

We found some differences between young Caucasian women and Oriental women as follows:

1. Young Oriental women's middle' and lower face is wider than young Caucasian women's face in horizontal dimension.
2. Young Oriental women's middle and lower face is shorer than young Caucasian women's face in anterior-posterior dimension.
3. The difference in vertical dimension between the two groups is minimal.
4. In angular relationship, young Oriental women's anterior and inferior angles are larger than young Caucasian women's angles.

These findings are coincident with other author's observations that Oriental face is wider and rounder than Caucasian face and Orientals have more prominent malar area and mandible angle. This three dimensional facial analytic system will be useful in surgical planning and research for middle and loser face

## A ROLE OF MASSETER MUSCLE IN THE FIXATION AND RAHABILITATION FOLLOWING SAGITTAL SPLITTING OF THE MANDIBLE

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We have developed a new strategy of the fixation, and rehabilitation method following sagittal splitting of the mandible. Purpose of this study is to present a new method of fixation utilizing the masseter muscle and discuss the usefulness of an early functional rehabilitation of the masseter muscle in sagittal splitting. After the completion of sagittal splitting, the accending ramus is kept in the place without fixation then anterior part of the fascia of masseter muscle is sutured to the mandibular body indicating drill holes along the osteotomized line. Intermaxillary fixation is applied with an aid of barton bandage. The motion of chewing well is started under intermaxillary fixation at one week postoperatively. In the two weeks postoperatively, privilege of intermaxillary fixation will be given and then the dental occlusion is supported in position by using a chin cap.

This principle has been indicated in twenty-one cases during past three years and a satisfactory resuit has been noted in our series of patients. The motion of chewing well is a phenomena resuited in a synchronized contraction of the masticatory muscle in the central occlusion. In the contraction of masseter muscle, the ascending ramus is strongly affected by the inward displacement. This mechanism can be utilized in the postoperative management of sagittal splitting. This may allow us to conclude that the motion of chewing well may lead to early stabilization of the dental occlusion and accelerate the bony union in sagittal splitting.



## SPLITTING ADVANCEMENT GENIOPLASTY: A NEW GENIOPLASTY TECHNIQUE

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A new genioplasty technique has been described and performed on 16 patients since 1995. According to our technique, a rectangular part of the outer table of mentum is split away from the mandible, advanced and fixated to the mandible. The technique can be used for advancement cases only not for reduction genioplasty. The technique was performed on 16 patients with minor complications which included one wound dehiscent, one hematoma and one osteomyelitis which was managed by systemic antibiotic therapy. Esthetic results were found to be satisfactory.

At the late postoperative period, the new bone formation between the advanced segment and mandible was seen on the x-ray films. Advantages of the technique include having more contact surfaces for bony healing, a natural position of the lower lip, more natural projection of the menturn, tri dimensional movement possibility of the mentum and improvement in the soft tissue of the neck. The disadvantages ofth technique are the potential risk of infection due to dead space from the advancement, manipulation problems during the surgery and possible mental nerve injury.

Splitting advancement genioplasty was found to be a useful technique for advancement genioplasty and more physiologic osteotomy technique than most of osseous genioplasty techniques.

## SLIDING GENIOPLASTY: PREVENTION OF MENTAL NERVE INJURIES AND MASCULINIZATION OF THE CHIN BY OSTEOTOMIES

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The sliding genioplasty is considered by this author and some others as the preferred method for altering chin shape and position. Those, however, who cannot or prefer not to work with bone (unfortunately, the majority) prefer chin implants and argue that osteotomies frequently cause nerve injuries, primarily affecting lower lip sensation. Unfortunately, implants only augment. They cannot reduce the size of a chin, make it vertically larger or smaller nor horizontally smaller and therefore markedly limited in what they can accomplish. Previous published articles by this author discuss how to avoid mental nerve injuries and also presents a large series of consecutive sliding genioplasties without nerve injuries. This series, new 150 consecutive patients without a permanent injury, will be presented. The principals of this research and the surgery will be reviewed (see figures 1 and 2) showing this surgery to be safe.

Presently, the large, prominent, angular chin is desired and considered in vogue by many males (see figure 3). The chin implant, cannot delivery such a desired result. An osteotomy technique for creating such a chin has been developed by the author (see figure 4) and has been successful in a small, consecutive series of patients with marked patient approval. Representative cases will be presented (see figures 5, 6, 7, 8). Potential but unrealized complications will also be discussed.

It is hoped that by this presentation to encourage others to use this safe technique for chin esthetic surgery.



## THE PATH OF THE INFERIOR ALVEOLAR NERVE THROUGH THE NORMAL ADULT MANDIBULAR RAMUS: A QUANTITATIVE THREE-DIMENSIONAL COMPUTERIZED TOMOGRAPHIC STUDY

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**Overview:**
This study was undertaken in order to quairtitatively describe the path of the inferior alveolar nerve in the normal human mandible. The objective was to relate the path of the nerve to anatomic landmarks which are frequently used by surgeons who perform osteotomies of the mandible. The goal was to provide information to assist the surgeon in avoiding injury to the nerve at time of surgery, and to provide normative data which could form the basis for the study of the course of the nerve in dysmorphic mandibles.

**Materials and Methods:**
Computed tomographic scans of 10 normal patients (6 male, 4 female) were used as the basis for the study. Using AnalyzePC 2.5™ imaging software, the mandibles were segmented, and the position of the nerve was recorded within its osseous canal in the mandibular ramus on each axial slice in which it was identifiable. Distances were calculated between the nerve and the anterior, posterior, lateral, and medial cortices. The positions of the antilingual prominence, and lowest point on the sigmoid notch were also recorded. The position of the mental foramen was recorded in relation to the nearest tooth, and the 3D surface distances from the foramen to the alveolar bone, inferior border of the mandible, and the mandibular symphysis were determined. The distance from the entrance of the the nerve into the mandible to the antilingual prominence and the lowest point on the sigmoid notch were calculated. Summary statistics were obtained, comparing differences in gender.

**Results and Conclusions:**
The nerve was identifiable in each ramus over a mean distance of 12.7 mm. On average, the antilingual prominence was 0.3 mm higher on the caudad-cephalad axis than the point at which the nerve enters the bone, while the location of the lowest point on the sigmoid notch was 16.6 mm above the nerve. The average distances from the nerve to the anterior, posterior, medial, and lateral cortices were 11.6, 12.1, 1.8, and 4.7 mm respectively. Gender differences were significant for all of these except the medial cortex to nerve distance. On average, the mental foramen exited the body of the mandible immediately below the second premolar, and the average surface distances from the foramen to the symphysis, the most cephalad alveolar bone, and the inferior border of the body were 30.9, 14.2, and 19.3 mm respectively.

## FAT GRAFTING FOR CRANIOFACIAL DEFORMITIES

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**Introduction:**
Fat grafting, a popular esthetic procedure, can be utilized to improve craniofacial deformities. We present our fat grafting experience in patiens with craniofacial asymmetry and contour deformity.

**Patients and Methods:**
From 1996 to present, 16 patients from Los Angeles Children's Hospital underwent fat grafting for soft tissue augmentation. There were 7 male patients and 9 female patients. The age ranged from 1 to 24 years old. Diagnosis included hemifacial microsomia, Treacher-Collins, hemifacial atrophy, facial clefts, cleft lip, post hemangioma scarring, and post radiation deformity. Fat injections were performed under general anesthesia, often in conjunction other rcconstructive procedures. Areas of grafting included brows, periorbital region, check, mandibular border, chin, and lip. Fat was harvested from either abdomen or lateral thigh using 2mm cannula after infiltrating with epinephrine solution. The aspirate was centriftiged to separate serum and oil. The free fat was transferred to a 1 cc syringe and injected via a 18G blunt tip needle. The fat was deposited in different tunnels at the level of supraperiostcum muscle, and subcutaneous tissue. Slight overcorrections were routinely performed. Postoperative photographs were taken at regular intervals and were compared to preoperative records. Length of follow up ranged from 6 to 24 months.

**Results:**
The average amount of fat injected ranged from 3 cc's for the vermilion in cleft lip to 30 cc's for half of the face in hemifacial microsomia. Regions without scarring were easily expanded by fat injection and the contour obtained was more uniform. In contrast, fat injections into scarred region was more difficult and results were less predictable. 2 patients with hemifacial microsomia required additional fat grafting. There were no cases of infection or fat necrosis.

**Conclusion:**
Fat injection is a useful primary or adjunctive procedure for augmenting wide range of craniofacial deformities to improve symmetry. The augmented volume appears to be stable 2 months following fat injection. Fat absorption varied with location of injection and local tissue condition.

## THE POSTERIOR LEDGE IN THE MANAGMENT OF ORBITAL FLOOR FRACTURES

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**Introduction:**
Identification of an adequate posterior ledge in the treatment of orbital floor defects is sometimes difficult potentially compromising the outcome of floor reconstruction. The purpose was to study the relationship of the posterior orbital floor before and after the creation of a floor fracture and to investigate the presence of a posterior ledge in patients presenting with orbital floor fractures.

**Materials and Methods:**
Part A: Fine cut CT scan (axial, coronal, and oblique sagittal views) were performed through both orbits often fresh frozen cadaver heads before and after creation of standard bilateral orbital-zygomatic complex (OZC) fractures followed by radiological and anatomic morphometric analysis. Part B: Patients presenting with orbital floor fractures requiring surgical intervention were sorted by fracture oattem into four groups (a) isolated; (b) orbital-zygomatic complex; (c) midface, and (d) cranio-orbital. Anatomic analysis of the fracture pattern was performed at the time of surgery with attention directed towards the posterior ledge and correlated with CT images.

**Results:**
Part A: CT scan analysis of OZC fracture patterns were similar to those seen clinically. A posterior ledge was identified in all 20 orbits and was located 28.6 ± 4.9 mm (mean ± SD) from the inferior rim with 9.2 ± 4.1mm anterior projection from the posterior maxillary antrum. Distance from posterior ledge to the optic foramen was 24.3 ± 4.2 mm and the vertical height of the orbit at the posterior ledge was 25.2 ± 3.2 mm. Part B: To date, 13 patients have undergone 14 orbital floor explorations. Identification a posterior ledge was easy in 6 orbits, moderate in 4 orbits, and very difficult in 3 orbits due to ledge dimensions. No ledge was identified in 1 orbit. Details of CT scan analysis and treatment outcome will be presented.

**Conclusions:**
This study describes the patterns of the fractured posterior orbital floor and suggests that a sub-population of patients exist in which the identification of the posterior ledge is technically difficult due to anatomic confines of the posterior orbit and the size of orbital floor intact remnant. Implications with respect to treatment outcome will be discussed.

## DEGREE OF ENOPHTHALMOS IN RELATION TO ENLARGEMENT OF EACH WALL OF THE ORBIT IN MALAR BONE FRACTURE

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Using 3D CT we measured the degree of enlargement of the lateral, inferior and medial orbital wall as well as total orbital enlargement in malar bone fracture. The degree of enophthalmos was measured on CT. As was expected, enophthalmos increased proportionally with the increase of total orbital volume. The degree of enophthalmos co-related well with the degree of enlargement of the orbital floor when the enlargement of the medial wall remained less than 1 ml in volume. However, when medial wall enlargement exceeded 1 ml in volume, enophthalmos had no co-relation with the degree of orbital floor enlargement. The degree of enophthalmos did not correlate with the enlargement of medial orbital wall, which is the striking difference in comparison with the situation in blowout fracture. The downward and medial displacement of the eyeball in relation to each orbital wall enlargement were also studied and will be reported briefly.



## LATE RECONSTRUCTION AND REPOSITIONING OF ENOPHTHALMOS OF ORBITAL BLOW-OUT FRACTURES

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**Objective:**
To evaluate the surgical technique for late reconstruction and repositioning of enophthalmos of orbital blow-out fractures (BOF) and its efficacy.

**Methods:**
Using the computer orbital three-dimensional measuring technique, Hertel exophthalmometer, Lancaster test and autogenous bone and hydroxyapatite composite material. Late reconstruction of orbit and repositioning of globe were performed on 21 patients with exophthalmos of BOF, follow-up for an average of 11 months.

**Results:**
There is high correlation between the increment of orbital volume and the degree of enophthalmos, with a 1 cm square increment in bony volume of orbit causing 0.9 mm enophthalmos. In 21 cases with enophthalmos of Bof, 15 patients presented moderate enophthalmos (3-4 m) and 6 patients presented severe enophthalmos (5-6 m). 3 months after operation, the measurements demonstrated that 3 patients had no enophthalmos, 15 patients had mild enophthalmos (1-2 mm), and 3 patients had no effect. 16 of 21 patients had diplopia preoperatively. 8 patients had satisfactory result, and 8 patients and no change, which followed up more than 3 months after operation.

**Conclusion:**
Late reconstruction of orbit and repositioning of globe for enophthalmos of BOF may obtain satisfactory results.

**Key Words:** orbital blow-out fractures, enophthalmos, reconstruction, repositioning

## DELAYED CORRECTION OF POST-TRAUMATIC OCULAR DISPLACEMENT

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**Purpose:**
Post-traumatic ocular displacement is a potential complication of orbital fracture. The purpose of this study is to review the causes of this sequelae, describe the preoperative evaluation and surgical techniques for delayed correction, and examine the long-term surgical results.

**Materials and Methods:**
This study includes 25 patients with post-traumatic ocular displacement in the last 3 years. For all patients, the records and the serial x-ray films were reviewed. We carefully evaluated the three-dimensional positions of the globes and assessed the deformed orbital conditions. In terms of surgical technique, periorbital scar tissue was released, the malunited zygomatic bone was osteotomised and repositioned in 14 cases, and an oculoplasty was done in 28 cases for the restoration of the balance between the orbital content and bony surroundings with permanent correction of traumatic ocular deformities.

**Results:**
Among the many causes, initial inadequate surgery, with resultant deformities, was the most common contributing factor (56.0%). There were various patterns of initial facial bone fractures, and the orbitozygomatic fractures were the major contributing pattern in post-traumatic ocular displacement (68.0%). The long-term (mean 9.5 months) surgical results were satisfactory. The aesthetic problems were corrected or significantly improved in all patients except one. Regarding functional results, among the 7 patients with diplopia. one was resolved and five were improved.

**Conclusion:**
It is concluded that most ocular displacement could be prevented if the surgeon carefully reconstructed the orbit in his first operation. And delayed correction of this deformity could be possible if the surgeon considered the pathologic condition preoperatively, operated with complete exposure of the facial skeleton and retease of scar tissue, and provided a safe and stable correction.



## LONG TERM PHYSICAL IMPAIRMENT AND FUNCTIONAL OUTCOMES FOLLOWING COMPLEX FACIAL FRACTURES

*John A. Girotto, Ellen MacKenzie, Bradley Robertson, Paul N. Manson*

**Introduction:**
This analysis focuses on the impact of facial fracture on overall health status and quality of life. Specifically, we hypothesize: 1. There is a difference in the general health status by severity of facial fracture; and 2. Comminuted midface fractures represent a subset most prone to long term complications.

**Methods:**
Study patients were required to 1. have sustained significant blunt trauma necessitating admission, 2. present with a Glascow comma scale score of > 8 and 3. have sustained a Le Fort fracture. These patients were matched to a control population by 1. year of injury 2. age and 3. by the ISS. All case and control patients were surveyed with the SF-36, GHQ, SAD, and BSS. This information was supplemented with questions focused on residual head and face symptoms.

**Results:**
Between July 1986 and July 1994, 25,758 patients were admitted to the University of Maryland Shock Trauma Center. 2,964 of these patients sustained facial and 190 meet inclusion criteria and were successfully contacted. When comparing overall Le Fort groups to the control matched population, there is a notable trend in all SF-36 scores toward decreasing social functioning, decreasing body/head satisfaction, and increasing anxiety with increasing severity of facial trauma. Patients sustaining comminuted Le Fort injuries reported lower scores in the SF-36 categories of physical limitations, social functioning, and mental health which reached statistical significance. There was a trend for those patients sustaining complex facial trauma to report a greater percentage of abnormal GHQ scores and a higher dissatisfaction with their facial appearance as indicated by the BSS. Similar relationships are found with SAD scores. Over half (53.3%) of the comminuted Le Fort patients reporting abnormal SAD scores.

**Residual Impairment:**
The percentage of patients reporting residual impairment increased with increasing complexity of facial fracture in the areas of visual problems (56%), alterations in smell (47%), difficulty with mastication (40%), and epiphora (45%) and these are statistically significant. Most importantly, 21.33% of patients sustaining comminuted Le Fort injuries report disability from their injury preventing work while only 11% of the control patients – with similar presenting ISS and GCS scores – report injury related disability.

**Conclusion:**
This work demonstrates that patients sustaining comminuted Le Fort level facial fractures report poorer health outcomes than patients with less severe facial injuries and substantially worse outcomes than population norms. It is also this population that reports the greatest percentage of injury related disability preventing employment at long term follow up. (21.33%)

## SOCKET CONSTRUCTION AND RECONSTRUCTION

*Jorge Latoni, S Anthony Wolfe*

Diagnosis m this series of 46 patients requiring socket construction or reconstruction included congenital malformation, and post-traumatic and oncologic deformities.

Congenital irucrophthalmias often required expansion of the bony orbit prior to eyelid reconstruction, and socket construction.

In cases where an eye was removed in infancy, particularly if followed by radiotherapy, bony expansion was also frequently required, often with a temporalis muscle flap.

Mucosal grafts (buccal, split palatal, pharyngeal) were preferred, but skin grafts were often necessary for larger defects due to size considerations.

An inadequate lower cul-de-sac was the most common cause for reoperation.

Close collaboration with an experienced ocularist is essential.



## SECONDARY NASOETHMOID REPAIR

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A series secondary nasoethmoid fracture repair is presented emphasizing a practical decision tree classifying bone and soft tissue problems. Techniques of reconstruction applicable to difficult defects are emphasized, such as those fractures with lining loss, cutaneous loss, lengthening the short nose, soft tissue management, repositioning and fixation of soft tissue and canthopexy. Options for replacement of lining and cutaneous reconstruction include galeal periosteal flaps and forehead flaps. Primary treatment errors and complications in the series emphasize lessons learned from failed reconstructions including soft tissue loss, bone resorption and subclinical infection. The case experience of 56 cases emphasizes the necessity to separately manage the bony and cartilaginous deformities from soft tissue skin and lining problems. This study is a cooperative craniofacial study involving several centers which has as its patient base the largest series presented of secondary nasoethmoid reconstruction.

## LENGTHENING OF THE NOSE, REVISITED

*S Anthony Wolfe, Jorge Latoni, Paul Tessier*

Lengthening of th6 nose is frequently necessary for a whole array of congenital facial malfonnations, as well as in post-traumatic deformities (NOE fractures, over-exuberant "aesthetic" procedures).

Elements which contribute to successful outcome of nasal bone grafts will be discussed, as well as methods of achieving adequate nasal lining. Lengthening of congenitally short noses by lining dissection alone, without bone or cartilage grafting, will be discussed.



## FRONTOORTNTAL PSEUDOENCEPHALOCELY—A RARE GROWING FRACTURE

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A 5-year-old Chinese girl presented a 2-year history of exophthalmus. Head injury was happened as she fell down from her parent bosom when she was 9 months. "Forehead fracture" was diagnosed in another hospital. 2 years late her parents found that her right eye sticking out slowly and gradually rotated to temporal side. First operation was performed for "removing right frontal lobe cyst" according to her parents' description. MRI imaging showed there were penencephalus on right frontal lobe as well as skull base encephalocele. 3-D CT scan showed there were right frontal skull base defect.

We performed subfrontal craniotomy and disected cysty brain tissue and C.S.F. which herroated into orbit through a big bone defect. Dura on bony defect were repaired during operation.

## POSTOPERATIVE CHANGES OF MANDIBULAR ANATOMY AND FUNCTION AFTER SAGITAL SPLIT RAMAL OSTEOTOMY

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Sagital split ramal osteotomy is one of effective procedure to correct maxillo-mandibular deformities. However many reports about postoperative changes of mandibular anatomy after sagital splitting of mandibular ramus have been fabricated. The purpose of this study is to investigate postoperative changes in morphology and position of ramus and condyle, gradual improvement of condylar movement within glenoid fossa and mouth opening in patient undergoing sagittal split ramal osteotomy with rigid screw fixation.

We reviewed forty two cases of mandibular prognathism underwent sagittal split ramal osteotomy from 1991 to 1998 for mandibular set-back. The morphology of ramus and mandibular angle was evaluated with pre-and postoperative cephalometric tracing. The position of ramus was assessed by angular measurement defined by sella-nasion and articulare-gonion. The change of condylar position was evaluated by measuring the angle of the condylar long axis (condylar angle) on submentovertex view. In the assessment of mouth opening and condylar movement, we use the interincisal distance and angular measurement defined by perpendicular line of centrale of articular eminence and centrale of condyle-centrale of articular eminence in given intervals.



## NEUROSURGICAL POINT OF VIEW IN COMPLICATED CRANIOSYNOSTOSIS

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Crouzon and Apert syndrome are classified in complicated craniosynostosis. In order to clarify the implication of neurosurgical condition, we review the cases from 1995 to 1998 in Chang Gung Medical Center. These cases are managed by the team composed of neurosurgeons and plastic surgeons. There are three Crouzon syndrome and Apert's syndrome. In the mean time we also had six cases of simple craniosynostosis which were used as control group.

In cases of Crouzon syndrome, two of three had abnormal findings in preoperative neuroimage studies including one obstructive hydrocephalus and digital sign indicating increased intracranial pressure. There was also high percentage of abnormal neuroimages in cases of Apert's syndrome, The brain echo in one patient revealed asymmetric lateral ventricle and increased intracranial pressure with patent cavum septum peducillum in another patient.

On the contrary, there was only one patient found incomplete corpus callosum agenesis with borderline intracranial pressure in simple craniosynostosis. In the aspect of surgical approach, we emphasis on the use of lumbar drainage in cases of IICP before craniotomy to make the brain slack. We did not meet postoperative epidural hematoma despite we did not do dural tentings.

In summary, the incidence of intracranial problems in cases of complicated is higher than those in simple craniosynostosis.

Therefore neurosurgeons have to pay special attention to complicated cases and lower down the ICP before craniotomy starts. The use of lumbar drainage is quit valuable in protection brain from possible damage during the procedure of craniotomy.

## BLINDNESS FOLLOWING EYELID SURGERY - UNEXPECTED COMPLICATION -

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A 55-year-old-female, with a complaint of a scarring and a foreign body of right upper eyelid that underwent a blephaloplasty 25 years ago, was referred from an opthalmologist for another blephaloplasty. Opthalmologically any eye problem, such as congestion, exopthalmos, opthalmoscopic disorder, had not been found.

The surgery was done under a local anesthesia. Via blephaloline scar, injected foreign body was excised with a part of orbicularis so that some orbital fat and a dilated looped vessel came out. The surgeon thought it was venous lake or a kind of venous malformation and ask a professor to see it. According as an instruction of a guide, a loop was dissected as deep as possible into the orbit. Cramp test was performed in order to search whether the vessel has not flow and cut off on both side of it. After a local cooling, the patient went home without a pain or bleeding.

The next day the patient come back with a extraorninary swelled lids, severe pain and nausea that had come over gradually since the previous night. Though she let hospitalized and intravenous steroid and glyceol (250mlx2/day) infusion were done for a week, the light perception was lost in 10th operative day. Post-operative CT showed that a winding dilated supraorbital vein in right orbit and a big pituitary tumor. So that a neurosurgeon did an angiography, in that a big aneurysm was detected in a right carotid artery (cavernous portion) and coiled dilated vein and an unusual blood flow 'to and fro' in its ligated distal end. In other words, an unsymptomatic carotid cavernous fistula has been revealed by means of angiography. The ligation of supraorbital vein which was found as a loop at the operation, caused interception of the intraorbital venous flow resulting the extraordinary intraocular edema and compression of the eye ball and the optic nerve lead to significant permanent visual loss.



## U.S. FDA REGULATION OF ORTHOTIC TREATMENT OF DEFORMATIONAL PLAGIOCEPHALY

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**Objective:**
The purpose of this poster is to provide information regarding the current medical regulation of cranial orthoses used for the treatment of deformational plagiocephaly, and to discuss the concerns raised by the U.S. FDA regarding the safety and effectiveness of these devices.

**Background:**
Orthotic treatment of nonsynostotic plagiocephaly was first introduced in Seattle, Washington in 1979. Although slow to gain acceptance, the recent increase in the number of infants presenting with deformational plagiocephaly has caused many craniofacial centers to establish programs of their own. Until recently, medical regulation of this industry was generally ignored.

**Regulation:**
In 1995 our office was served notice that our orthosis would require submission of a premarket notification [510(k)]. After three years of working closely with the U.S. FDA to provide data regarding the efficacy of these devices, approval was finally granted. Since no device had ever been approved for treatment of deformational plagiocephaly, the FDA created a new device category known generically as the 'cranial orthosis'. A cranial orthosis is considered a Class II, neurology device, requiring both general and special controls in order to ensure safety and effectiveness. The general controls require 1) facility registration, and 2) manufacturing under current Good Manufacturing Practices (cGMP). Special controls require 1) prescription use, 2) biocompatability testing, 3) quantitative tracking of improvement of head shape and circumferential growth, 4) neurological assessment, 5) dermatologic assessment, 6) methods for reducing potential restriction of cranial growth, 7) treatment of torticollis 8) quality assurance, 9) written clinical instructions for casting, fitting, and care of the device, and 10) written parental instructions.

**Summary:**
Orthotics used for treatment of deformational plagiocephaly in the United States are categorized as Class II devices requiring premarket notification [510(k)], and are subject to both general and special controls in order to provide reasonable assurance of safety and effectiveness.

## DISTRACTION OF THE CRANIAL VAULT FOR CRANIOSYNOSTOSIS IN INFANTS; PRE- AND POSTOPERATIVE ASSESSMENTS BY DIGITAL X-P AND 3D-CT

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Distraction is quite useful for selected cases of maxillofacial deformities, and satisfactory results are reported. However, the reports of distraction of the cranial vault are limited in number. We present the assessment by digital X-P and 3D-CT of the fronto-orbital advancement and cranial vault remodeling by gradual distraction.

Five patients with craniosynostosis underwent fronto-orbital advancement and reshaping assisted by gradual distraction; 3 primary and 2 secondary cases. The average age was 11.6 months (from 6 to 22 months of age). The distraction was initiated a week after the operation. The degree of advancement was determined by our criteria we have reported before and simulated surgery with 3D-solid model was performed. Pre- and postoperative assessments were made by digital X-P at weekly interval during the distraction. 3D-CT was taken preoperatively and at the completion of the distraction.

Predetermined advancement and remodeling were possible in almost all cases. A shorter operating time and less amount of blood loss were the advantage by distraction procedure. In two cases osteogenesis was confirmed on digital X-P three weeks postoperatively. We report these results in detail with the change of pre- and postoperative intracranial volume.



## CASE REPORTS IN MIDFACIAL DISTRACTION: IN SEARCH OF THE OPTIMAL TECHNIQUE

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In performing a midfacial distraction at the LeFort III level, options include internal and external distraction devices. We report one case each of midfacial distraction using the Rigid External Distraction system (R.E.D., KLS Martin), and the Modular Internal Distraction system (M.I.D., Howmedica Leibinger), and indicate the advantages and disadvantages of each technique in our hands.

**Methods:**
A) M.I.D. System: A 4½ year old male with Apert Syndrome underwent LeFort III osteotomy and application of the M.I.D. system for the correction of midfacial retrusion. Following distraction of 1 mm per day for 15 days, the patient demonstrated a left malar complex fracture with shift of the midface to the left. The M.I.D. system was removed and the midface stabilized using rigid fixation. B) R.E.D. system: An 11-year-old male with Pfeiffer Syndrome and extensive bone loss of the midface and cranium secondary to infection following craniofacial reconstruction underwent LeFort III osteotomy and application of the R.E.D. system. A transverse K-wire was placed just below the nasion for control of midfacial rotation. Anterior distraction proceeded at a rate of 1 mm per day. During this process the patient developed a left infraorbital rim abscess, which was drained without interruption of the distraction process. Progressive counterclockwise rotation of the maxillary occlusal plane occurred until corrected by applying additional traction force at the nasion. The patient achieved 19 mm anterior distraction resulting in end-on occlusion.

**Conclusions:**
M.I.D. System – *Advantages*: 1) suitable in the young patient and less dependant on patient cooperation. 2) Less cumbersome than external devices. *Disadvantages*: 1) Only unidirectional distraction can be achieved. 2) All distraction forces are translated to the malar complex, which may be prone to fracture in young patients with syndromic craniosynostosis. 3) A second operation will full coronal incision is needed for device removal. R.E.D. System – *Advantages*: 1) The direction may be adjusted during distraction. 2) May be performed despite thin or absent bone segments in the zygoma. 3) Distraction may proceed in the presence of postoperative infection without the need for hardware removal. 4) Device removal does not require a second operative procedure. *Disadvantages*: 1) Difficult in young non-cooperative patients. 2) Requires a secure dental arch, which is often deficient in young patients with syndomic craniosynostosis. 3) Control of distraction forces for lengthening or rotation of the midface is less predictable.

## RAPID DISTRACTION OF THE SEVERELY HYPOPLASTIC MANDIBLES IN THE HEMIFACIAL MICROSOMIA

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During the last 3 years, we distracted the severely hypoplastic mandibles at the rate of 1 mm every 12 hours in 8 patients of an age of 3 to 7 years with the hemifacial microsomia. Long Pennig Minifixator (Orthofix®, Bussolengo, Italy) maintained the mandible in fixation for 4 days after the curvilinear osteotomy at the hypoplastic ramus through intraoral incision along the oblique line with two pins inserted at the proximal segments along the posterior border of the ramus and another two pins inserted along the lower border of the body of the mandible distal to the osteotomy. Following this period, the device was lengthened serially 1 mm every 12 hours by turning the nut. After the period of active lengthening, the mandible was maintained in external fixation for approximately 10 weeks.

The amount of vertical distraction was from 25 to 32 mm and horizontal distraction was from 2 to 18 mm. The period of active distraction was from 13 to 27 days. There was no perioperative complications and the length of the clinical follow-up ranged from 1 to 3 years. Early reconstruction with minimal morbidity on rapid mandibular distraction in the hemifacial microsomia was successful.



## CEPHALOMETRIC EVALUATION OF THE CRANIOFACIAL COMPLEX IN PATIENTS TREATED WITH AN INTRA-ORAL DISTRACTION OSTEOGENESIS DEVICE—A PRELIMINARY REPORT

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The purpose of this pilot study was to evaluate cephalometrically, the changes occurring in the craniofacial morphology following distraction of the mandible with an intraoral distraction osteogenesis appliance. Five patients diagnosed with hemifacial microsomia were enrolled in this pilot study and underwent unilateral distraction. PA and 45° lateral oblique cephalometric radiographs were used to quantify the changes occurring in craniofacial morphology and assess any relapse. Our analysis showed a significant change in mandibular length after distraction with $p < 0.001$. There was no statistically significant relapse at the follow up visits. A significant improvement in the position of menton to the facial skeletal midline was also seen following distraction with $p = 0.01$. The data does show menton moving away from the facial skeletal midline over time. The cant of the occlusal plane improved significantly as well with $p < 0.05$. Our data suggests that the bone lengthening appears stable and that the distracted mandible maintains a growth rate roughly similar to the normal side. The chin point should be made coincident with the facial skeletal midline or even overcorrected slightly to maintain facial symmetry. The occlusal plane changes appear to be due to dentoalveolar adaptations following distraction.

## CORRECTION OF CORONAL SUTURE SYNOSTOSIS USING BONE DISTRACTION TECHNIQUE

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One years old baby of left hemicoronal synostosis and six months old baby of coronal synotosis were treated by distraction osteogenesis using internal distraction device. In the case of hemicoronal synostosis, frontal bone and upper half of orbits were osteotomized en-block with lateral one forths of the frontal bone in unaffected side intact, and internal distraction device was placed in the left temporal area. In the case of coronal synostosis, frontal bone including upper half of orbits were osteotomized en-block and internal distraction devices were placed bilaterally. A 0.5 mm to 1 mm/day rate of distraction was performed after a 5-day latency period, and the devices were removed after consolidation period of 2 months. The result obtained was satisfactory in hemicoronal synostosis. However, inadequate advancement was achieved in the case of coronal synostosis because osteotomized segment was pivoted at the patent metopic suture.

The distraction osteogenesis of craniofacial skeleton has some merits such as no extradural dead space after the operation, minimum epidural dissection, simple operative procedure and minimum blood loss, and no fixation device was left after the second operation. However, timing of the operation and indication of the procedure must be considered in further experiences.



## MIDFACIAL ADVANCEMENT BY DISTRACTION OSTEOGENESIS FOR TREATMENT OF OBSTRUCTIVE SLEEP APNEA IN CROUZON'S SYNDROME

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Midfacial hypoplasia is one of recognized causes of obstructive sleep apnea syndrome in Crouzon's syndrome. Craniofaciostenosis like Crouzon's syndrome is associated with midfacial hypoplasia and has been treated traditionally by using Le Fort advancement osteotomies and bone grafts. This surgical technique requires a prolonged operation time, several osteotomies with significant blood loss, and wide surgical exposure.

**Case:**
We performed Le Fort III midfacial advancement with gradual distraction using internal devices in 2 year and 5 month old Crouzon's syndrome boy associated with obstructive sleep apnea to reduce the operation time, blood loss, and other complications. We placed the distraction device on the malar bone and temporal bone bilaterally. The device was not activated for 7 days after surgery, after which the distraction was initiated 1 mm per day, and the final midfacial advancement was 12 mm.

**Result:**
The obstructive sleep apnea was improved clinically after surgery.

**Conclusions:**
In the child Crouzon's patient associated with obstructive sleep apnea syndrome, distraction osteogenesis is an initial therapy to reduce the severity of the deformity, and to improve the upper airways obstruction.

## BENEFIT OF THE HALO-FRAME IN CRANIOFACIAL DISTRACTION

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During the last decade distraction osteogenesis has enriched the therapeutical concepts in craniofacial surgery. There is always a controversy about the most useful device, and most surgeons suggest the use of small internal working devices. As the forces strengthen with increasing lengthening, in severe cases an accurate guidance of the lengthening device should guarantee for a good result. Using distraction osteogenesis in our department since about 8 years we early considered about the benefit of the halo-frame in the distraction field.

The halo-frame serves as a base. While in small children taking a closed ring made out of special plastic, in elder patients we prefer a open u-bow made out of stainless steel. All parts for lengthening- passive and active elements – are taken out of our modular system for distraction. This technique was used for midfacial advancement in one patient each suffering from Crouzon syndrome, Pfeiffer syndrome and midfacial hypoplasia as a sequelae of bilateral facial clefts, for mandibular augmentation in one patient each with bilateral Microsomia and hemifacial microsomia. In the Pfeiffer syndrome the pre-distraction Le Fort III osteotomy was combined with a cranioplasty. The distraction protocol was as usual: in children below 3 years distraction was started at the third postoperative day with a lengthening rate of 1 mm every 12 hours, in all other patients 1 mm daily starting at the fourth postoperative day. In every patient the intended augmentation was achieved, in no case problems with the device occurred. The clinical use will be demonstrated, the pros and cons are discussed.



## TREATMENTS OF THE ORBITAL HYPERTELORISM

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**Materials and Methods:**
In recent 10 years, 7 patients with orbital hypertelorism were treated at Kanazawa Medical University Hospital. They were included 3 patients with Tessier No. 0 cleft, one patient with No. 1 cleft, one patient with No. 1, 2, 3 cleft, one patient with No. 10 cleft and one patient with No. 14 cleft. Total mobilization of the orbit were performed in 5 patients. Bilateral mobilization were done in 3 patients with No. 0 cleft. Unilaterl mobilization were done in 2 patients with No. 1 cleft or No. 10 cleft. These patients were reconstructed in one stage operation. On the other hand, one patient with No. 14 cleft were reconstructed in two stage. When she was one year old, the first operation was performed in order to repair the naso-ethmoidal encephalocele and supraorbital components were mobilized simultaneously. The second operation was carried out in 6-year-old in order to reconstruct infraorbital components which were not mobilized at the first operation.

**Results and Discussion:**
The follow-up period were the 10 years from 2 years. Severe complications were not occurred postoperatively. Facial aesthethic improvement was acquired in all cases, however, scars of the forehead which were made at soft tissue reconstruction of the forehead were conspicus in some cases. In this report, our representative cases are presented and our technical contrivances are discussed.

## OSTEOPLASTY OF EXTENDED JAW DEFECTS BY USE OF PROTECTED BONE REGENERATION WITH MACROPOROUS RESORBABLE SHEETS

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This study is performed to demonstrate a new method of protected bone regeneration using macroporous resorbable sheets for the treatment of extended lower and upper jaw defects. For the reconstruction of such defects basic requirements of bone healing have to be assumed such as good vascularisation, immobilisation and prevention of soft tissue prolapse. By applying mechanical protection of bony defects with e.g. membranes or Titanium mesh, soft tissue prolapse as well as pressure on bone transplants which contributes to partial resorption, can be avoided.

Previously Titanium mesh for Osteoplasty has been used with the disadvantage of the difficulty of removal after bone healing was complete.

The use of a pressure- resistant, resorbable, macroporous sheet combines the advantage of protected bone regeneration and complete resorption of the implanted sheet. The macroporous structure facilitates capillary ingrowth from the surrounding soft tissue.

The sheet itself is made of 70:30 Poly(L-co-DL)-lactate with thermoplastic character and can be used as a container for autologous spongiosa or other osteoinductive and – conductive bone graft substitutes.

In a pilot study, seven patients with lower jaw defects resulting from large cysts or tumor resections, some affecting the continuity of the mandible were treated with this method. In these cases, the good practicability in the use of these sheets could be established. Following a protocol, x-rays were obtained to document the bony regeneration. Histological growth of vessels from surrounding soft tissues as well as bony reconstruction could be demonstrated.

The positive experience with this pilot study encouraged a multicenter- project involving five University hospitals and fifty patients. First, the application of resorbable sheets in combination with transplantation of mersilized autologous spongiosa is currently being investigated. In future studies fillings of the sheet with osteoconductive and – inductive materials are planned.



## EFFECT OF BIOABSORBABLE OSSEOUS FIXATION MATERIALS ON DURA MATER AND BRAIN TISSUE

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**Introduction:**
Recent clinical reports of passive cranial translocation of plates and screws have raised questions about the effects of nonabsorbable materials on dura mater and brain tissue. This is especially important in pediatric craniofacial surgery, where extensive postoperative bone remodeling and intracranial growth occurs. Previous studies compared the biocompatibility and morphometric properties of nonabsorbable plates and screws in soft tissues, cranial bone, dura mater and brain. The purpose of this study is to assess the histologic alterations on dura mater and brain tissue, caused by bioabsorbable plates, composed of polylactic and polyglycolic acid with potential of hydrolytic resorption over a period of one year. The exact timing of bioabsorption of the plates and screws and their local effects on osteocytes are not clear.

**Methods:**
Forty-five male Sprague-Dawley rats of 15 weeks old and weighing 350-400 grams are evaluated in two groups. Bioabsorbable plates, composed of 80% polyglycolic acid and 20% polylactic acid were used in dimensions of 5x3x2 mm. *Group I:* Sham operation (n=15). Following a sagittal scalp incision and periosteal elevation, a biparietal cranial window was created and closed without placement of bioabsorbable plates. Five rats were sacrificed after each 3, 6 and 12 month follow-up periods. *Group II:* (n=30) Using the same operative technique, a biparietal cranial window was created and PGA/PLA plates were placed in the epidural space, followed by the replacement of the cranial bone segment. Ten rats were sacrificed after each 3, 6 and 12 month follow-up periods. The rats were assessed weekly for neurologic or behavioral changes suggesting neural damage. For histologic analysis, total block resection of overlying cranium, dura mater and brain was performed. All tissue sections were examined by standard H&E stains.

**Results:**
None of the animals showed any behavioral changes or neurologic deficits suggestive of systemic or localized toxicity from the implants. Histologically, non-specific minor inflammatory changes were identified and there was no obvious evidence of transdural intracranial effect in the early postoperative period.

**Conclusions:**
The neurological and histological effect of bioabsorbable plates on neural tissue may be considered as negligible in the early postoperative period.

## SURGICAL MANAGEMENT OF CRANIOFACIAL DISEASES WITH 3D IMAGING AND THE IOO

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Surgical management was presented with the aid of three dimensional (3D) imaging to patients with Crouzon Anomaly, exophthalmos for Graves Endocrinopathy and enophthalmos after facial trauma. A spatial relationship in the orbital region was identified to each case. Auxiliary surgical data as the placement of the intra-conal fat, the relationship among the extraocular muscles with the eye and the optic nerve were also presented. The Orbit-Ophthalmic Index (IOO) was determined for each case. It was the mathematical expression of the volumetric relationship of the orbit contends with its eye, with Normal Distribution pattern, satisfying the criteria of skewness, kurtosis and Shapiro-Wilk test accounting IOO = 4.02 (± 0.51) n = 28. The accuracy of such methodology was developed in 1995 by comparing linear and volumetric measurements on cadaver matching against the same 3D image. All the images were processed by 3D VIEWNIX software (Philadelphia, Pennsylvania). It was discussed the resources and the limitations of the manual segmentation method that we used in this software, on identifying and studying the proper orbital elements.



## ENDOSCOPICALLY ASSISTED SURGICAL MANAGEMENT OF FACIAL BONE FRACTURES

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Endoscopy is one of fields of rapidly growing interest and improvement, and also its techniques are now widely applicated in many fields of aesthetic surgery. The superior advantage is its easy approach with even small incisions on the face. We have applied endoscopic instruments to the surgical management of facial bone fractures, especially the area that can not be easily approached. The endoscope can be used helpfully in the reduction of proximal fracture to the mandible angle, orbital blow-out fracture and corrective rhinoplasty or other procedures.

We successfully experienced 27 cases of endoscopically assisted surgical management of facial bone fractures: reduction of mandible angle fracture (7 cases), subcondyle fracture (3 cases) or condyle fracture (4 cases) of mandible, inferior blow-out fracture (4 cases) or medical blow-out fracture (3 cases) of orbit, obliteration of nasofrontal duct (2 cases), and intraoral approach of corrective rhinoplasty (4 cases). These approaches need delicate techniques and well-trained assistants but sometimes minor inattention can cause moderate or severe functional problems such as malocclusion or diplopia after reduction of mandible or periorbital fracture. So, we recommend minor skills during these operations: 1) in angle fracture, reduction of retromolar trigone at first and then inferior angular border with compression plate under the endoscopic assist, 2) in condyle or subcondyle fracture, careful detachment of lateral pterygoid muscle from the condyle and then fixation under the endoscopic assist, and 3) in inferior blow-out fracture, the transantral approach with endoscope. 4) In the operation of corrective rhinoplasty, the exact site and level of ostetome can be confirmed with the help of endoscope through the intraoral vestibular approach. Under the endoscope, the symmetrical nasal osteotome can be easily done with the reciprocating saw in a short time.

In spite of technical difficulty of hand-eye coordination at first, after acquiring endoscopic skills, comparable results can be obtained in the field of trauma.

## TISSUE EXPANSION IN CRANIOFACIAL RECONSTRUCTION

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This paper presents the authors experience in the use of tissue expansion technique in eleven patients requiring cutaneous and skeletal craniofacial reconstruction. The expanded flaps were used for nose, cheek and scalp reconstruction usually in conjunction with bone or cartilage grafts for skeletal restoration. The approach and techniques used will be discussed as also the problems faced at various stages. All the patients showed favorable results after the completion of staged reconstruction.



## ENDOSCOPICALLY ASSISTED REDUCTION OF THE ZYGOMATIC FRACTURE

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Zygomatic arch can be employed as a key landmark to the accurate alignment of a displaced malar fracture; it has been traditionally avoided because of the need for a bicoronal incision. Zygomatic arch exposure by conventional bicoronal incision has several possible disadvantages, including an increased risk of blood loss, alopecia, loss of sensation posterior to the incision and traction palsy of the facial serve in addition to the longstanding operative time.

Since 1994, endoscopic technique was used in facial rejuvenation than this technique was used broadly by other surgeon in facial skeleton surgery.

Endoscopic assisted zygomatic fracture repair without a bicoronal incision has been developed. This procedure can largely obviate the disadvantages of a bicoronal incision and yield this site accessible to reduction and fixation in the routine treatment of displaced malar fracture. Main disadvantages of this procedure are the learning curve of endoscopic surgery and the expense of endoscopic instruments.

Endoscopic assisted for zygoma fracture reduction technique with minimal incision wound was used in 16 cases. Usually the incision wound locate at the intraoral, inside the sideburn or/and subcilliary region to explore zygoma and arch. The length of sideburn incision wound usually no more than 3.5 cm, which we undermine below the superficial temporal fascia and then explore the zygoma arch and fix it under endoscope after alignment is well done.

This method just has a small incision wound, that it can replace bicoronal incision to prevent sequales. Since 1996 to 1999, we performed this method in 16 patients including thirteen zygoma and arch fracture, 3 pure zygoma arch fracture (2 fracture sites) and follow up for a long period (3 months ~ 33 months) with a good alignment. Our experience in this method will be presented.

## CLINICAL EXPERIENCE IN THE CRANIOFACIAL BONE RECONSTRUCTION WITH MEDPOR® (HIGH DENSITY POROUS POLYETHYLENE)

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**Purpose:**
The purpose of this study is to describe the experience of using high density porous polyethylene (Medpor®) for reconstructive facial bone surgery because of its perceived increased biocompatibility.

**Materials and Methods:**
A total of 77 Medpor® implants were placed in 65 patients over a one and half year period. Implants were used for orbital reconstruction and correction of craniofacial contour deformities such as forehead, malar and chin area. Before surgery, adequate photographic and radiologic examinations were performed to carry out operation plan. Within 65 patients, 38 operations were due to acute trauma, 23 surgeries were due to posttraumatic facial deformity, and 4 were cosmetic problems.

**Results:**
The results were satisfactory: 95.4% of the patients showed post-operative improvement. One complication occurred due to previously existed sinusitis and it was cured with conservative treatment.

**Conclusion:**
On the basis of our results, we believe the Medpor® implant is an excellent alternative to existing implant materials. It is easy to handle, shape, contour, position, and fixate. If applied under adequate circumstances, this implant will permit predictable and stable results with few complications.



## TRAUMATIC ORBITAL EMPHYSEMA: CLASSIFICATION AND TREATMENT

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**Purpose:**
Orbital emphysema commonly results from medial or inferior orbital wall fractures, which can be easily detected by CT scan. It may be a self-limited condition, or may result in potential ocular injury, or cerebral sequelae. This study was designed (1) to classify orbital emphysema observed on CT scans as a guide to treatment and (2) to compare potential ocular and cerebral injuries between groups with and without orbital emphysema.

**Materials and patients:**
A total of 105 peri-orbital fractures were analyzed over a 7-year period. A classification of orbital emphysema types was developed from patterns observed on CT scans. Three types of orbital emphysema were identified: type I, pre-orbital emphysema; type II, intra-orbital emphysema; type III, orbital emphysema associated with pneumocranium.

**Results:**
50 patients (47.61%) had orbital emphysema. The incidence of occular injury was 62% in the group with orbital emphysema and 40% in the group without orbital emphysema respectively. Type I orbital emphysema occurred concurrently with subconjunctival hemorrhage, eyelid laceration, globe rupture, nasolacrimal duct disruption, and traumatic lens subluxation. Type II often occurred concurrently with optic neuropathy, vitreous hemorrhage, retrobular hemorrhage and oculomotor paralysis. Type III occurred with associated subarachnoid hemorrhage, dural tear, epidural hemorrhage and brain swelling. One case with ball-valve orbital emphysema causing optic neuropathy required urgent decompression.

**Conclusions:**
The incidence of ocular injuries in the orbital fracture was 50.47%, but higher incidence (62%) occurred in the group with orbital emphysema. The ball-valve orbital emphysema might case optic neuropathy. Poor prognosis of orbital emphysema was associated with optic neuropathy, globe rupture, lens subluxation and vitreous hemorrhage. Type I and type II orbital emphysema require early consultation by an ophthalmologist. Type III needs early consultation by a neurosurgeon. Awareness and treatment of associated cerebral and ocular injuries are important in addition to repair of orbital fracture.

## A SINGLE TRANSCONJUNCTIVAL INCISION FOR THE ZYGOMA-ORBITAL FRACTURE REPAIR

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**Background:**
A transconjunctival incision is one of the recognized approaches for access to orbital fracture. As it provides excellent exposure to the inferior orbit and produces no visible scar, the transconjunctival approach has become increasingly popular. A lateral canthotomy is often added to the transconjunctival incision to obtain adequate exposure of the zygo-orbit fracture. The lateral canthotomy incision may negate the advantage of this potentially invisible scar in those patients. For improving the aesthetic results, a single transconjunctival incision without combining lateral canthotomy has been attempted to approach the zygoma-orbital fractures.

**Patients and Methods:**
From July 1997 to December 1998, the single transconjunctival incision was utilized to approach 54 consecutive zygoma-orbital fractures. The anterior limb of the lateral canthal ligament, the periosteal attachments of the frontal process and zygomatic process of the frontal bone should be incised and mobilized. Access was adequate for the procedures without combining a lateral canthotomy incision.

**Results:**
Temporary ectropion or intropion were present in 8 patients but resolved sufficiently with conservative management. There were 5 cases of conjunctival granuloma, and 2 of these required excision. There was no intraoperative eyelid avulsion. Our clinical experiences reveal that lateral canthal reattachment is not necessary.

**Conclusion:**
It is our belief that the same incision used for a patient seeking elective aesthetic eyelid surgery should be used for the facial fracture patient. Through a single transconjunctival incision, the zygoma-orbital fracture can be repaired. Complications of this approach are uncommon, and most may be prevented by careful surgical technique.



## TRAUMATIC SUPERIOR ORBITAL FISSURE SYNDROME

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**Purpose:**
The superior orbital fissure syndrome (SOFS) is caused by impairment of the contents that cross the superior orbital fissure. It can be caused by tumor, infection, or trauma. It is relatively rare when associated with craniofacial fractures. The treatment and prognosis are still not well established.

**Material and Method:**
24 patients were reviewed retrospectively between January 1981 and July 1988. They were selected from those having craniofacial fractures. 23 patients received conservative treatment without steroid. 1 patient received surgical decompression of SOF. 14 patients received ORIF for facial fractures.

**Result:**
Follow-up periods were from 6 months to 24 months. 22 patients were in incomplete recovery, and 2 patients were in complete recovery. 14 patients have zygomatico-orbital fracture. 3 patients have Carotid-Cavernous fistula (C-C fistula). Cranial nerve VI seemed easier to get injured. The recovery of cranial nerves seemed no difference. 1 patient received resection of levator muscle for ptosis. 1 patient received ophthalmic surgery to correct diplopia.

**Conclusion:**
SOFS was an uncommon injury with craniofacial fractures. It should be recognized and given proper treatment. Steroid may play a role. Surgical decompression should be done whenever compression of SOF is suspected. There is no reason to delay the facial fracture repair. Arteriovenous fistula should be worked up and embolized.

## ANALYSIS OF THE CANDIDATE GENES ASSOCIATED WITH NONSYNDROMIC CLEFT LIP AND PALATE IN JAPANESE

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**Purpose:**
Cleft lip and palate is one of the most common birth defects. But its etiology and pathogenesis is still obscure. The frequency of cleft lip and palate in Japanese is higher as compared to caucasian and black race. Epidemiologic studies suggest that exogenous factors and genetic factors play a role in the etiology of cleft lip and palate. Specifically, we studied the relationship between nonsyndromic cleft lip and palate and polymorphism of the candidate genes, such as transforming growth factor alpha (TGFA) gene, transforming growth factor beta 2 (TGFB2) gene, transforming growth factor beta 3 (TGFB3) gene, and γ-aminobutyric acid receptor beta 3 (GABRB3) gene.

**Materials and Method:**
Among the Japanese, after consent was obtained, blood samples were collected from cases and controls. Patients who had cleft lip with or without cleft palate, or cleft palate only, and had no other congenital anomalies were selected for case group. Persons who had no craniofacial anomaly, other congenital disease, or no family history of craniofacial anomaly were used as control. DNA extracted from whole blood was amplified and the amplified DNA fragments were screened for DNA sequence variants by SSCP, DNA restriction enzymes, and direct sequences. In statistical examination, $X^2$ analysis was performed at a significant level of $p < 0.05$.

**Result and Discussion:**
In this study, as candidate genes, we selected TGFA, TGFB2, and TGFB3 of which local occurrence in the developmental stage of palate was demonstrated in the study of rodents. In the present study, we demonstrated significant differences in the polymorphism of TGFA K-primer region ($p = 0.027$) and TGFB2 ($p = 0.019$) between nonsyndromic cleft lip and palate group and control group. Mice lacking GABRB3 gene showed phenotype include cleft palate and the study suggest GABRB3 gene is necessary in craniofacial development as GABA-receptor but our analysis did not show any significant ones. We hope the result of the present study will help clarification of the etiology, gene diagnosis and gene therapy of cleft lip and palate in the future.



## PURE ORBITAL BLOWOUT FRACTURE: NEW CONCEPTS, AND IMPORTANCE OF MEDIAL ORBITAL BLOWOUT FRACTURE

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Pure orbital blowout fracture first occurs at the weakest point of the orbital wall. Although the medial orbital wall should be theoretically involved more frequently than the orbital floor, the orbital floor has been reported as the most common site of pure orbital blowout fractures. We evaluated 82 orbits in 76 patients with pure orbital blowout fracture diagnosed by using computed tomographic scan in all patients with any suspicious clinical evidence. Isolated medial wall fracture was most common (54.9 percent), followed by combined (medial and inferior wall) fracture (26.8 percent). The most common facial fracture associated with medial wall fracture was nasal fracture (50.8 percent), not orbital floor fracture. It suggest that the force causing nasal fracture is an important causative factor of pure medial wall fracture as the bucking force. Of patients with medial wall fractures, 25 percent had diplopia and 40 percent had enophthalmos. Diagnostic signs in plain radiograph were found in 79.1 percent of medial wall fractures on Caldwell view. On computed tomographic scan, late enophthalmos was expected in 76.1 percent of medial wall fractures. It suggest that the medial wall fracture is an important etiology of late enophthalmos because it has a highest occurrence rate, a low diagnosis rate, and a high severity rate. The medial and inferior orbital walls are clearly demarcated by the bony buttress which is an important structure supporting these orbital walls. Most of orbital blowout fractures without the collapse of the bony buttress has a trapdoor type with or without small fragments of punched-out type. In the medial wall fractures, muscle traction of the connective septa and direct muscle injuries were frequently found, but true incarceration of the extraocular muscles was not found.

## OPTIMIZED MULTISPIRAL 3D CT TECHNIQUE FOR LONGITUDINAL POSTOPERATIVE ESTIMATION IN CRANIOFACIAL SURGERY

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3D-CT has become essential tool in the field of cranio-maxillo-facial surgery. It is used not only for preoperative diagnosis and surgical simulation but also for longitudinal postoperative estimation. However, for the latter purpose, several times of volume CT scanning usually is necessary in the first postoperative year, and such frequent scanning has the possibility of deteriorating lenses, which are susceptible to X-ray. The purpose of this study is to establish an optimized multispiral 3D CT technique for longitudinal postoperative estimation in craniofacial surgery.

A Hispeed-advantage spiral CT scanner was used to conduct axial scanning of a head phantom. 1, 3, and 5 mm slice thickness spirals were obtained with pitches of 1 and 0.5. Exposure doses during each scanning were measured by probes attached to the position of lenses in the phantom. Three-dimensional osseous surface images and coronal plane tomographies were then reconstructed from these accumulated volume data and compared with each others.

The results suggested that, for the purpose of postoperative estimation, helical volume CT should be performed with a slice thickness of less than 3 mm and pitch of 0.5.



## ANOMALOUS INTRACRANIAL VENOUS DRAINAGE IN FGFR-ASSOCIATED CRANIOSYNOSTOSIS

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**Introduction:**
The aetiology of raised intracranial pressure in complex multisutural craniosynostosis is multifactorial. A group of patients do not respond to expansion cranioplasty, the management of the airway or treatment of hydrocephalus and persist in exhibiting intracranial hypertension.

**Methods:**
Cerebral digital 4 vessel angiography was performed in a sequential series of 20 children (3 months to 9 years, 9 months), with documented intracranial hypertension and a variety of multisutural synostoses. In a parallel study, the expression of the fibroblast growth factor (FGFR) genes was accessed in human intracranial vessels at various stages of embryonic development.

**Results:**
Abnormal venous phase angiography was present in all cases. Intracranial venous drainage was attenuated asymetrically via the transverse and sigmoid dural sinuses, with increased flow through cortralateral system. In patients who underwent 2 sequential studies, progressive attenuation of flow in the stenosed sinuses was noted with commensurate increase in collateral venous blood flow. This was accompanied by worsening intracranial hypertension. FGFR genes 1, 2 and 3 were found to be differentially expressed in vascular endothelia of the intracranial compartment of human embryos from 8-14 weeks, in company with their expression, and that of selected FGF ligands, in the human choroid plexus.

**Discussion:**
Anomalous venous drainage is an important factor in producing intracranial hypertension in these patients. Its pathogenesis is discussed with respect to fibroblast growth factor (FGFR) mutant biology and the expression of these genes in vascular endothelia, and intriguingly, in the choroid plexus.

## COULD CHEILOPLASTY IMPROVE SUCKING FUNCTION IN UNILATER CLEFT LIP AND PALATE INFANTS

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Surgical repair of cleft lip was traditionally aimed to restore the anatomical alignment of upper lip. However, functional restoration of upper lip during sucking has not been studied. The sucking patterns of 80 infants with unilateral complete cleft lip and palate were monitored at the age of 1, 3, 6, 12, 18 and 24 months. Electromyographic (EMG) activities of perioral muscles, masticatory muscles and suprahyoid muscles as well as the sucking force were recorded and analysis. During sucking, the EMG activities of upper lip were decreased right after the lip repair and remained low throughout the entire observation. It is concluded the infants avoid using the repaired upper lip to suck even the orbicularis muscles was repaired during the cheiloplasty.



## LONG TERM OBSERVATION ON A PEDIATRIC PATIENT WITH RADIATION INDUCED FACIAL DEFORMITY: REPORT OF A CASE

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Radiation therapy for facial region employed in childhood often remains a serious prognosis such as osteoradionecrosis and facial deformity. We have experienced long term observation on a pediatric patient with radiation therapy induced facial deformity for buccal hemangioma. The patient is female, now 26 years old of Japanese. She was irradiated by lineac extra-beam at total dosage of 21 Gy for her buccal hemangioma when she was 1 and half years old. After irradiation, red-colored dermal change and hard scar on mid-portion of buccal area, severe trismus and facial deformity was remained then referred us at 4 years old in our department. She was received first surgery at 9 years 6 months old. In this surgery, buccal scar was excisioned full thickness preserved buccal branch of fasical nerve and reconstructed by combination of D-P plus island forehead flap at the same time of excision. However, trismus was unfortunately remained. Second surgery was performed at 17 years old. In this surgery, segmental mandibulectomy for her osteoradionecrosis and excision of intra-oral buccal scar was performed and immediately reconstructed by vascularized scapular osteocutaneous flap plus parascapular cutaneous flap. Result of the second surgery showed that trismus and facial deformity was cosmetically much improved. However, gradual resorption of cutaneous flap can be observed under the long term observation. Operative procedures and problems in this patient will be introduced.

## MAXILLARY RECONSTRUCTION COMBINED WITH VASCULARIZED ILIAC CREST FREE FLAP AND OSSEOINTEGRATED IMPLANT - TO ACHIEVE IDEAL SOFT TISSUE ENVIRONMENT OF IMPLANTS

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The advent of both microvascular free tissue transfer techniques and osseointegrated implant-retained prostheses has greatly improved the potentiality of functional and aesthetic results for maxillectomy patients. The iliac crest is an excellent source of bone to achieve and adequate height and width for the reconstruction of maxillary defects and to install a dental implant. Recently some authors have recommended the use of muscle flap instead of skin paddle for lining mucosal defects because the muscle epithelializes rapidly, providing an excellent oral appearance with minimal bulk. We presented a case of maxillary reconstruction who was tried to prepare ideal soft tissue environment for implants.

**Case report:**
A 34-year-old man was referred to our department for diagnosis and treatment of swelling of right upper canine region. The tumor was confirmed as odontogenicmyxoma. Via extraoral approach a right nearly hemimaxillectomy including tumor was performed in March 1995. He was underwent reconstructive surgery by a vascularized iliac crest free flap with neither muscle flap nor skin paddle in May 1997. Exposed surface of iliac bone placed into oral cavity were rapidly covered with acquired epithelium like a oral mucosa. In April 1998, four dental implants were installed in grafted bone and two implants placed remaining maxilla. Six months later, second abutment surgery was done and simultaneously acquired epithelium was thinned because covered tissue was mobile and thick. As hyperplasia around implant was postoperatively occurred penetrated through the acquired epithelium, palatal mucosa was transferred to provide a greater area of attached mucosa.

**Conclusion:**
In just a vascularized iliac bone, acquired epithelium changed as a oral mucosa, uneventfully covered iliac bone, but it's quality was considered to be unsuitable for dental implant encompassing. To solve this problem, we should make muscle cuffs thin as possible as we do at harvesting.



## CLINICAL APPLICATION OF DENTAL IMPLANT IN REVASCULARIZED MAXILLARY AND MANDIBULAR BONE FOLLOWING ORAL CANCER ABLATION

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Not only cosmetics but also functional reconstruction ha been required for the patient's satisfaction and quality of life when radical excision is needed for the treatment of oral cancer. In recent, revascularized osteocutaenous flaps have often employed to reconstruct both soft tissue and maxillo-mandibular bone defects because of several advantages following oral cancer ablation. However, satisfactory functional repairments for phonetics and mastication are a big challenge after surgery. We have employed combination reconstruction by osteocutaenous flaps and dental implant placement in grafted bone since 1987 and treated for total number of eight patients respectively. Subjective patients were 6 males and 2 females, and 7 lower gum, 1 maxillary gum at primary sites. Delayed bony reconstruction of maxilla or mandible was performed following several kind cutaneous or musculocutaneous flap had been used for the reconstruction of soft tissue defect at primary surgery. Employed osteocutaneous flaps were 6 fibular, 1 iliac, and 1 scapular. Installed dental implants were 6 endosseous implants and 2 Bosker's Transmandibular Implants. Results and problems of this treatment will be presented.

## MANDIBULAR RECONSTRUCTION FOLLOWING SUBTOTAL MANDIBULECTOMY FOR ORAL CANCER TREATMENT

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Reconstructive procedure using revascularized osteocutaneous flap following oral cancer ablation have various advantages that is possible to reconstruct three-dimentionally even wide bony and soft tissue defects remained and for poor recipient condition such as high-dose of irradiation and previous surgical scars. We have employed in clinic as one of reconstructive surgery since February, 1985. Until 1998, total number of 28 vascularized osteocutaneous flaps, 10 iliac, 7 scapular and 11 fibular, were employed in our department. In this presentation, two representatives from five patients who were reconstructed by revascularized osteocutaenous flaps following subtotal mandibulectomy will be introduced. Case 1; 53-y.o., male, lower gingival squamous cellcarcinoma, T4N2bM0, who was performed subtotal mandibulectomy and reconstructed by combination of D-P and pectoral major musculocutaneous flap for his soft tissue defects as primary surgery. Two stage of delayed reconstruction of the mandible using bilateral scapular flaps were employed for his wide bony defect; 11 cm in length of scapula was harvested and same length of scapula was harvested from another site one year late. After bone union, Bosker's TM-Implant was placed in grafted scapula directly. Case 2; 44-y.o., male, recurrent carcinoma of flow of the mouth who was reconstructed by forearm flap at primary surgery but found recurrent tumor two years later. Subtotal mandibulectomy, total glossectomy and pharyngectomy was carried out and reconstructed by combination of pectoral major and latissimus dorsi musculocutaneous flaps for his soft tissue defects. Five years later, mandible was reconstructed by fibular osteocutaneous flap for his cosmetics. Five endosseous dental implants were placed in grafted fibula two years later. Fibular osteocutaneous flap is enable to reconstruct whole mandible by one stage.



## A CASE OF LARGE CRANIOFACIAL ARTERIOVENOUS MALFORMATION

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Facial arteriovenous malformation (AVM) is the most difficult to treat of all the vascular lesions and is potentially lethal. In this paper, we present a case in which large facial AVM resulted from several palliative treatment could be subtotally excised.

**Case Report:**
On Nov, 1991. A 36-years-old woman was referred to our department because of the deformity of her left cheek. She had previously and partial resection of a facial AVM which included hemimandibulectomy and external carotid artery ligation in 1980 at another institute. Our first repair was reconstruction of the mandible using a free osteocutaneous parascapular flap and coverage of AVM in parotid region by dacron sheet which was expected to inhibit expansion of the lesion. However, the lesion rapidly enlarged in a year and she sometimes suffered bleeding from the lesion. She refused radical excision of the lesion and had only palliative treatment including embolization and tumor ligation until she suffered massive bleeding and hemorrhagic shock. At that time, the AVM had envolved left temporoparietal region, left ear, left cheek and left eyelids. On Oct 1997, subtotal excision of the AVM was performed. In spite of the preoperative repeated embolization by the neurosurgeon, the feeding vessels in sphenopalatine fossa could not have been occluded and it took 15 hours to remove most of the lesion because of the profuse bleeding. Simultaneous repair could not be done and the defects was tentatively covered with STSG. Two weeks later, the reconstruction was done with a combined serratus muscle and latissimus dorsi myocutaneous free flap. The postoperative angiogram revealed the presence of the small vascular lesion.

This case may indicate the importance of the radical excision of the facial AVM in early stage.

## TRANSFORMING GROWTH FACTOR-BETA1 IS ASSOCIATED WITH THE TRANSITION FROM SCARLESS REPAIR TO HEALING WITH SCAR FORMATION IN THE FETUS

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**Introduction:**
Fetal wounds transition from a scarless repair to healing with scar formation in the rat model, occurs between 15 to 18 days gestation (term = 21 days). The mechanism regulating this transition remains unknown. Transforming growth factor-beta 1 (TGF-β1) induces scar formation in wounds while TGF-β3 is thought to decreases scar. The purpose of this study are to determine the temporal and spatial expression of TGF-β and TGF-β3 during the transforming stages of fetal repair.

**Method:**
Full-thickness excisional wounds were made on the dorsum of 14.5 and 18.5 day gestational age Sprague-Dawley rat fetuses (term = 21 days) and adult animals (N=6). The wounds were harvested at 12 and 36 hours, and immunostained for TGF-β1, TGF-β3. 14.5 and 18.5 gestation day represent the scarless (pre-transition) and scar (post-transition) formation respectively.

**Results:**
A distinct temporal and spatial pattern of TGF-β1 expression was found as a function of gestation age. As a control, minimal amount of TGF-β1 was present in the 14.5 gestation day unwound skin while higher level of TGF-β1 was observed on dermal fibroblasts and the basal epithelial cells layer of the unwounded 18.5 day gestation and adult skin. In the wounding experiment, we observe minimal elevation of TGF-β1 in the 14 day gestation scarless wound. In the 18.5 day gestation scar wound, we observed a dromatic increase of TGF-β1 expression in the wound adjacent basal epithalial cell layer, dermal firoblast layer, the wound cellular infiltrate and fibroblasts. Similar level of TGF-β1 up-regulation was observed in the adult wounds. The analysis of TGF-β3 is in progress and a pilot study did not suggest a significant change during the transitional stages.

**Conclusions:**
TGF-β1 has been well indicated in the ontogeny of adult scar formation. Our data clearly demonstrate for the first time *in vivo*, a distinct temporal and spatial up-regulation of TGF-β1 during the transition from scarless repair to scar formation in the fetal rat wound. Thus, we suggest that the minimal induction of TGF-β1 during wounding in 14.5 day gestation may be responsible for the scarless wound repair. Further studies are in progress.



## UPREGULATION OF OSTEOGENIC CYTOKINES IN DURA MATER: MECHANISMS FOR ENHANCED CALVARIAL OSTEOGENESIS IN IMMATURE ANIMALS

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**Introduction:**
In contrast to adults, young children can re-ossify large calvarial defects. Numerous experimental studies have implicated the dura mater (DM) as the critical determinant of calvarial re-ossification in immature animals. To date, however, the exact biomolecular differences responsible for this age-related phenomenon remain unknown. The purpose of these studies was therefore, to elucidate the physiologic and biomolecular differences between DM from immature and mature animals.

**Methods:**
Cell cultures were established from parietal DM of 6 and 60-day-old rats (ie. immature and mature animals). 1st passage cells were harvested and used to assess cellular proliferation, alkaline phosphatase (AP) production (a marker of osteoblastic activity), *in vitro* bone nodule formation, and production of TGF-$\beta$1, and FGF-2. In addition, in order to relate our *in vitro* findings to *in vivo* events, RNA isolated from the parietal DM of an additional group of animals was probed with radiolabeled DNA probes against TGF-$\beta$1, collagen I, and osteocalcin.

**Results:**
Dural cells from immature animals proliferated nearly twice as fast as mature dural cells (*$p<0.05$). In addition, immature DM cells demonstrated significantly increased osteoblastic activity as assessed by alkaline phosphatase production (2x increase; *$p<0.01$) and produced significantly greater amounts of TGF-$\beta$1 (6x increase', *$p<0.01$) and FGF-2 (4x increase; *$p<0.01$) when compared to mature DM cells. These findings mirrored northern blot analysis of immature and mature DM demonstrating 10 fold, 22 fold, and 20 fold increases in TGF-$\beta$1, collagen I and osteocalcin expression, respectively, in immature as compared to mature DM. Changes in cellular proliferation, differentiation, and cytokine production translated to marked differences in osteogenic potential such that immature dural cells formed large, calcified bone nodules *in vitro* where as mature dural cells did not.

**Conclusions:**
We have, for the first time, demonstrated physiologic and molecular differences between dural cells and DM derived from immature and mature animals. These studies suggest that up-regulation of osteogenic growth factor and ECM production, as well as differences in cellular proliferation and differentiation underlie differences between the osteoinductive capacity of immature and mature DM

## HUMAN CARTILAGE TISSUE ENGINEERING: CHONDROCYTE EXTRACTION, PROLIFERATION, AND CHARACTERIZATION

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**Introduction:** We evaluated human rib chondrocyte extraction for tissue engineered cartilage construction. We also evaluated human auricular chondrocytes to compare the respective properties of these two cartilage types. Additionally we examined chondrocyte proliferation in tne unstimulated state and in response to transforming growth factor beta-1, fibroblast growth factor-2, and bone morphogenetic protein-2.

**Methods:** Portions of human rib cartilage (0.3-2.1 g/specimen) and auricular remnants from ear reconstructive surgery for microtia were sterilely stripped of their perichondrium and minced into 1mm pieces. These pieces were then subjected to 0.3% bacterial collagenase II digestion for 2, 6, 12, or 18 hours. The liberated chondrocytes were filtered with a 400 $\mu$m filter mesh and centrifuged. After washing, the cells were resuspended in media and were plated in flasks or chamber slides. Determination of cell number was achieved with a hemocytometer. Samples of extracted cells were subjected to S-100 immunofluorescence staining. Density at confluence was determined after trypsinization of chondrocytes maintained at confluence for 3 days. TGF-$\beta$1 (1ng/ml), FGF-2 (0.4ng/ml), and/or BMP-2 (60ng/ml) in serum-free media were added to pre-confluent chondrocytes and, after 24 hours, cellular proliferation was determined with a colorimetri acid phosphatase assay.

**Results:** Maximum viable cell yield occurred after a 6 hour digestion ($3x10^5$ cells/g). Further digestion yielded more cells, but decreased survival. Renewed digestion of undigested cartilage remaining after 6 nours of digestion did not increase cellular yield. The rapid growtn of the auricular remnant cells (doubling time 3 days) suggested fibroblast overgrowth, likely from unstriped perichondrium and, indeed, S-100 staining confirmed the lack of the chondrocyte phenotype. In contrast, S-100 staining of the rib cellular extract was consistent with the chondrocyte lineage. The chondrocytes were large with a density at confluence of 2.5x10 (+/- $5x10^5$) cells/T75. Initially, the chondrocytes grew with a doubling time of 8 (+/- 1) days. As the cells were passaged, however, proliferation decreased such that by passage six, the cells stopped proliferating and adopted a large, spindle-shaped morphology. Interestingly, TGF-$\beta$1, FGF-2, and BMP-2, alone or in combination, did not significantly alter proliferation of the cells.

**Conclusion:** The theoretical appeal of autologous chondrocyte harvest, culture expansion, and tissue engineered matrix construction is met with challenges at each step. We have identified that the major obstacles to the use of autologous rib chondrocytes in matrix construction are the low cell yield from a small piece of rib and the limited proliferation these cells will undergo. Further investigation of mitogenic cytokines may help resolve this problem. Additionally, TGF-$\beta$1 may provide a means to prevent terminal differentiation of these chondrocytes thereby allowing for continued proliferation. Autologous auricular cartilage has little practical application in tissue engineering because of the limitea donor source and the insufficient rigidity required of a framework for ear reconstruction. Furthermore, our work has demonstrated that the microtic auricular remnant is not easily suitable to chondrocyte extraction due to the technical difficulty of fully removing the enveloping perichondrium resulting in fibroblast overgrowth in culture.



## THE EFFICACY OF OSCILLATING DISTRACTION OSTEOGENESIS OF THE RODENT MANDIBLE

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**Purpose:**
Distraction Osteogenesis (DO) is a powerful technique for mandibular reconstruction. The alternative lengthening and compression of the distraction regenerate ("pumping the regenerate") has anecdotally been reported to enhance osteogenesis and bone stock. The purpose of this experiment was to systematically test this hypothesis using a rat model of mandibular DO.

**Methods:**
The mandibles of 12 rats were osteotomized and a distraction device was applied. Animals were divided into two groups. Group I animals (n=6) were distracted to 2.5mm over 5 days, compressed 1.0mm over a two day period, followed by gradual distraction to a Final length of 5mm. Group H animals (n=6) were distracted 5mm without compression. All animals were allowed to consolidate for 5 wks. Regenerate cross-sectional area and volume were evaluated using 3-D CT reconstruction and histologic sections.

**Results:**
Both groups demonstrated excellent regenerate bone formation and complete closure of the distraction gap. Analysis of regenerate volume and cross sectional area using 3-D CT and histological sections failed to demonstrate significant differences between groups. In addition, histologic sections failed to demonstrate marked differences in regenerate bone thickness or remodeling.

**Discussion:** We have shown that oscillating DO has, contrary to popular belief, little effect on the volume, cross-sectional area, and quality of the distraction regenerate. Biomechanical testing of these specimens will shed more light on the potential utility of these techniques in enhancing osteogenesis during DO.

## OSTEOGENESIS DURING MEMBRANOUS BONE HEALING: A COMPREHENSIVE GENETIC INVESTIGATION OF TEN CRITICAL MOLECULES

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**Introduction:**
A wide variety of molecules have been sought to improve bony wound healing in craniofacial procedures. Three important groups of molecules: the TGF-β super-family, angiogenic cytokines such as VEGF, and extracellular matrix proteins play active roles in bone regeneration and remodeling. However, the temporal expression of these molecules during membranous bone healing remains unknown. Therefore, in this comprehensive study, we examined the temporal expression of ten genes thought to be important in rat mandibular bone repair to determine molecules that may be candidates for therapeutic manipulation.

**Methods:**
Adult male rats (n = 28) underwent identical right mandibular osteotomies and rigid external fixation. Four animals were sacrificed on 3, 5, 7, 9, 23, and 37 days post-operatively. Four control animals underwent soft-tissue sham procedures. RNA from each mandible was extracted and pooled. Northern blot analysis was performed using probes specific for VEGF, TGF-β1 and β3, TGF-βR-II, IGF-I and II, collagen I and II, osteocalcin and TIMP-1.

**Results:**
IGF I and II mRNA expression demonstrated the most activity with increases of 2.1 and 2.3 respectively in mid-repair. Expression of VEGF was downregulated to 60% of baseline after fracture then rose later in repair. Interestingly, neither TGF-β1 nor TGF-β3 showed dramatic increases. In contrast, TGF-βR-II initially decreased, then became upregulated at day 9. Collagen I and II became elevated in mid-repair and remained increased. Osteocalcin was dramatically downregulated, then incrementally returned to baseline by 37 days. TIMP-1 also increased 2-fold later in repair.

**Conclusions:**
In this study we have demonstrated the complex temporal pattern or symphony of gene expression during membranous bone repair. The temporal analysis of gene expression during this process has significant clinical implications since the elucidation of these mechanisms may lead to biomolecular approaches to accelerate and improve bone healing.



## FRACTURE HEALING: THE EFFECTS OF STRAIN ON OSTEOBLASTS

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**Introduction:**
Wolff's Law defined the importance of mechanical stimulation on bone remodeling. More recently, distraction osteogenesis (gradual stretching of a fracture callus) has been shown to stimulate osteogenesis. In these experiments, we sought to define the cellular effects of mechanical stimulation (strain) on isolated osteoblasts by analyzing the expression of cytokines critical to bone repair and angiogenesis.

**Methods:**
Osteoblastic cells underwent cyclic strain (low/10Hz or high/20Hz) for 24 or 48 hours using a Flexcell Strain Unit. Control cells were unstrained. Cellular proliferation, as well as alkaline phosphatase, VEGF and TGF-β1 protein production was determined.

**Results:**
Low strain (10Hz) resulted in more than a 2x decrease in cell number (*$p<0.05$). This decrease was accentuated in cells subjected to high strain (2.5x to 5.5x after 24 and 48 hrs, respectively; $p<0.05$). Interestingly, alkaline phosphatase production, a marker of osteoblastic activity, was increased in strained osteoblasts. In addition, cellular VEGF and TGF-β1 protein production were dose dependently elevated by strain (*$p<0.05$).

**Conclusions:**
We have demonstrated a simultaneous and dose-dependent increase in cellular VEGF and TGF-β1 protein production by osteoblastic cells in response to mechanical strain. These changes may reflect a potential mechanism by which angiogenesis and osteoblastic recruitment occur during fracture repair and distraction osteogenesis. In addition, increased alkaline phosphatase activity in response to mechanical strain implicates this stimulus in the regulation of osteoblastic differentiation, which may, in turn, underlie alterations in cellular proliferation.

## NEL-2 ASSOCIATED WITH BONE FORMATION IN CRANIOSYNOSTOSIS

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**Introduction:**
Surgical correction of unilateral coronal synostosis (UCS) offers a unique opportunity to examine the molecular differences between an abnormal versus a normal cranial suture. Previously, we reported the isolation and identification of a cDNA fragment whose expression was upregulated in the premature fusing and fused coronal sutures, as compared to normal coronal sutures in UCS. The nucleotide sequence of the full length cDNA of this gene has approximately 61% homology with the chicken *Nel* gene, and therefore we named this cDNA human *NEL-2*. Both chicken Nel and human NEL-2 are comprised of 6 EGF-like repeats.

**Methods and Results:**
*In situ* hybridization of human *NEL-2* on UCS sutural samples showed that *NEL-2* messages were localized primarily in the mesenchymal cells and osteoblasts at the osteogenic front, along the parasutural bone margins, and within the condensing mesenchymal cells of newly formed bone in sites of premature sutural fusion. Human multi-organ tissue mRNA blot showed that *NEL-2* was specifically expressed in fetal brain but not in fetal kidney, liver, or lung. We also showed that *Nel-2* was expressed in rat calvarial osteoprogenitor cell cultures and was largely absent in rat tibiae and fibroblast cell cultures. Transient transfection of *Nel-2* gene with a mamalian expression vector in MC3T3 osteoblast cell line demonstrated increased deposition of collagen and mineralized nodules of up to two fold as compare to control.

**Conclusions:**
Our data suggest that the *NEL-2* gene is preferentially expressed in cranial intramembranous bone and neural tissue (both of neural crest cell origin) and is up-regulated during unilateral premature closure of the coronal suture. The precise role of this gene is currently unknown.



## A LONG TERM CONTROLLED OUTCOME ANALYSIS OF *IN UTERO* VS. NEONATAL CLEFT LIP REPAIR USING AN OVINE MODEL

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**Introduction:**
Successful open repair of a cleft lip *in utero* has the advantage of scarless wound healing in the fetus. Unfortunately, no long-term outcome studies have been performed to evaluate the efficacy of these repairs. No study to date has compared the long-term results of an *in utero* cleft lip repair to a similar, control matched newborn cleft repair. This study was performed to evaluate the 9-month outcome of *in utero* cleft lip surgery compared to an identical cleft lip repair performed on infant lambs.

**Methods:**
*In utero* epithelialized cleft lips were created through an open hysterotomy in 16, 65 day old fetal lambs (term = 140 days) using methods described by Longaker et al. Eight of 16 animals underwent subsequent *in utero* repair of these clefts at 90 days gestational age. The repair of the remaining 8 animals was delayed until 1 week post partum. At 9 months, the animals were analyzed for changes in lip contour and degree of scarring by Hematoxylin & Eosin and Masson's Trichrome collagen staining.

**Results:**
Two animals in each group died from preterm labor. Of the animals that survived to term, all repaired lips had some degree of abnormality postoperatively. One out of 6 lips repaired *in utero* dehisced prior to delivery. Three out of 6 neonatal repairs dehisced in the first post-operative month. In the remaining animals with intact lip repairs, the vertical lip height on the repaired side was an average of 9-12 mm shorter than the normal lip in both the *in utero* and neonatal repaired animals. Phenotypically, the postnatal repaired animals has more lip distortion and visible notching. Histologically, the *in utero* repair was scarless while the neonatal repairs had scar throughout the entire vertical height of the lip with an associated loss of hair in this region. Maxillary growth was also evaluated. There was no inhibition of maxillary growth in the animals that underwent *in utero* cleft lip repair. However, in the neonatal repair group, significant maxillary retrusion was evident. Compared to the cleft side of the maxilla, horizontal growth was decreased by 11% ($p=0.01$). Compared to the intrauterine repair group, there was a 17% decrease in horizontal maxillary width ($p=0.01$).

**Conclusion:**
Straight-line *in utero* repair of a cleft lip produces a better long-term result of maxillary growth than a similar repair performed postnatally in the ovine model. There was no diminution in maxillary growth in the *in utero* treated animals. Histologically, *in utero* repair of clefts was indeed scarless. However, both lip repairs produced lips that were significantly shorter than their contralateral non-cleft sides. This degree of lip shortening would require a secondary lip revision, thereby defearing the purpose of performing an intrauterine repair. Comparisons now need to be made between *in utero* and neonatal repairs using a Millard-type rotation advancement technique before intrauterine treatment can be considered to be more beneficial than our current treatment modalities.

## TREATMENT OF POTENTIALLY FATAL EXTENSIVE RADIONECROSIS OF NASOPHARYNX

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**Case report:**
We reported a 61-year-old gentleman with radionecrosis of nasopharynx after combined courses of external irradiation and iridium mould brachytherapy for nasopharyngeal carcinoma. He presented with intractable headache and neck pain five years after the radiotherapy. Endoscopic examination revealed extensive ulceration over the whole nasopharynx, extending to left lateral oropharyngeal wall. C1 and C2 vertebral bodies were exposed. This potentially fatal complication was managed by operative intervention. The nasopharynx was accessed by split-palatal approach, which allowed wider exposure for adequate debridement of necrotic tissues. The defect was resurfaced with a vascularized rectus abdominis muscular flap. Full epithelization of the muscular flap was documented endoscopically four months after the surgery. Despite the symptom of nasal blockage attributed to the velopharyngeal stenosis, complete relief of his headache and neck pain were achieved. There was no evidence of ongoing sepsis around the skull base two years after the intervention. The potentially fatal complication of radionecrosis of nasopharynx can be remedied by the microvascular transfer of muscular flap following adequate debridement.



## HEMIFACIAL AUGMENTATION USING COMBINED PEDICAL SUPERFICIAL TEMPORAL FASCIAL FLAP WITH DERMAL AND SUBDERMAL GRAFT

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**Objective:**
In order to improve the thickness of the transplanted tissue flap and also increase the flap survival rate.

**Method:**
Designed the pedicled superficial temporal fascial flap involving dermal fat to treat hemifacial depression resulting from different causes.

**Results:**
Twenty one cases have been accomplished with satisfactory therapeutic results.

**Conclusion:**
The new technique is a simple safe and effective procedure for treatment of facial depression deformity.

**Key words:**
Superficial temporal fasica, Dermal fat, Tissue flap, Repair face

## HYDROXYAPATITE CEMENT FOR RECONSTRUCTION OF CRANIOFACIAL DEFECTS – A MULTI-INSTITUTIONAL REVIEW

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**Introduction:**
Reconstruction of craniofacial defects remains a significant challenge despite the numerous synthetic bone substitutes which have been developed. Hydroxyapatite cement (HAC) is a relatively new biomaterial which has been demonstrated in clinical trials to have wide application and excellent results in craniofacial reconstruction. Clinical trials have demonstrated HAC to be easily contourable, osseointegrated with bone, and minimally reactive.

**Methods and Results:**
In a multi-institutional retrospective study involving three craniofacial centers, HAC was implanted in 89 patients over the past 2.5 years. HAC was commonly used for post-synostosis defects, post craniotomy defects, and traumatic defects involving the skull, frontal sinus, orbits or facial skeleton. Average age ranged between 2 and 63 years with a mean skull defect of 58 $cm^2$ and average HAC volume used of 34 gms. In follow-up of 1 to 2.5 years, 90% of patients had excellent skull contour. 10% of patients developed complications requiring operative intervention including seromas (5), extrusion (1), wound problems (2).

We have subsequently found that 1) meticulous hemostasis with a dry surgical field, 2) mixing of HAC with sodium phosphate solution instead of water, 3) allowing adequate time for complete drying and setting of the compound, 4) the use of an underlying titanium mesh or absorbable plates to provide for the support of HAC and to dampen dural pulsations, 5) the use of a closed suction surgical drain, and 6) careful watertight wound closure to prevent infection all greatly facilitate the ease of use and the successful outcome for the patient.

**Conclusions:**
In our institutions, HAC has replaced methyl methacrylate and autologous bone as the material of choice in craniofacial reconstruction because of its contourability, ease of use, biocompatibility, osseointegrative properties, and its low complication rate.



## INHIBITION OF MOUSE CALVARIAL SUTURE FUSION BY TGF-β ANTIBODY AND ANTISENSE PLASMID DNA

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**Introduction:**
We have shown that TGF-$\beta_1$ is actively expressed during mouse calvarial suture fusion. The present study was performed to investigate whether modulation of suture fusion can be achieved by blocking the bioavailability of TGF-β.

**Methods:**
*In vitro*: The posterior frontal (PF) sutures from 24 day old mice were harvested and cultured for 2, 3, or 4 weeks in the presence of 20 μg/ml of pan-specific TGF-β polyclonal antibody or rabbit IgG as a control (n=3). Culture media were changed every 48 hours and fresh antibody or rabbit IgG were added upon each media change. Suture fusion was evaluated by histometric analysis. *In vivo*: TGF-β cDNA was cloned and inserted in opposite direction into the expression cassette of the plasmid vector RLDN. This TGF-β antisense containing 10 μg plasmid DNA in 50 μl lipofectamine was then injected into the subgaleal layer along the frontal suture of 22 day old mice under anesthesia (n=3). For control groups, 50 μl lipofectamine alone was used (n=3). The PF sutures were harvested at various time points and examined by histometric analysis, immunohistochemistry, and reverse transcription and PCR for the detection of mRNA.

**Results:**
*In vitro*: Histology showed that the presence of TGF-β antibody in culture media delayed PF suture fusion. By three weeks in culture new bone area was only 20% of that in control groups as determined by histometric analysis. By the end of the $4^{th}$ week suture fusion was only 25% completed compared to controls. *In vivo*: Histology showed that PF suture fusion was significantly delayed in the antisense group compared to control. By postnatal day 45, suture fusion was complete in control animals. There was a 70% inhibition of suture fusion (new bone area) in antisense groups measured with an imaging system. Four days after antisense injection (age 26 days), mRNA expression for TGF-β was 77% lower in the anitsense group (2.3±1.8) than in the control group (9.8±3.1). Immunohistochemistry studies also showed decreased TGF-β1 expression in the antisense group.

**Conclusions:**
We have successfully established *in vitro* TGF-β antibody delivery system and *in vivo* TGF-β antisense plasmid DNA transfection system in mouse calvarial sutures. These data indicate that TGF-β plays a causative role in mouse PF suture fusion and that modulating TGF-β gene expression *in vivo* can alter the natural history of cranial suture fusion.

## A NOVEL MURINE MODEL FOR STUDY OF CLEFT LIP AND PALATE: MOLECULAR CHARACTERIZATION OF THE *TWIRLER* MICE

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**Introduction:**
The *Twirler (Tw)* mouse model offers an excellent system to analyze the genetic contributions of cleft lip and palate as all homozygous *Tw* mice *(Tw/Tw)* present with clefts of the palate and lip. The ability to genotype *Tw/Tw* before any phenotypic manifestation of clefts makes the *Tw* a powerful system to begin the dissection of the genetic pathway of the etiology of clefts of the lip and palate. With this in mind, we characterized the expression of a number of genes shown to play critical roles in the development of the facial primordia and palatal shelves in the *Tw* mice.

**Methods:**
*Tw/Tw* embryos at gestational ages 10.5, 11.5, 12.5, 13.5, and 14.5 days post-coitum were obtained by breeding *Tw/+* pairs. Genotyping was performed on these embryos by polymerase chain reactions with primers complementary to microsatellite markers on the *Tw* locus. Wildtype and homozygous *Tw* mice were processed for histology and *in situ* hybridization. Whole mount *in situ* hybridization was performed using antisense digoxigenin labeled murine *Msx-1*, *Msx-2*, or TGF-β1 RNA probes.

**Results:**
At day 11.5 dpc *Msx1* expression was reduced in the *Tw/Tw* medial and lateral nasal processes compared to the wildtype. At 13.5 dpc TGF-β1 expression was also reduced in the medial epithelial region of the secondary palate in *Tw/Tw* mice compared to the wild type.

**Conclusion:**
Increasing evidence suggests that *Msx-1* and TGF-β play significant roles in midfacial and palatal development. Reduced expression of *Msx-1* and TGF-β1 in the facial primordia and palatal shelves respectively in *Tw/Tw* compared to that of wildtype mice suggests that these genes play an important role in the etiology of clefting in the Tw mice. Further characterization of other important regulatory genes in the craniofacial region of *Tw* mice would yield more information on the role of such genes in the etiopathology of clefts of the lip and palate. Once the molecular basis of clefts of the lip and palate is understood, better strategies, such as gene therapy, can be designed to treat and prevent this birth defect in humans.



## FGFR3-ASSOCIATED CORONAL SYNOSTOSIS SYNDROME: PREVALENCE AND CLINICAL PRESENTATIONS

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A mutation in the FGFR3 gene that converts Proline 250 into Arginine has been recently identified in apparently non syndromic coronal synostoses. Eighty-seven patients with sporadic or familial coronal craniosynostosis were investigated for this mutation. In familial cases, the mutation was identified in 24/33 families (73%), while only 7/54 sporadic cases (13%) harbored the mutation.

There was a inter- and intrafamilial variability of the coronal synostosis and bone anomalies of the hands and feet in affected individuals. Females were significantly more severely affected than males.

The most typical presentation was the association of bicoronal synostosis with hypertelorism and marked bulging of the temporal fossae. The comparison of morphological and functional results in patients positive or negative for the P250R mutation suggest that molecular identification is necessary in all patients with coronal craniosynostosis.

## EVALUATION OF FREE FLAPS APPLICATION IN DEFECTS AFTER CRANIOFACIAL EXTENDED RESECTION

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**Objective:**
This clinical survey investigates value of free flaps in the tissue reconstruction after craniofacial extended resection.

**Methods:**
From July, 1976 to July, 1998, forty-nine cases of advanced malignancy tumors involved base of skull, underwent craniofacial extended resection combined with reconstructive surgery using myocutaneous flaps in one-stage. From 1980, all cases were treated by free myocutaneous flaps to reconstruct their defects. There were 24 cases undertaken many flaps included 15 cases of pectoralis major myocutaneous flaps (PMMF), PMMF combined with radical forearm flaps in 5 cases, and latissimus dorsi myocutaneous island flaps in 4 patients.

**Results:**
All twenty-nine flaps in twenty-four operation survived completely. Both contours and effects were satisfactory. Apart from one case dead from cerebral edema, most of patients survived without severe complications.

**Conclusions:**
It is regarded that free flaps is excellent for reconstructing tissue defects after craniofacial extended resection without influence next radio-therapy or chemotherapy. It elevated patients' life quality and is recommended for clinic.



## THE EFFECT OF EARLY LEFORT III SURGERY ON PERMANENT MOLAR ERUPTION IN PATIENTS WITH CRANIOSYNOSTOSIS

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**Purpose:**
The study was completed to evaluate the extent to which an early LeFort III surgical procedure affects the position and eruption of the maxillary first and second permanent molars in patients diagnosed with craniosynostosis syndromes.

**Methods:**
This is a retrospective study of 33 patients diagnosed with craniosynostosis (14 Crouzon's, 9 Apert's, 9 Pfeiffer's and 1 Carpenter's). Mean age at the time of surgery was 5.32 years (S.D. 1.3 years). All patients underwent a LeFort III surgical procedure in order to correct the anatomical deformity for functional and psychosocial reasons. A single surgeon performed all surgeries. Eighteen unoperated patients with craniosynostosis (11 Crouzon's, 4 Apert's and 3 Pfeiffer's) served as controls. Mean age in this group at the time that the last radiograph was taken was 21.18 years. Molar position and eruption pattern was assessed by panoramic radiographs by multiple observers (N=3). The criterion for adequate molar eruption was its presence in the oral cavity, or if not erupted, proper position of the molar and adequate space for eruption. First and second molars were evaluated separately. Cases where molars were severely displaced, rotated (more than 45 degrees), ankylosed or impacted were considered non-functional with no possibility for eruption. If any doubt existed in the adequacy of eruption, it was classified in the erupting molar group.

**Results:**
For the 33 patients who underwent surgery, long-term evaluation showed that 77.4% (48/62) of all first permanent molars and 22.6% (14/62) of all second permanent molars erupted or would erupt. The control group showed 100% (36/36) of all first permanent molars erupted and 89% (32/36) of second permanent molars erupted.

**Conclusions:**
It appears that in the majority of cases, an early LeFort III osteotomy affects first and second maxillary permanent molar tooth bud position by decreasing the probability of their eruption.

# Index for Participants

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