New Surgical Concepts Resulting From Cranio-orbito-facial Surgery

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The authors have defined the subspecialty of craniofacial surgery and described the organization of the multi-disciplinary team required to care for such patients. Common features of the craniofacial patient have been summarized and three major categories of patients have been proposed. These are: I. Syndromes associated with hypertelorism; II. Syndromes associated with premature synostoses or growth arrests; III. Syndromes associated with primarily mid- and lower face anomalies. Growing out of an experience with 242 operations on 106 patients, the authors have listed 9 relatively new surgical "principles." Each has led to a current surgical approach that is now being employed by the craniofacial team at The University of Virginia. A number of examples are given to show ways in which the lessons learned from the craniofacial patients are now being applied, with improved results, to patients with neoplasms, traumatic injuries, or other conditions.

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CRANIO-ORBITO-FACIAL SURGERY is a rapidly developing surgical subspecialty. Dr. Paul Tessier[12–15] of Paris deserves enormous credit for developing and spreading the new approaches to the treatment of these severely deformed children.

In the United States, reports by Converse, Murray,[11] Lewin, Munro,[9,10] Salyer,[7] as well as our own[2,3,6] have documented the success of these operations.

Initially, it was thought that the rarity of these deformities would limit the application of the newly learned surgical techniques. However, many more children were referred than expected. We also saw large numbers of children with relatively mild degrees of deformity who are appropriate candidates for these surgical techniques, now that reasonable safety has been demonstrated.

Perhaps no medical condition or disease seen today in a large medical center requires the active involvement of so many different highly skilled health professionals as does the child with major facial clefting, involving displaced eyes, bifid nose, absent ears, and malformed jaws. Team coordination is critical (Tables 1 and 2).

Medical students, residents, nurses, and paramedics also find the complexities of diagnostic analysis and treatment planning for the child with a major craniofacial deformity to be a classic demonstration of the need for intercommunications among medical specialities. Such programs tend to reverse the traditional tendencies of medical specialties to become more isolated from one another. The needs of the craniofacial patient involve many connecting bridges between medical disciplines and serve to unify concepts.

A second characteristic of the "craniofacial" patient is

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TABLE 1. *Craniofacial Team–Diagnostic*

Pediatrician	Orthodontist
Geneticist	Pedodontist
Plastic Surgeon	Neurologist
Neurosurgeon	Speech Pathologist
Otologist	Audiometrist
Opthalmologist	Endocrinologist
Radiologist	Medical Photographer
Oral Surgeon	Computer Scientist

TABLE 3. *Syndromes with Major Degrees of Hypertelorism (Preventing Stereopsis)*

1. Primary Hypertelorism (Greig's Syndrome)
2. Median Fronto-Nasal Clefts
3. Bilateral Para-Median Facial Clefts
4. Unilateral Para-Median Facial Clefts
5. Frontal or Nasal Encephaloceles
6. Frontal or Nasal Meningoceles
7. Lateral Nasal Clefts (? Goldenhar's)

that often the deformity has been left untreated in early childhood, or, if surgery was attempted, only superficial and relatively unsatisfactory corrections of the defects were accomplished. Many of these children are now being discovered in institutions for the mentally retarded, where a baby would be placed shortly after birth because of a grotesque facial appearance with no one realizing, until years later, that the child might have a perfectly normal intelligence.

A third feature of the "craniofacial" patient is that surgical re-arrangement of the orbital and skull base architecture is usually best, or most safely, accomplished if the cranial cavity is opened. This improves surgical exposure, protects the brain and dura, reduces the danger of postoperative meningitis, and makes possible the synchronous correction of cranial and facial aspects of certain deformities.

Frequent Diagnoses at the Virginia Craniofacial Center

There are several major categories of deformity that make up the patient material referred to a craniofacial center. Over the past 5 years (June 1970 to June 1975), at The University of Virginia, we have carried out 242 operations on 106 patients with major craniofacial deformities. Although there is a significant degree of overlap of syndromes, most of the patients can be considered primarily in one of the following groups:

I. *Hypertelorism (or Hyperorbitism):* This is a symptom and not a specific disorder. It includes all anomalies that result in wider than normal separation of the medial bony walls of the orbits. The diagnosis is based on the measurement of the bony intercanthal distance by x-ray tomography or by caliper at operation. The most common syndromes associated with hypertelorism may be conveniently divided into two groups, depending on the degree of hypertelorism encountered with each condition. (Tables 3 and 4). Other rare conditions are also associated with increased bony intercanthal distances.

Many syndromes also cause hypercanthorum (or increased width of soft tissues between the eyes—with or without the formation of epicanthal folds). These conditions are not listed as they do not involve true increase in width of the ethmoid or interorbital bony complex. They may be corrected surgically by *extra cranial* reconstructions of orbital walls and canthal ligaments.

II. *Deformities Associated with Early Arrest of Growth in Cranial and Facial Bone Suture Lines.* (Table 5): Craniostenoses and faciostenoses (Delaire) are quite serious when multiple suture lines are involved. Characteristically, the lack of growth is accompanied by symmetrical or asymmetrical distortions of the shape of the skull, orbits, face, nose and jaws. The intracranial cavity may be small and the patient may develop increased intracranial pressure or hydrocephalus. Several of these syndromes are familial and hereditary.

When orbital or maxillary bones are involved, the eyes may be proptosed out of the shallow bony orbits, separated more widely than normal, or lie at different horizontal levels in the face. Eye function may be disturbed due to optic atrophy, strabismus, or keratitis associated with poor lid closure.

In many of these syndromes, multiple bones of the mid-face are small and have little growth potential, causing the associated deformities to become progressively more grotesque with each passing year. Secondary deformities of the teeth, tongue, lips, and lower jaw seem to

TABLE 2. *Craniofacial Team–Therapeutic*

Plastic Surgeon	Pedontist
Neurosurgeon	Prosthodontist
Anesthesiologist	Speech Pathologist
Oral Surgeon	Resp. Physiologist (ICU)
Opthalmologist	ICU Nurse Team
Otolaryngologist	Pediatric Psychiatrist
Audiologist	Orthopticist
Orthodontist	Bioengineer

TABLE 4. *Syndromes Associated with Mild Ocular Hypertelorism*

1. Acrocephalosyndactyly (Apert's Syndrome)
2. Larsen's Syndrome (with cleft palate and multiple dislocations)
3. Craniocarpotarsal dystrophy (Whistling-Face syndrome)
4. Craniofacial Dysostosis (Crouzon's Syndrome)
5. Hurler's Syndrome (Gargoylism or mucopolysaccharidosis)
6. Sprengel's Deformity (Klippel-Feil and Wilderranck's Syndrome)
7. Dyscraniopygophalangy (Micrognathia, polydactyly, and genital anomalies)
8. Orodigitofacial Dysostosis (OFD Syndromes)

TABLE 5. *Growth Arrest Deformities of Cranial and Facial Bones*

1. Premature synostoses of cranial bones
2. Crouzon's craniofacial dyostoses
3. Apert's acrocephalosyndactyly
4. Sprengel's deformity (with oculo-auriculo-dysplasia)
5. Idiopathic infantile hypercalcemia
6. Oculomandibulodyscephaly (Hallermann-Streiff syndrome)

appear and increase as the child makes efforts to adapt to the hypoplastic parts of the face. Defective muscle patterns of speech, mastication, and expression are inevitable. Breathing may be so labored that sleep is difficult and general nutrition suffers.

The most common clinical conditions in this group of *growth arrests* are: A) Combinations of premature synostoses of the cranial bone suture lines (sagittal, coronal, metopic, and lambdoidal). We do not believe that fusion of a *single* cranial suture line will cause threatening restriction to brain enlargement. It may, however, produce progressive, visible, asymmetric deformity if not corrected early. B) Crouzon's craniofacial dysostosis (optic nerve involvement is noted in 80% of untreated patients); C) Apert's acrocephalosyndactyly; D) Sprengel's deformity (with oculo-auriculo-dysplasia); E) Idiopathic infantile hypercalcemia; F) Oculomandibulodyscephaly (Hallermann-Streiff Syndrome).

Other rare conditions are seen with premature cranio- or faciostenoses that result in orbital dystocias or asymmetries of the face or cranium.

III. *Deformities Involving Primarily the Bones of the Mid-face, Orbits, and Auriculo-mandibular Regions* (Table 6): These conditions require early bone grafting to maintain facial symmetry and jaw elongation. Soft tissues are frequently hypoplastic and may need pedicle flaps to provide skin and fat. Missing external ears and facial paralyses are common. Eyelid colobomas, epibulbar dermoids, and palate deformities are common in this group of syndromes. Malocclusion and open bite deformities may be severe. Some of these syndromes are associated with aplasia of the corresponding cranial nerves.

Grob[5] has described 13 different patterns of oblique facial clefts that may occur in humans. All radiate from the oral stoma and 8 of these meloschises lie lateral to the attachment of the nose.

TABLE 6. *Deformities Involving Primarily Midfacial Orbital and Mandibular Regions*

1. Hemifacial microsomia
2. Mandibulofacial dysostosis (T-C)
3. Oculo-auriculo-vertebral-dysplasia (G.)
4. Congenital facial diplegia (moebius)
5. Romberg's disease
6. Oro-facial digital dysostosis
7. Arrhinencephaly and cebocephaly
8. Micrognathia and glossoptosis (Robin)
9. Larsen's syndrome (dishface and CP)
10. Oblique facial cleft (meloschisis)

TABLE 7. *Classification of Cranio-facial Anomalies Based on Treatment Requirements*

1. Syndromes with hypertelorism
2. Growth arrest deformities of cranial and facial bones
3. Deformities involving primarily the midfacial skeleton and mandible

The conditions listed in Tables 3 to 6 are those most commonly seen in the Virginia craniofacial program.

Anesthesiology techniques are of special importance in children undergoing craniofacial surgery in three major areas: 1) the cardiovascular system and blood volume, 2) the respiratory and thermoregulartory system, and 3) the central nervous system.

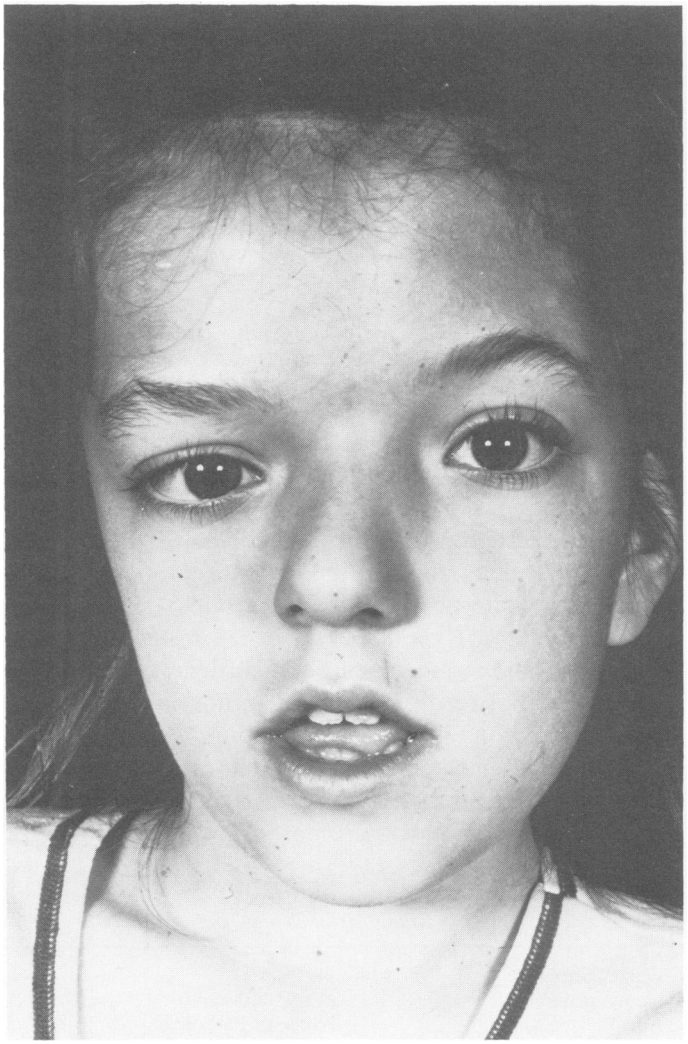

FIG. 1. Case A: This 8-year-old girl had premature synostosis of the left coronal and the anterior sagittal suture. At four months of age, synectomy of the left coronal suture provided additional room for growing brain, but persistent deformity of the forehead with dystocia of the orbits remained as the child grew older.

An arterial line is used on a continuous basis to measure blood pressure, blood gases, and hematocrit. Its presence has made monitoring of vital signs sufficiently accurate that we now routinely use hypotensive techniques—even in infants. The only operative death in our 106 patients occurred early in the series in a small child, before we instituted the routine use of this essential monitoring technique.

This reduces blood loss and operating time. Deep halothane or sodium nitroprusside has been used to keep the systolic blood pressure at 50 to 70 mm Hg. If tissue perfusion is adequate, no base deficits develop. Using this technique, we have had no postoperative deficits.

The addition of humidification to the anesthetic gases (12-16 mg H_2O/l) prevents epithelial necrosis of the tracheobronchial tree. To reduce heat loss, we now also warm anesthetic gases to 35-37 C and warm all intravenous fluids and blood. Even the operating room is maintained at 22-24 C.

We will not attempt in this paper to describe the details of the multiple surgical procedures that have been undertaken to correct these deformities. The surgical exposure of almost every case has led to a better appreciation of the basic anatomical deformity and, consequently, each operation tends to be modified to improve the resultant correction (Figs. 1-22).

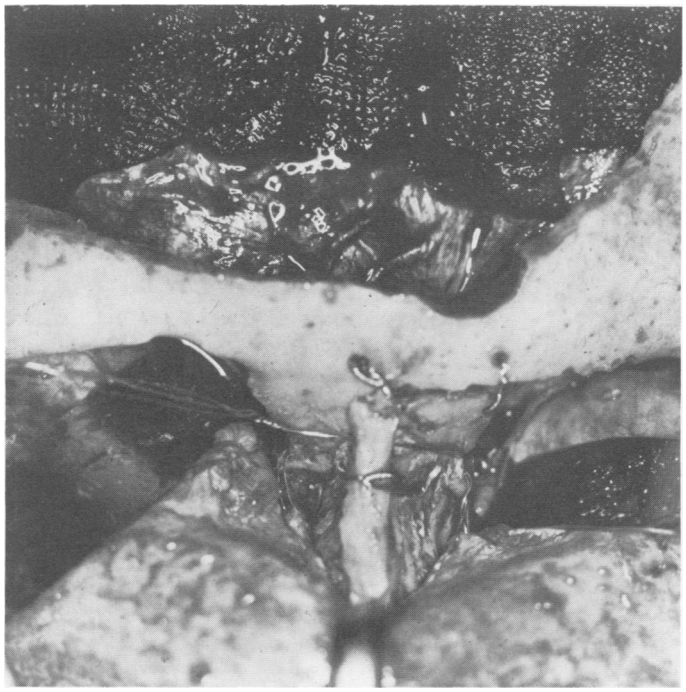

FIG. 3. Case A: After removing the triangular right frontal bone graft and freeing the dura from the floor of the anterior cranial fossa, the left orbit has been mobilized medially and inferiorly to correct the hypertelorism. A graft of rib bone has been fixed to the bridge of the nose with steel wire.

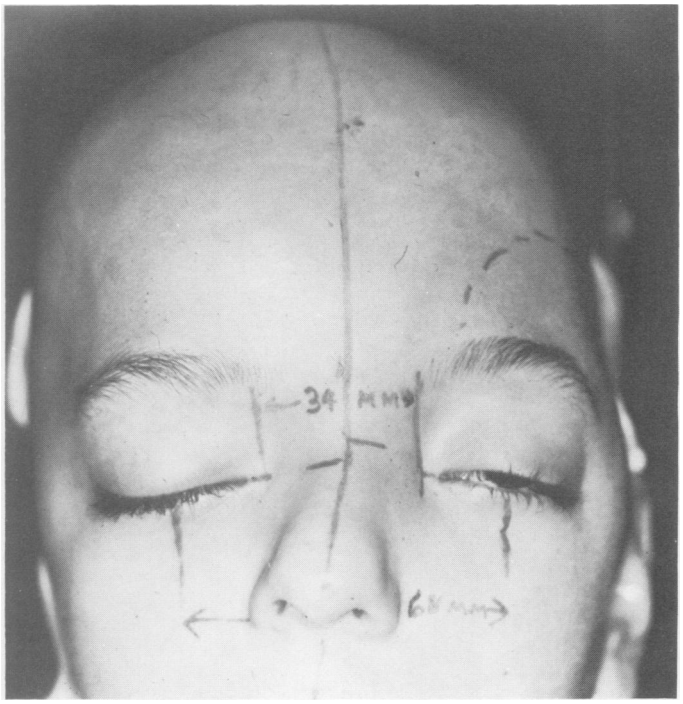

FIG. 2. Case A: At operation, the dye markings indicate a moderate degree of hypertelorism with angulation of the nasal skeleton and downward displacement of the right orbit. Bone is also deficient above the left eyebrow.

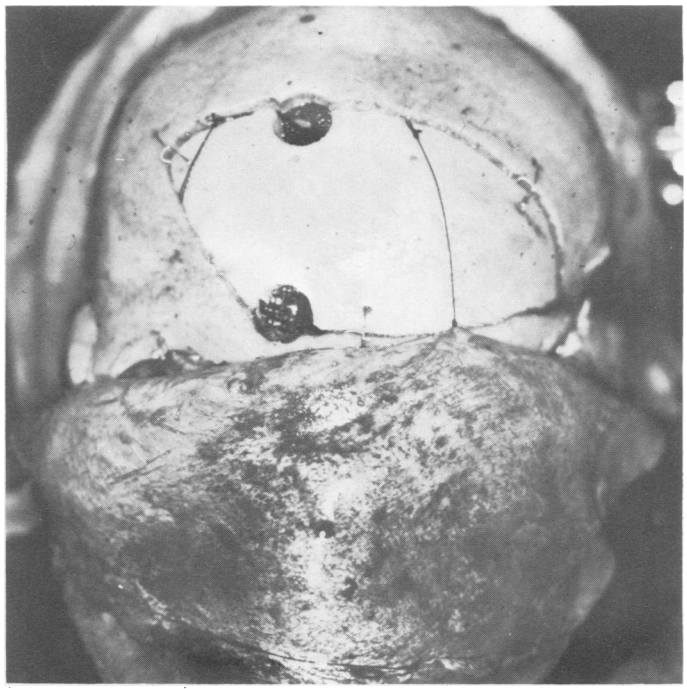

FIG. 4. Case A: The free frontal bone graft has been replaced. Silk suspension sutures hold the dura forward against the under surface of the cranial bone. Periosteum was elevated along with overlying forehead flap.

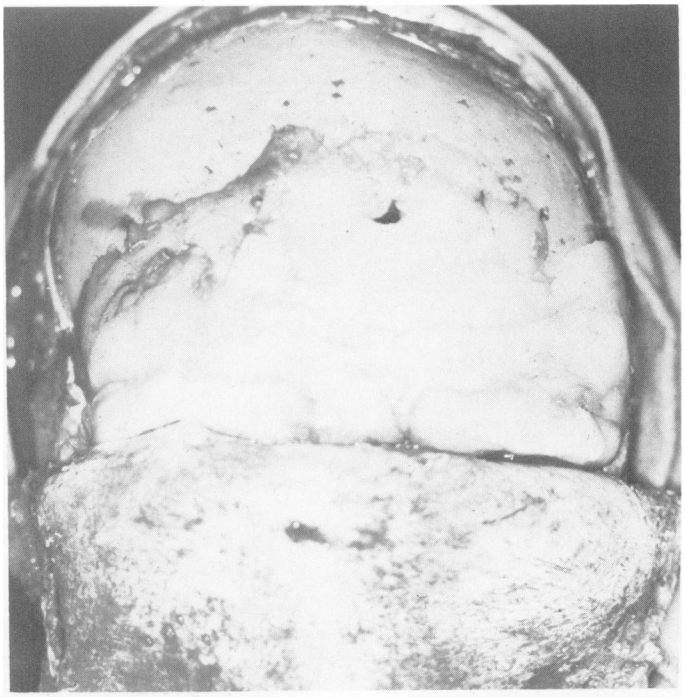

FIG. 5. Case A: Left frontal deficiency has been corrected by immediate application of methylmethacrylate applied over the anterior surface of the frontal bone graft. The blood supply to this bone is furnished by the underlying dura.

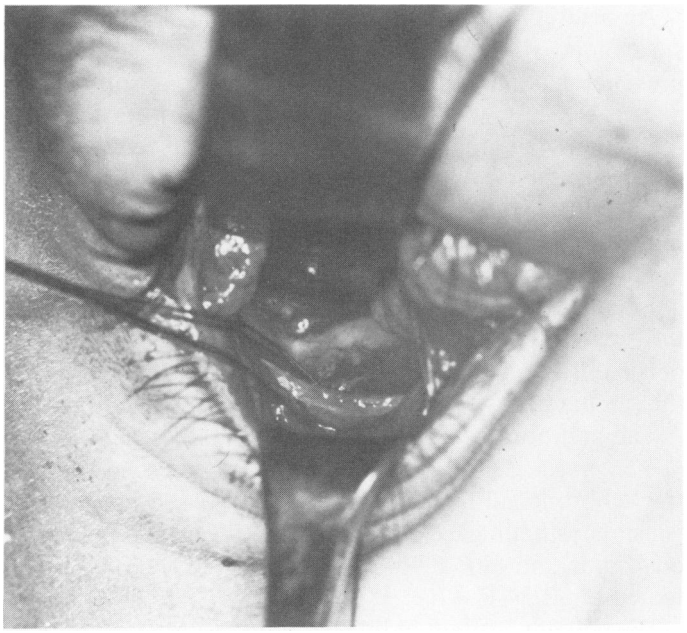

FIG. 7. Case A: The thin inferior orbital rim is identified and a mersilene suture is placed through a drill hole in its margin.

Current Surgical Approach to Cranio-orbito-facial Deformities

We have selected 9 relatively new concepts or principles that have emerged from our operative experiences and followup studies over the past 5 years. Firm data for some of these principles are still lacking. New measurement techniques involving radiopaque metallic markers, implanted in key points of the human craniofacial skeleton at the time of surgery, will give us some of the evidence over the next decade. Animal experiments will help to answer some of the other questions. Careful post-

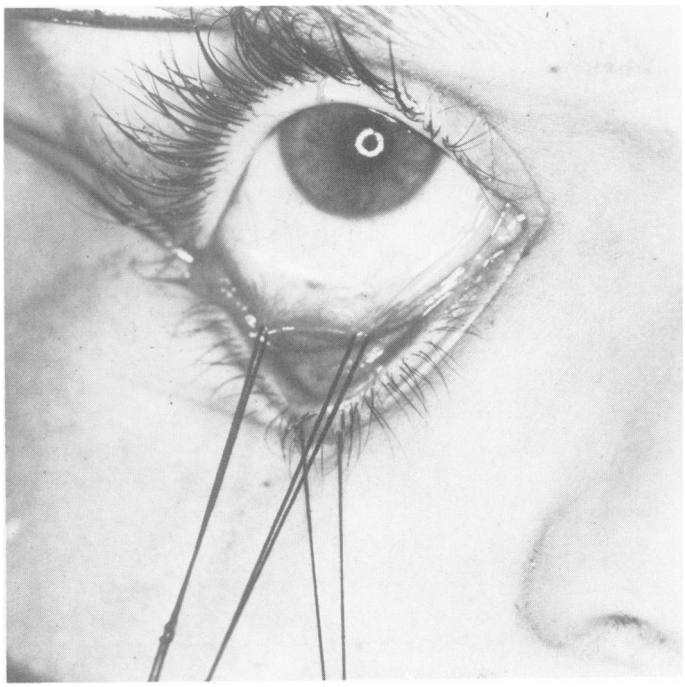

FIG. 6. Case A: In order to raise the level of the right eye, an incision is made through the conjunctiva in the sulcus behind the right lower eyelid.

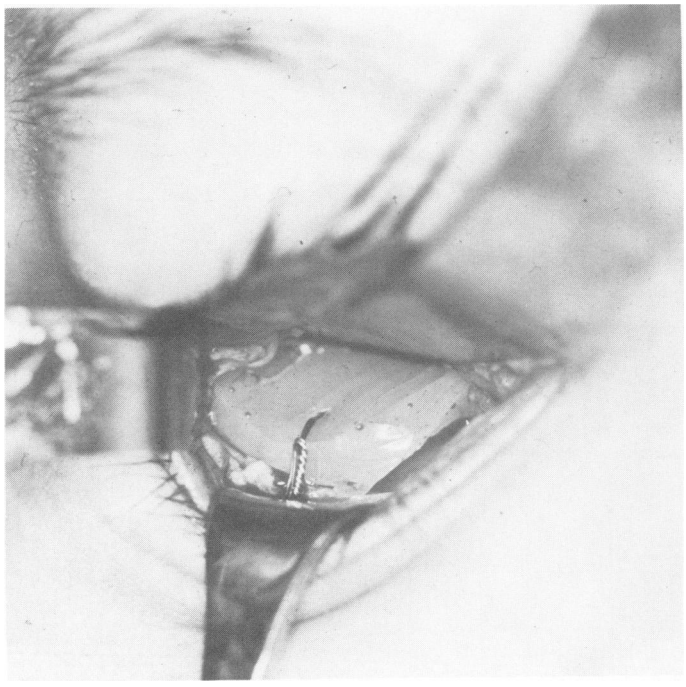

FIG. 8. Case A: An appropriately sized and shaped silicone rubber platform is fashioned to elevate the globe and secured firmly with steel wire.

operative examinations with computerization and pooling of data from several centers will guide us in the future. In the meantime, it is our responsibility to proceed with the care of those children in dire need of reconstruction by use of the methods that represent the most carefully considered facts and intuitions of the present. We would predict that future data will support the following contentions:

1. **Proposition:** Growth and shape of the bones of the skull (and face) are highly dependent on the growth of the brain (and attached muscles of mastication).[18,8]

Corollary: The release and reshaping of the dura may be one of our best surgical methods for control of ultimate head size and shape.

2. **Proposition:** Many growth arrest malformations (craniostenoses and faciostenoses) of the face and skull progressively increase with age due to the lack of growth of the affected suture lines and bones.[4]

Corollary: Earlier surgical corrections carry less danger of inducing additional growth arrests in children with this type of deformity than in the case of similar operations when performed on bones with normal growth potentials. Indeed, early release of soft tissue restrictions may even reduce the expected degree of deformity.

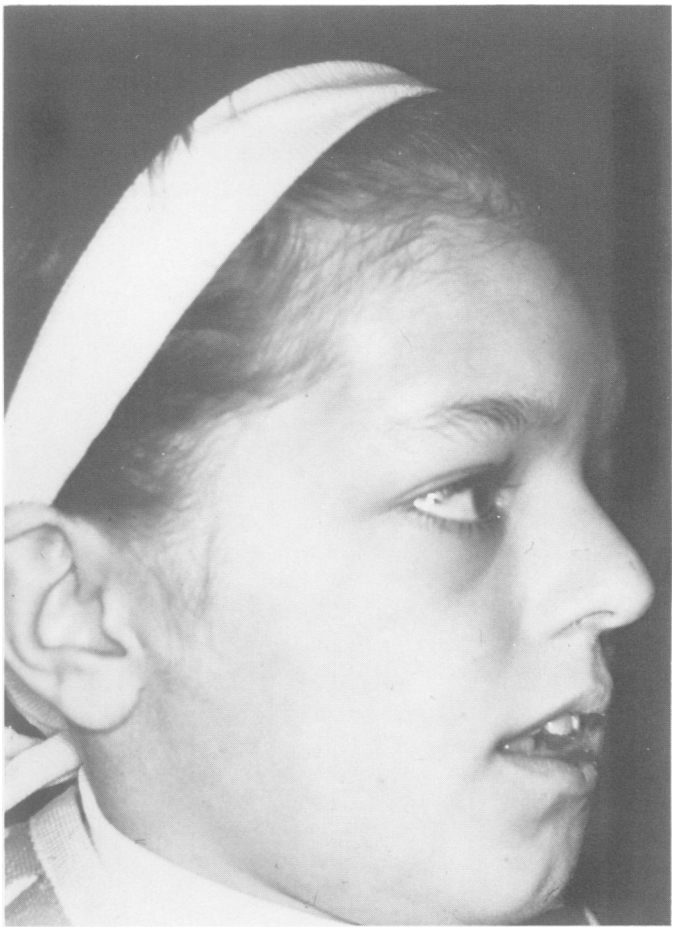

FIG. 9. Case A: The profile one year after surgery shows satisfactory prominence of the supraorbital ridges provided by the methacrylate and without complications.

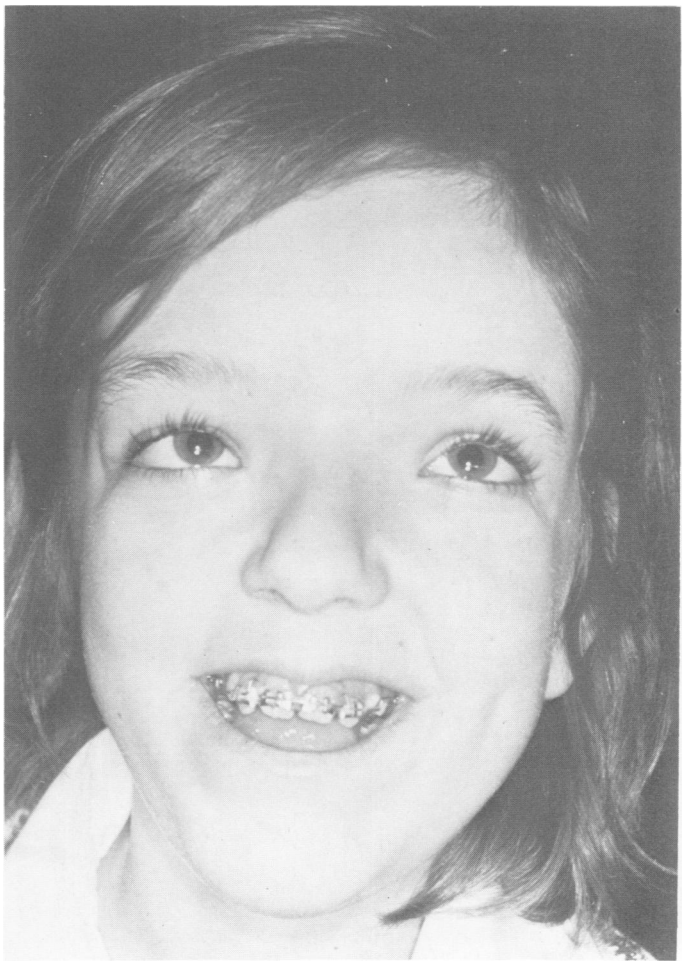

FIG. 10. Case A: Front view of the patient, one year following surgery, with marked improvement in facial symmetry, in eye level, and in the degree of extraocular muscle coordination. There is no dyplopia. This may be compared with the preoperative view in FIGURE 1.

3. **Proposition:** Modern pediatric anesthesiology, with the aid of monitoring and regulatory techniques makes acceptably safe the performance of extensive and prolonged craniofacial operations—even in infants.[1,3]

Corollary: Craniofacial reconstruction should be performed as soon as indicated by the physiological and psychological impacts of the deformity, regardless of age.

4. **Proposition:** The correction and release of the restraints and abnormal tensions produced by muscles and other soft tissues may prove to be the most difficult and essential requirement at operation if the surgeon expects to maintain, postoperatively, the new positions of the craniofacial skeleton and avoid tethering effects on those bony parts with subsequent growth.

Corollary: Operative techniques must place more emphasis on release of dural tensions, mobilization and

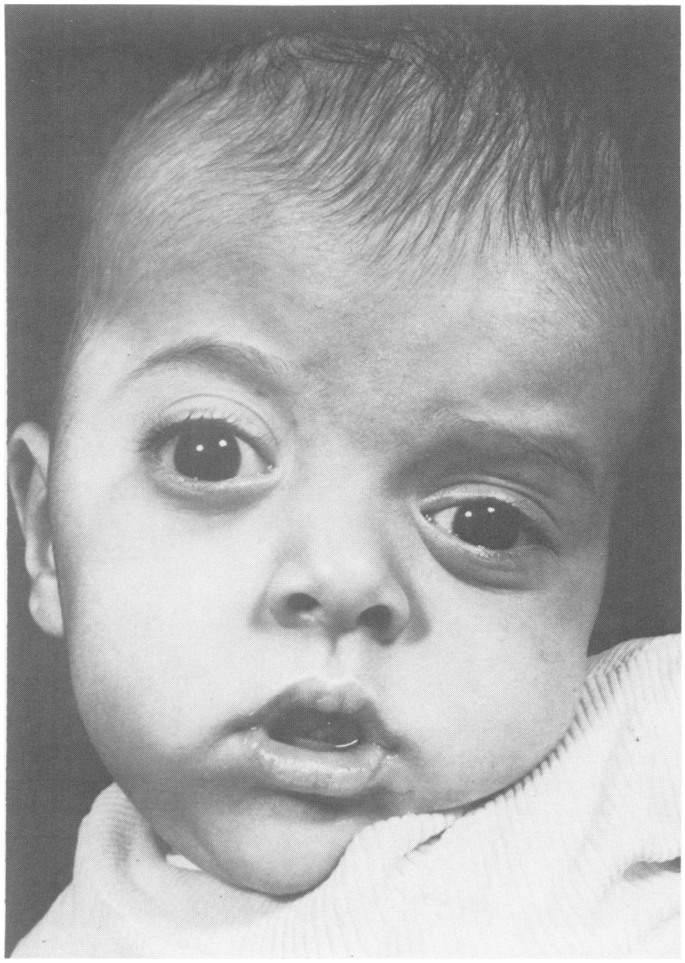

FIG. 11. Case B: This four month old baby boy's mother has typical symmetrical Crouzon's deformity. Since the growth arrest involves the suture lines at the base of the skull, one may predict that the deformity will progress with age.

section of periosteum, freeing of muscle attachments, and relaxation of overlying skin and fascia if postoperative "relapse" of repositioned bones is to be prevented. Bone grafts will absorb if wedged into position under excess pressure. The role of postoperative skeletal traction needs further study.

5. **Proposition:** Most craniofacial reconstructions require large amounts of autogenous bone and/or alloplastic material to augment the huge congenital skeletal deficiencies and bony gaps seen after osteotomies and repositioning.

Corollary: Late donor site deformities are enormous if large amounts of iliac bone are removed from the hips of small children; thus, the use of alloplastic materials for skeletal augmentation becomes an attractive choice when operating on small children.

6. **Proposition:** Large surgical implants of methylmethacrylate have proved to be well-tolerated (3 wound complications in 34 craniofacial patients). This is true even though there has been routine opening of the paranasal, nasal, pharyngeal, and oral cavities. Such implants may be used alongside autogenous bone grafts.[7]

Corollary: Alloplastic materials will play a steadily increasing role in contour augmentations ("Float-on-bone" technique) of the skull and face. Where structural wedging is needed, autogenous bone grafts remain our primary choice.

7. **Proposition:** The accuracy of postoperative neurological evaluations for increased intracranial pressure is significantly reduced after craniofacial surgery as a result of lid edema, facial dressings, and alterations of normal eye reflexes by surgical manipulations.

Corollary: A newly designed "pressure screw"[16,17] may be inserted in the subarachnoid space at the time of surgery. This will give direct, continuous, and reassuring monitoring of intracranial pressure, P_{O_2}, P_{CO_2}, and CSF pH in the postoperative period.

8. **Proposition:** Binocular vision with image fusion may be possible if the displaced orbits of children are brought back into relatively normal positions by the time the child is four years of age.

Corollary: Clinical evidence would suggest that pa-

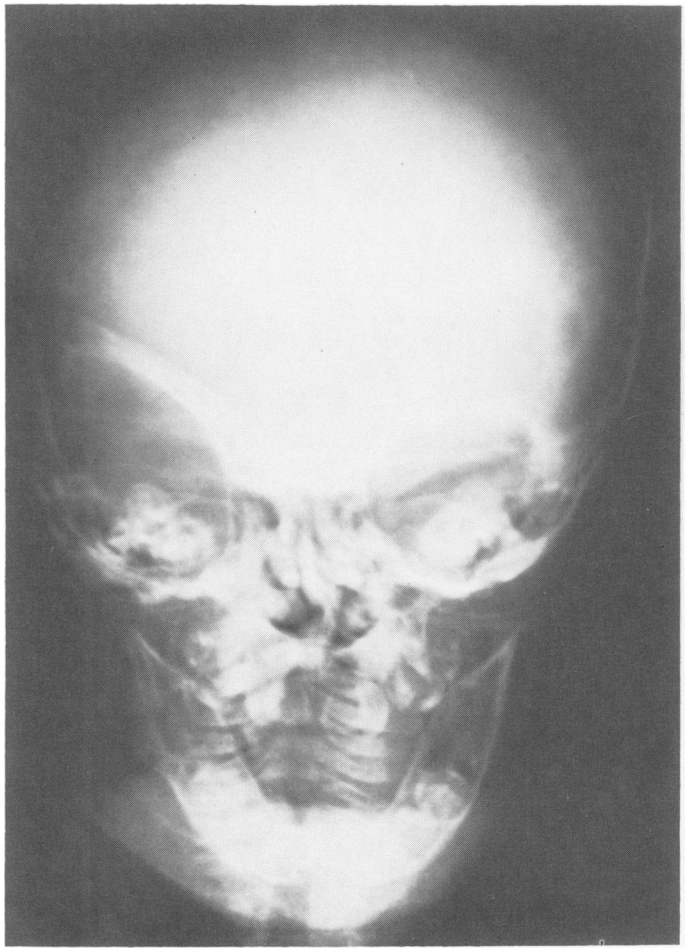

FIG. 12. Case B: X-ray of the skull, showing orbital deformity. Mild hypertelorism is present and the right orbit is in an elevated position.

tients with marked hypertelorism should receive orbital repositioning during infancy if optimum visual function is to be obtained.

9. **Proposition:** Massive craniofacial deformity obscures neurologic and psychologic potentials, making current methods of psychometric testing especially difficult and unreliable.

Corollary: The prognosis for development and function of a deformed child after craniofacial surgery is proving to be considerably better than preoperative testing or performance would suggest.

Discussion

The above propositions represent the conclusions and working hypotheses of the Virginia Craniofacial Surgical Team. They are based on the experiences involved in the treatment of 106 craniofacial patients.

The detailed review of the diagnoses, results, complications, and followup of these patients will be reported in

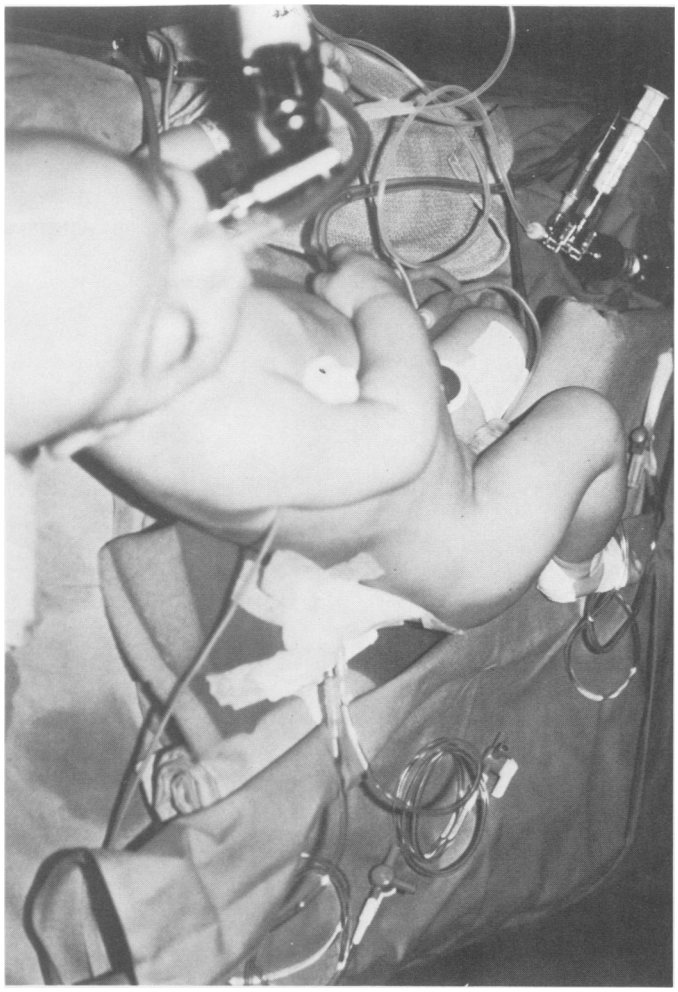

FIG. 13. Case B: Shows the baby at the time of surgery with monitoring devices including an arterial line, venous pressure, two lumbar space needles, a bladder catheter, and an intravenous line. The anesthetic gases are being warmed and humidified before reaching the trachea.

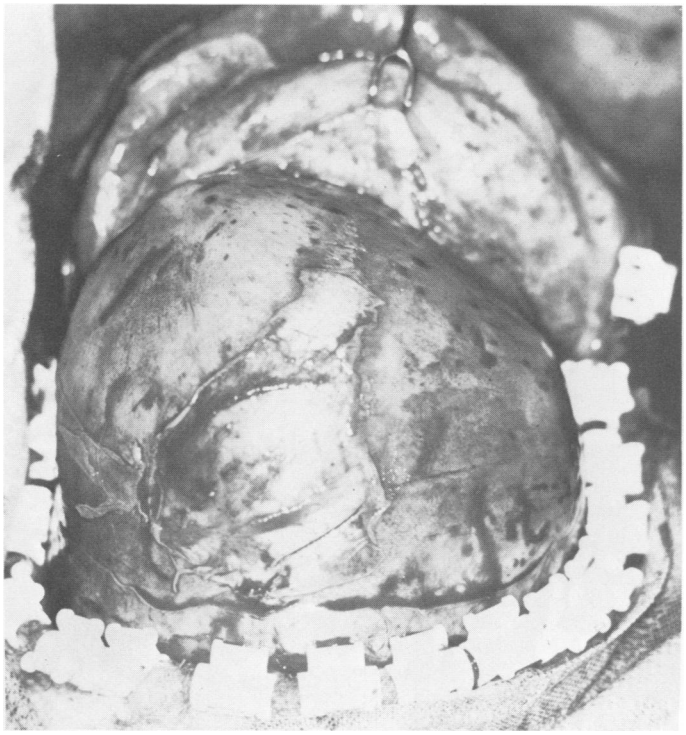

FIG. 14. Case B: Reflection of the scalp and frontal flap shows the appearance from the region of the vertex, looking inferiorly toward the frontal bulge on the left side.

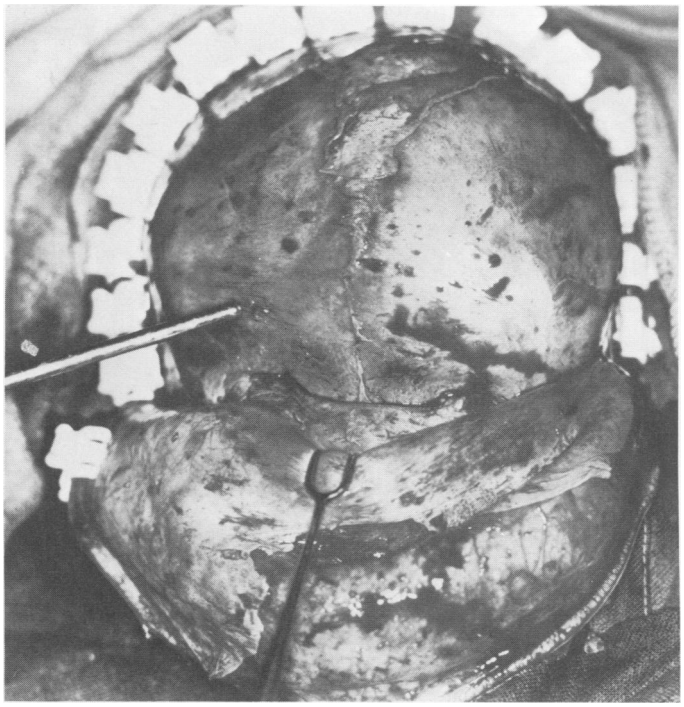

FIG. 15. Case B: A view from anteriorly shows the marked deficiency in the right supraorbital ridge region as indicated by the tip of the suction tube.

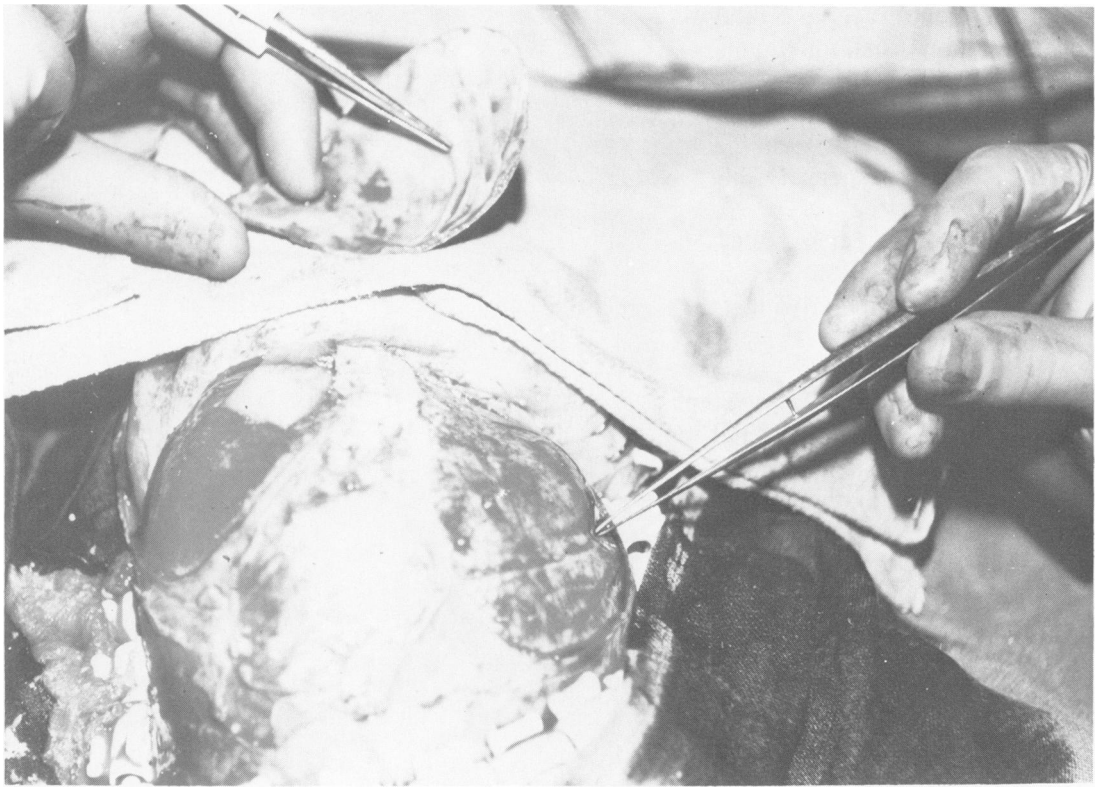

FIG. 16. Case B: The abnormal bulging left frontal cranial bone has been removed as a free graft, and the fused coronal suture is seen on its undersurface. The forceps indicate where the dura will require release and patch grafting in the flattened right frontal region.

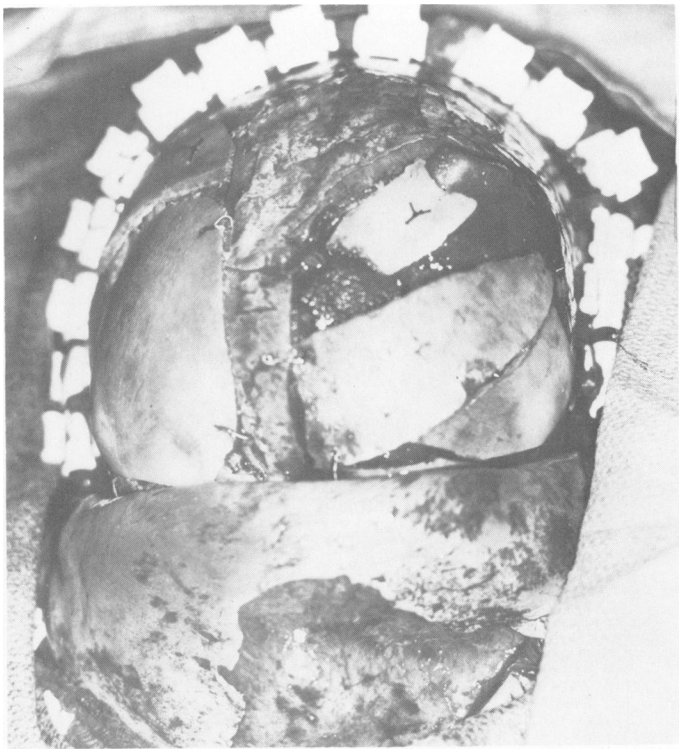

FIG. 17. Case B: Prior to closure, the left frontal dura has been plicated and covered with three island bone grafts. Periosteum has been removed from the undersurface of the left side of the scalp flap and used as a free graft to release the dural constrictions in the right frontal region. This periosteal graft has also been covered with bone grafts, anchored to the strip of midline bone.

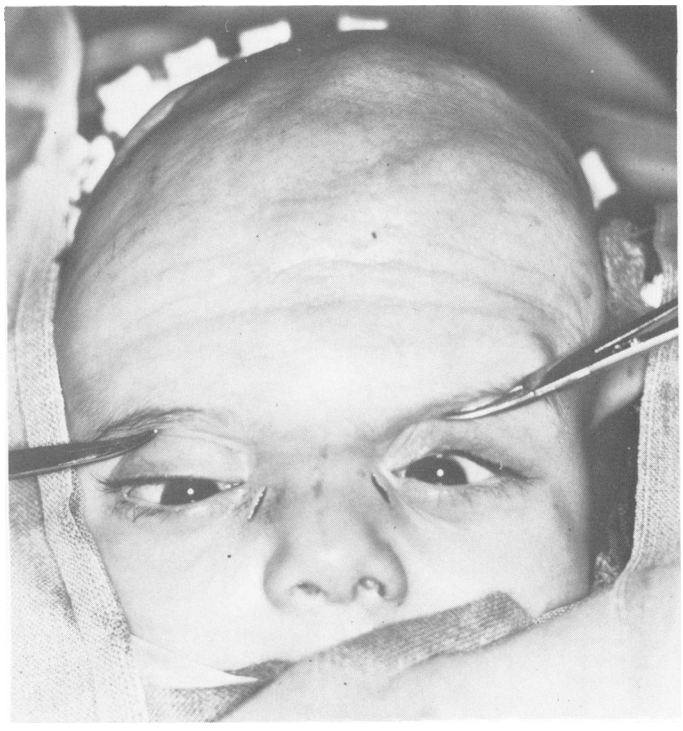

FIG. 18. Case B: Returning the scalp and forehead flap allows a check on the position of the eye level prior to closure. Small incisions were made on either side of the nose in order to straighten the nasal bony pyramid.

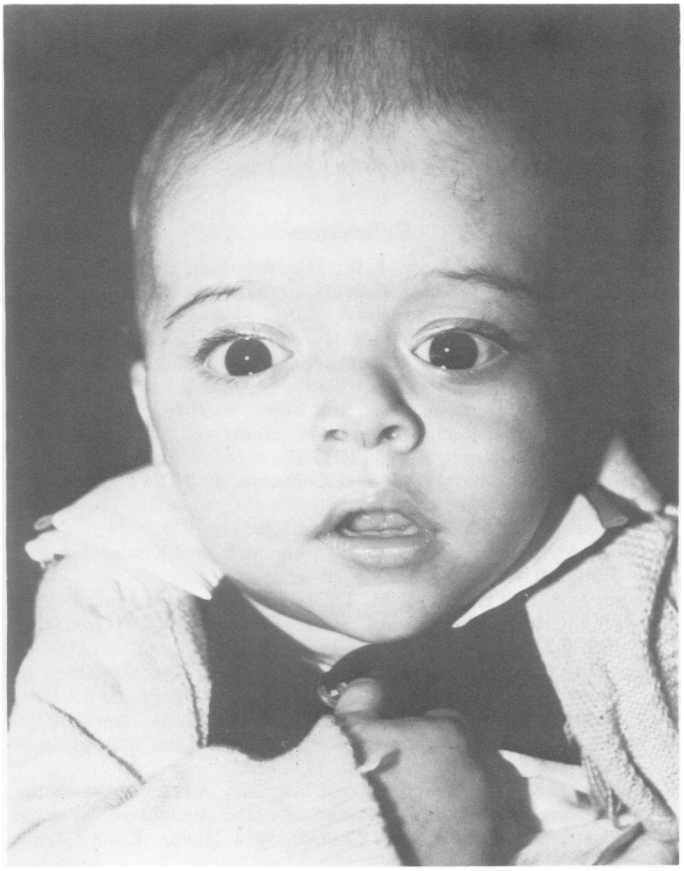

FIG. 19. Case B: This boy's appearance twelve months after surgery shows marked improvement in symmetry of the skull, eyebrows, orbits and nose. This view may be compared with the preoperative condition shown in FIG. 11.

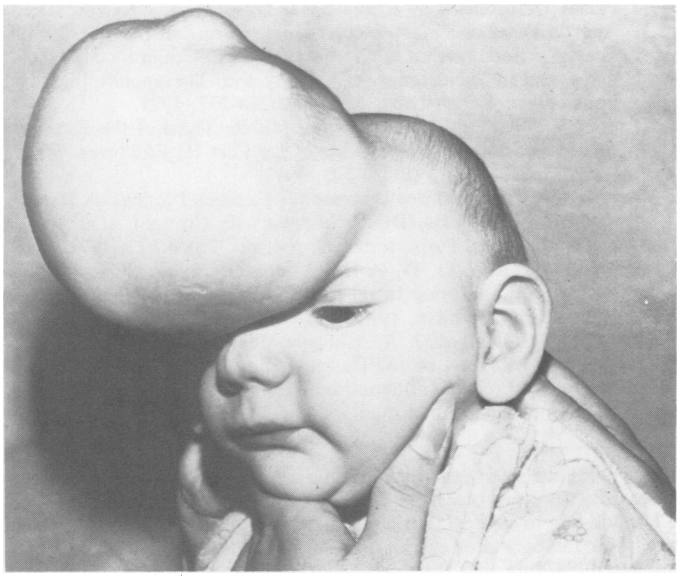

FIG. 20. Case C: This eight month old baby had a severe dysplasia with absence of the frontal bones and at eight months of age, the mass was still enlarging. Pneumoencephalograms revealed brain and ventricles in the herniated mass. Orbital hypertelorism and nasal deformity was present and microcephaly is evident in the posterior cranial region.

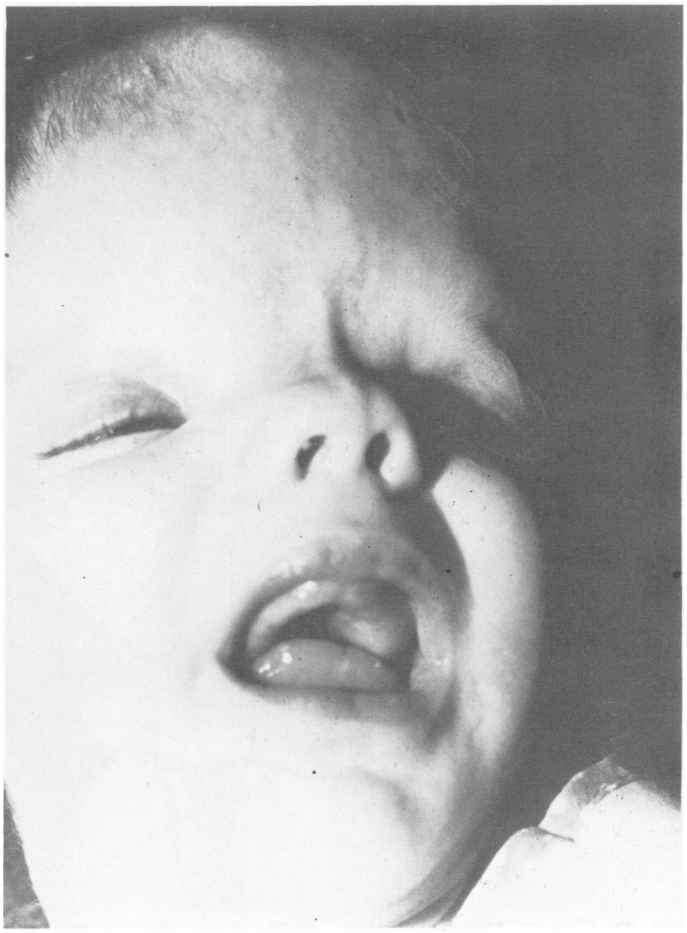

FIG. 21. Case C: After a single stage correction, the forehead has been remodeled using a bone graft from the occipital region and pedicle flaps of dura and fascia cover the brain after its return to a more acceptable position. Note the small median raphe at the nasal bridge.

a series of papers. The new insights resulting from this work have already altered our actual management of many non-craniofacial patients. Several examples will illustrate the "spin-off" value to other problems.

Use of the "pressure screw" has become almost routine in our emergency room for the diagnosis of increased intracranial pressure in unconscious patients with suspected head injury.

Surgical exposure of tumors in patients with malignancies of the orbit or paranasal sinuses is now obtained by the combined intra-extra cranial technique. This improves the accuracy of dissection and reduces the danger of unexpected dural injury.

As a result of the findings with orbit repositioning in craniofacial patients, the opthalmology profession is having to take an entirely new look at the theories about the causes and treatment of ordinary strabismus.

The significance and treatment of blow-out fractures of the orbit take on a different perspective in view of the massive defects of the orbital floor, deliberately created

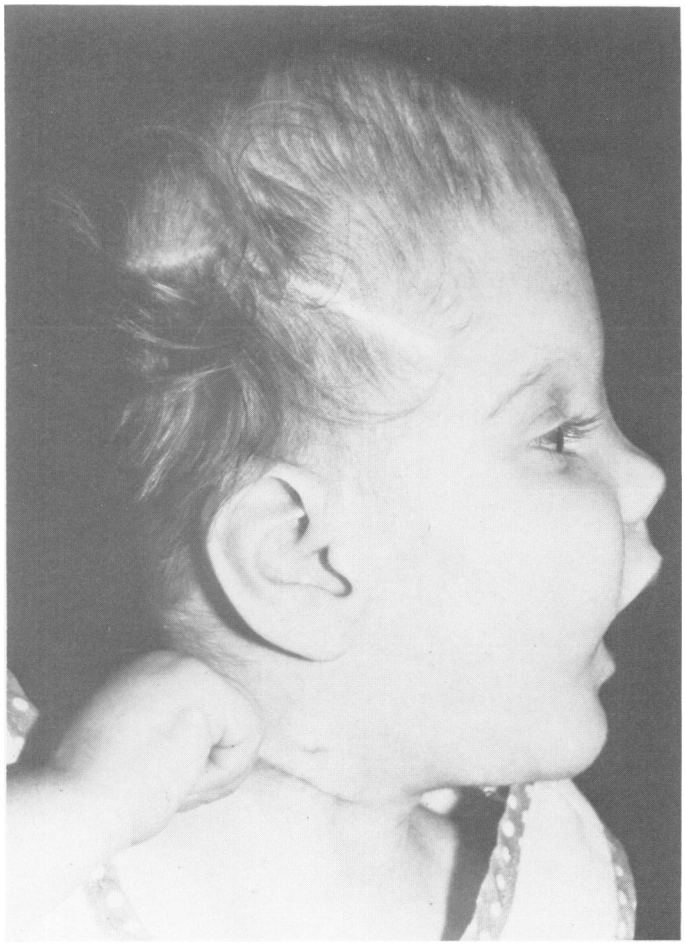

FIG. 22. Case C: The postoperative profile demonstrates a 70% enlargement of the cranial cavity. Bone grafts and hair-bearing flaps were used to reconstruct the occipital region. The child continues to gain weight and may need a plastic procedure to the nasal bridge at a later date. The use of hypotensive anesthesia makes it possible for such small children to stand long (14 hours in this case) operations.

by orbital rim advancements, used to correct the exophthalmus of Crouzon's deformity.

We have had one child with maxillary hypoplasia, who experienced unexpected improvement in hearing after mid-face advancement. Two others have developed temporary hypernasality of speech. These observations are leading to better understanding of the causes of conductive deafness and the dynamics of cleft palate speech. Thus, a number of new techniques and concepts have emerged that will benefit surgery "in general" as well as the deformed child "in particular."

Deformity is itself a serious disease, but meaningful rehabilitation for these children has had to wait until new skills in anesthesiology, plastic surgery, and neurosurgery made possible and safe the methodical and extensive dismantling and reassembly of virtually all of the bones and soft tissues of the face and cranium.

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DISCUSSION

(Note: Some of the discussants' remarks refer to both this paper and to the following one by Dr. Joseph E. Murray. Such remarks appear following both articles.)

DR. PAUL W. GREELEY (La Jolla, California): These two papers are very difficult to discuss and may I say that I congratulate the energy that both Dr. Edgerton and Dr. Murray had in going ahead and attacking the problems of this type.

One thing we must remember is that all children grow and many of these things are performed in children of a very young age group.

I would like to suggest that Dr. Edgerton and Dr. Murray come back here 10 or 15 years later and show us what has happened to these same individual children that they've shown tonight.

DR. BRADFORD CANNON (Boston): I should like to speak for two deceased members of this Association who would have greatly enjoyed Dr. Edgerton's and Dr. Murray's reports.

Dr. Barrett Brown who introduced each of the authors to the chal-