Surgical Management of the Anophthalmic Orbit, Part 1: Congenital

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Congenital microphthalmos and anophthalmos are rare conditions in which orbital growth is deficient. Hypoplasia of the globe affects the bony orbit (micro-orbitism), the conjunctival sac, and eyelids (microblepharism), and it may be associated with abnormalities of the entire hemifacial skeleton (hemifacial microsomia). In the present article, the authors review a series of 19 patients with microphthalmos (nine had right-sided, one had bilateral, and nine had left-sided microphthalmos) who were treated in the Orbitopalpebral Unit at Hospital Foch over a period of 15 years (follow-up, 5 months to 18 years).

Orbital expansion was achieved using spherical implants ($n = 13$), orbital osteotomies ($n = 4$), and orbital expanders ($n = 2$). Both expanders were removed within 6 months because of failure (one infection and one rupture). The current preferred method for orbital expansion is to use serial implants in the growing orbit and osteotomies in cases of late referral or insufficient orbital volume in the older child. The target proportions of the reconstructed orbit are not planned to mirror the healthy side exactly. The inferior orbital rim is kept higher to support the orbital implant, and the orbit is kept shallow to avoid a sunken appearance.

Cranial bone grafts were used to augment deficient orbital contours; they were assisted by anterior transposition of the temporalis muscle ($n = 5$) when additional orbital volume was required. Conjunctival sac reconstruction was achieved by the use of serial conformers placed in the conjunctival sac during the neonatal period, followed by grafts of buccal mucosa and full-thickness skin maintained in place with a tarsorrhaphy for 3 to 6 months. Eyelid reconstruction using local flaps and skin grafts proved to be necessary in cases treated by osteotomy expansion, although reconstruction was not required after expansion using serial solid shapes. The results illustrate an evolution in approach and concepts of reconstruction of the microphthalmic orbit and emphasize the need for an integrated craniofacial approach for this complex deformity. (*Plast. Reconstr. Surg.* 108: 817, 2001.)

Clinically, the phrase "congenitally anophthalmic orbit" refers to any orbit that contains an absent or severely hypoplastic eye at birth. The diagnosis is most frequently microphthalmos, in which arrested growth of the eye during the first few weeks of gestation leads to a small globe and a hypoplastic orbit. It occurs with varying degrees of severity, with a minority of cases retaining useful visual acuity. True anophthalmos describes a complete absence of the globe through failure of formation of the optic vesicle, and it is an extremely rare condition. The overall prevalence of congenital anophthalmos and microphthalmos has been estimated at 1 to 1.5 per 10,000 births,[1,2] with a normal sex distribution. Microphthalmos is unilateral in three-quarters of cases and, although extrinsic causes such as maternal rubella or environmental teratogens are often suspected, no consistent hereditary basis has been found.[3] Systemic abnormalities may coexist with the congenitally anophthalmic orbit in more than 30 syndromic conditions,[4] yet anophthalmos presents most often as an isolated finding or within the spectrum of otomandibular dysostosis. The globe plays a role of essential importance in orbital growth,[5,6] and its diminished contribution in the congenitally anophthalmic orbit is the cause of microorbitism (Fig. 1), conjunctival sac atresia, and short, phimotic lids in the majority of cases (Fig. 2).[4,7]

In severe microphthalmos, the eye is grossly hypoplastic and blind. It is eviscerated whenever possible (removal of its contents, preserving the sclera with its extraocular muscle attachments and Tenon's capsule) or enucleated (removal of the globe, preserving Tenon's capsule) and replaced by a spherical implant (Fig. 3). The implant serves as a support for the

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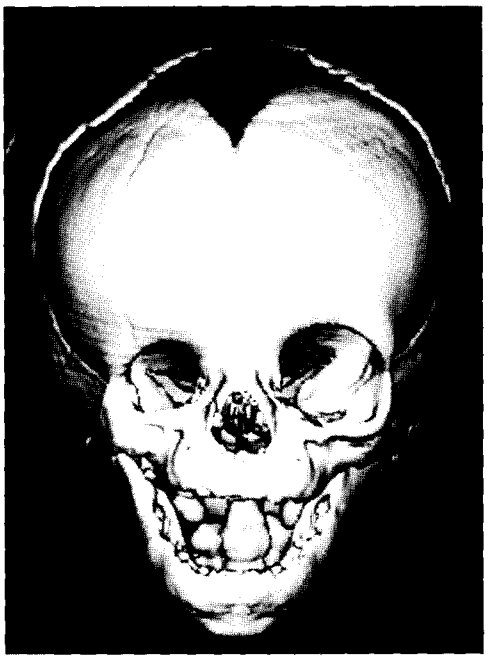

FIG. 1. Three-dimensional computed tomography reconstruction of 1-year-old female patient with severe right microphthalmos. Characteristic features on the affected side include a blunted superior orbital rim, a short lateral orbital rim, a malar bone higher than that on the opposite side, and mild facial scoliosis.

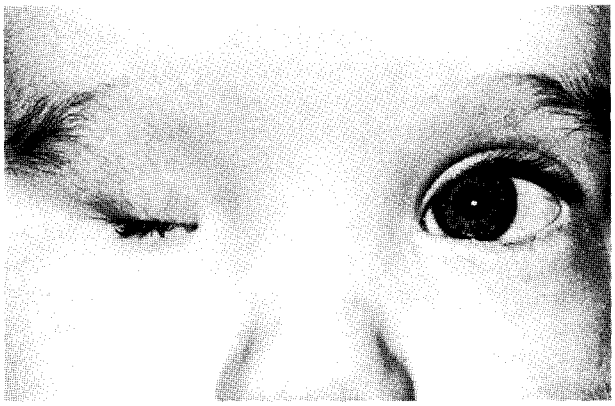

FIG. 2. This 2-year-old male patient had severe right macrophthalmos, with the characteristic features of congenital microblepharism: short and phimotic lids, a narrow palpebral fissure, telecanthus, and absent eyelashes.

eyelids and conjunctival sac anteriorly and maintains the volume of the orbit. In a second stage, a removable ocular prosthesis that mimics the eye is fitted to the contours of the conjunctival sac (Fig. 4).

In moderate microphthalmos, the eye is present but smaller than normal. In certain rare cases it may be partially sighted, but a prosthetic reconstruction is often warranted to motivate the growth of the orbital framework.

We are indebted to Paul Tessier and the foundations he laid in craniofacial surgery fo an improved understanding of orbital tissu relationships in microphthalmos. Neverthele achieving an acceptable, lasting degree of fun tional symmetry in these patients is a challen ing endeavor from which we are continual learning. This report outlines the evolution i concepts and techniques developed on th unit over the past 15 years for reconstructin the congenitally anophthalmic orbit. A stage sequential approach has been developed th considers the "container" (the bony orbit) se arately from its "contents" (the soft-tissue co stituents of orbital volume).

PATIENTS AND METHODS

Patients

Over a 15-year period in the Cranio-Orbit Palpebral Unit at Hospital Foch, 19 patien (nine male and 10 female) were operated o by the senior author (D.K.). Five cases we seen after treatment had already been initiate by the referring center (mean referral age, years), and four cases were initially managed Dr. Tessier. Eighteen patients presented wi unilateral microphthalmos (nine right-side and nine left-sided), and one patient had bila eral microphthalmos. Two unilateral cas were classified as severe according to the crit ria above. In 10 of the patients, the microp thalmic orbit was an isolated finding. The r mainder consisted of four cases of Goldenh variant, three cases of simple hemifacial micr somia, one case of bilateral number 4 Tessi facial clefts, and one forme fruste of a numb 4 Tessier facial cleft. No family history of t condition was detected in this series. In or case, maternal pregnancy was complicated by toxoplasmosis infection.

Microphthalmos, micro-orbitism, and m croblepharism were present in all cases, ar two-thirds of the patients had other signs hypoplasia in the head and neck. Systemic a normalities were present in four patients in t form of esophageal atresia and cardiac ar vertebral anomalies.

Methods

The surgical protocol that has been dev oped in our unit for the treatment of the patients takes the form of a staged reconstru tion that relies on fundamental concepts cranio-orbitopalpebral surgery. Two goals a considered: (1) creating an aesthetic orbit ar

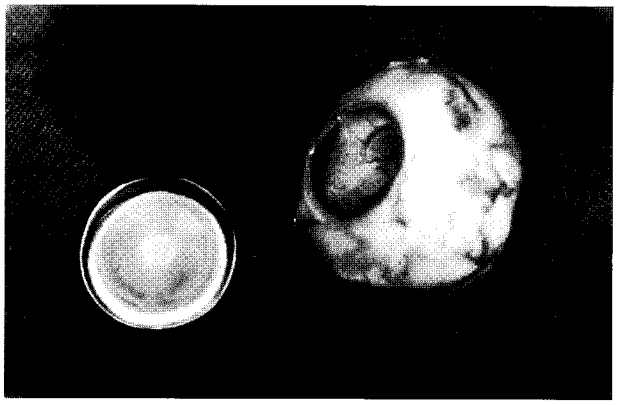

FIG. 3. Materials used in composite reconstruction of the congenitally anophthalmic orbit include a solid silicone implant, which is placed within a cadaveric scleral graft (*right*). Alternative autologous tissues, such as fascia lata or pericranium, may also be used to wrap the implant and reduce its likelihood of expulsion from the orbit.

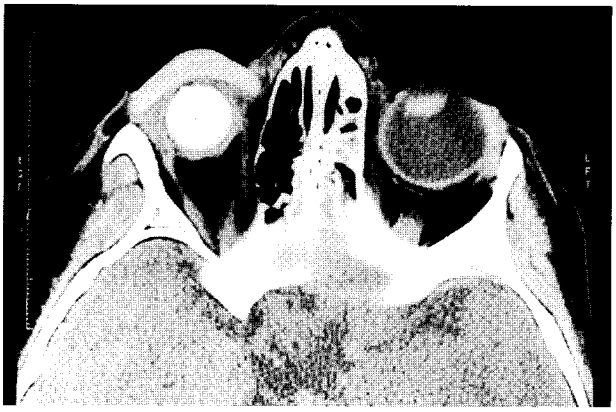

FIG. 4. Computed tomography scan of a completed reconstruction in a patient with right-sided microphthalmos. After expansion of the conjunctival sac and orbital frame using serial solid shapes, a silicone implant is placed within Tenon's capsule. The centric position of the implant in the orbit maximizes the efficiency of the extraocular muscles and ensures a symmetrical projection of the reconstruction with the opposite, seeing eye. After matching the contours of the conjunctival sac, a removable ocular prosthesis is then fitted that mimics the appearance of a healthy eye. The scan also illustrates the balance in volume distribution between the elements forming the reconstruction, which must be carefully adjusted to optimize symmetry and minimize the weight of the ocular prosthesis.

(2) restoring harmony to the face. To achieve these goals, each of the following components of the reconstruction must be addressed individually: orbital volume, orbital contour, the implant and its placement, the conjunctival sac, prosthetics, and the eyelids (both in terms of aesthetics and function).

Orbital volume. Managing the volume of an anophthalmic orbit is a key element in its reconstruction. A dynamic balance exists between the bony orbital volume and the soft tissues that occupy it. The growth of these tissues, in particular the globe, provides an essential impetus for bony growth of the orbit.[5] Little is known about the regulation of orbital growth, but it is likely that humoral and neurotrophic influences are modulated by the forces of the soft-tissue matrix,[8] which are abnormal in the anophthalmic orbit. The composite anatomy of the bony orbit may, in part, explain why some of these patients are born with a facial scoliosis that requires frontal remodeling and mandibular and maxillary surgery or distraction to be incorporated into the treatment plan.

Paradoxically, the ideal dimensions required for the reconstruction of a congenital anophthalmic orbit do not match those of the healthy side. Rather, the depth of the orbit should be targeted to be slightly shallower, which narrows the error in achieving a balance between size of the implant and protrusion of the prosthesis. A deep socket buries an implant, resulting in an enophthalmic appearance on the reconstructed side. Increasing the implant diameter to counter this phenomenon restricts any mobility of the prosthesis, and attempts to compensate with a thicker prosthesis overload and eventually fatigue the lower eyelid.

The frame of the orbit, however, should mirror the healthy side, such that the brows and medial and lateral canthi lie symmetrically opposed. The only exception is the inferior orbital rim, which benefits from being fashioned more superiorly to provide a shelf of bony support for the implant. Failure to uphold the implant with the rim or a bone graft placed on the orbital floor burdens the palpebral sling excessively, which in time may cause an inferior displacement of the prosthesis. The same argument applies to the volume correction of an excessively large orbit (as might follow an overzealous tissue expansion or osteotomy), which we feel is best achieved with autologous tissue (temporalis flap or bone grafts) rather than by increasing the volume of alloplastic materials in the orbit.

Spherical implants of fixed diameter are a time-tested, effective method for stimulating bony orbital growth, and they remain our preferred method for primary expansion of the orbit. Although they may be criticized on the grounds of providing an unphysiological, static drive for orbital growth, very satisfactory results can be achieved when expansion is started early (within weeks of birth) and with frequent

upsizing of the implant (every 6 months or less). The implant is wrapped in an autograft of fascia lata, temporalis fascia, or pericranium that is closed with a pursestring suture and placed within, or immediately posterior to, Tenon's capsule.[9] Placement of the implant anywhere other than centric within the extraocular muscle cone reduces the mobility of the implant and induces a radial asymmetry in the orbit that makes it unstable. The conjunctival layer is closed without tension over the implant, which itself is chosen to be as large as possible (16 to 20 mm in diameter) when placed during the period of orbital growth. This maximizes its expansile force within the orbit and on the soft tissues of the conjunctival sac and palpebral fissure anteriorly. Numerous implant materials and types are available, but in the growing anophthalmic patient, silicone or methylmethacrylate are preferred in our unit because of their smooth contour and low density. Hydroxyapatite has enjoyed a recent popularity in the adult anophthalmic orbit because of its tissue integration properties.[10] These properties may account for its low rate of implant extrusion, but they make it a poor choice in the young patient in whom frequent implant changes are required.

Autogenous bone grafts serve a versatile function in the congenital anophthalmic orbit, and they are used to correct volume defects (intraorbital placement), to support the implant (orbital floor placement), or to bolster the stock of deficient orbital rims (lateral, superior, or inferior orbital rim or lateral wall placement). We prefer to use cranial bone because it closely resembles the recipient site embryologically and morphologically and carries low postoperative resorption and morbidity rates. Harvest is parietooccipital through the bicoronal incision used to access the orbits. Burring achieves a reduction and softening of orbital contours, which is used to "open" a constricted micro-orbit using the concepts of orbital remodeling previously reported in the "mask lift."[11] In the case of a late referral in which growth has ceased, expansion of the orbital frame requires a patterned osteotomy.[12] Care must be taken not to produce an excessively voluminous orbit using this technique, for the reasons outlined previously.

Orbital contents. Aside from implants, we avoid the use of foreign materials in the orbit altogether, because there is neither a substitute for nor a shortage of autologous tissue in the vast majority of cases. Implant rejection can oc cur and, if repeated, justifies the use of a de epithelialized dermafat graft harvested fron the buttock or inguinal fold. Using this tech nique, one must anticipate a degree of graf shrinkage from atrophy of the fat, which in turn may result in a decreased projection of th prosthesis.

Supplementing the volume of a micro-orbi is primarily achieved with transposition of th temporalis muscle and the use of cranial bon grafts. The anterior two-fifths of the temporali muscle are used to line the orbit through window osteotomy made in the lateral orbita wall.[13] In the severely hypoplastic orbit, thi supple, highly vascularized tissue provides lining for the fashioning of a conjunctival sa and adds volume to the contents. The poste rior three-fifths are then transposed anteriorl to prevent an unattractive hollow in the tem poral region.

Conjunctival sac. The conjunctival sac play a crucial role in the maintenance and mobilit of the ocular prosthesis,[9] yet it is perhaps th most vulnerable structure in the orbit: contrac tures within the fornices shrink the cavity an threaten to expel the prosthesis unless the prob lem is corrected with grafts. Early expansion c the conjunctival sac to dilate the existing vo ume and to expand the orbital rim using seria conformers should be started shortly afte birth.[14,15] The atretic sac in the neonate is ofte too small to retain a conformer, and hydro philic expanders may be used to create space.[16] Despite serial static expansions, graft are often required to create fornices of suff cient depth, and a choice exists between bucca mucosa and skin. The former is preferable i the long term, because it moisturises the pro thesis and keeps it clean. However, buccal mu cosa is a finite source, and its harvest carries th risk of contractures that may be troublesome. The morbidity of these can be kept to a min mum with frequent massages and the chewin of gum during the period of scar maturation Skin grafts carry low donor-site morbidity, bu are less popular with our patients because of th frequent maintenance they require to clear th sac of malodorous secretions.

In refashioning the sac, the pretarsal con junctiva is usually preserved and the recipien site for the grafts is dissected to the margins c the orbital rim. Bolster sutures through th eyelid skin maintain the angles of each fornix and a ring conformer acts as a splint that i

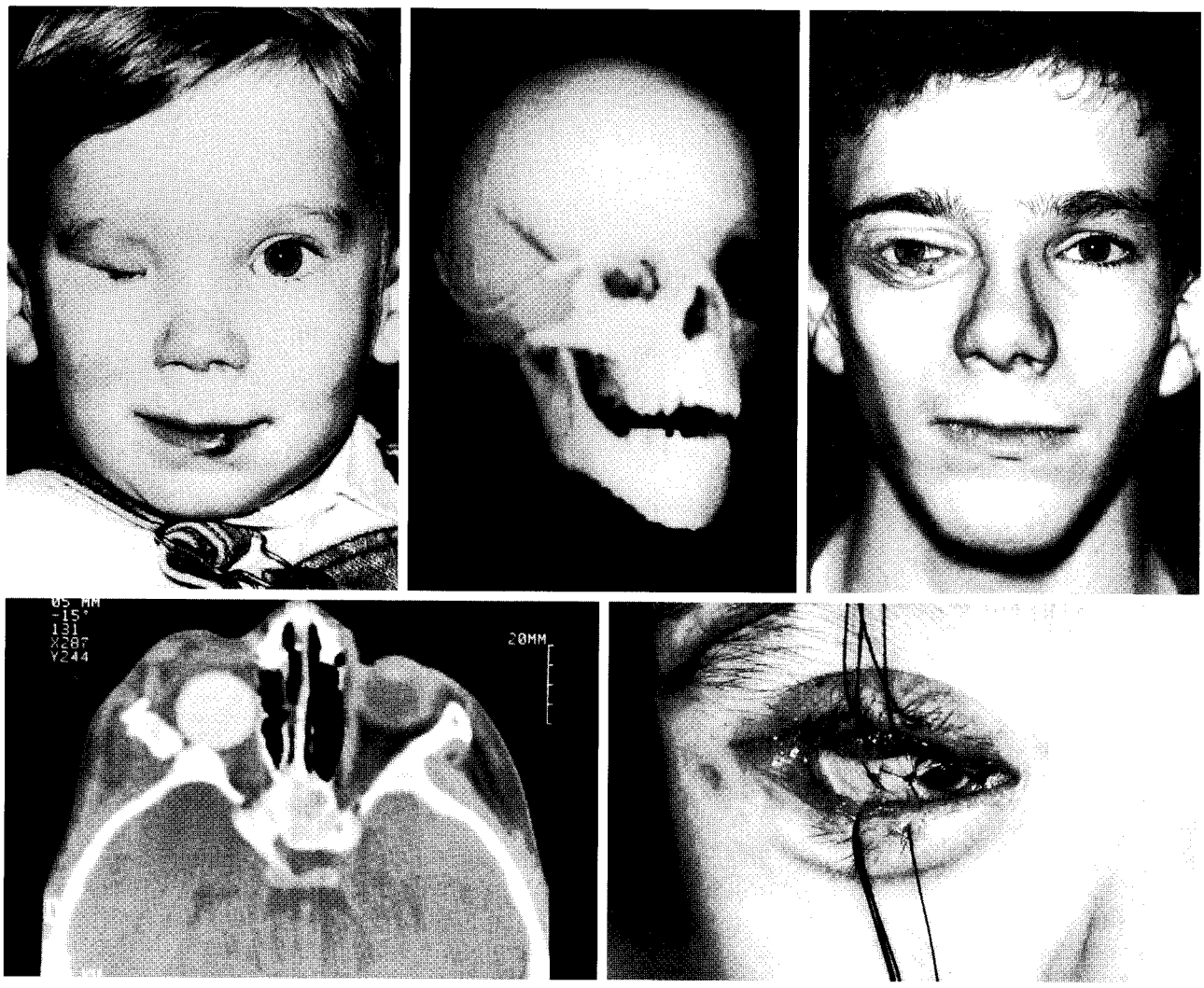

FIG. 5. Examples of an initial attempted orbital expansion using a sperical tissue expander. In this case, the reconstruction was set back after rupture of the expander, and a further orbital expansion by osteotomy was required. (*Above, left*) Male patient aged 2 years, with right microphthalmos, before evisceration and placement of a sperical silicone expander in the orbit. (*Above, center*) Three-dimensional computed tomography reconstruction showing the expander in place (patient aged 3 years). (*Above, right*) The patient at 16 years, after having also undergone a medial and lateral canthopexy, lower eyelid reconstruction with an upper lid flap, upper lid reconstruction with a full-thickness skin graft, correction of ptosis, and fitting of an ocular prosthesis. (*Below, left*) Rupture of the expander occurred within 6 months of its placement. A severe contraction within the orbit ensued, which necessitated orbital osteotomies, transposition of the temporalis muscle, and the placement of a silicone implant within the orbit (shown here on an axial computed tomography scan taken when the patient was 5 years old). (*Below, right*) Reconstruction of the conjunctival sac using buccal mucosal graft sutured over a conformer (patient aged 6 years).

kept in place by a two-layer median tarsorrhaphy kept in place for 3 to 6 months. There may be insufficient orbicularis muscle to support a graft destined to enlarge the sac, which is an indication for a transposition of the temporalis muscle before lining the sac with mucosa or skin.[18]

Eyelids. Congenitally short and stiff eyelids, a narrow palpebral fissure, canthal dystopia, and eyelash deficiencies are encountered across a spectrum of severity in microphthalmos. Unlike the bony orbit, which has reached most of its adult proportions by the age of 8 years, the palpebral fissures undergo a gradual increase in size until puberty. Good results may be obtained with serial conformers that are started soon after birth and changed daily and then weekly in increasing sizes until temporary prosthetics can be fitted at around the age of 2 years. Surgical maneuvers to widen the palpebral fissure should be discouraged unless absolutely necessary, because they scar the lids and, in our opinion, destroy the aesthetics of the lateral canthus forever.

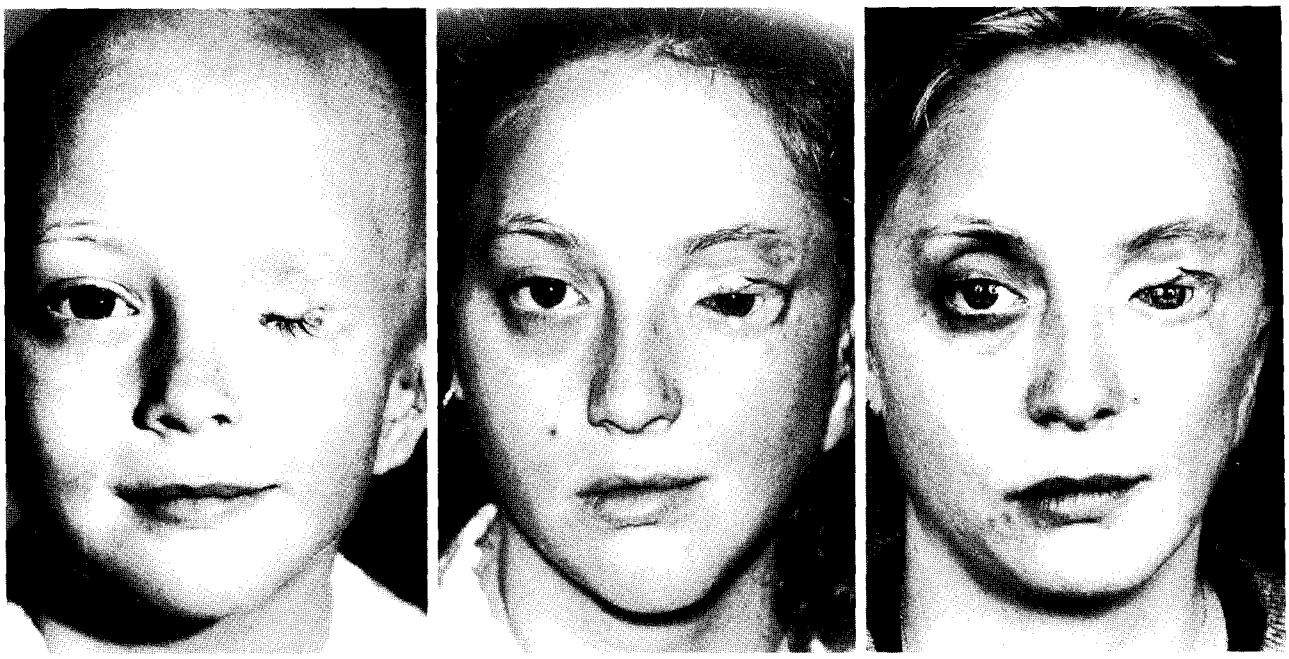

FIG. 6. Example of a case in which primary osteotomies were used to expand the orbit (Tessier pattern). (*Left*) A 5-year-old female patient with left-sided hemifacial microsomia that manifested as left microphthalmos, maxillary hypoplasia, and microtia. Microblepharism is also evident. (*Center*) The patient at 12 years. Orbital frame osteotomies were performed at the age of 5 years; this was accompanied by a complete orbital reconstruction including evisceration, implant placement, and cranial bone grafts. Eyelid reconstruction was achieved with an upper eyelid flap and full-thickness skin grafts, medial and lateral canthopexies, eyelash grafts, and an ocular prosthesis. (*Right*) The patient at 16 years. She is now more concerned with her appearance and has agreed to undergo bimaxillary distraction and address her residual facial asymmetry.

The function of the lower lid in the anophthalmic socket is primarily one of support, and an upper lid myocutaneous flap (unipedicled or bipedicled) is our preferred choice for reinforcing it when it is deficient. The upper lid donor unit is resurfaced with a full-thickness skin graft, usually taken from behind the ear, which resurfaces the orbital unit of the lid and blends it into the brow along a serrated line. During transposition of the upper lid flap, the anatomy of the levator complex should be explored. Although hypoplastic, the levator serves an important function in creating a supratarsal fold and preventing entropion.[15] Deficient levator function is treated with a frontal suspension if necessary, whereas lid retraction is corrected with the interposition of autologous fascia grafts (fascia lata or temporalis fascia) in a ratio that approximates 2:1 to the desired distance of lid descent.

Facial harmony. The final step in rehabilitating the microphthalmic patient consists of restoring harmony between the affected orbit and the rest of the face. The subperiosteal mask lift provides the access required to recontour the upper and middle third of the craniofacial skeleton and to shape the palpebral fissure and correct brow malposition. Rhinoplasty is preferably delayed until adolescence, whereas mandibular distraction and maxillary surgery are coordinated with orthodontics during childhood. Microtia reconstruction, facial reanimation techniques, and lipofilling all contribute to optimizing symmetry with the unaffected side.

RESULTS

Mean follow-up was 10.1 years (range, 5 months to 18.1 years). Orbital expansion was performed with the use of implants and solid shapes in 13 patients in the series. Two patients underwent orbital expansion using a spherical tissue expander in the early part of this study. In both cases, the expander had to be removed within 6 months, in one case for infection and in the other because of rupture. Orbital expansion by osteotomy was performed in four other patients, in two before referral and in two because the period of orbital growth had passed.

Four patients received an upper lid myocutaneous flap (heteropalpebral flap) to reconstruct the lower eyelid, five required transposition of the temporalis muscle into the orbit, and one required correction of eyelid ptosis.

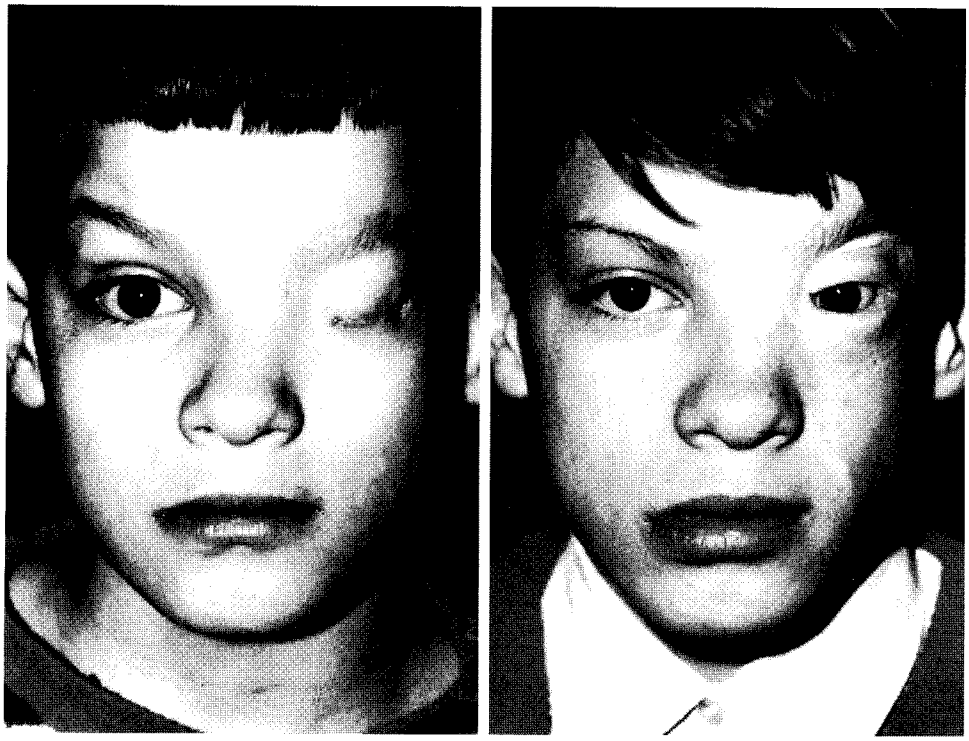

FIG. 7. Example of another patient who underwent bony orbital remodeling with expansion osteotomies for left-sided microphthalmos. As shown in Figure 6, results after osteotomies are most often moderate, and a total reconstruction of the eyelids is always required. This case required the use of a nasolabial flap, a bipedicled upper musculocutaneous lid flap, and full-thickness skin grafts.

Other procedures performed included the mask lift (which included frontal remodeling, lateral canthopexy, and onlay malar cranial bone grafts), transnasal medial canthopexy, mandibular distraction, iliac and rib mandibular grafts, rhinoplasty, nasal reconstruction with a forehead flap, frontal suspension using fascia lata, lower lid reconstruction with a nasolabial flap, open brow lift, cartilage grafts to the lower eyelid, eyelash grafts, and excision of a coloboma.

Complications included the extrusion of two silicone expanders, which were replaced with solid implants once inflammation had subsided. One patient extruded a silicone orbital implant that was judged to have been too large 1 month postoperatively. An orbital osteotomy performed in a 3-year-old patient failed; a revision of orbital remodeling was required within 1 year. Cicatricial retraction of the conjunctival sac occurred at some time during treatment in more than half of the patients; this required the repeated use of buccal mucosa or skin grafts to deepen the deficient fornices. Figures 5 through 9 show representative results.

DISCUSSION

The orbital level of the face is a challenging anatomical area to reconstruct, not least because of the scrutiny it bears from others. As a result, attempts to surgically restore a microphthalmic orbit to any pleasing degree of symmetry have often been frustrating, and the patient may find it preferable to wear an eye patch. In our experience, however, the results seen in patients who have begun treatment as infants more than justify the rationale for a composite reconstruction. None of the patients in this series conceal their surgical result when in public, although it must be noted that they receive emphatic and continued psychological support as a routine part of their management.

Mimicking the growth impetus of normal intraorbital contents remains a technological challenge. Spherical tissue expanders[19–21] have been applied to the problem, but in our opinion they are too unreliable to justify their routine use. Experimental studies suggest that a direct relationship exists between intraorbital pressure and orbital volume,[6,22] yet control over the direction and degree of pressure ex-

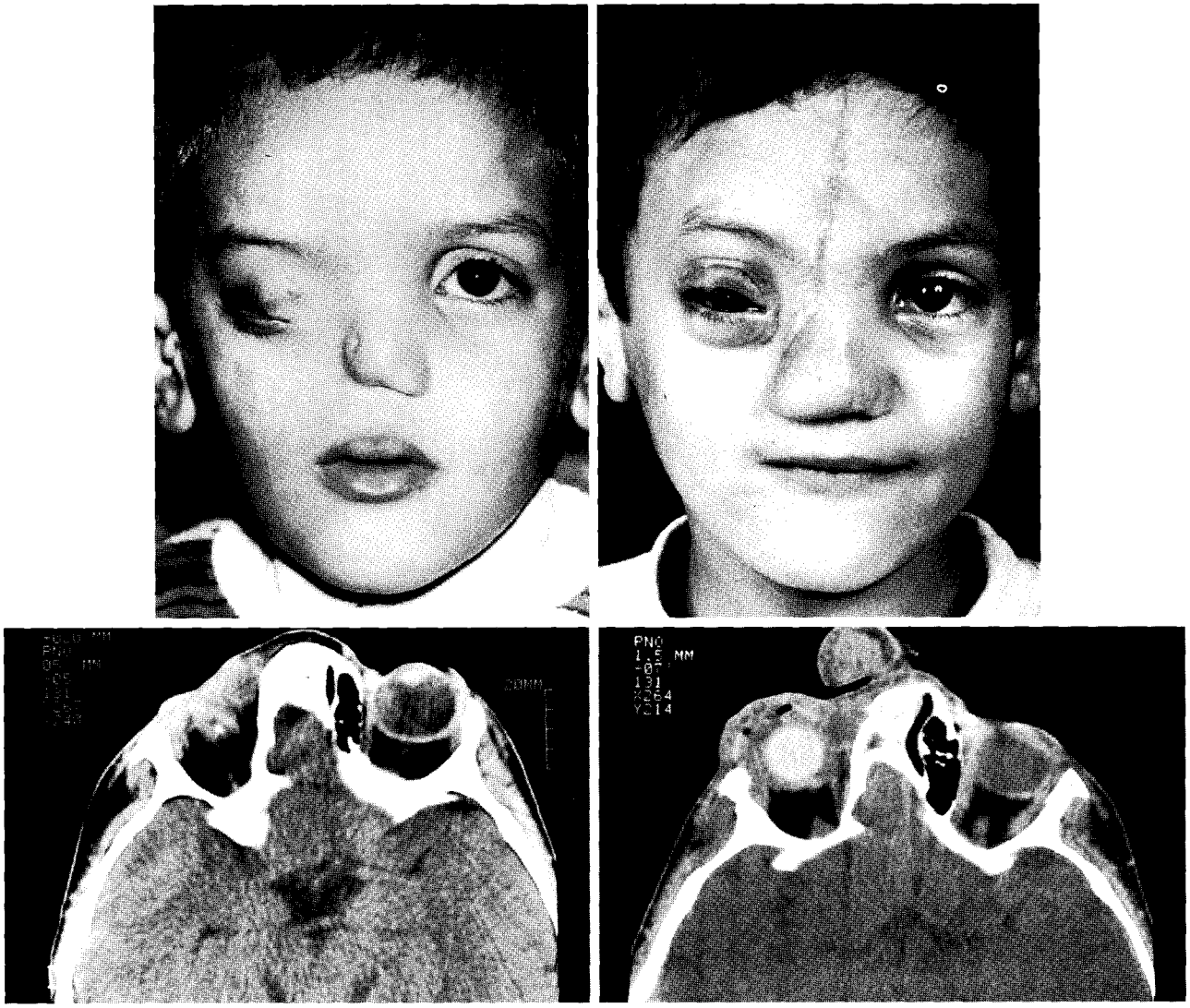

FIG. 8. Example of expansion achieved using orbital implants alone in a patient referred late. (*Above, left*) A 4-year-old male child with right microphthalmos in the context of a hemifacial malformation that included a right frontal encephalocele and right heminasal agenesis with proboscis and atresia of the nasal airway. (*Above, right*) Patient at 7 years, after evisceration and placement of a silicone implant, bone grafting of the superior orbital rim, conjunctival sac reconstruction, medial and lateral canthopexies, ptosis correction, first stage heminasal reconstruction with a forehead flap, and fitting of an ocular prosthesis. (*Below, left*) Axial computed tomography scan taken when the patient was 4 years old showed a cystic, rudimentary right eye within a hypoplastic orbit. (*Below, right*) Axial computed tomography scan taken when the patient was 5 years old, showing the expansion in right orbital volume achieved after evisceration and placement of a silicone implant.

erted by traditional soft-tissue expanders is imprecise. Although some good results have been obtained,[23,24] overexpansion of the posterior orbit rather than the frame is of concern, and the long implantation time often results in expanders extruding or rupturing before term. We are of the opinion that an expansion failure, such as one complicated by infection or extrusion, induces a state of scarring in the orbit that sets back the reconstructive process beyond its starting point. We hope that modifications to the design of expanders will overcome these shortcomings in the future, but as yet there is a paucity of published long-term results to support their use.

A continuous struggle exists to maintain the volume of the conjunctival sac in microphthalmos reconstructions. Contractures occur insidiously and predictably after an acute infection or episode of periprosthetic inflammation, and much ground previously gained with the placement of grafts can be lost.[25] We have observed the pubertal patient to be precocious in that graft contraction tends to occur more frequently and more severely during this period. In these cases, we prefer to secure the con-

FIG. 9. Example of a case in which orbitopalpebral expansion was achieved using serial solid shapes and orbital implants of increasing size. Over time, we have found this method provides the most reliable results, with little or no need for reconstructive procedures of the eyelids. (*Left*) An 18-month-old female child with right-sided microphthalmos. Expansion of the conjunctival sac was started a few weeks after birth with conformers that simultaneously acted to enlarge the orbital frame. Orbital volume expansion was achieved using a silicone implant wrapped in scleral allograft that was placed during the first year of life. (*Right*) Patient at 5 years, after being fitted with a definitive ocular prosthesis. The width of the palpebral fissure and symmetry of the orbital level of the face are satisfactory and seem to be superior to results achieved with the use of other techniques of orbital expansion.

former that carries the graft with a tarsorrhaphy that is kept in place for 6 months instead of the usual 3 months. Finally, a heavy prosthesis damages the conjunctival sac by stretching the fornices inferiorly, which reduces their mobility and, hence, that of the prosthesis itself.

The subperiosteal dissection of the mask lift provides ideal access for forehead remodeling, restoring malar projection and orbital contours with cranial bone grafts, and placing a durable lateral canthopexy on the affected side. Eyebrow position is adjusted at the time of closure by scoring the periorbita and frontal galea; if necessary, this is done with an asymmetric resection of the anterior scalp. Orthognathic and ancillary plastic surgery procedures are frequently required to harmonize the orbital proportions with the rest of the face; in our opinion, they are best deferred until adolescence when the growth of the face is near complete.

The striking asymmetry of an anophthalmic orbit weighs heavily on a young patient, both physically and psychologically. Rehabilitation demands a fully dedicated, long-term commitment from the multidisciplinary team, who should expect to follow-up these patients well into their adult life. The dynamic relationship between prosthetics and bony and soft tissues in the orbit means that multiple adjustments to the cavity may be required before adequate symmetry can be achieved. Satisfactory results with orbital expansion can be obtained using early and frequent serial solid shapes, but we hope that future technological advances will lend themselves to a more complete and physiological simulation of normal growth in the congenital anophthalmic orbit.

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