Correction of Hypertelorbitism: Evaluation of Relapse on Long-Term Follow-Up

Derrick C. Wan, MD, Ben Levi, MD,† Henry Kawamoto, MD, DDS,* Neil Tanna, MD,* Christina Tabit, BA,* Cassio Raposo do Amaral, MD,‡ and James P. Bradley, MD**

Background: Hypertelorbitism has been associated with a variety of congenital deformities. Appropriate timing for surgical correction remains controversial. We present our long-term experience of 33 patients with hypertelorbitism undergoing facial bipartition or orbital box osteotomy.

Methods: Patients with hypertelorbitism treated with either facial bipartition or orbital box osteotomy and repositioning who had long-term follow-up were studied (n = 33). Age at the time of first surgery, preoperative interdacryon distance, and immediate postoperative interdacryon distance were recorded. Relapse was determined on postoperative follow-up, and the need for secondary correction was noted. Physician satisfaction score (range, 0–4) was also assessed.

Results: Patients had a mean total follow-up of 14.0 years. With regard to age at the time of initial procedure, patients younger than 6 years were all noted to have relapse, and 83% underwent revision surgery. In patients 6 years or older, only 11% had relapse and required a second operation. Yet, satisfaction scores were similar (3.2 versus 3.5). With regard to the severity of hypertelorbitism, there was no relapse noted among patients with mild hypertelorbitism (interorbital distance [IOD], 30–34 mm). Among those with moderate hypertelorbitism (IOD, 35–40 mm), 29.4% developed relapse. By contrast, all patients with severe hypertelorbitism (IOD, >40 mm) were noted to have relapse requiring repeat correction. Satisfaction scores were similar (3.4 versus 3.3 versus 3.1).

Conclusions: Relapse after surgery for hypertelorbitism is related to the age of the patient at correction and the preoperative severity. When possible, surgical repositioning of the orbits should be delayed until later childhood.

Key Words: Hypertelorbitism, surgical timing, facial bipartition, orbital box osteotomy

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From the *Division of Plastic and Reconstructive Surgery, Department of Surgery, University of California, Los Angeles School of Medicine, Los Angeles; †Hagey Laboratory for Pediatric Regenerative Medicine, Plastic and Reconstructive Surgery, Stanford University School of Medicine, Stanford, California; and ‡Institute of Craniofacial Plastic Surgery, Sobrapar, Campinas, Brazil.

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Address correspondence and reprint requests to James P. Bradley, MD, Division of Plastic and Reconstructive Surgery, 200 Medical Plaza, Suite 465, Los Angeles, CA 90095; E-mail: jpbradley4@mac.com

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Hypertelorbitism was first described by Greig in 1924 with his presentation of 2 craniofacial malformations resulting in "great breadth between the eyes."[1,2] In subsequent years, arbitrary use of the term *hypertelorbitism* resulted in significant confusion. By definition, hypertelorbitism refers to an abnormal increase in the bony interorbital distance and must be distinguished from other deformities associated with telecanthus, the latter of which may also give the illusion of hypertelorbitism but warrants an entirely distinct surgical approach.[1] The dacryon, defined as the most medial osseous part of the orbit, has been most frequently used as a landmark to objectively determine orbital distance. Although studies have shown interdacryon distance to vary with age, measurements in excess of approximately 25 mm in the growing child have been described as abnormal.[3–6]

Hypertelorbitism has been observed to be associated with a variety of deformities. Both syndromic and nonsyndromic craniosynostoses may present with increased interorbital distance. Furthermore, some of the most severe cases of hypertelorbitism may be seen in patients who fall within the spectrum of median craniofacial dysplasia. Also referred to as a Tessier no. 0 to no. 14 cleft, internasal dysplasia, median cleft face syndrome, or frontonasal dysplasia, patients with this deformity may demonstrate a duplicated anterior nasal spine and spectrum, broad and flattened nasal bones, enlarged ethmoid and sphenoid sinuses, and a frontal encephalocele all associated with dramatic hypertelorbitism.[6,7]

Although the underlying etiology may vary, from a surgical perspective, hypertelorbitism can be distilled down to an anatomic deviation with excess tissue resulting in abnormal spacing between the orbits. Frequently, the actual size of the orbits fall within the norm, and it is the surfeit of intervening bone and soft tissue that must be removed to restore a proper interdacryon distance.[1] Surgical procedures aimed at correction of hypertelorbitism have undergone significant evolution during the last century, with Tessier laying the foundation for contemporary combined intracranial-extracranial approaches.[8–10] His original description in 1967 led to subsequent reports on rates of early postoperative complications and long-term persistence of stigmata.[11] McCarthy,[12] however, provided one of the earliest studies on the durability of correction for hypertelorbitism, showing that surgery could be performed safely at 7 years or younger and that the orbits could be repositioned with stability. This was subsequently confirmed in another study consisting of patients averaging 3.9 years at the time of surgery.[13] In addition, there may be psychosocial benefits for operating at a young age.

Despite such reports, appropriate patient age for correction of hypertelorbitism remains controversial. Mulliken et al[2] found that in younger patients, recurrence of increased interorbital distance was noted. Furthermore, midface surgical procedures at an early age interfere with normal anterior facial growth. To address some of the debate over timing of surgery and postoperative relapse, we therefore evaluated our own experience with correction of hypertelorbitism. We present 33 patients undergoing facial bipartition or orbital box osteotomies for hypertelorbitism and report on their degree of relapse seen after a mean follow-up of 14.0 years.

PATIENTS AND METHODS

Patients presenting with hypertelorbitism at UCLA Medical Center between the years 1975 to 2009 were identified and their medical charts were obtained. Level 1 review by the institutional review board committee was performed for exempt status. Patients undergoing surgical correction with either facial bipartition or orbital box osteotomy had a minimum follow-up of 5 years and had complete records were included in the study. We also excluded patients with asymmetric or nonmidline rare craniofacial clefts and hypertelorbitism in this study. Patients' demographics, namely age at the time of surgery, diagnosis, and preoperative interdacryon distance, were noted. Measurements were made based on computed tomographic scans and intraoperatively with calipers. For analysis, patients were grouped according to age (<6 or ≥6 y) and preoperative interdacryon distance: mild, 30 to 34 mm; moderate, 35 to 40 mm; severe, greater than 40 mm.[6] Medial canthal (intercanthal) distances were also directly measured.

Patients were subsequently evaluated after 6 weeks and then at 1 year. Clinical follow-up on an annual basis was performed until patients reached skeletal maturity. Radiographic evaluation was obtained when clinical evidence for relapse was present or when indicated for other procedures. Physician satisfaction with operative correction of hypertelorbitism was also measured by physical examination and images at 1 year by 2 surgeon examiners on a 5-point scale (0–4: 0 = dissatisfied, no improvement; 1 = minimally satisfied, mild improvement; 2 = moderately satisfied, moderate improvement; 3 = mostly satisfied, good improvement; 4 = totally satisfied, complete improvement). Perioperative complications including wound infection, meningitis, and seizure were recorded. Relapse was defined as an interdacryon distance greater than 15% above the norm, as determined by Costaras et al,[3] Costaras and Pruzansky,[4] McCarthy et al,[13] and Sukonpan and Phupong.[14] All patients undergoing repeat operation for correction of hypertelorbitism were noted.

Operative Technique

Both facial bipartition and orbital box osteotomy were used for hypertelorbitism correction; however, most cases had a facial bipartition, and the orbital box osteotomy was generally reserved to skeletally mature patient without a malocclusion. The orbital box osteotomy was performed through a coronal incision to allow dissection of the orbital cavity and nasal process. A gingivobuccal sulcus incision was also used to complete the exposure. A frontal craniotomy was performed to allow safe access to the orbital roof, cribiform plate, and crista galli. Calipers were used to measure the interdacryon distance after exposure. The lateral orbital wall was then osteotomized using a reciprocating saw and continued through the frontal bandeau and orbital roof. A 3-mm osteotome was used to mobilize the orbital floor as far posteriorly as possible to ensure translocation of the globe without jeopardizing the optic nerve. Full mobilization of the orbit was completed by osteotomizing the zygomatic arch and maxilla through a combination gingivobuccal sulcus and coronal incision. To move the orbits medially for hypertelorbitism correction, a V-shaped osteotomy was carried out in the midline toward the crista galli. This allowed removal of the frontal bone, ethmoid, and nasal septum. The orbits were then stabilized with rigid skeletal fixation employing titanium plates and screws. Medial canthopexy was performed using transnasal wiring and soft tissue bolsters.

Facial bipartition was similarly performed through combined coronal and gingivobuccal sulcus incisions. Facial bipartition was ideal for patients with hypertelorbitism and midface hypoplasia in need of palatal arch widening. Frontal craniotomy was performed, and the lateral orbital wall, frontal bandeau, and orbital roof were osteotomized using a reciprocating saw (Fig. 1). The orbital floor and medial wall were cut using a 3-mm osteotome to complete the "doughnut osteotomy." A curved osteotome was lastly used to mobilize the pterygomaxillary buttress. Before down-fracture, the interdacryon distance was measured using calipers and a V-shaped excision was marked between the orbits. Once full mobilization of the midface and orbits was performed, the midline V was excised, and the bipartition halves were brought together by rotating them toward the midline. Rigid fixation was obtained using a "box" titanium plate. Medial canthopexy was performed as described above for box osteotomy. Redundant soft tissue in the glabellar region was reefed together using horizontal mattress sutures to position the eyebrow just medial to the medial canthus.[5]

Statistical Analysis

All patient data were expressed as mean ± SEM. Comparisons made between age groups were performed using a 1-way analysis of variance or unpaired t test to calculate a 2-tailed P value. A 2-tailed Fisher exact test was used to evaluate rates of relapse when assessing the effect of age. $^*P < 0.05$ was considered statistically significant. To compare interdacryon distance and percent relapse based on preoperative severity of hypertelorbitism, a linear regression was used to assess the overall variance. Significance was assessed using a 2-sided Cochran-Armitage trend test. $^*P < 0.05$ was considered statistically significant.

RESULTS

Between the years 1975 and 2009, 33 patients were identified at UCLA Medical Center who underwent surgical correction of hypertelorbitism. Diagnoses included Crouzon syndrome (n = 13), Apert (n = 8), Pfeiffer (n = 3), Saethre-Chotzen (n = 1), Jackson-Weiss (n = 1), Antley-Bixler (n = 1), nonsyndromic craniosynostosis (n = 1), and median craniofacial dysplasia (n = 5). The mean age at time of surgery was 9.2 ± 3.0 years. The mean preoperative interdacryon distance was 36.5 ± 4.8 mm, and the immediate intraoperative distance achieved was 18.3 ± 1.4 mm. Intercanthal distance changed from preoperative of 46.5 ± 3.9 mm to a distance of 28.5 ± 2.3 mm with a mean reduction of 39%. There were 9 relapses (>15% wider than the norm) noted or 27.2%, with 8 patients undergoing subsequent repeat operation. Total follow-up for these patients averaged 14.0 ± 5.9 years (Table 1).

Of the 33 patients, 6 were younger than 6 years when they underwent surgery and 27 were 6 years or older at the time of hypertelorbitism correction (Figs. 2 and 3). There were 12 patients with mild hypertelorbitism (32.2 ± 1.5 mm), 17 patients with moderate hypertelorbitism (37.2 ± 1.3 mm), and 4 patients with severe hypertelorbitism (46.5 ± 3.7 mm). Twenty-eight patients underwent

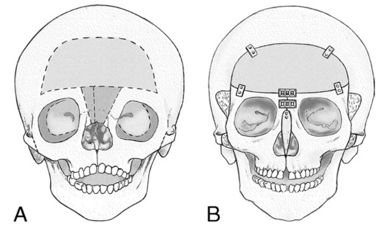

FIGURE 1. Facial bipartition intraoperative correction. A, Illustration of frontal view of osteotomy lines including craniotomy, midline asymmetric "V" wedge excision of frontonasoethmoidal bone, and midface buttresses (zygomatic arch, circumferential orbital walls, pterygomaxillary). B, Illustration of frontal view with rigid fixation and cranial bone graft to nasal dorsum.

TABLE 1. Patients' Demographics

No. patients	33
Mean age at surgery, y	9.2 ± 3.0
Age <6 y	6
Age ≥6 y	27
Mean preoperative interdacryon distance, mm	36.5 ± 4.8
Mild hypertelorbitism (30–35 mm)	12
Moderate hypertelorbitism (36–40 mm)	17
Severe hypertelorbitism (>40 mm)	4
Mean postoperative interdacryon distance, mm	18.3 ± 1.4
Mean follow-up interdacryon distance, mm	22.2 ± 2.6
No. relapses	9

facial bipartition and 5 patients underwent orbital box osteotomy. Between these 2 procedures, there was similar preoperative (facial bipartition [FB] = 35.7 ± 3.1 and OB = 34.2 ± 2.7) and follow-up (FB = 22.1 ± 2.3 and orbital box osteotomy [OB] = 22.9 ± 1.7) interdacryon distances. Overall mean physician satisfaction score was 3.4 ± 0.4.

Patient Age

For patients undergoing surgery younger than 6 years, the average age at surgery was 4.7 ± 0.5 years. The mean preoperative interdacryon distance was 42.0 ± 6.7 mm, and the immediate correction achieved in the operating room was 18.2 ± 1.7 mm (Fig. 4). The mean intercanthal distance changed from 50.8 ± 3.1 preoperatively to 26.1+1.9 after the procedure to 32.4 ± 2.2 during follow-up. On follow-up at 3.3 ± 0.8 years, the average interdacryon distance was found to be 26.2 ± 3.0 mm. There were 3 postoperative complications in this group noted, including 1 patient with meningitis, 1 with a seizure, and 1 with a wound infection. On the basis of normal measurements for age, all 6 patients undergoing surgery younger than 6 years were noted to have relapse (Fig. 4). Five of the 6 patients underwent subsequent repeat operation for correction at an average age of 13.0 ± 2.9 years. Total follow-up for these patients was 20.0 ± 5.6 years, and physician satisfaction score was 3.2 ± 0.5.

Of the 27 patients undergoing correction of hypertelorbitism 6 years or older, the average was 10.2 ± 2.3 years for surgery. The mean preoperative interdacryon distance measured was 35.3 ± 3.3 mm, and the immediate postoperative distance was 18.4 ± 1.4 mm (Fig. 4). There were 2 complications noted in this group; one patient was taken back to the operating room for postoperative bleeding and another patient had a wound infection. At an average follow-up of 3.8 ± 1.7 years, the interdacryon distance observed was 21.3 ± 1.5 mm. The mean intercanthal distance changed from 46.2 ± 3.9 preoperatively to 27.6 ± 2.3 after the procedure to 29.7 ± 2.0 during follow-up. There were 3 patients with relapse (11.1%), and all 3 were again operated on at 18.7 ± 2.9 years (Fig. 4). Total follow-up

FIGURE 2. Frontal images of patient with midline cleft and moderate hypertelorbitism. A, Preoperative view of patient with intradacryon distance of 36 mm at age 4.5 years. B, Postoperative view of the same patient after facial bipartition correction at age 10 years. C, Follow-up image after 12 years at age 17 with mild relapse. Patient did not elect to have another procedure.

FIGURE 3. Frontal images of a patient with midline cleft and moderate hypertelorbitism. A, Patient seen with intradacryon distance of 37 mm at age 3 years. B, Preoperative view of the same patient before facial bipartition correction at age 9 years. C, Follow-up image at 12 years with no relapse.

for these patients was 12.7 ± 5.1 years after initial correction, and the physician satisfaction score was 3.5 ± 0.4.

Severity of Hypertelorbitism

Twelve patients with mild hypertelorbitism were operated on at an average age of 10.4 ± 2.2 years. The immediate correction achieved was 17.7 ± 0.9 mm. At 3.8 ± 1.1 years of follow-up, the average interdacryon distance measured was 20.4 ± 1.2 mm (Fig. 5). The mean intercanthal distance changed from 40.2 ± 3.1 preoperatively to 24.6 ± 2.3 after the procedure to 28.0 ± 2.0 during follow-up. There were no relapses in this group, and the physician satisfaction score was 3.5 ± 0.3. Patients with moderate hypertelorbitism were operated on at an average at of 9.0 ± 3.1 years. The immediate interdacryon distance achieved was 18.5 ± 1.5 mm. At 3.8 ± 1.9 years of follow-up, the mean interdacryon distance measured was 22.4 ± 1.6 mm. The mean intercanthal distance changed from 44.8 ± 3.1 mm preoperatively to 26.7 ± 1.8 mm after the procedure to 32.8 ± 3.0 mm during follow-up. There were 5 patients (29.4%) among the 17 who had relapse, and the overall satisfaction score for this group was 3.4 ± 0.4 (Fig. 5). Among the 4 patients with severe hypertelorbitism, the average age of operation was 6.3 ± 3.2 years. The interdacryon distance measured in the operating room after correction was 19.5 ± 1.3 mm. At an average of 3.3 ± 1.0 years of follow-up, the measured interdacryon distance increased to 26.8 ± 3.8 mm, which was significantly greater than in patients with mild and moderate hypertelorbitism. The mean intercanthal distance

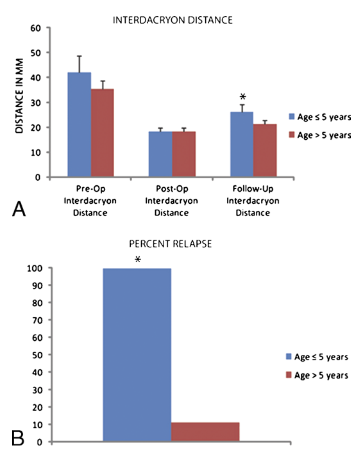

FIGURE 4. Hypertelorbitism relapse based on patient age. A, Interdacryon distance (mm) measured preoperatively, postoperatively, and on follow-up for younger patients (<6 y; left) and for older patients (≥6 y; right) showed similar preoperative and improved postoperative intradacryon distances. Follow-up intradacryon distances increased for the younger but not older group. B, Percent relapse (>15% above norm) was also significantly greater on follow-up for patients undergoing surgery at younger than 6 years (left) compared to patients 6 years or older (right). *$P < 0.05$.

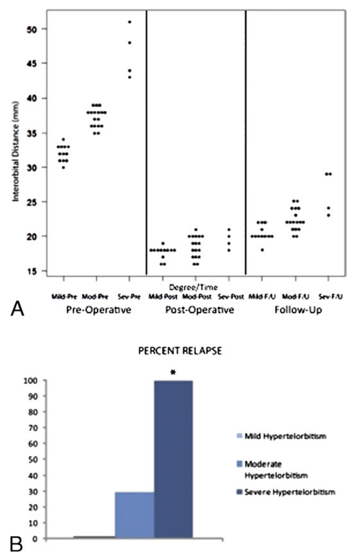

FIGURE 5. Severity of hypertelorbitism. A, Interdacryon distance in millimeters measured preoperatively (left column), postoperatively (middle column), and on follow-up (right column) for patients with mild, moderate, and severe hypertelorbitism. Linear regression with two-sided Cochran-Armitage test demonstrated significantly greater distance on follow-up for patients with severe hypertelorbitism. B, Percent relapse was significantly greater on follow-up for patients who started with severe hypertelorbitism (greatest interdacryon distance). *$P < 0.05$.

changed from 51.4 ± 3.5 mm preoperatively to 25.3 ± 1.1 mm after the procedure to 35.2 ± 2.8 mm during follow-up. All 4 patients in this group had relapse, which was statistically greater than in those with mild and moderate hypertelorbitism, and the final physician satisfaction score was 3.1 ± 0.3.

DISCUSSION

Hypertelorbitism is often among the most severe and readily apparent of all congenital deformities.[15] A variety of surgical approaches for correction have been elaborated in the literature. Early attempts described resection of just the central bony portion of the nose and medial movement of the inner eyebrow in an attempt to camouflage the underlying abnormality.[1,16] Subsequent attempts to move the bony orbit included direct excision of the midportion of the nasal bones and translocation of the medial orbital walls with wire fixation. To compensate for enlargement of the orbital cavity, silicone implants were placed inside the lateral wall.[17] Contemporary techniques for correction of hypertelorbitism (facial bipartition and orbital box osteotomy) take root in the work done by Tessier et al, who first introduced a 2-stage craniofacial osteotomy technique, followed later by a 1-stage combined intracranial and extracranial approach.[1,9] Converse et al[1] demonstrated this strategy to be safe, successfully performing encephalocele reduction, dural repair, orbital osteotomy, and median bone resection in 3 patients as a single procedure.

With facial bipartition and orbital box osteotomies now established as the procedures of choice for correction of hypertelorbitism, attention has shifted toward the optimal timing to perform these surgeries. At the root of this controversy lies the appropriate age to perform orbital repositioning, taking into account continued growth of the midface and risk for relapse. In our study, we use the term *relapse* to take into consideration either a relapse of bony fixation or failure to keep up with normative facial growth. In an early series of 19 patients with ages ranging from 3 to 11.5 years, Mulliken et al[2] evaluated a variety of factors associated with favorable and unfavorable results. Average follow-up of 6.7 years was reported for this group. Of note, an unfavorable outcome was defined as greater than 5 mm relapse in interorbital distance. Greater preoperative and immediate postoperative interorbital distances were found to be significantly associated with unfavorable results.[2] Interestingly, initial conclusions did not find age at the time of surgery to be an important parameter. Careful evaluation of the data, however, demonstrated a disproportionate distribution of younger patients with relapse, particularly those ages 5 or younger (Fig. 3).[2] Furthermore, midfacial growth was found to be adversely affected by surgery to reposition the orbits.

In contrast to these findings, McCarthy et al[12] reported on a series of 20 patients all younger than 5.3 years at the time of hypertelorbitism correction and all with a minimum of 5 years of follow-up. Relapse in this study was defined as greater than 2 SDs above age-adjusted bony interorbital distance mean.[3,4] Interestingly, despite the younger demographic in this series, 16 of the 20 patients were found to have no relapse. Among the 4 patients with relapse, the average preoperative interorbital distance was 31 mm, and the intraoperative reduction was 13 mm.[12] Although these data may argue that facial bipartition or orbital box osteotomy can be performed safely in younger children, the relapse rate was still 20%. In addition, close analysis of the findings revealed that, as a whole, the study population comprised patients with less severe hypertelorbitism. The greatest preoperative interorbital distance was only 38 mm, falling within range of mild to moderate for classification of hypertelorbitism.[12] In comparison, the series of Mulliken et al[2] included an average distance of 39.8 mm for their unfavorable group, and preoperative interorbital distances reached as high as 59 mm. Considering such disparate reports, appropriate timing for hypertelorbitism correction remains a highly controversial subject in the craniofacial literature.

In this present study, we have reported our experience with 33 patients undergoing correction for hypertelorbitism in association with syndromic craniosynostosis, nonsyndromic craniosynostosis, or median craniofacial dysplasia. Our data showed a distribution of severity for hypertelorbitism ranged from a preoperative interdacryon distance of 30 to 51 mm.[2] Among patients undergoing surgery at younger than 6 years, all patients failed to keep up with the norms or experienced relapse and 83.3% were again operated on for correction. One patient elected not to pursue further surgery. This contrasts with the rate of relapse among patients who were operated on at 6 years or older (11.1%). All 3 of these older patients elected to undergo repeat operation. A 2-tailed Fisher exact test demonstrated this difference in relapse rate to be statistically significant. Although there was a trend toward higher physician satisfaction scores among patients operated on later, this difference was not significant.

Our data also showed that preoperative severity of hypertelorbitism was a predictor of long-term relapse. Relapse was greater for patients in the severe group (interdacryon distance >40 mm) when compared with patients with mild (30–35 mm) or moderate (36–40 mm) hypertelorbitism using a Cochran-Armitage 2-sided trend test. Interestingly, there was a trend toward operating earlier on patients with the most severe condition; however, this difference was not significant. There was also a trend toward lower physician satisfaction scores as the severity of hypertelorbitism worsened, but again this was not significant.

A multitude of factors undoubtedly contributes to the decision as to when surgical correction should be attempted. Although delaying surgery until skeletal maturity would most likely yield the lowest rates of relapse, other mitigating variables often emerge to accelerate the timeline. Exigent concerns from the patient's family to correct the deformity as early as possible even before school age may compel early attempts at correction. Finally, as interdacryon distance increases, orbital positioning and eye alignment diverge further from the norm. Thus, with a delay in surgical correction in these severe cases, there may be impairment in the development of stereoacuity, adversely affecting long-term binocular vision after correction.[18–20] Although it has been argued that early surgery may not restore single binocular vision, we did note, in one of our most severe patients, good postoperative bilateral vision with coordinated ocular movement at 1 year of follow-up.[2] A combination of these factors may have therefore contributed to the *slightly* earlier age at surgery noted in this present series for those with the most severe hypertelorbitism.

On the basis of the significantly higher rate of relapse noted for patients operated on younger than 6 years, we advocate delaying surgery, when possible, until later childhood. By waiting until at least 6 years, the child would be at the stage of early mixed dentition and the orbits and midface would have attained 85% to 90% of their adult size.[21–24] Nonetheless, a decision should be balanced with the psychosocial benefits of earlier correction, particularly in a child with a severe facial cleft and hypertelorbitism. In patients with less severe forms of hypertelorbitism, waiting until after the age of 6 before surgical repositioning of the orbits will likely yield the most durable results.

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