chapter 16

# Craniosynostosis

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## PATIENT EVALUATION AND SELECTION

Craniosynostosis is defined as the premature closure of a cranial suture which causes abnormal calvarial growth. Virchow's law states that skull growth is arrested in the direction perpendicular to the fused suture and expanded at the sites of unaffected sutures, leading to characteristic calvarial deformations.[1] This condition can be classified into simple (single suture) versus complex (multiple sutures) and nonsyndromic versus syndromic subtypes (Tables 16-1 and 16-2).

Preoperative assessment for craniosynostosis includes a detailed medical history, physical examination, and radiographic imaging. Medical history should elicit skull irregularities, associated syndromes, family history of calvarial deformities, and symptoms of intracranial hypertension (headache/vomiting, developmental changes, irritability, and oculomotor paresis). Physical examination should evaluate for characteristic calvarial shapes and asymmetries, premature closure of the anterior fontanelle (normally open until 12–18 months of age), perisutural ridging, and signs of intracranial hypertension (papilledema, supraorbital retrusion, severe towering (turricephaly), and severe frontal/occipital bossing). Head circumferences, cranial indices, and anthropometric measurements should be documented.

Radiological investigation may be necessary to corroborate the diagnosis and/or rule out any associated intracranial abnormalities. Computed tomography remains the most sensitive barometer of bony fusion and may provide evidence for elevated intracranial pressure, as noted by erosion of the inner calvarial table ("copper-beaten" appearance also referred to as "thumb printing") (Figure 16-1). CT imaging and MRI are also helpful in evaluating the underlying brain for structural or functional abnormalities, including hydrocephalus, holoprosencephaly, cortical dysplasias, and Chiari malformations.

Surgical intervention is indicated in craniosynostosis both for the correction of calvarial contour deformities and the prevention of psychosocial dysfunction, intracranial hypertension, and/or mental retardation. Studies have shown that the presence of intracranial hypertension is dependent on the number of affected sutures, ranging from approximately 14% for single-suture synostosis to approximately 47% in multisuture synostosis.[2–3] Sutural release in simple craniosynostosis has been advised due to the concerns regarding increased intracranial pressure as well as the mild but significant developmental delay in the aging child. Patients with complex synostoses present with increased severity of physical and neurological symptoms; therefore, surgical intervention is even more imperative.

## PATIENT PREPARATION

Preoperative considerations include the optimal type and timing of surgical correction for craniosynostosis. A broad range of surgical options exist in the armamentarium of contemporary craniofacial surgical reconstruction, all with the primary objective of releasing the affected suture(s) to permit normalization of skull growth in the setting of accelerated cerebral growth. An open craniofacial approach remains the mainstay of therapy, relying on wide scalp dissection, extensive calvarial osteotomies, and skull reconfiguration that is individually tailored to each cranial vault deformity.[4] To address concerns regarding incision length, operative blood loss, and length of stay for open craniofacial procedures, minimally invasive techniques have been proposed. These techniques include endoscopic sutural release,[5] spring-assisted cranioplasty,[6] and distraction osteogenesis.[7]

The optimal surgical age has been a source of contention, given its differential effects on intraoperative hemodynamics, postoperative cranial growth, and subsequent mental

development. While the literature is inconclusive regarding the appropriate timing for correction of craniosynostosis, the majority of craniofacial surgeons operate between 3 and 12 months of age. Surgical age is also dependent on the type of surgical approach employed. Minimally invasive techniques, which rely on dynamic cranial vault alteration during rapid calvarial growth, are generally performed at an earlier age than open surgical correction.

Given the senior authors' extensive experience with open craniofacial reconstruction for craniosynostosis, surgical considerations, operative steps, and outcomes shall be described for this type of surgical approach in the accompanying sections. In preparation for open craniosynostosis surgery, measures for ensuring adequate blood resuscitation should be undertaken. These include obtaining a blood type and cross, autologous blood donation, and/or allogenic blood direct donation from a family member. Parents should be instructed that open craniosynostosis surgery is performed under general anesthesia. For most types of open craniosynostosis surgeries, patients are placed in the supine position to facilitate calvarial exposure. However, if the posterior vault is being addressed, as in a lambdoidal synostosis or some sagittal synostoses, the patient is ideally placed in the prone position. In the prone position, care must be taken to ensure adequate protection of the globes, facilitated by bilateral tarsorrhaphies and periorbital cushioning.

A plating system is used by most surgeons to maintain the proper position of the osteotomized bone segments after they have been placed in the desired position. Historically, titanium was used but fell out of favor when it was noted that the plates translocated with continued calvarial growth. Resorbable plates eliminate the problems associated with translocation. They maintain strength across the osteotomy long enough for the bone to heal but are resorbed by the body after several years. Most surgeons today employ some type of resorbable fixation for cranial vault remodeling.

## TECHNIQUE

### Metopic Synostosis (Trigonocephaly)

Metopic synostosis is marked by a variable degree of phenotypic severity, ranging from mild ridging to the formation of a triangular shaped head (trigonocephaly) or prominent "keel" forehead with or without hypotelorism. Although an endocranial ridge is not commonly seen in patients with metopic synostosis, an endocranial notch can be observed on axial CT images and is virtually diagnostic of premature suture fusion. Weinzweig termed this radiographic finding the *metopic notch*, a morphologic abnormality that is seen in 93% of synostotic patients (Figure 16-2).[8] A metopic notch is not seen in *any* nonsynostotic patients and, therefore, can be used to diagnose metopic synostosis even *after* the period physiologic suture closure. This notch represents the anatomic site of attachment of the falx, a dural reflection off the crista galli with basicranial origins, and suggests a role for the cranial base in metopic suture fusion.

Whereas a metopic notch describes the endocranial finding on axial CT images in patients with metopic synostosis, a corresponding three-dimensional groove is found on the endocranial surface of the actual skull that extends from the nasion to the anterior fontanelle in these patients. Weinzweig termed this clinical finding the *metopic groove*, an anatomic abnormality that can reliably be found in patients with metopic synostosis (Figure 16-3).

In general, the goals of surgery are the normalization of the forehead with reconstitution of a normal supraorbital rim when necessary. Individuals presenting solely with a prominent midline keel may be best served by simple contouring of the frontal bone or by removal of the frontal bone flap followed by reconfiguration. Conversely, patients with significant trigonocephaly and hypotelorism will require a fronto-orbital reconstruction, recontouring the frontal bone, and laterally expanding the orbits, often with cranial bone graft placed in the midline of the frontal bandeau at the level of the nasion.

The essentials of fronto-orbital reconstruction involve a standard "stealth" (zig-zag) coronal incision, providing for adequate exposure of the fronto-orbital region while minimizing any postoperative scar. Perioperative antibiotics and steroids are given prior to the start of surgery. The incision is infiltrated with 0.5% lidocaine and 1:400,000 parts epinephrine to minimize intraoperative bleeding. The frontal and temporal regions are dissected in the subgaleal plane and care is taken to preserve the periosteum on the surface of the bone, which helps minimize blood loss and may be used to stabilize osteotomized bony segments. The dissection is taken down to the level of the periorbital tissues, with caution taken to avoid any injury to the underlying globes. Following exposure of the frontal and orbital regions, the frontal bone is removed, providing access to the intracranial compartment. The supraorbital rim is then removed in one piece to facilitate reconstruction of the previously triangular-shaped supraorbital bar. Care is taken to remove sufficient bone in the region of the sphenoid bone to allow for growth at the midface and orbits. If the orbits require correction of hypotelorism, it will be necessary to displace the lateral walls of the orbit as well as to split the midline and interpose a calvarial bone graft. Reconfiguration of the supraorbital bar often requires a midline osteotomy to facilitate a flattened forehead with additional partial-thickness bone cuts at the lateral (pterional) angle to promote normalization of the lateral supraorbital angle. The supraorbital reconfiguration is maintained by the utilization of intervening bone grafts as well as resorbable hardware. Following placement of the supraorbital bar as a foundation, the frontal bone is reconstructed using the remaining portions of bone. It is often possible to reverse the original frontal bone flap (posterior portion is now in an anterior position) to obtain an adequate width and contour with the new frontal bone flap. It is important to provide adequate enhancement at

the pterional region to avoid long-term supratemporal hollowing or recession.

## Sagittal Synostosis (Scaphocephaly, Dolichocephaly)

Children with sagittal synostosis will present with a narrow, elongated skull (*dolichocephaly*, long headedness; *scaphocephaly*, boat shaped). Depending on the region of greatest premature fusion of the sagittal suture, the child may manifest frontal or occipital bossing, or a combination of both. Some children will also demonstrate a "towering" skull, also known as *turricephaly*.

The treatment for sagittal synostosis remains controversial, with surgical approaches ranging from minimal removal of the involved suture and bone to extensive total calvarectomy and reconfiguration. Simple synostectomy or simple strip craniectomy is safe and well tolerated, providing adequate cosmetic results in select patients with mild deformities. This procedure, however, has several disadvantages stemming from the fact that it strictly addresses the fused suture and not the compensatory changes in skull shape. It also leaves a large unprotected area over the vertex of the skull, an area with a high rate of restenosis and renewed growth restriction.

An extended synostectomy provides immediate restoration of normal skull contour by shortening the anteroposterior dimension, expanding the biparietal dimension, and addressing the frontal and occipital prominences. Amongst extended synostectomy techniques, the pi procedure has become a widely utilized approach for older infants (3–12 months of age) with scaphocephaly. The essential steps involve removal of bone along both sides of the sagittal suture as well as over the coronal suture, in the design of the Greek letter pi (Π). Cranial bone overlying the sagittal sinus is left intact to minimize bleeding. Drill holes are placed in the adjacent osteotomized bone flaps, allowing for placement of an absorbable suture that helps narrow the anteroposterior dimension while expanding the biparietal length.

Staged or partial vault remodeling procedures also work well in indicated patients. In some cases the majority of the deformity occurs anteriorly or posteriorly. In these cases, vault remodeling may be limited to the most involved area (posterior vs anterior) of the cranium. In properly selected cases this approach is less morbid and produces excellent results.

To address older infants beyond the period of maximal cerebral growth (>12–18 months of age), as well as those children with significant frontal or occipital prominence, procedures with more aggressive craniectomies and reconstruction must be undertaken.[9] Total calvarectomy and reconstruction in the setting of severe or late presentation scaphocephaly has been proposed to offer superior cosmetic results with a minimal increase in morbidity. This may be accomplished by removal of the frontal, occipital, and both parietal bones. Subsequently, reconfiguration is carried out and aimed at providing a shortened anteroposterior dimension in addition to a widened biparietal diameter. The use of rigid fixation is indispensable in these cases, providing greater three-dimensional conformational stability and decreased intraoperative time, bleeding, and postoperative infection.

## Coronal Synostosis (Anterior Plagiocephaly/ Brachycephaly)

Patients with unicoronal synostosis present with anterior, or frontal, plagiocephaly whereas those with bilateral coronal involvement demonstrate brachycephaly. Phenotypic features of anterior plagiocephaly include ipsilateral perisutural ridging, forehead flattening, and orbital recession, coupled with contralateral compensatory frontal bossing. Facial deformities are also common, including nasal root displacement toward the ipsilateral side, anterior displacement of the ipsilateral ear, increased orbital aperture, and chin deviation toward the contralateral side. Pathognomonic radiographic findings diagnostic of unicoronal synostosis include elevation of the ipsilateral orbit which is seen secondary to superior displacement of the greater wing of the sphenoid, also known as the "harlequin" deformity (Figure 16-4A–C). Phenotypic features of brachycephaly include forehead retrusion and flattening, frontal towering, and biparietal widening. The nasal dorsum can be low and hypertelorism may be present. AQ3

Surgical intervention in unicoronal and bicoronal synostoses aims to correct both the frontal and orbital asymmetries. With the current understanding that unilateral coronal synostosis presents with bilateral dysmorphic changes, bilateral correction is now believed to be the optimum approach. For anterior plagiocephaly, a bilateral fronto-orbital advancement procedure is employed for expansion of the affected forehead and orbit with concomitant recession of the contralateral orbit (Figure 16-4E–I). Excellent symmetry can be achieved in this manner (Figure 16-4J,K). Bilateral fronto-orbital advancement also serves in the correction of bicoronal synostosis.

The bilateral fronto-orbital advancement reconstruction involves the release of both coronal sutures while providing bilateral frontal and orbital correction (Figure 16-5). A standard coronal skin incision is made. A bifrontal bone flap is removed typically in one piece, leaving a 1 to 2 cm wide supraorbital bandeau. Extending the osteotomy posterior to the coronal suture will often provide adequate width and a satisfactory new frontal reconstruction when this bone flap segment is inverted. The sphenoid wing is osteotomized and the coronal sutures are opened to the level of the skull base, serving to prevent continued growth restriction and resultant postoperative hollowing of the pterional regions. Caution must be exercised when performing the osteotomy across the sphenoid wing on the affected side, as its superior displacement may cause technical difficulty upon frontal bone flap removal and lead to dural laceration if caution is not exercised. After removal of the frontal bone flap, the orbital bandeau is freed with osteotomies performed across the lateral orbit

at the frontozygomatic suture, the orbital roof, and the nasion just above the nasofrontal suture. Care is taken to protect the underlying brain as well as orbital contents with judicious placement of retractors. The bandeau is then reconfigured using partial osteotomies at the midline while using absorbable hardware on the interior surface to maintain the newly contoured shape. The newly configured supraorbital bar is replaced, its lateral aspects and midline fixed to the calvarial foundation with resorbable fixation for improved healing and postoperative maintenance of the surgical construct. It is frequently necessary to overcorrect the expansion of the affected side by 5% to 10% while also providing a convex shape at the lateral border for a satisfactory reconstruction. Often, the unaffected orbit is mildly set back to correct for preoperative compensatory overgrowth. The frontal bone flap is then attached to the supraorbital bar, taking care to match it to the previously overcorrected (5%–10%) orbital bandeau on the affected side. Remaining portions of bone are fixed with absorbable plates or suture, and closure is performed in a routine fashion with placement of subgaleal drains.

### Lambdoid Synostosis and Posterior Deformational Plagiocephaly

Posterior plagiocephaly due to lambdoid suture synostosis is rare, with the majority of observed posterior plagiocephaly secondary to positional molding. Understanding the phenotypic differences between lambdoid synostosis and posterior deformational plagiocephaly is critical toward making the appropriate diagnosis and designing the proper course of treatment. Children with lambdoid synostosis characteristically have a trapezoid-shaped head in association with posterior displacement of the ipsilateral ear, contralateral occipital bossing, and frequent ridging of the affected lambdoid suture. In contrast, posterior deformational plagiocephaly is marked by a parallelogram-shaped head, anterior displacement of the ipsilateral ear, and ipsilateral frontal bossing in the absence of palpable ridging along the lambdoid sutures.

Infants with true lambdoid synostosis may benefit from a variety of surgical approaches, aiming to release the affected suture(s) and normalize the posterior calvarial vault contour. Options include simple synostectomy, unilateral reconfiguration of the affected occipital region, and bilateral occipital reconstruction with or without the use of an occipital bandeau. The majority of lambdoid surgical candidates have significant parietal and frontal compensatory changes in addition to their occipital deformation; therefore, they are best served by a more extended calvarectomy and reconstruction.

For an extended calvarial reconfiguration, the globes are protected with bilateral tarsorrhaphies and patients are placed in a prone position. Perioperative antibiotics and steroids are administered. Intraoperative bleeding is minimized with the use of local anesthetic mixed with epinephrine as well as controlled hypotensive anesthesia. A coronal incision is then undertaken followed by subgaleal dissection to expose the occipitoparietal regions. Both parietal bone flaps are subsequently removed with osteotomies taken posterior to the coronal and anterior to the lambdoid sutures. A midline strip of bone is then left to protect the underlying sagittal sinus. After removal of the parietal bone flaps, dissection is carried out at the level of the lambdoid suture under direct visualization, taking great caution at the level of the transverse, sagittal and sigmoid sinuses. Osteotomies are brought to within 1 cm on either side of the midline, with the final cut made after the underlying dura and sinus have been clearly dissected free under direct visualization. Inadvertent entry into the sinus, particularly at the region of the asterion, may lead to significant blood loss over a short period and constitutes the greatest risk encountered with this approach. Nevertheless, with appropriate care and meticulous dissection, this complication may be avoided in the majority of individuals. The removed calvarial plates are reconfigured to provide adequate reshaping of the occipital contour and stabilized with resorbable hardware, deliberately leaving open the region of the prior lambdoid suture. Subgaleal drains are then placed and closure is carried out in routine fashion.

Treatment of posterior deformational plagiocephaly is a function of both the age and severity at presentation. When presenting before the age of 6 months, therapy consists of positional modifications combined with physiotherapy in the case of constrained neck movements or asymmetric neurological development. In cases of no improvement or progression of the deformity despite repositioning after 2 to 3 months, cranial orthotic (helmet) therapy is indicated.

## COMPLICATIONS

Perioperative morbidity may include wound infection, dural laceration, superficial brain injury, cerebrospinal fluid leak, encephalocele formation, subgaleal hematoma, and ocular injury. Intraoperative blood loss and transfusion requirements, leading to hemodynamic instability, constitute great dangers to the patient and should never be underestimated. It is imperative to accurately gauge the extent of blood loss and match accordingly with packed red blood cells. Other serious perioperative complications consist of ischemic brain injury, venous air embolism, epidural and subdural hemorrhage, and severe transfusion reactions.

Long-term postoperative concerns include recurrent calvarial deformities, cranial bone defects that fail to fill in over time, and hardware-related problems. Depending on the extent of morphologic asymmetry, intracranial hypertension, and developmental delay, recurrent calvarial deformities may require a minor or major reoperation. Defects larger than 2 cm in patients older than 18 to 24 months will often persist and may need eventual correction with split calvarial bone graft or bone substitute. Persistent hardware may also be problematic, particularly in patients in whom resorbable hardware was used. It is not uncommon for the polylactic/polyglycolic constructs to remain

in place for 12 to 18 months before eventual resorption. In rare individuals, sterile abscesses may develop at sites of hardware resorption and subsequently require exploration for debridement. Postoperative mortality rates are low and continue to decline with technological advancements and experience.

## OUTCOMES ASSESSMENT

Based on outcomes data on open craniosynostosis surgery, estimated blood loss spans 25% to 500% of estimated blood volume (EBV), transfusion requirement range from 25% to 500% of EBV, complication rates are between 6.8% and 23.3%, and mortality rates center around 0% to 1.1%. Hospital length of stay is typically between 4 to 7 days, with longer admissions for complex synostoses. Reoperation rates range from 7.2% to 23.3% and are indeed higher for complex synostoses.

Outcome studies have also demonstrated that conservative therapy is beneficial in the reduction of calvarial deformities secondary to positional molding. Postural changes and helmet therapy are most effective between 4 and 12 months of age, during the period of rapid brain growth. Approximately 95% of infants can be expected to have satisfactory cosmetic improvement with conservative management. Counterpositioning with or without physiotherapy or helmet therapy may reduce skull deformity, with better results noted with increasing compliance.

AQ3 ## REFERENCES


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**TABLE 16-1.** Classification of Cranioynostosis

| *Affected Suture* | *Phenotypic Presentation* |
|---|---|
| Sagittal | Dolichocephaly, scaphocephaly |
| Coronal (unilateral) | Anterior plagiocephaly |
| Coronal (bilateral) | Brachycephaly |
| Metopic | Trigonocephaly |
| Lambdoid | Posterior plagiocephaly |
| Multiple sutures | Cloverleaf (Kleeblatschädel), acrocephaly, oxycephaly |

**TABLE 16-2.** Craniofacial Dysostosis Syndromes

| *Syndrome* | *Involved Suture* | *Morphological Presentation* |
|---|---|---|
| Crouzon | Coronal, sagittal | Midface hypoplasia, shallow orbits, proptosis, hypertelorism |
| Apert | Coronal, sagittal, lambdoid | Midface hypoplasia, shallow orbits, proptosis, hypertelorism, *symmetrical syndactyly of hands and feet*, choanal atresia, ventriculomegaly, genitourinary/cardiovascular anomalies |
| Pfeiffer | Coronal, sagittal | Midface hypoplasia, proptosis, hypertelorism, *broad great toe/thumb* |

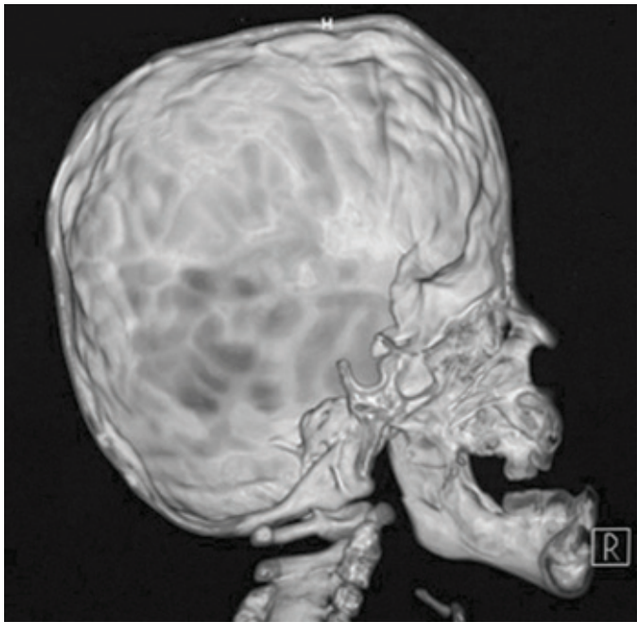
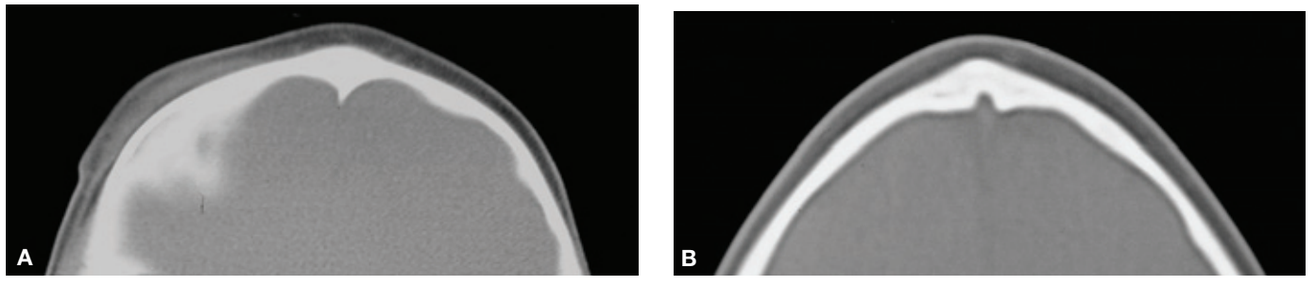



**FIGURE 16-1.** CT findings of elevated intracranial pressure. This sagittal CT image demonstrates the classic "copper-beaten" appearance, also referred to as "thumb printing," resulting from increased intracranial pressure.

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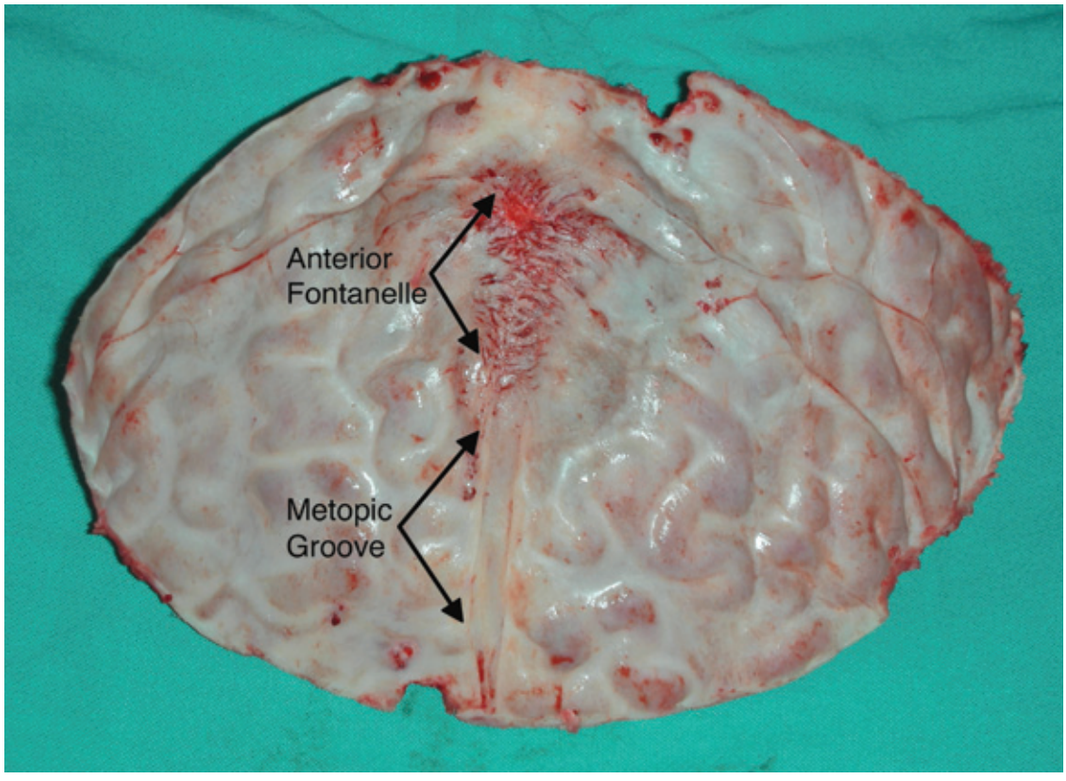
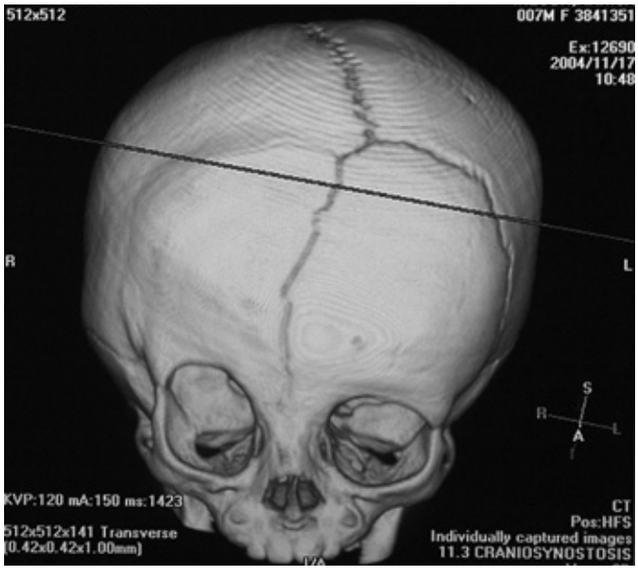
● **FIGURE 16-2.** The metopic notch. Axial CT images demonstrate the endocranial bony spur associated with normal metopic suture fusion (A), and the Ω (omega)-shaped *metopic notch* (B). Moderate ectocranial ridging is also appreciated in this patient.




● **FIGURE 16-3.** The metopic groove. Intraoperative evaluation of the inner table of the anterior cranial vault of a previously untreated 9-year-old child with metopic and bicoronal synostosis and Crouzon syndrome demonstrates a *metopic groove*. While Crouzon syndrome is commonly associated with bicoronal synostosis, this child demonstrated both a metopic notch and metopic groove, pathognomonic findings consistent with metopic synostosis. The metopic groove typically extends from the nasion to the anterior fontanelle. Note the severe depressions of the inner table of the skull indicating increased intracranial pressure in this older child.

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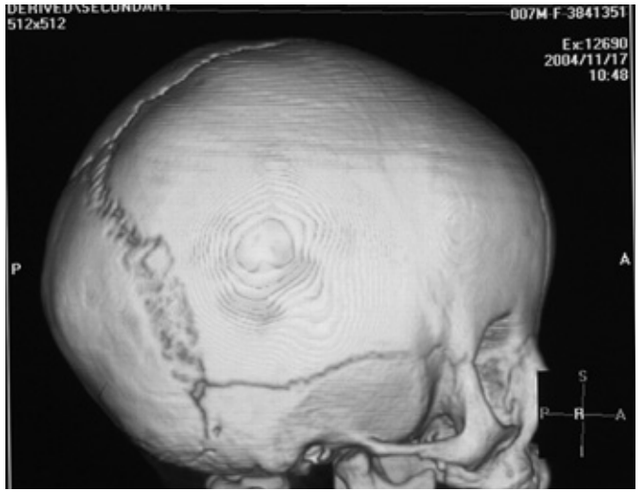
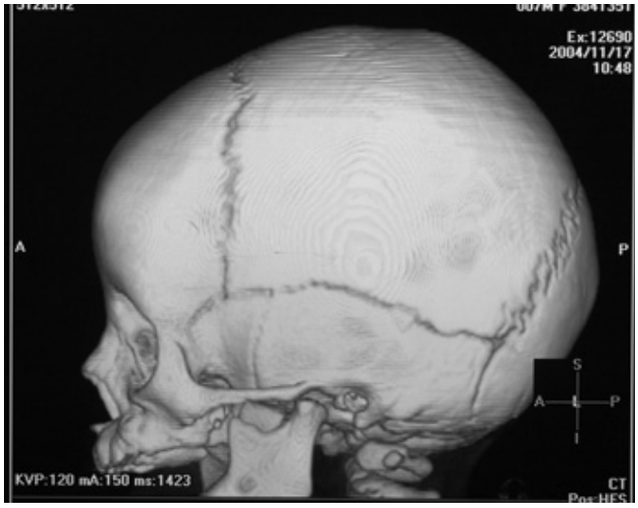
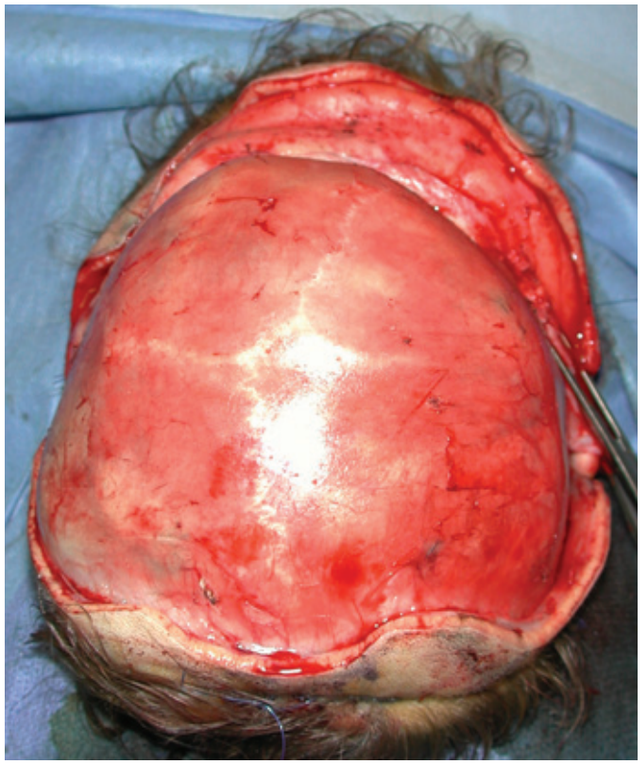



B




C

● **FIGURE 16-4.** Bilateral fronto-orbital reconstruction for unicoronal synostosis. Three-dimensional CT scans demonstrate right coronal synostosis and a harlequin deformity (A, B), and a normal, patent contralateral coronal suture (C). AQ5



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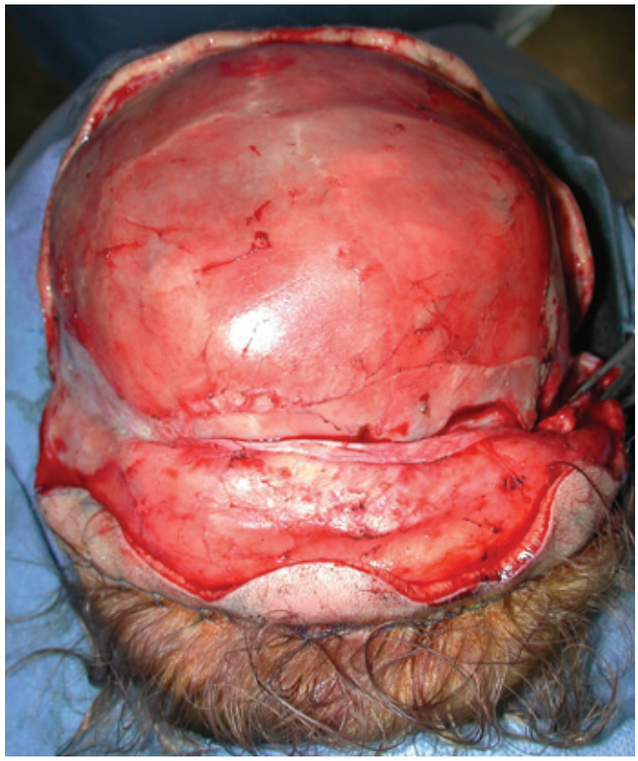

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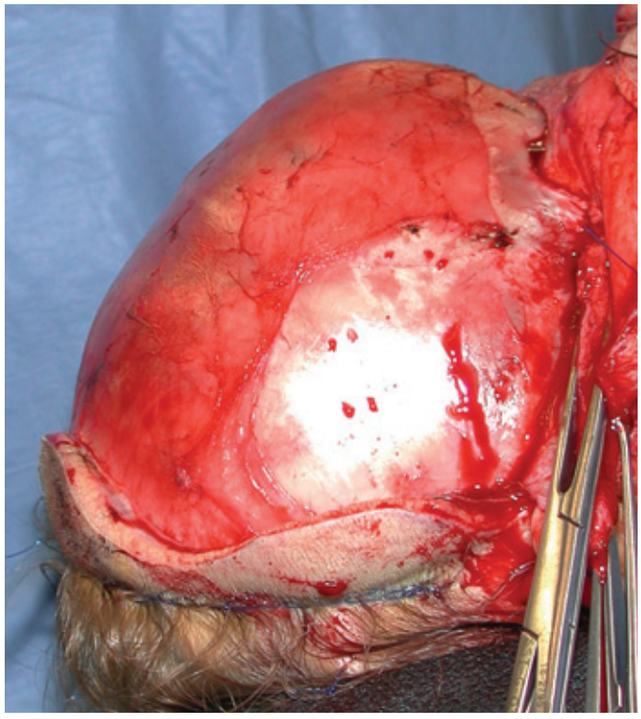
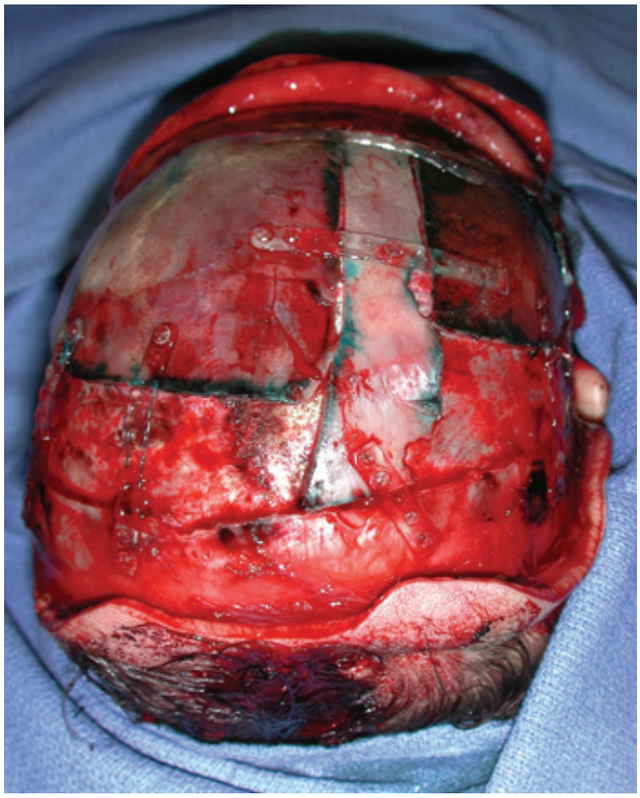
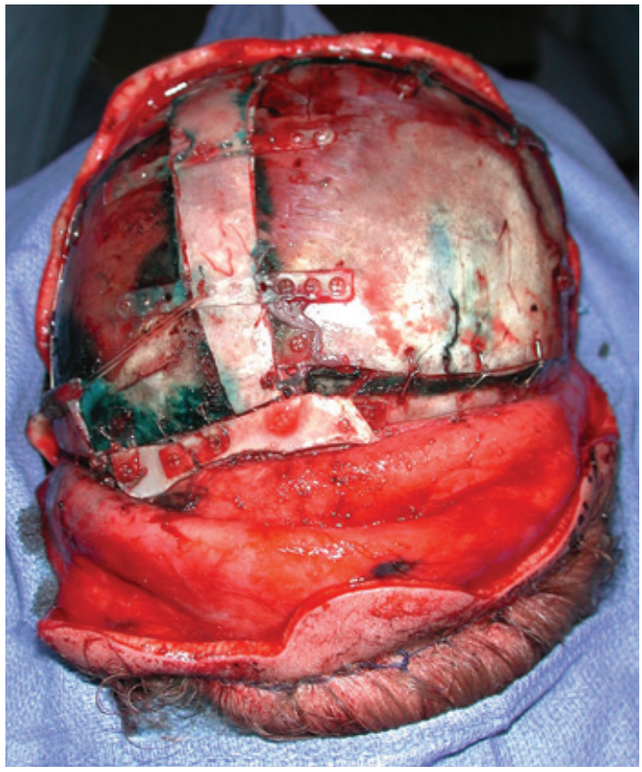


F

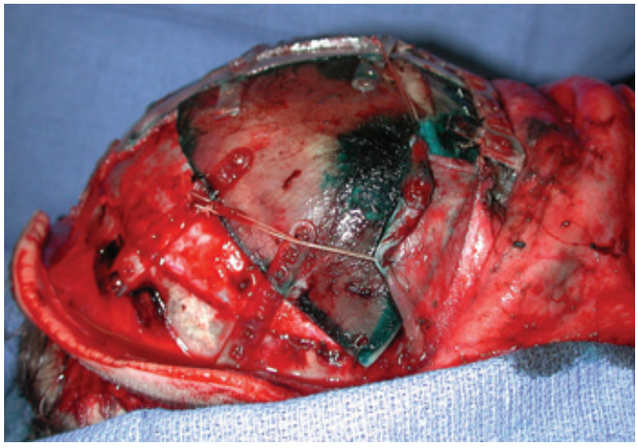
● **FIGURE 16-4.** (*continued*) Intraoperative exposure demonstrates right coronal synostosis, ipsilateral frontal flattening, and contralateral frontal bossing (D–F).



G

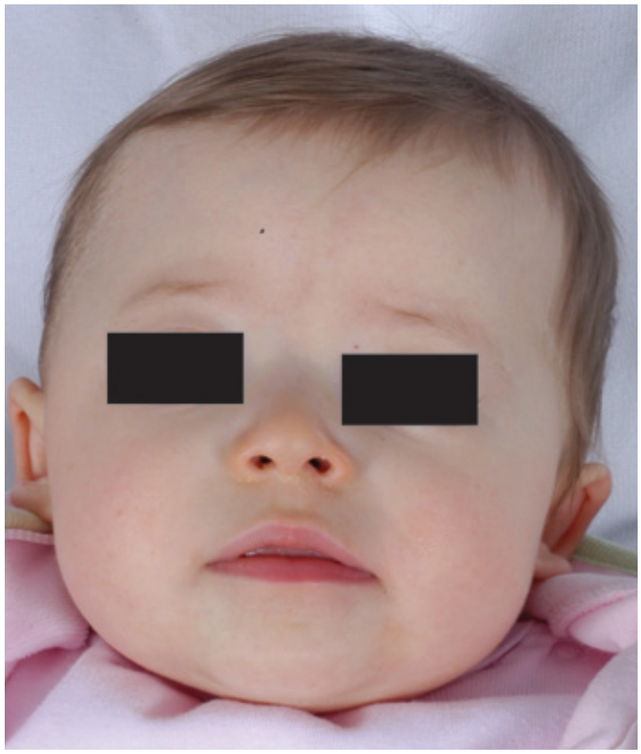


H

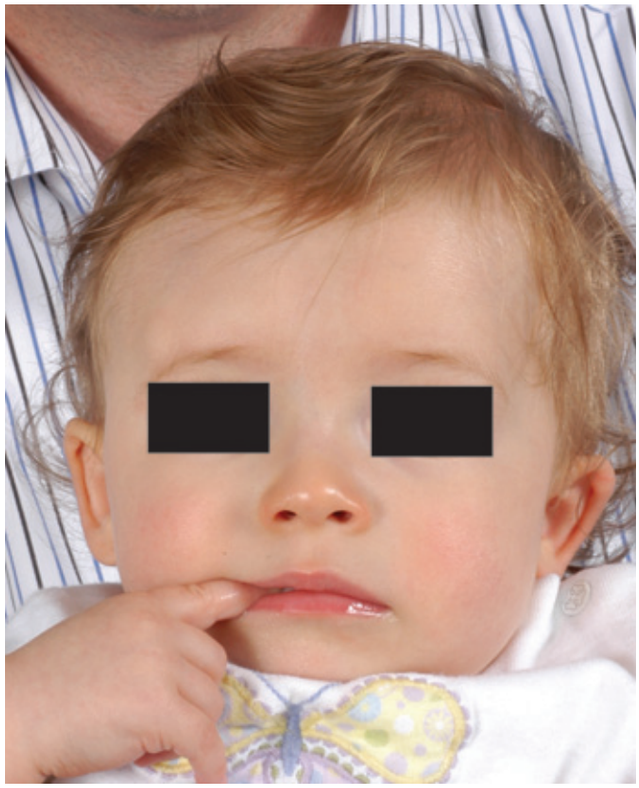


I

● **FIGURE 16-4.** (*continued*) Bilateral fronto-orbital advancement is performed for expansion of the affected frontal bone and orbit with concomitant recession of the contralateral orbit. Fixation is performed using resorbable plates and screws (G–I).



J



K

● **FIGURE 16-4.** (*continued*) Preoperative (J) and postoperative (K) photographs demonstrate correction of the orbital and frontal bone asymmetries. Note the increased aperture of the right eye in the preoperative photo and subsequent correction.

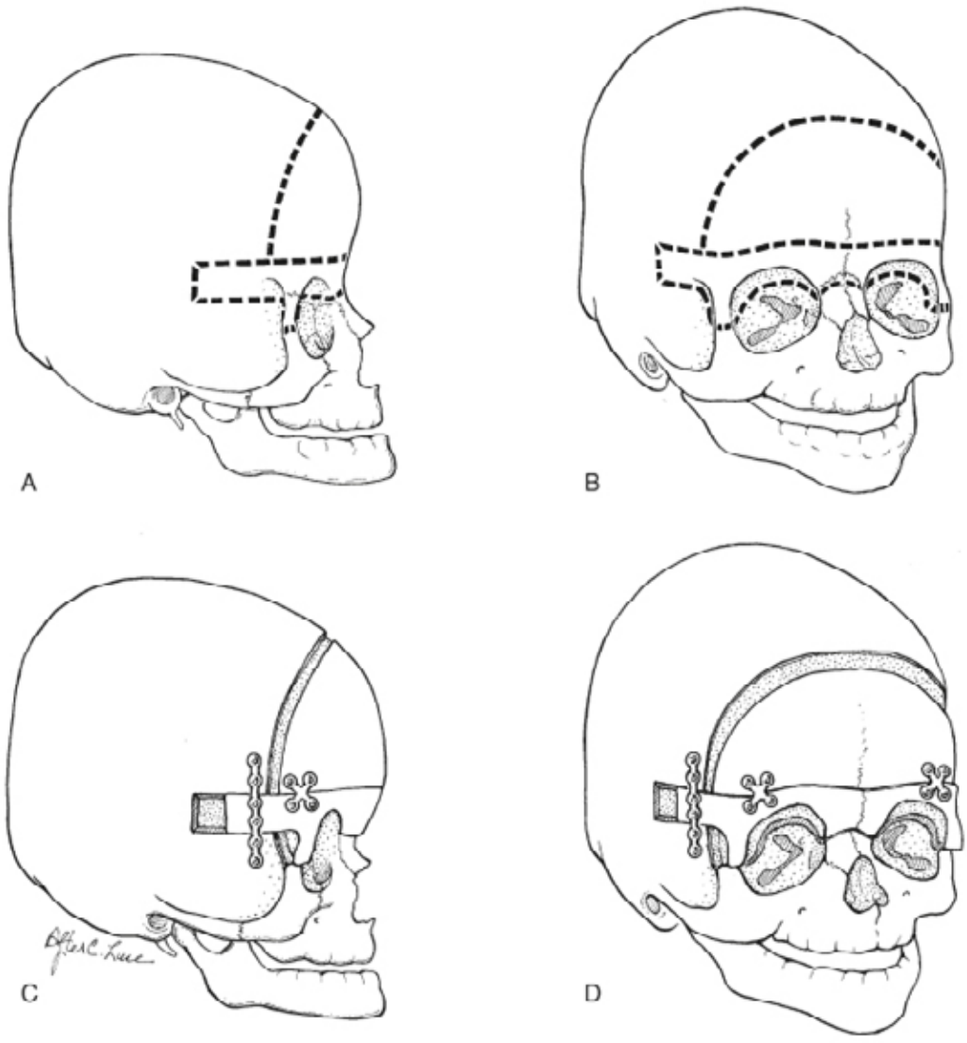



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**FIGURE 16-5.** Bilateral fronto-orbital advancement reconstruction (FOAR) for bicoronal synostosis. The FOAR procedure involves release of both coronal sutures while providing bilateral frontal and orbital correction. A bifrontal bone flap is removed, typically as a single segment, leaving a 1–2 cm wide supraorbital bandeau. A tongue-and-groove osteotomy of the bandeau facilitates advancement and fixation (A, B). Fixation of the frontal bone to the supraorbital bar is then performed (C, D).

## AUTHOR QUERY

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