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The evolution of surgical management for craniosynostosis

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Craniosynostosis, the premature closure of cranial sutures, has been known to exist for centuries, but modern surgical management has only emerged and evolved over the past 100 years. The success of surgery for this condition has been based on the recognition of scientific principles that dictate brain and cranial growth in early infancy and childhood. The evolution of strip craniectomies and suturectomies to extensive calvarial remodeling and endoscopic suturectomies has been driven by a growing understanding of how a prematurely fused cranial suture can affect the growth and shape of the entire skull. In this review, the authors discuss the early descriptions of craniosynostosis, describe the scientific principles upon which surgical intervention was based, and briefly summarize the eras of surgical management and their evolution to present day. *(DOI: 10.3171/2010.9.FOCUS10204)*

Key Words • **craniosynostosis** • **surgical management** • **suture**

Early Descriptions of Cranial Deformity

The aberrant congenital deformities of the skull have been known to exist for centuries and were well-recognized and described as early as the time of antiquity. In the Iliad, Homer describes the warrior Thersites as "the ugliest man who came before Troy…his head ran up to a point..," a description characteristic of oxycephaly.[20] The recognition of cranial vault deformities by the ancient physician Galen, and some early understanding of the role of cranial sutures by Hippocrates, have also been reported.[6,55] By the 16th century, it appears that anatomists appreciated the existence of cranial sutures and had documented a broad range of the characteristics of the deformity, from an appreciation of suture pattern and premature suture fusion in a variety of configurations by Hundt,[21] specific abnormal varieties of sagittal and coronal sutures by Dryander,[13] and what would now be described as oxycephaly and brachycephaly by della Croce[10] and Vesalius[58] (Fig. 1). However, von Sömmering[61] in the late 1790s was the first to go beyond simple descriptions and apply scientific principles to the study of abnormal cranial suture growth. He recognized the importance of sutures in active skull growth and the consequences of premature fusion, thus laying the foundation for our modern understanding of craniosynostosis and the subsequent surgical and nonsurgical interventions.[62,63]

Early Scientific Exploration

In his first scientific descriptions of craniosynostosis, von Sömmering sought to not only describe the primary defect and the cosmetic consequences, but also to elucidate the secondary global cranial impact. He aimed to develop a unifying mechanistic theory describing the pathogenesis with a clear intent on developing a treatment. Soon after von Sömmering, Otto[43]—based on his observations in humans and animals—proposed that a consequence of premature suture fusion was a compensatory cranial expansion along another trajectory in the skull, providing the first explanation for the global cranial abnormalities observed. In 1851, Virchow[59] published a landmark paper in the history of craniosynostosis in which he described the fundamental aberrant growth patterns in this condition, which he termed Virchow's law. Virchow's law stated that the observed deformities occurred as a result of "cessation of growth across a prematurely fused suture," with "compensatory growth" along nonfused sutures in a direction parallel to the affected suture, causing obstruction of normal brain growth.[3] This was the first accurate and generalizable principle applicable to all patterns of premature suture fusion.

Virchow[59] initially described this disorder in 1851 as craniostenosis, meaning a structured or narrowed skull, but was convinced by Sear[47] to instead call this entity

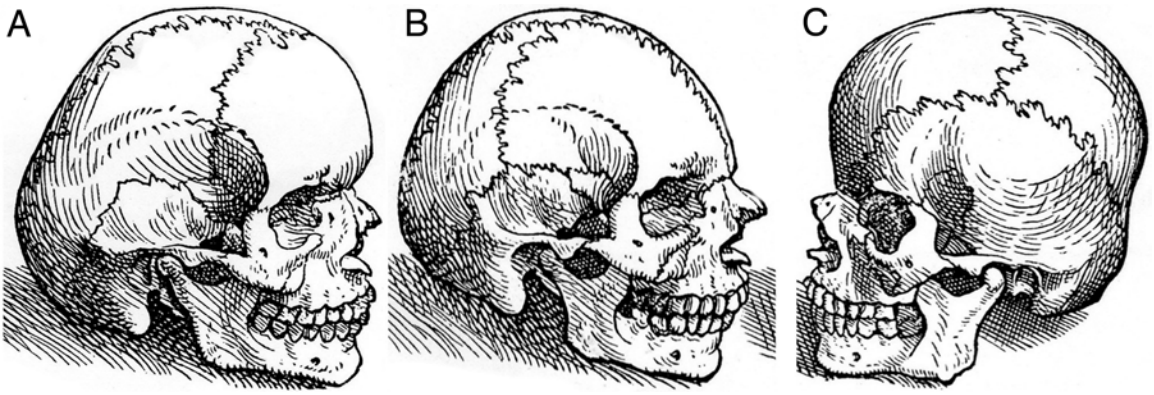


Fig. 1. Illustrations of abnormal skull shapes by Vesalius in 1543 in *De Humani Corporis Fabrica.* Abnormalities were described in terms of prominences, not sutures. **A:** Loss of posterior eminence. **B:** Loss of anterior and posterior eminence. **C:** Loss of eminence at sides. Image credit: William H. Welch Medical Library, Johns Hopkins Medical Institutions.

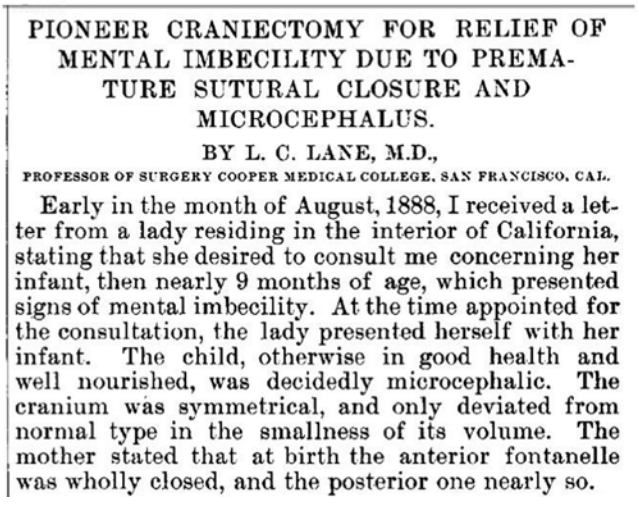
craniosynostosis, which more accurately indicated suture involvement and encompassed all varieties of suture disease. Virchow's impact was significant, as the first surgical interventions and subsequent iterations were based directly on his observations and principles. By the early 1900s, craniosynostosis was recognized as one component of complex syndromic deformities, most notably by Apert[4] in 1906 and Crouzon[7] in 1912, whose names bare two of the most well-known syndromic deformities, of which there are now more than 60.

Nearly a century later, Moss became very interested in craniosynostosis but categorically rejected Virchow's law. Instead, in an attempt to unify all types of craniosynostosis, he proposed that the cranial base, not the suture, was the primary site of abnormality, with suture fusion being a secondary consequence.[12,40,41] He based this theory on 4 observations: 1) sutures were often patent at surgery, even when there was a high degree of preoperative suspicion of suture fusion; 2) there were characteristic abnormalities at the cranial base that occurred with certain suture fusion patterns; 3) excision of the fused suture did not always improve the cranial shape; and 4) embryologically, skull development occurred after cranial base development.

Moss' theory fell out of favor as surgical treatment in humans directed at the prematurely fused sutures demonstrated reversal of the deformity. Unfortunately, some surgeons attempted complex cranial base expansion based on these theories. Moss's theory was later definitively disproven when it was shown that the suture itself was the primary site of abnormality in craniosynostosis, that cranial base and facial abnormalities responded to opportunities for suturing,[44,46] and that abnormalities in the cranial base could resolve completely with suture release.[36] However, a lasting contribution of Moss' work was his recognition that the active growth of the underlying brain dictated the passive cranial growth along the suture lines. He termed this the "functional matrix theory" and it would later form part of the justification for the minimally invasive approach early in life.

Early Attempts at Surgical Intervention

By the late 1800s, the understanding of sutures and the consequences of premature fusion was growing, but surgical intervention was not attempted until it was reliably recognized that craniosynostosis could lead to impaired neurological and cognitive growth, blindness, and hydrocephalus.[18] The first reported surgical interventions for craniosynostosis were strip craniectomies, first by Lannelongue in Paris in 1890[33] followed shortly by Lane in San Francisco in 1892.[31] Lannelongue performed bilateral strip craniectomies for sagittal synostosis and strongly advocated for release, not resection, of the fused suture. Lane reports being approached by the mother of a child who pleaded to him: "Can you not unlock my poor child's brain and let it grow?" Lane performed a strip craniectomy with removal of a stenosed sagittal suture along with lateral strip of parietal bone bilaterally (Fig. 2). However, the patient died 14 hours postoperatively, reportedly from complications of anesthesia. Despite these isolated reports with limited outcome data, it appears that this technique was quickly adopted and used for the treatment of craniosynostosis. An atlas with figures demonstrating a variety of craniectomies for craniosynostosis was published just 5 years after Lannelongue's first report,[11] along



PIONEER CRANIECTOMY FOR RELIEF OF MENTAL IMBECILITY DUE TO PREMATURE SUTURAL CLOSURE AND MICROCEPHALUS.

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Early in the month of August, 1888, I received a letter from a lady residing in the interior of California, stating that she desired to consult me concerning her infant, then nearly 9 months of age, which presented signs of mental imbecility. At the time appointed for the consultation, the lady presented herself with her infant. The child, otherwise in good health and well nourished, was decidedly microcephalic. The cranium was symmetrical, and only deviated from normal type in the smallness of its volume. The mother stated that at birth the anterior fontanelle was wholly closed, and the posterior one nearly so.

Fig. 2. Lane's early report of surgical repair for presumed craniosynostosis published in the *Journal of the American Medical Association.* Reproduced with permission from Lane LC: **JAMA XVIII:**49–50, 1892.

with many surgical texts illustrating techniques for treatment of fused sutures.[32,34]

However, the outcomes of these early interventions were limited by 2 major challenges. First, it later became clear that many of the children operated on at this time were more likely to have microcephaly, rather than craniosynostosis, a distinction that was either not diagnosed or considered at that time. Second, in patients with true craniosynostosis, these procedures were performed late in the course of the disease after neurological deficits developed. Their outcomes were therefore accompanied by significant reossification, and only served to temporize the constriction for a short period of time. At this time, Jacobi,[23] the father of American pediatrics in this era, reviewed a series of 33 children treated for presumed craniosynostosis, and found alarming results of surgery with a high mortality rate (15 of 33 children dying), and publicly denounced the practice to an audience at a meeting of the American Academy of Pediatrics, marking the end of surgery for nearly 3 decades. He famously said:

> The relative impunity of operative interference accomplished by modern asepsis and antisepsis has developed an undue tendency to, and rashness in, handling the knife. The hands take too frequently the place of brains…Is it sufficient glory to don a white apron and swing a carbonized knife, and is therein a sufficient indication to let daylight into a deformed cranium and on top of the hopelessly defective brain, and to proclaim a success because the victim consented not to die of the assault? Such rash feats of indiscriminate surgery…are stains on your hands and sins on your soul. No ocean of soap and water will clean those hands, no power of corrosive sublimate will disinfect the souls.

Revival of Surgical Treatment

Surgical intervention for craniosynostosis was revived decades later when Mehner[39] reported on the first successful craniectomy for complete removal of a fused suture. A few years later, Faber and Towne[15]—now presumably with the capability to accurately differentiate microcephaly from craniosynostosis—also reported excellent preservation of neurological function with minimal morbidity and mortality. Additionally, based on their observations of outcomes, they pioneered the concept of early and prophylactic linear synostectomy for preservation of neurological function and improvement of cosmesis, commenting "it is probable that the evil effects of synostoses are largely preventable by a suitable operation performed sufficiently early in life."[16] By the 1940s, strip craniectomies and suturectomies were once again widely accepted and the critical importance of early intervention—which they describe as the period before 2 months of age—leading to better functional and cosmetic outcomes was beginning to be appreciated. However, despite advances in surgical management in this era, a new challenge in the management of these children became apparent. In older children, reossification with rapid bridging of the artificial suture was a commonly observed complication, which often required multiple extensive cranial vault remodeling procedures with limited efficacy.[25] The outcomes in these complex patients with mature and delayed fusion led Harvey Cushing to question the indication of late linear craniectomies in these patients[8] and presented surgeons of the next era with major challenges to solve.

Advances at Children's Hospital Boston

Once an effective approach for early craniosynostosis was in widespread use, much of the research focus, which was primarily taking place at Children's Hospital Boston, shifted to addressing the limitations of surgical intervention for children who presented late in the disease course or in children who had experienced reossification at the synostectomy site. The primary concern was that surgery at suture sites that have recurrent fusion required extensive and difficult secondary cranial reconstruction operations that were technically challenging and associated with high morbidity and mortality rates.

In one of the first attempts to minimize reossification, Donald Matson and Frank Ingraham[22] proposed the use of a polyethylene film at the edges of cut bone following strip craniectomy. One year prior to Matson and Ingraham's report of polyethylene film, Simmons and Peyton[49] reported on the use of tantalum foil between the newly cut bone, but both techniques fell out of favor due to reports of infection and reossification. However, these techniques became widespread as more effective modern surgical options became available. Interestingly, Anderson and Johnson[2] developed a technique in 1956 in which Zenker's solution was applied directly onto the dura. This served to cauterize the ossifying elements within the meninges and increased the incidence of suture patency, but was found to cause seizures.[35] Matson and Ingraham's simple craniosynostectomy became widely popular and replaced strip craniectomies as the treatment of choice in most pediatric neurosurgery texts and was one of the most common approaches of this era.[37,42] Their technique consisted of removing a 1-cm strip of bone at the site of the involved suture, extending the craniectomy across the adjacent normal sutures, and excising the pericranium to prevent reossification. The importance of restoring natural skull shape early to allow a proper rate of expansion for normal brain development was again recognized in this era, particularly by Shillito and Matson.[48]

By the mid-1950s, there had been significant advances in anesthesia, blood transfusion, and surgical technique at high-volume centers such as Children's Hospital Boston, and surgery for craniosynostosis became very safe. In one of their largest series, Shillito and Matson reported only 2 deaths in 394 operations, a stark contrast to the results reviewed by Jacobi just decades prior.[63] This allowed the consideration of cosmesis (which Shillito and Matson strongly argued for) as a primary indication for surgical intervention,[48] inviting Paul Tessier and other craniofacial surgeons to pioneer techniques with more focus on cosmesis and the correction of associated facial anomalies.[52–54] Although simple craniosynostectomy and strip craniectomy produced excellent results in very young infants, these were inadequate approaches for older children with advanced disease, and the pioneers of the next era were challenged to develop procedures to treat their disease.

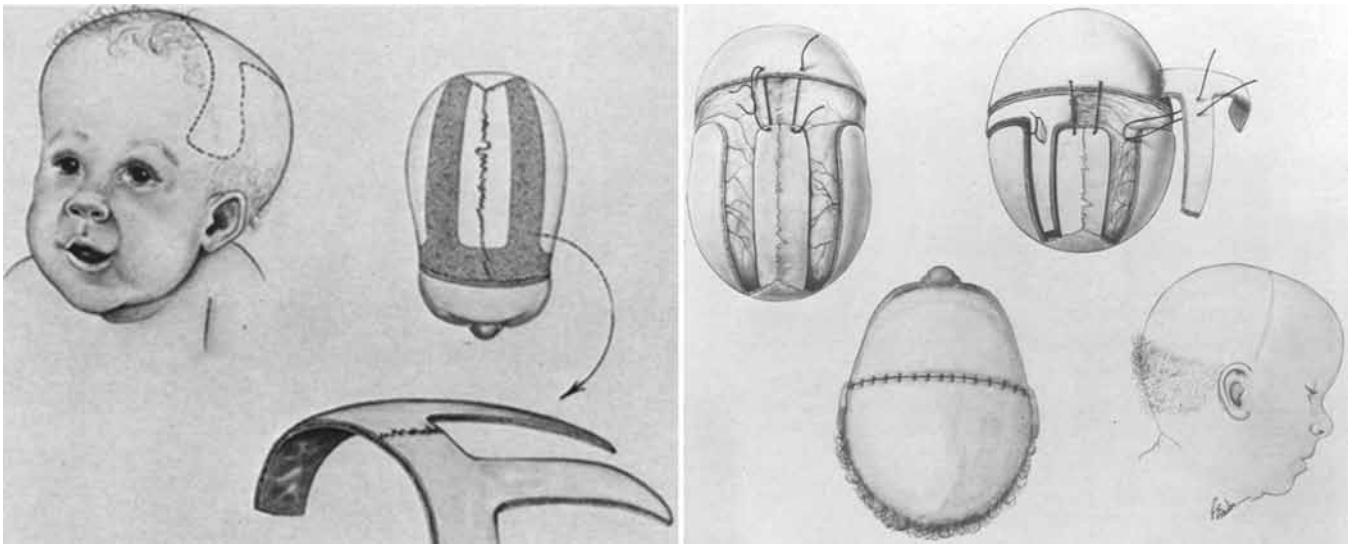

Fig. 3. Illustrations of the pi procedure developed by Jane and colleagues. **A:** Schematic of site and shape of bone removed. **B:** Schematic depicting the technique to replace bone flap to reduce anterior-posterior diameter. From Jane JA et al: **J Neurosurg 49:**705–710, 1978.

Extensive Calvarial Remodeling

The early 1960s to mid-1990s marked an era in which the limitations of simple suturectomies and strip craniectomies for advanced late disease were recognized, challenging surgeons to develop novel procedures for complex calvarial vault remodeling. The innovation of these procedures was driven by the need for immediate deformity correction to prevent impending neurological dysfunction in nonneonates, as well as the need to treat the secondary compensatory changes at sites away from the diseased suture that had taken place. Also fueling this movement was the discovery by Jane et al. that the major cause of the global cranial deformity was compensatory overgrowth at adjacent sutures.[9] Some of the most popular procedures included wide-strip craniectomy with bilateral wedge parietal craniectomy,[1] sagittal craniectomy with biparietal morcellation,[19] extended vertex craniectomy, midline craniectomy with occiput resection,[57] and complete calvarial remodeling via the pi procedure for advanced sagittal synostosis and orbitofrontal advancement for metopic, unicoronal, or bicoronal synostosis. For the most common form of craniosynostosis—isolated sagittal synostosis—Jane and colleagues[5,24] developed the pi procedure, named after the shape of bone that is removed (Fig. 3). In this technique, the sagittal, bilateral coronal, and lambdoid sutures are first removed and the parietal bones are outfractured to increase the skull width. The sagittal suture is then removed and used as a strut to maintain the outward position of the parietal bones. Finally, the frontal and occipital bones are secured to the parietal bones with adjustments of anterior-posterior dimension and frontal bossing. The pi procedure and its variations and modifications have the advantages of addressing the primary suture fusion and also the global cranial deformity, including anterior-posterior dimension and frontal bossing, and providing immediate correction without the need for a postoperative helmet.

In 1982, Epstein et al.[14] described total vertex craniectomy, a hybrid of the midline craniectomy procedure of Stein and Schut[51] and the occipital and coronal prominence excision of Venes and Sayers.[57] Epstein et al. further modified this approach by extending the width of bone removed to 6–8 cm, which eliminated the need for interpositional Silastic, a common technique of the time. They reported excellent cosmetic and functional outcome without any morbidity and deaths.[14] Summarizing the experience of this era, McComb and colleagues at Children's Hospital Los Angeles published one of the most significant articles on contemporary surgical management in the journal *Pediatrics*, describing their institutional experience with 250 patients over 6 years.[50] Of the many important findings, they describe the prevalence of fused suture patterns, the morbidity and mortality of extensive calvarial remodeling, and describe a novel 7-category outcome classification system for more scientific analysis of outcomes. Additional contributions during this era from Children's Hospital Los Angeles include McComb's approach for management of sagittal synostosis in the older infant, in which a reduced rate of brain growth provides an insufficient force for passive cranial vault expansion. In this technique, termed "occipital reduction–biparietal widening," the occipital protuberance is reduced, the biparietal diameter widened, and the height of the vertex is lowered (Fig. 4). Employing this technique in a large series of infants > 6 months of age, McComb[38] reported excellent cosmetic outcomes, no bone defects, and no need for reoperation.

As the efficacy of these procedures became apparent, they gained widespread acceptance as the preferred method of treatment for nonneonates, despite the perioperative limitations. Another significant advance during this era was the contributions of Paul Tessier, who is widely regarded as the father of craniofacial surgery. He developed the principles that define modern craniofacial surgery, as well as sophisticated techniques and tools that

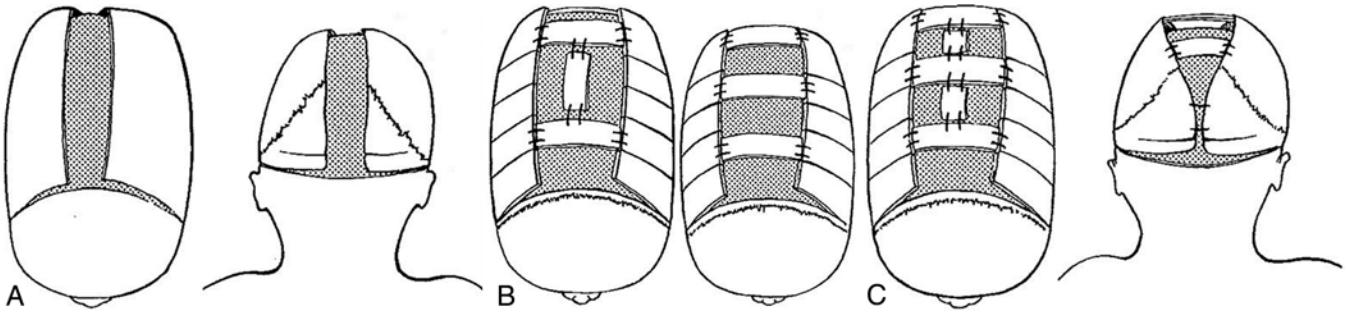

Fig. 4. Schematic illustrations of McComb's occipital reduction–biparietal widening technique for sagittal synostosis. **A:** Posterior and vertex view of midline bone strip removed and cuts parallel to the skull base in the occipital bone. **B and C:** Vertex **(B)** and posterior **(C)** views of methods to graft the midline bone removed. Reproduced with permission from McComb JG: **Pediatr Neurosurg 20:**99–106, 1994, S. Karger AG, Basel.

led to significant improvements in cosmetic outcomes, particularly for those with facial abnormalities.[52–54] The advances of extensive calvarial remodeling allowed for normal neurological development and excellent cosmetic results, particularly for those with the most complex of multiple-suture disease.[60] However, these advances were associated with significant operative time, hospital stay, blood loss requiring transfusion, and complications that were well described even in modern literature of the 1990s.[17,30,56] These limitations became the impetus for the most recent endoscopic advances.

Modern Endoscopic Strip Craniectomy

In the early 1990s, Jimenez, a pediatric neurosurgeon, and Barone, a plastic surgeon, recognized the limitation of the approaches of the past quarter century, including extensive operations in young children, prolonged operative time, blood loss and need for blood transfusion, significant scalp mobilization, and need for subsequent reconstructive procedures. They proposed a novel technique: simple suturectomy via an endoscopic approach (Fig. 5). The success of this approach can be attributed to

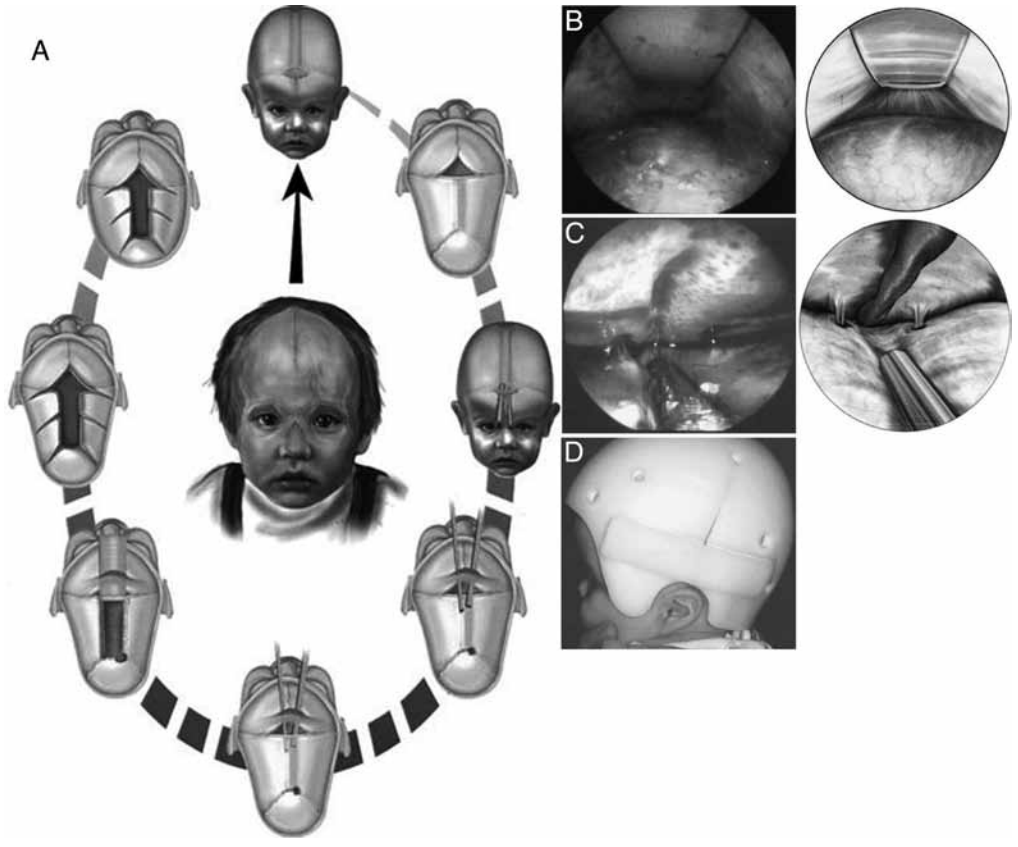

Fig. 5. Images from the work of Jimenez and Barone on endoscopic craniectomy for early surgical correction of sagittal craniosynostosis. **A:** Schematic of sequential steps in endoscopic strip craniectomy. **B:** Endoscopic *(left)* and schematic *(right)* views of the subgaleal space. The scalp is retracted superiorly away from the cranium. **C:** Endoscopic *(left)* and schematic *(right)* views of the stenosed sagittal suture. **D:** Photograph of a postoperative molding helmet that partially restricts anteroposterior growth and allows bitemporal and biparietal expansion. From Jimenez DF and Barone CM: **J Neurosurg 88:**77–81, 1998.

Jimenez and Barone's consideration of 3 basic principles of craniosynostosis. First, as recognized by Farber and Towne, they recommended surgery early in life. Second, as described by Moss's functional matrix theory, they recognized that if timely intervention occurred, the rapidly growing brain would cause expansion of the skull into a normal shape. Third, to counteract the tendency of the cranial vault to revert to a premarid shape as described by Otto and Virchow, they employed an adjunct vault remodeling helmet introduced by Persing et al. in 1986,[45] into which the brain would shape the skull. They first presented this work in a small series of 4 patients, all with sagittal synostosis, treated with early endoscopic strip craniectomy with adjunct postoperative cranial molding helmets. They were able to demonstrate minimal blood loss, short operative times, early hospital discharge, and excellent functional and cosmetic results, although with limited follow-up.[26] Subsequent studies with progressively larger patient samples and a wide variety of fused suture patterns with long-term follow-up confirmed the efficacy and safety of this approach.[27–29]

The collective outcomes from these studies show remarkable results with short operative times, minimal blood loss, early hospital discharge, and minimal operative and perioperative risks, including extremely rare cases of infection, dural sinus tears, CSF leaks, or neurological injury. A collective review of their technique from the past 11 years in infants with a variety of multiple-suture nonsyndromic craniosynostoses demonstrates results superior to those achieved with more invasive procedures of the prior era.[28] This most recent advancement, founded upon the principles of the natural history and pathophysiology of craniosynostosis, has led to dramatically improved outcomes and has fundamentally changed the treatment of these patients.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Bettegowda, Mehta, Ahn. Acquisition of data: Bettegowda, Mehta, Ahn. Analysis and interpretation of data: Bettegowda, Mehta, Ahn. Drafting the article: all authors. Critically revising the article: Bettegowda, Jallo, Ahn. Reviewed final version of the manuscript and approved it for submission: Bettegowda, Jallo, Ahn.

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