2 History of Craniosynostosis Treatment

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The history of modern craniosynostosis surgery can be understood through the evolution of cranial and maxillofacial surgery. Cranial surgery can be traced back to neolithic period, whereas maxillofacial surgery developed much later. Both surgeries developed into different surgical specialties: neurosurgery and maxillofacial/plastic surgery. The skull base as the separating structure between the cranium and the face was surgically touched by both disciplines. The wall separating the face and the cranium was broken by Paul Tessier and Gérard Guiot [1] in the 1960s, making it possible to perform a combined operation around the orbits and forehead and opening up close cooperation between maxillofacial/plastic surgeons and neurosurgeons, especially for the treatment of major craniofacial malformations. The distraction of craniofacial bone and the subsequent gain in soft tissue structures (introduced by Ilizarov for the limbs and applied to the face at the mandibular level by McCarthy [2]) have overcome one of the problems of skull shaping: the retraction of soft tissues with the risk of relapse of facial retrusion. Computer-aided planning and execution of craniosynostosis surgeries are now on the way to refine surgical results. The way from the beginning of skull surgery to modern craniofacial procedures gives insight into milestone developments in this surgical specialty.

History of Craniotomy and Cranioplasty

Craniotomy in Ancient Times

The evolution of craniotomy and cranioplasty can be traced back over thousands of years. Craniotomy procedures and also cranioplasty procedures were done out of cultural, religious, or medical reasons. The surgical procedure of a craniotomy has been practiced longer than any other and certainly that for which we have, by far, the oldest tangible evidence. The first findings related to perforation of the skull date back to the neolithic period (8000–5000 BC) and were found in France already in 1685 [3]. According to the most popular theory, the very first cases of craniotomy were probably performed, by prehistoric man, for reasons related to magic or religious rituals (Fig. 2.1), as an initiation practice, or as part of a ritual related to exorcism. As proof of the great religious importance attached to those who had been subjected to drilling, it is worthwhile recalling that, from some of these skulls, diskettes of bony tissue were removed postmortem (Fig. 2.2), which

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Neolithic trephining. Making a hole in the skull to let the devils out.

Fig. 2.1 Illustration of neolithic trephination making a hole in the skull to let the devils out (Behind the doctor/ Logan Clendening from https://wellcomecollection.org/images?query=ueceazt9, licensed under CC-BY-4.0)

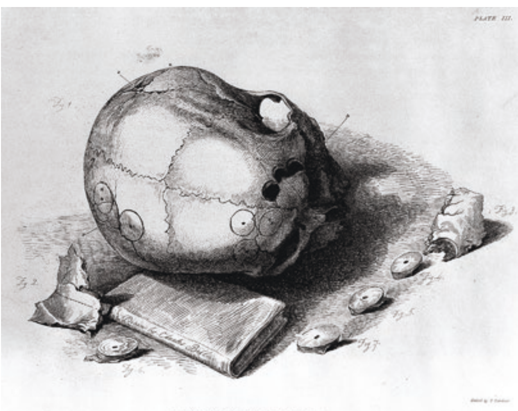

Fig. 2.2 Human skull with sections removed (Illustrations of the great operations of surgery, trepan, hernia, amputation, aneurism, and lithotomy/[Sir Charles Bell], from https://wellcomecollection.org/works/nx7xdmf5/images?id=xxd8rtdk, Public Domain)

were then worn as amulets around the neck (the so-called rondelles, described for the first time, by Prunières, in 1783) [4, 5].

Cranioplasty in Ancient Times

It is not definitively solved when and why cranioplasty operations were done instead of craniotomy operations. The earliest cranioplasty operation is dated to 3000 BC in the Inca civilization, where precious metals, gourds, and shells were found next to trepanned skulls in graveyards, suggesting that cranioplasty had been performed [6]. In the Paracas region of present-day Peru, a skull from 2000 BC with a thin plate of gold covering a cranial defect was found [7]. Moreover, defective skulls were found covered with coconut shells or palm leaves in ancient tribes of the Polynesian Islands [8]. Sanan and Haines stated that the materials being used for cranioplasty were associated with the status of the patient [9]. Research and practice on trephination were documented in ancient Greece and Rome, but cranioplasty procedures are not emphasized among early surgical authors in ancient Asia, Egypt, Greece, and Rome.

Evolution of Medically Driven Craniotomies

Indications and surgical approaches changed during the centuries. Whereas early craniotomy procedures were based on cultural or religious reasons, opening of the skull was later used for treatment purposes: the finding that this procedure could, thanks to encephalic decompression, lead to an improvement in certain preexisting neurological symptoms probably led to it being employed in the presence of these symptoms and, in particular, in traumatic lesions. The techniques used for such surgeries evolved over time (Fig. 2.3). Even the medicine men of neolithic times had an incredible technical ability in performing a craniotomy procedure despite the fact that they only had primitive tools [10, 11]. The most ancient technique of craniotomy consisted of thinning down the bony wall with abrasive instruments; later, circular incisions were progressively made deeper, or a series of small holes were made in a circle, after which the bony bridges between them were broken down. The Egyptian physician Imhotep performed craniotomy, presumably related to head injuries that would have resulted from the numerous battles fought by the ancient Egyptians. The Edwin Smith Surgical Papyrus,

Fig. 2.3 Edinburgh skull, showing trepanning hole in the back of the skull (Edinburgh Skull, trepanning showing hole in back of skull from https://wellcomecollection.org/works/hk22bxbp, licensed under CC-BY-4.0)

which provided a scientific approach to craniotomy, dates between the sixteenth and seventeenth centuries BCE but is believed to have been originally written by Imhotep around 2900 BCE [12]. Much later were metallic instruments used, made of copper or bronze, such as gouges, curettes, scalpels, and knives of various forms, some of which very special, such as the "tumi," or scalpel, in ancient Peru [13]. The ancient Greek civilization saw a further understanding of pathologies of the central nervous system. By the fifth century BCE, Hippocrates codified guidelines on the use of craniotomy for the treatment of intracranial pathology [14]. When craniotomy was performed, the crown drill ("trupanon") and perforating drill were employed, instruments that Hippocrates does not describe but only mentions, as if it was in common use at that time. Hippocrates advised performing the craniotomy without delay, in fact within the first 3 days of the trauma, in the case of severe contusions or of simple fractures, whereas in the case of the comminute type or with embedded fragments, he suggested that they be removed, paying particular attention to preserve the meninx.

During the Middle Ages, the Arabic surgeon Abul-Qasim Al Zahrawi, known in Western literature as Abulcasis, wrote extensively on early depictions of neurosurgical diagnosis and treatments, including the treatment of head injuries, skull fractures, hydrocephalus, and subdural collections [15].

Following a long period of decline, surgery involving craniotomy began to be performed again on a vast scale during the Renaissance period due to the widespread use of firearms, which greatly increased the incidence of fractures and trauma involving the skull. Moreover, it should also be pointed out that in the second half of the seventeenth century, studies performed by Vieussens, Malpighi, and Willis led to a better understanding of the neurophysiological aspects and, in particular, stressed the importance of the cerebral cortex, which had not been clearly understood until that time, inasmuch as the humoral theory took into consideration only the ventricles as essential structures of the brain [16–18]. From the end of the eighteenth century onwards, the use of craniotomy gradually decreased, mainly on account of the increase in the incidence of complications due to infections. Infections in hospital surroundings and suppuration of wounds had become so frequent.

Cranioplasty in Modern Times

The earliest modern description of cranioplasty was written by the surgeon Ibrahim bin Abdullah of the Ottoman Empire, in his surgical book Alâim-i Cerrâhîn in 1505. The book mentioned the use of xenografts from Kangal dogs or goats as materials for cranioplasty. Such materials were used due to the accessibility of these animals near battlefields, where the procedure is likely to be performed [19]. The first true description of cranioplasty in Europe was made by Fallopius in the sixteenth century (Fig. 2.4), stating that the fractured cranium should be removed and be reinserted with a gold plate if the dura was damaged. This was questioned by other practitioners at his time, concerning that surgeons may keep the gold instead of using it for surgery. The first cranioplasty was reported by the Dutch surgeon Job Janszoon van Meekeren. The report described the use of a segment of a canine cranium as a mate-

Fig. 2.4 Portrait of Gabriele Fallopius, 1523–1562 (Profili bio-bibliografici di medici e naturalisti celebri italiani dal sec. XVo al sec. XVIIIo/[P. Capparoni] from https://wellcomecollection.org/images?query=v3f8dc3a, licensed under CC-BY-4.0)

rial for cranioplasty on a nobleman in Moscow. The operation was successful; however, the use of canine bone in the operation was not accepted by the church and the man was forced to leave Russia [20]. Since the first operation, bones from more animal species were used as xenografts for cranioplasty. These include dogs, apes, geese, rabbits, calves, eagles, oxen, and buffalos. In 1917, William Wayne Babcock reported the use of "soup bone," a piece of cooked and perforated animal bone as a xenograft [21, 22]. Autografts, allografts, and synthetic materials are the main types of materials used for cranioplasty.

During the second half of the nineteenth century, after the advent of antisepsis and general anesthesia, the use and technique of cranioplasties evolved (Fig. 2.5). By the twentieth century, neurosurgery became an autonomous discipline, and the modern era began [23, 24]. At first, neurosurgical approaches were performed with

Fig. 2.5 Drawing of skull trephination in the operation theatre (Le chirurgie françoise recueillie des antiens médecins et chirurgiens. Avec plusieurs figures des instrumens necesseres pour l'opération manuelle/Par Iacques Guillemeau, from https://wellcomecollection.org/works/x7j3tpvn/images?id=anyvs5a6, Public Domain)

extended craniotomies. The development of neurosurgical techniques and approaches was greatly aided by the evolution of advanced diagnostic imaging. The ability to combine high-definition computed tomography images (developed in the 1970s) with magnetic resonance imaging (MRI) (developed in the 1980s) has improved pre-procedural planning.

Detection of Craniosynostosis Biology

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 [25]. The recognition of cranial vault deformities by the ancient physician Galen, and some early understanding of the role of cranial sutures by Hippocrates, has also been reported [26, 27].

By the sixteenth century, it appears that anatomists appreciated the existence of cranial sutures (Fig. 2.6) and had documented a broad range of characteristics of the deformity, across an appre-

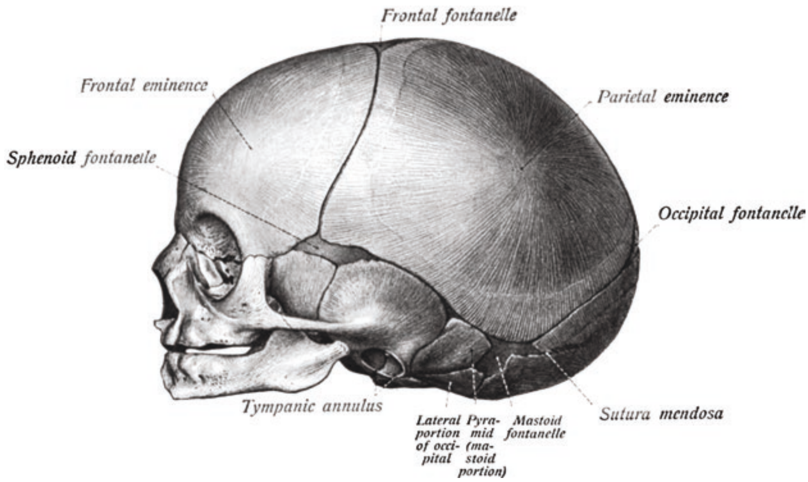

Fig. 2.6 Sutures of a child's head. (Sobotta's Atlas and Text-book of Human Anatomy 1909)

ciation of suture pattern and premature suture fusion in a variety of configurations by Hundt [28], specific abnormal varieties of sagittal and coronal sutures by Dryander [29], and what would now be described as oxycephaly and brachycephaly by della Croce [30] and Vesalius [31]. However, von Sömmerring [32] in the late 1790s was the first to go beyond simple descriptions and apply scientific principles to the study of abnormal cranial suture growth.

In 1851, Virchow [33] 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 (Fig. 2.7). 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 non-fused sutures in a direction parallel to the affected suture, causing obstruction of normal brain growth [34].

By the early 1900s, craniosynostosis was recognized as one component of complex syndromic deformities, most notably by Apert [35] in 1906 and Crouzon [36] in 1912, whose names bare two of the most well-known syndromic deformities. Moss stated that the active growth of the underlying brain dictated the passive cranial growth along the suture lines [37]. He termed this the "functional matrix theory," and it would later form part of the justification for the minimally invasive approach early in life.

Fig. 2.7 Virchow, observing a skull operation at a Paris clinic (from https://wellcomecollection.org/works/pnc-n9yjq, licensed under CC-BY-4.0)

History of Craniosynostosis Surgery

Craniectomies and Suturectomies

Patients with craniosynostosis were not properly treated until the twentieth century, so that disfigured persons lived their lives under special circumstances (Fig. 2.8). The first reported surgical interventions for craniosynostosis were strip craniectomies, first by Lannelongue in Paris in 1890 [38] followed shortly by Lane in San Francisco in 1892 [39]. Lannelongue performed bilateral strip craniectomies for sagittal synostosis and strongly advocated for release, not resection, of the fused suture. Lane performed a strip craniectomy with

Fig. 2.8 Girl aged 17 years with marked proptosis (St. Bartholomew's Hospital Archives & Museum, from https://wellcomecollection.org/works/d6nnjbay, licensed under CC BY 4.0)

removal of a stenosed sagittal suture along with lateral strip of parietal bone bilaterally. An atlas with figures demonstrating a variety of craniectomies for craniosynostosis was published just 5 years after Lannelongue's first report, along with many surgical texts illustrating techniques for treatment of fused sutures. Surgical intervention for craniosynostosis was revived decades later when Mehner [40] reported on the first successful craniectomy for complete removal of a fused suture. A few years later, Faber and Towne [41]—now presumably with the capability to accurately differentiate microcephaly from craniosynostosis—also reported excellent preservation of neurological function with minimal morbidity and mortality. 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. In one of the first attempts to minimize reossification, Donald Matson and Frank Ingraham [42] proposed the use of a polyethylene film at the edges of cut bone following strip craniectomy.

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. 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. Some of the most popular procedures included wide-strip craniectomy with bilateral wedge parietal craniectomy, sagittal craniectomy with biparietal morcellation [43], extended vertex craniectomy, midline craniectomy with occiput resection [44], and complete calvarial remodeling via the pi procedure for advanced sagittal synostosis and orbitofrontal advancement for metopic, unicoronal, or bicoronal synostosis.

History of Craniofacial Surgery

The modern era of craniofacial surgery started in the 1960s with Tessier, who first established multidisciplinary craniofacial teams in Paris [45]. In 1967, he showed a procedure of fronto-orbital advancement with cranial vault remodeling (Fig. 2.9), with reshaped removal bone pieces stabilizing back to the cranium, and established new protocols that followed and consisted of Moss's functional matrix theory in 1959 and the concept of compensatory cranial vault growth by Vollmer and Delashaw [46, 47]. The principles of mobilization of the orbits to correct hypertelorism or orbital dystopia are recalled with reference to the different variations and with clinical exam-

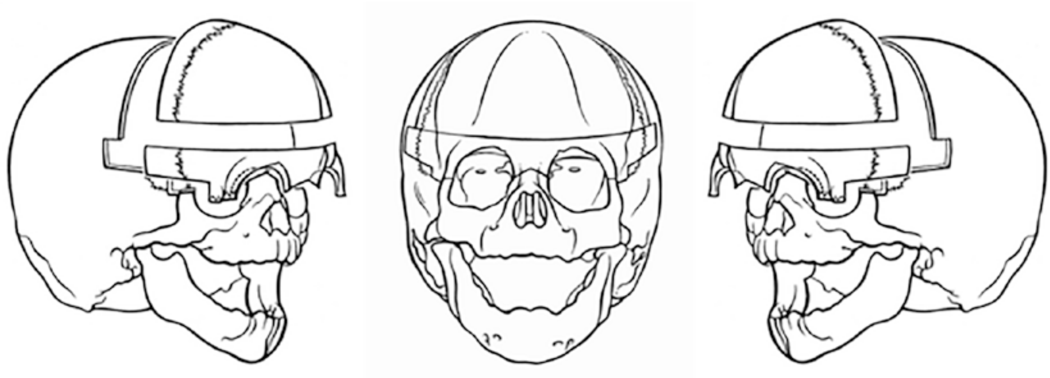

Fig. 2.9 Schematic drawing of a fronto-orbital advancement procedure (Xxjamesxx, Location of the incisions in fronto-supra-orbital advancement, from https://commons.wikimedia.org/wiki/File:Incision_locations_advancement.jpg, licensed under CC BY-SA 3.0)

ples. The wall separating the face and the cranium was broken by Paul Tessier and Gérard Guiot in the 1960s [1], making it possible to perform a combined operation around the orbits and forehead and opening up close cooperation between maxillofacial/plastic surgeons and neurosurgeons, especially for the treatment of major craniofacial malformations. Facial advancement to correct the retrusions created by faciocraniosynostosis is explained with the many possible variants, combined with an intracranial approach or not, with or without a bipartition. The indications are discussed as is the risk linked to combined advancement of face and forehead. In 1978, Marchac reported a frontal advancement procedure [48] and followed and established the method as a common treatment for the craniosynostosis in all over the world. After Tessier, various craniofacial surgeons developed an extensive and more whole cranial construction approach [49–52]. Plastic surgeons had started this work in association with neurosurgeons during this period, when the International Society of Craniofacial Surgery was founded by Tessier and his disciples in 1983.

Craniofacial Distraction

In 1904/1905, the Italian Codivilla already reported about the possibility of lengthening the lower limb by continuous traction [53]. However, the first successful callus distraction of a human femur after bilateral diaphyseal fractures was performed in 1923 by August Bier in Berlin [54]. Within the field of maxillofacial surgery, distraction approaches were already described between 1920 and 1930. In 1926, Wassmund [55] reported about the possibility of closing an open-bite situation by applying elastic traction to the upper jaw after its subtotal surgical mobilization. Almost at the same time, Rosenthal [56] in Leipzig managed to reconstruct the lower face of a female patient affected by mandibular hypoplasia by applying a tooth-borne expansion device to the anterior lower jaw after bony separation. The progressive bone elongation principle introduced by Ilizarov for the limbs has been applied to the face with an external distractor at the mandibular level by McCarthy, with great success [2]. The distraction of bone structures is now also applied at the level of the whole skull and makes it possible to overcome the retraction of soft tissues and lower the risk of relapse of facial retrusion. Many applications of the distraction principle have been developed for the craniofacial, midface, and mandible levels. The surrounding structures, including the developing tooth germs, must be taken into consideration when planning the osteotomy cut. The process of DO in the craniofacial region consists of both linear and rotational movements as opposed to only linear movements in the case of epiphyseal lengthening. This is because of the morphology of the structures present in the head and neck region. The vector produced by the distraction device is based on its position in relation to the surrounding bony structures. The expansion of the fronto-orbital skeleton by distraction was able to address the anterior cranial volume as well as the retruded orbital bandeau [57–61]. However, the degree of cranial volume expansion is limited by globe-to-orbit proportion. The introduction of distraction to expand the posterior calvarium [62] addressed many of these shortcomings. It permitted the scalp to be closed without tension and facilitated a controlled expansion. In addition, it obviated the need for secondary bone grafting of the residual bony defect. Dural injury, device failure and loosening, infection, and wound dehiscence were all reported in the initial study by White et al. in 2009 [62]. White's report of the posterior calvarial expansion has been rapidly accepted by other surgeons. The method is thought to be a good indication for the syndromic craniosynostosis, because the amount of cranial expansion is much more effective than frontal distraction advancement or conventional procedure [63, 64].

Endoscopic Suturectomy

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 [65]. They proposed a novel technique: simple suturectomy via an endoscopic approach. The success of this approach can be attributed to Jimenez and Barone's consideration of three 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 pre-marid shape as described by Otto and Virchow [66], they employed an adjunct vault remodeling helmet introduced by Persing et al. in 1986 [58], into which the brain would shape the skull. Endoscopic strip craniectomy followed by orthotic helmeting has since then been shown to be a successful treatment option for single-suture craniosynostosis [67–71]. This procedure is associated with significantly lower blood loss, fewer transfusions, shorter operative time, decreased length of stay, and fewer ICU admissions [72]. However, patients are required to wear the orthotic helmet for 23 h per day, until approximately 1 year of age. This requires frequent follow-up with a trained cranial orthotist. Jimenez and Barone have reported on using this technique to treat bilateral coronal craniosynostosis [73]. However, to date, there has been no direct comparison between the FOA procedure and the endoscopic strip craniectomy followed by orthotic therapy for the treatment of bilateral coronal craniosynostosis.

Computer-Assisted Craniofacial Surgery

The advent of computer-assisted technology has revolutionized planning for complex craniofacial operations, including craniosynostosis surgery. Recent advances in the field of three-dimensional (3D) imaging using computed tomography (CT) or cone-beam computed tomography (CBCT) have led to the development of computer-assisted craniofacial surgery, in which detailed presentation of the craniofacial complex and enhanced analysis of surgical planning lead to improved predictability of surgical outcomes. The application of computer-aided design and computer-aided manufacturing (CAD/CAM fabrication of surgical guides and osteosynthesis plates) has rapidly developed and spread widely from research to routine clinical medicine. Craniofacial reconstruction is ideally suited for virtual planning and execution, as it allows the surgeon to assess the complex three-dimensional bony anatomy and

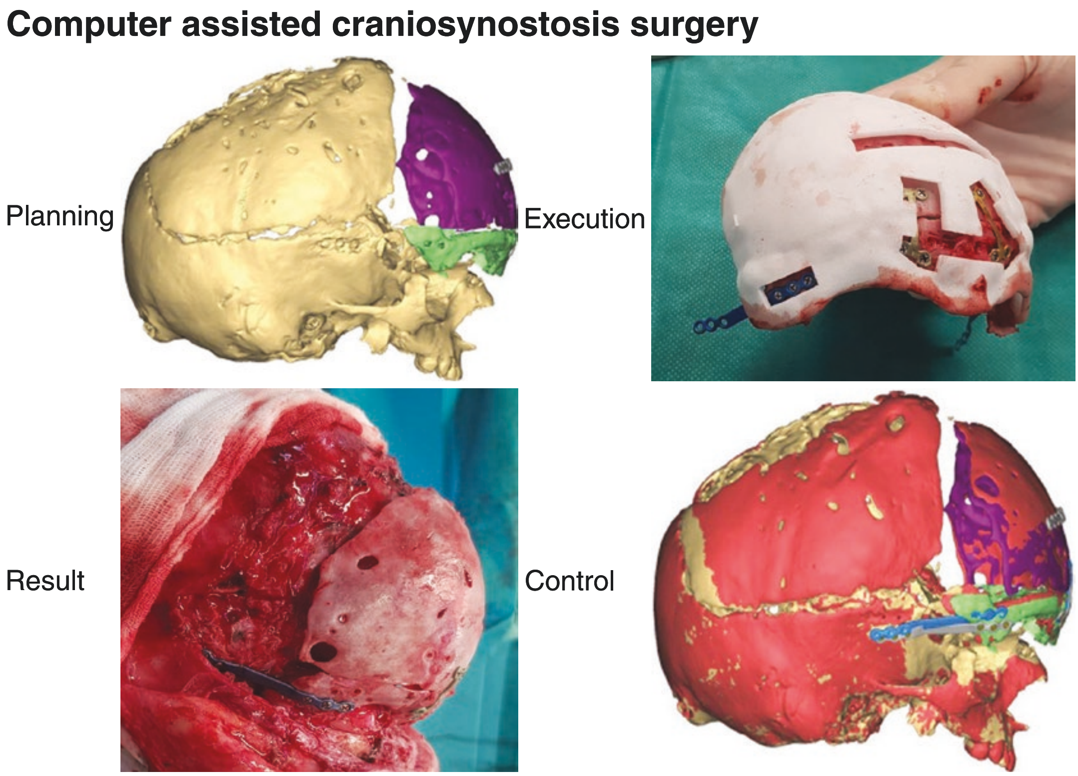

Fig. 2.10 Computer planning and guided surgery in craniosynostosis surgery. (Source: Ulrich Meyer)

critical neurovascular structures within the skull, the skull base, the orbit, and the midface and plan osteotomies, bone movements, and osteosynthesis plate placement with high predictability and accuracy. Additionally, the accuracy of the surgical result can be evaluated by matching of simulation and postoperative datasets (Fig. 2.10).

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