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3D visualization and simulation in surgical planning of orbital hypertelorism

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Abstract--Background: simulation and three-dimensional visualization of object motion is a prerequisite for any surgical planning system. Orbital hypertelorism is a disease, which is most commonly associated with craniofacial malformations. We have developed a surgical planning system for planning and evaluation of orbital hypertelorism surgery. In our system CT-based virtual surface models fitted by oriented bounding boxes (OBB) are manipulated. Three-dimensional motion as well as a correction surgery can be simulated. Both are controlled by collision detection. The computer-based interactive surgery simulation systems (CISSS) presented here can take virtual surgical operation and forecast facial features after the correction of orbital hypertelorism.

Keywords--surgery simulation, orbital hypertelorism, oriented bounding boxes, osteotomy simulation.

Introduction

Orbital hypertelorism (ORH) is defined as an increased distance between the two orbits with true lateral displacement. Anthropometric measurements will yield

increased inner canthal distance (ICD), outer canthal distance (OCD) and interpupillary distance (IPD), while increased inner canthal distance alone is more properly described as telecanthus. It is not a disease in itself rather than a physical finding (R. K. Sharma, 2014). David Greig was the first to say the phrase “ocular hypertelorism” in 1924 to describe a craniofacial deformity associated with “a great breadth between the eyes. Tessier then defined that true hypertelorism represented a congenital divergence of the bony orbits (ie, both medial and lateral walls) and not simply an increased interpupillary distance that could result from various acquired injuries. Van der Meulen and Vaandrager further refined the term to describe a “true lateralization of orbits”. Hence, true orbital hypertelorism represents increased interorbital and outer orbital distances (Shakir, Hoppe, & Taylor, 2019). Van der Meulen et al. distinguish between orbital and inter-orbital hypertelorism. He defined orbital hypertelorism as a true lateralization of the entire orbit and inter-orbital hypertelorism as displacement of the medial orbital wall known as “bony telecanthus. Orbital Hypertelorism may be unilateral or bilateral, symmetric or asymmetric (Shakir et al., 2019).

Incidence

The overall estimated incidence of hypertelorism is rare, with some sources estimating it to be about 1 in 20,000 births (Stevens & Emam, 2012).

Etiology

Hypertelorism is rare. Genetic inheritance is likely sporadic with just a few reported cases that suggest autosomal dominance (Soldanska & Taub, 2019). Orbital hypertelorism is not a syndrome in itself, but it may present in many craniofacial syndromes. It may be present in conditions such as craniofacial dysplasia, encephaloceles and craniosynostosis syndromes (Shakir et al., 2019). The increased inner intercanthal distance (ICD) can also occur following trauma or tumors in the naso-orbital region; however, there is no change in the position of the lateral wall of the orbit. This ‘pseudo-hypertelorism’ is now a days referred to as ‘telecanthus (R. K. Sharma, 2014).

Craniofacial syndromes with ORH **Frontonasal Malformation**

Frontonasal malformations encompass the terms median cleft face syndrome, frontonasal syndrome, and frontonasal dysplasia (FND). These malformations do not represent a single developmental localized defect, but rather present as a heterogeneous category of variable facial appearances. There are no documented reports of familia transmission or gender predominance (Soldanska & Taub, 2019). FND is a rare condition characterized by hypertelorism, nasal abnormalities and associated with variable midline facial defects (S. Sharma, Sharma, & Bothra, 2012). FND includes 2 or more of the following: hypertelorism, broad nasal root, median facial cleft of the upper lip or palate, clefting of the nasal alae, poorly formed nasal tip, cranium bifidum occultum and a widow’s peak hairline. It may be associated with heart anomalies, mental delay, optic disc anomalies, and central nervous system anomalies demonstrating a wide spectrum of the disease (Weathers, Wolfswinkel, Albright, Hollier, & Buchanan, 2013).

Craniofrontonasal Dysplasia

Unlike frontonasal malformations, craniofrontonasal dysplasia (CFND) represents a rare familial craniofacial syndrome that is sex-linked with an unusual pattern of inheritance. There is a strong gender preference favoring female patients who invariably present with more severe phenotypes than their male counterparts. Coronal synostosis (either unilateral or bilateral) represents the primary feature of CFND in addition to a variety of frontonasal deformities including a broad nasal bridge and tip and down-slanting palpebral fissures (Shakir et al., 2019). The orbital hypertelorism found in CFND may be asymmetric. Unique extracranial features include thick “frizzy” hair, shoulder and hip girdle abnormalities and longitudinal ridging of finger or toenails. Additional associated anomalies include cleft lip or palate, high-arched palate, maxillary hypoplasia, strabismus, soft tissue syndactyly of the fingers and toes, and broad thumb or great toe (Figure I). Surgery involves cranosynostosis correction usually achieved by fronto-orbital advancement during infancy and later orbital translocation. Facial bipartition may be used to simultaneously reposition the bony orbit and correct malocclusion (Shakir et al., 2019).

Encephalocele

An encephalocele is defined as a herniation of meninges (meningocele) through a congenital bony defect with or without brain (encephalomeningocele). The internal defect lies at the junction of the frontal and ethmoidal bones, whereas the external or facial defect can vary (Velho et al., 2019). Its classification system is based on characteristics of the external bone defect including type and location:

- Nasofrontal (between nasal and frontal bones).
- Nasoethmoidal (between nasal bones and cartilages).
- Naso-orbital (through medial orbital wall).
- Combined (nasoethmoidal and nasoorbital coexist).
- Abortive (external bone defect unidentifiable).

It should be noted that encephaloceles can occur as a secondary feature in frontonasal malformations, craniofrontonasal dysplasia, and paramedian facial clefting (Shakir et al., 2019).

Tessier Clefts and Hypertelorism

Paul Tessier introduced the Tessier classification system of facial clefts based on his clinical examination and surgical experience in 1976. He first described “clefting” as a process involving “interruption of either soft tissue or skeleton” and used the eyelid and orbit as his frames of reference, as these structures are common to both the cranium and face. He recognized that facial clefts are found around the orbit and eyes and jaws and lips, with the skeletal providing more consistent landmarks compared with the overlying soft tissues. The classification system uses a numbering system (no. 0–14) (Figure II) (Shakir et al., 2019). Embryologic malformation may cause facial clefting and prevent medialization of the orbits during development. Facial clefts can be associated with true hypertelorbitism, described in Tessier's classification as clefting numbers 1-13, 2-

12, and 3-11. The midline widening demonstrated in frontonasal dysplasia corresponds to 0 to 14 clefts (Shakir et al., 2019).

The true expansion of midline or paramedian facial structures as a result of a cleft can result in widening of the interorbital distance. This can be owing to cleft expansion of the median or paramedian bony structures or can be owing to such clefts leading to local encephalocele, which lateralizes the orbits without creating excess bone between them. Depending on the causative cleft, there will be a variable degree of vertical dystopia, enophthalmos, and associated ocular abnormalities. Correction of these anomalies is an integral part of repairing the underlying Tessier clefts, although such procedures carry an additional risk to the orbital structures and optic nerve as the orbits and globes are repositioned (Morris & Tatum, 2016). In correcting hypertelorism, excess bony tissue between the orbits is reduced via paramedian wedge osteotomies to allow corrective rotation of the orbit, and box osteotomies are combined with circular osteotomies to free the orbits individually from the remainder of the caniofacial skeleton. These units are rotated and medialized into the desired position and secured with absorbable plates (Morris & Tatum, 2016).

Craniosynostosis

Craniosynostosis is defined as premature fusion of one or more of the cranial sutures. It leads not only to secondary distortion of skull shape but to various complications including neurologic, ophthalmic and respiratory dysfunction. Ocular hypertelorism, proptosis, beaking of the nose and midface hypoplasia are the common facial features of the craniosynostosis. The epidemiology of craniosynostosis is thought to be approximately 1 of 2,000 live births, though not all craniosynostosis syndromes have hypertelorism. The overall estimated incidence of hypertelorism is rare, with some sources estimating it to be about 1 in 20,000 births (Ko, 2016).

Hypertelorism associated with craniofacial synostosis may be syndromic or nonsyndromic. Of the craniosynostosis syndromes, Apert syndrome (Figure III) is associated with a sunken appearance of the midface, beaked nose, teeth crowding second to the underdeveloped maxilla, vision problems second to shallow eye sockets, mild to moderate intellectual disability, syndactyly or polydactyly, hyperhidrosis, and cleft palate (Begum, Khatun, Rayhan, & Rahman, 2012). Crouzon syndrome can present with midface hypoplasia, hypertelorism, beaked nose, dental abnormalities, hearing loss, and normal intelligence (Bowling & Burstein, 2006).

Classification of Orbital Hypertelorism

Orbital hypertelorism can be classified according to the interorbital distance in adults as in Tessier Classification (Patel & Ghali, 2022).

- 1st degree: 30 – 34 mm ICD
- 2nd degree: 35 – 40 mm ICD
- 3rd degree > 40 mm ICD (47)

In infants, it is classified based on standard deviation (SD) of (IOD) from age and matched data (Patel & Ghali, 2022):

- 1 year: 18.5 mm
- 2 years: 20.5 mm
- 3 years: 21 mm
- 5 years: 22 mm
- 7 years: 23- mm
- 9 years: 25 mm

Clinical Manifestations

Patients with ORH will be presented by widening of the orbital complexes. The frontal hairline may be V-shaped, and patients often have dystopia or eyebrow distortion. Abnormal ocular findings may include strabismus, optic atrophy, blepharophimosis, exotropia, microphthalmos, amblyopia, and corneal abrasions (Hwang, Baek, & Lee, 2012). Many children with hypertelorism lack binocular vision, unless their hypertelorism is mild. Binocular vision is completely formed by age 2; therefore, unless surgery is performed at a very early age, recovery of stereoacuity is rare (Okumoto, Inoue, & Yoshimura, 2012; Wan et al., 2012). In evaluating a child with hypertelorism, therefore, vision, hearing, airway, and cognitive development should all be evaluated. Different measurements have been used to determine the presence and severity of the hypertelorism. These include the interpupillary distance, medial intercanthal distance, lateral interorbital distance, and interdacyon distance (Soldanska & Taub, 2019). Osseous measurements tend to be utilized more often as they differentiate hypertelorism from telecanthus (Yang et al., 2009). Medial intercanthal distance is a useful external landmark especially without available radiographic imaging. It is inapplicable, however, in cases with soft tissue alterations such as abnormal epicanthal folds. Interpupillary distance in hyperteloric patients is seldom useful as there are often differences due to ocular deviation such as strabismus (Soldanska & Taub, 2019).

Diagnosis

The bony interorbital or interdacyon distance is most often used for surgical planning as it is a good bony landmark easily measured on cephalograms or CT scans. The dacryon is the most medial osseous part of the orbit and marks the junction of the lacrimal bone, frontal process of the maxilla, and frontal bone. Assessment should include: interdacyon distance, intercanthal distance and interpupillary distance. Normal variants in adults were measured as 20–26 mm in females and 21–28 mm in males. Increases in interdacyon distance reflect hypertelorism, measured as mild (first degree), 30–34 mm; moderate (second degree), 35–39 mm; and severe (third degree), 40 mm or greater. These measurements vary with age from about 15 mm at birth to about 25 mm in adulthood and are therefore only a guideline in children (Soldanska & Taub, 2019).

Treatment

The corrective surgery is indicated primarily for cosmetic reasons. In the majority of the patients mental and physical development is normal. The patients may have issues with the body image and find it difficult to get integrated into the peer group and the society (R. K. Sharma, 2014). The goals of surgery in hypertelorism are to:

- Bring the two orbits closer together.
- Correct any orbital dystopia.
- Narrow the nasal dorsum and create a normal nose with adequate projection.
- Correct any soft tissue blemishes such as excess skin over the nose, and any nasal clefts (R. K. Sharma, 2014).

It was not possible to correct the hypertelorism before the landmark work of Paul Tessier in late 60's when he developed the combined intra-cranial and extra-cranial technique for the correction of the deformity. The challenge in surgical correction was to move the orbits in three dimensions without adversely affecting the vision. Tessier suggested that this was possible because of the inherent laxity of the optic nerve in the socket. He also demonstrated that large areas of the craniofacial skeleton can be devascularised and these would still survive completely provided healthy lining and cover is present. He visualised the bony orbit consisting of two parts, the outer square shaped box containing the globe and the inner cone shaped part housing the optic nerve. If these parts can be separated, the outer box can be moved in three dimensions without adversely affecting the vision

Timing (Age) of Surgery

Few topics generate more heated discussions regarding when or at what age to do these operations. As said, "As late as possible but as early as necessary" (van den Elzen et al., 2014). One would like to operate as early as possible to avoid peer teasing and emotional sequelae of ORH (R. K. Sharma, 2014). The ideal timing for this surgery is between two and five years of age in order for the psychological trauma involved with the deformity to be minimized while maximizing the ophthalmological benefits (Xie, Yang, & Zhu, 2011).

However, others advise not to perform this surgery in a child <5 years of age. Since the osteotomy lines during the surgical procedures is going through the inferior orbital rim, it is likely to injure the un-erupted tooth roots with resultant maxillary growth disturbances. Moreover, the bone stock may not be very good for holding the osteotomies together with fixation, thereby predisposing to relapse of the deformity. Hence, majority of the craniofacial centres would delay the correction of ORH to about 5-7 years of age. The results of surgery performed in adults are more reliable, stable, and the operation is technically much simpler than in a child. However, waiting till the adulthood can lead to a lot of psychological and body image problems (R. K. Sharma, 2014).

Surgical Correction

Hypertelorism surgery may be performed through an intracranial or extracranial approach; however, the extracranial approach should only be used in mild hypertelorism. Preoperative measurements should be performed based on CT imaging, as an intraoperative guide, including the medial and lateral interorbital distances, orbital wall length. The globe has a natural tendency to maintain its original position; therefore, a block movement of the orbital osseous complex is required. The decrease in interocular distance with a hypertelorism surgery is less than the decrease in interorbital distance, possibly due to periocular fat counteracting forces, and correlates better with movement of the lateral orbital rim than medial. Tessier proposed mobilizing the ventral 2/3 of the orbit, within about 7–8 mm of the orbital apex (Soldanska & Taub, 2019).

Operative Techniques

The two major surgical procedures to address hypertelorism are box osteotomy and facial bipartition. The choice between the two is dictated by maxillary width: when it is normal, box osteotomy is indicated or, when it is short, facial bipartition is recommended. Soft tissue management is more difficult and depends on the classification of the malformation (Urrego, Garri, O'Hara, Kawamoto Jr, & Bradley, 2005).

Box osteotomy

The surgery may be performed as a “box osteotomy” which permits a translational movement along the horizontal or vertical axes as per needs of the ORH deformity. This entails bony cuts going through the infraorbital rim. The classic orbital box osteotomy involves an en-bloc movement of the orbits medially into the space created by resection of abnormally wide nasal and ethmoidal bones. The procedure is best performed by a combined intracranial and extracranial approach. The access to orbital roof and cribriform region is gained after a frontal craniotomy has been performed, and the frontal lobes have been retracted gently. The bi-coronal incision gives a good view of the lateral and medial orbital walls. However, the addition of subciliary incisions is recommended for better access to the infraorbital rim and floor of the orbit (R. K. Sharma, 2014).

All the soft tissue is separated from the orbital roof, lateral wall, medial wall and the floor. The proposed markings for the orbital osteotomy are shown in the (Figure IV). The cuts are made in the orbital roof staying about 2 cm from the supraorbital margin, and the cut stops short of the cribriform region so as to protect the olfactory nerves. The medial wall cut is posterior the lacrimal crest so that the nasolacrimal duct and the lacrimal sac can be protected. The lateral cut can either split the orbital wall sagittal or simply go through the greater wing of sphenoid. The cut in the infraorbital region is below the infraorbital nerve exiting from the infraorbital foramen and this extends on to the floor of the orbit (R. K. Sharma, 2014).

The desired bony resection to be performed is marked on the dorsum of the nose and the excess nasal bone, nasal septum and the enlarged ethmoid sinuses are

removed. After removal of the central excess tissue that is causing hypertelorism, the bony orbits can be moved medially into this space and held in place either with plates and screws or by wires. The orbital shift medially results in bony gaps in the lateral orbital wall that need be filled with bone graft and split cranial bone graft. Since all the walls of the orbit are moving as one single unit, the orbital volume remains unchanged. Tessier advocated keeping a supraorbital bar to stabilize and guide the mobilized orbits. Orbital box osteotomy procedure is generally chosen whenever the dental occlusion is normal (R. K. Sharma, 2014).

Facial bipartition

If the dental occlusion is angulated as in cases of Apert's syndrome or some craniofacial clefts, "facial bipartition" is the surgery of choice. It involves medialisation of the two hemi-faces. For the facial bipartition, the medial resection of the nasal dorsum is in a "V" shaped form, and the palatal bone is split in the midline. Pterygomaxillary, septal, and median palatal osteotomies were added to the bone cuts described above to allow complete midface mobilization. Rowe disimpaction forceps were used to downfracture the midface. A wedge of central nasal, frontal, and ethmoid bone was removed, and the hemifacial segments were rotated toward the midline. The inferomedial aspects of the bipartition halves were rigidly fixed to one another with plates and screws. Autologous bone grafts were placed at the advanced portions of the lateral orbital rims and zygomatic arches. Where indicated, a cantilever autologous nasal bone graft was placed and rigidly secured (Figure V) (C. E. Raposo-Amaral et al., 2011).

The ensuing rotation allows correction of the narrow maxillary arch and also widens the nasal fossae and improves breathing. It would also result in a change of the orbital axis. This procedure is very useful for severe deformities. Facial bipartition would also increase the orbital volume and may improve the proptotic eyes seen in many of these cases (R. K. Sharma, 2014). This technique allowed rotation of each hemi-face, which in turn allowed appropriate orbital correction and simultaneous transverse widening of the maxillary (Batut et al., 2019).

Inverted U-Shaped Osteotomy

Inverted-U osteotomy by the intracranial and extracranial approaches is suitable option for orbital hypertelorism. The advantages of Inverted-U osteotomy are as follows: first, compared with the orbital box osteotomy by the intracranial and extracranial approaches, this method has a similar effect as correcting moderate and severe widening of the orbit, but it is less difficult to operate and less invasive, and the operative time is shorter. Second, the suborbital wall is not osteotomized, therefore, not damaging the maxillary sinus and un-erupted tooth roots. It reduces the risk of infection and does not affect the maxillary development in children. Third, all surgical procedures can be completed through a coronal incision without incising the infraorbital margin, thus preventing facial scarring (Huang, Yang, Li, & Ma, 2021).

Through bilateral Z-shaped coronal scalp incision, we could turn the scalp flap with exposure of the upper border of the zygomatic arch. The lower orbital rim was not exposed before osteotomy. Orbital osteotomy approached the following

purposes: (1) to raise the frontal bone flap; (2) to remove interorbital expansive part of anomalies; (3) inverted U-shaped orbital osteotomy, to retain the infraorbital rim (Figure VI) (Shen, Cui, Chen, & Ying, 2015).

Soft-tissue management

Soft tissue has posed a constant barrier to achieving optimal results in hypertelorism surgery. Soft-tissue management is the greatest challenge in the correction of hypertelorism. The three major corrections concern are:

- The midline with excess soft tissue after bone shortening,
- Canthi positioning
- Nasal correction (R. K. Sharma, 2014).

Tessier believed that hypertelorism correction has its real difficulties, not much in reducing an exaggerated interorbital distance as in correcting the other malformations associated with it (Shakir et al., 2019).

Midline soft tissue excess

The ideal timing and type of reconstruction to address midline soft tissue excess following bony hypertelorism correction remains to be determined (Urrego et al., 2005). The excess frontal, glabellar, and nasal skin can be respected, however, with poor cosmetic results and midline scar which extend from the central forehead to the nasal tip. Excision of soft tissue also appeared to interfere with nasal development. McCarthy advises against excising excess skin at the time of initial surgery, as it may be needed for secondary nasal augmentation procedures. Kawamoto proposed a circumferential dermal purse suture of the redundant soft tissue of the glabella to contract the excess skin and position the eyebrow just medial to the medial canthus (Soldanska & Taub, 2019).

Canthi and periorbital

The prevailing stigmata associated with hypertelorism correction relates to persistence of telecanthus, epicanthal folds, and loss of the “naso-orbital valley.” Soft tissue laxity in the medial canthal region leads to the illusion of bony relapse or inadequate correction, despite appropriately medialized bony orbits. Initial attempts at hypertelorism correction involved medial canthal detachment; however, this led to notable canthal drift in the postoperative period. Postoperative medial canthal drift after canthus release led to the preference for canthus preservation during medial orbital wall osteotomy and transnasal wiring, which led to notable improvement (Shakir et al., 2019). Epicanthal folds may secondarily be addressed through Y-V plasty or double-opposing Zplasty (C. E. Raposo-Amaral, Denadai, Ghizoni, & Raposo-Amaral, 2017).

Nasal reconstruction

Tessier stated that “the correction of hypertelorism is surgery of the nose.” The importance of nasal augmentation cannot be understated, as midline forehead and glabellar scars can often be justified in the setting of an aesthetically pleasing reconstruction (C. E. Raposo-Amaral et al., 2017). Most (if not all) patients undergoing hypertelorism reconstruction require some form of secondary nasal reconstruction. McCarthy advocated for a sizable iliac corticocancellous nasal bone graft at time of index correction to not only augment the nose, but also to use midline soft tissue excess. Various grafting sites have been described, including cranial, rib, iliac, and midline bony excess at time of resection in addition to various surgical approaches (i.e., open rhinoplasty, coronal incision). Tessier advocated for the nasal bridge to be reconstructed with a rib bone graft bolted to the frontal bone and an auxiliary strut at the anterior nasal spine to support the graft. The bifid nose, a common presentation in patients with craniofrontonasal dysplasia, requires simultaneous bone grafting, nasal ala, and alar cartilage reshaping, namely the divergent medial crus (Marchac, Sati, Renier, Deschamps-Braly, & Marchac, 2012).

Raposo-Amaral (C. E. Raposo-Amaral et al., 2017) advocate for primary midline soft tissue excision at index procedure in the setting of redundant soft tissue with adequately positioned nasal ala and alar cartilages. Paramedian forehead or scalping flap reconstruction combined with medialization of the divergent medial crura via interdomal suture technique can sufficiently correct more severe nasal phenotypes, in contrast. When performing the Converse scalping flap, the superficial temporal artery must be preserved, while the K stitch cannot be combined with paramedian forehead flap reconstruction to protect the vascular pedicles for total nasal reconstruction (Shakir et al., 2019).

Complications

Hypertelorism surgery carry many risks and may lead to some postoperative complications. These possible complications are divided into acute and delayed events (Bradley et al., 2006). Acute complications such as excessive bleeding, intracranial infection, and cerebrospinal fluid leakage, epilepsy, nasal tip flap necrosis (Hidalgo, Romo, & Estolano, 2009) and less common, but devastating, complications such as blindness and death have been reported. Delayed events, such as insufficient correction, palpable implants, an overly long nasal tip after rhinoplasty using an autologous bony and chondral graft, illusion of relapse due to soft tissue laxity in the medial canthal region and temporal hollowing amenable to fat grafting (Bradley et al., 2006). Bleeding, cerebrospinal fluid (CSF) leak and infection remain the major postoperative acute complications associated with hypertelorism correction (Shakir et al., 2019).

Preoperative 3D planning in hypertelorism surgery

Orbital structure is complicated as there are many important organs in that area. The surgical operation that involves orbital areas is difficult, and dangerous, and the facial features as well as functions must be taken into account, so preoperative surgical planning is important. Surgical procedures for orbital

hypertelorism have undergone changes since the second half of the 20th Century because of the recent techniques like: 3D printing and computer-assisted surgical planning (Xie et al., 2011). The use of digital technology in the treatment of orbital hypertelorism aims to achieve accurate osteotomy, reduce surgical risk, and optimize surgical technique. Traditional methods depend on the experience of the surgeons, so it is possible to damage the lacrimal apparatus and olfactory nerve. Through three-dimensional surgical design, important nerves and vessels can be preserved, accurate osteotomy can be achieved, and the symmetry of patients' eyes can improve (Huang et al., 2021).

A presurgical planning technique of high prediction for the correction of hypertelorism was first described in 1989 by Ortiz-Monasterio et al based on the location of anatomic points and planes on a posteroanterior x-ray to obtain the measures and location of the osteotomies (Hidalgo et al., 2009). Hypertelorism correction could be also simulated on anteroposterior cephalograms by geometrical planning (Queiros et al., 2017). Then new technologies appeared when an anatomical model with detailed geometric characteristics was constructed from CT-scans. This technology was used to produce rough physical models of the head and face. More effective techniques, such as stereolithographic models, enabled 3D printing of solid cranial models and computer-assisted virtual simulation for surgical planning have been developed to be used in various aspects of craniomaxillofacial surgery (Batut et al., 2019). 3D printing is generally superior to stereolithography for better accuracy, quicker printing time and lower cost (Engel, Hoffmann, Castrillon-Oberndorfer, & Freudlsperger, 2015).

Stereolithography is a precise tool for planning hypertelorism correction, as it enables any malformation to be observed from various angles. Therefore, the surgical team has a more tangible approach, with good prediction of otherwise unexpected events that could occur during surgery (Batut et al., 2019). Sailer et al. were the first to use a stereolithographic model in surgical simulation for facial bipartition, in the 1990s. (Batut et al., 2019). Stereolithography is a technique of manufacturing with computerized support that is used to make a high-precision model. Originally developed in the aerospace industry, this technology uses the detailed surface description to create a plastic model in layers. An ultraviolet laser controlled by computer catalyzes the polymerization of the curable plastic in a tomographic way to create a solid model, layer by layer. Combining the scanned information of the tomography with this manufacturing technology is possible to create exact anatomic models of not only the external surface but also the complete representation of the internal structures (Figure VII) (Hidalgo et al., 2009).

3D printing skull model provides the possibility to observe a disorder from an infinite way of angles. The space realism and the tactile capacities of these models offer a type of virtual reality for the surgeon to have a visual and tactile approach towards the coming surgery without touching or knowing the patient. Furthermore, 3D model techniques are helpful to minimize surgical approaches, save operation time and help the families to understand the surgical procedure. It is possible to precontour miniplates or microplates and to precisely define the later position of those plates or any prefabricated implant. The technique is

further used to determine the osteotomy lines, to decide where to raise bone grafts and how to shape them (Figure VIII) (Engel et al., 2015).

Computer-assisted surgery (CAS) and virtual simulation became a prominent tool in surgical planning, and its use has been widening to more craniofacial applications in recent years. CAS displays various advantages such as better handling and positioning of the bone fragments, a shorter operative time and safer than conventional surgery (Hidalgo et al., 2009). The simulation of the surgery can be carried out from the preoperative CT scanned images. The appropriate osteotomy lines, bone removal, and orbital correction can be simulated (Figure IX). The design of the customized surgical cutting guides and osteosynthesis plates can be performed using the 3-matic software (Materialise) (Batut, Paré, Kulker, Listrat, & Laure, 2020).

Virtual simulation begins by transfer all the appropriate CT data in DICOM format (Digital Imaging and Communications in Medicine) to be used in craniomaxillofacial (CMF) Planner software. Then, designs of the osteotomy delineation are done. Osteotomies vary depending on the shape of the hypertelorism (symmetrical, asymmetrical) and on the nature of the osteotomy (box osteotomy, facial bipartition). This virtual simulation helps the surgeon to visualize the expected outcome of surgery. More specifically, the interorbital distance can be defined with very good precision for final outcome. Figures (X) provide an example of simulated images used in a case of hypertelorism using facial bipartition to shorten the inter-orbital distance and to increase maxillary width (Laure et al., 2019).

Conclusions

3D printed models have been used to replicate a broad spectrum of tissues, ranging from hard tissue structures such as the skull, spine, limbs, and ear bones, to soft tissue structures including the heart, lungs, liver, and vasculature. 3D printing in surgical assistance domain has seen a tremendous rise in the last two decades. The use of 3D printing to form soft tissue structures has been largely unexplored. Very few attempts have been made to capture the mechanical properties of soft tissue along with high resolution anatomical detail. Previous attempts utilized flexible polyurethane or silicone but did not achieve the correct mechanical properties such as elastic modulus, and do not provide a true feel of the wet and soft biological tissue. The ability to 3D print hydrogel materials which mimic *in vivo* soft tissue properties has been widely utilized for tissue engineering applications, but these techniques have yet to be applied to the fabrication of anatomical models for surgical simulation. Considering recent advances in 3D printing softwares and model materials together with the FDA's approval of this technology for diagnostic use, the applications of 3D printed anatomical models will continue to expand and further improve healthcare.

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Figures

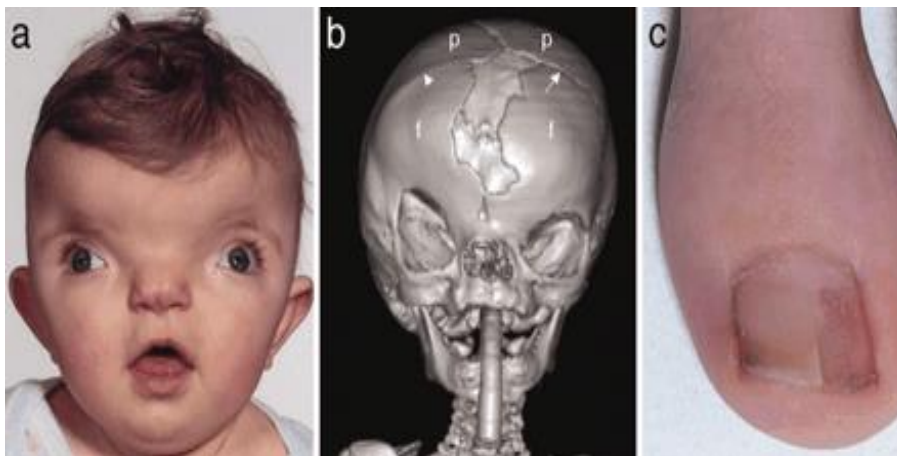


Figure I. Clinical features of CFNS. (a) Facial view showing marked hypertelorism. (b) Three-dimensional computed tomographic skull reconstruction showing right unicoronal synostosis, lateral displacement of orbits, and central defect between frontal bones. (c) Longitudinal splitting of the nails is frequent (van den Elzen et al., 2014)

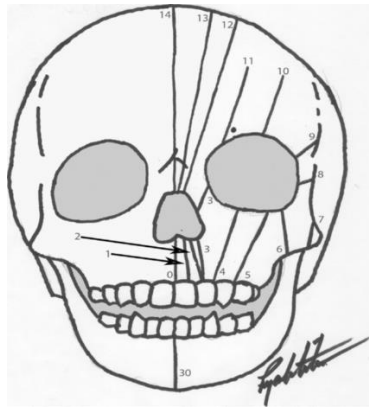


Figure II. Clefts numbered according to Tessier (Winters, 2016)



Figure III. Patient with Apert syndrome, Extraorally: Hypertelorism, vertical excess of the lower third of the face (Kaya et al., 2012)

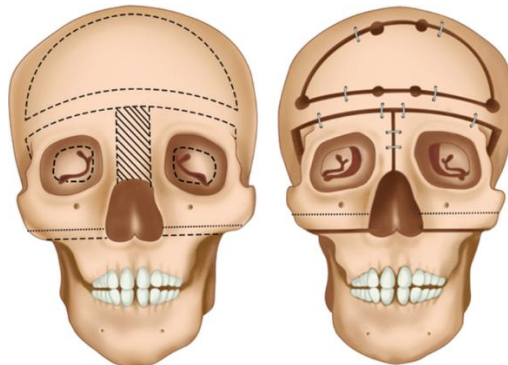


Figure IV. Schematic drawing of a box osteotomy to medialize the orbits and corrects hypertelorism (Cassio Eduardo Raposo-Amaral, Jarrahy, Lim, & Alonso, 2018)

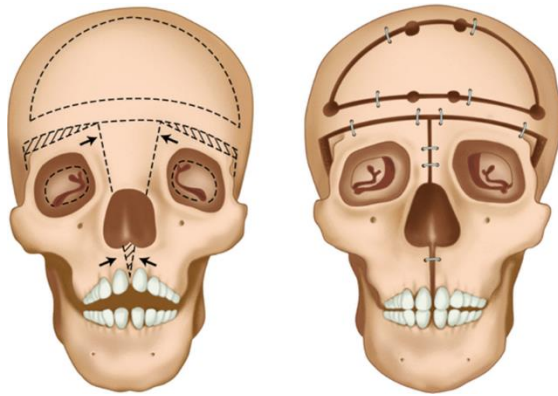
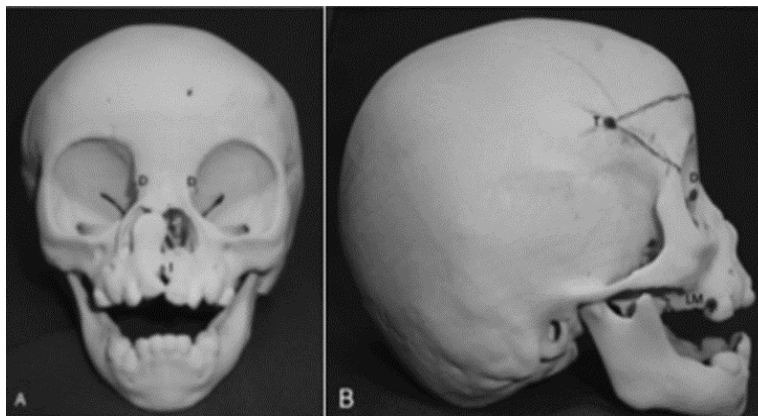


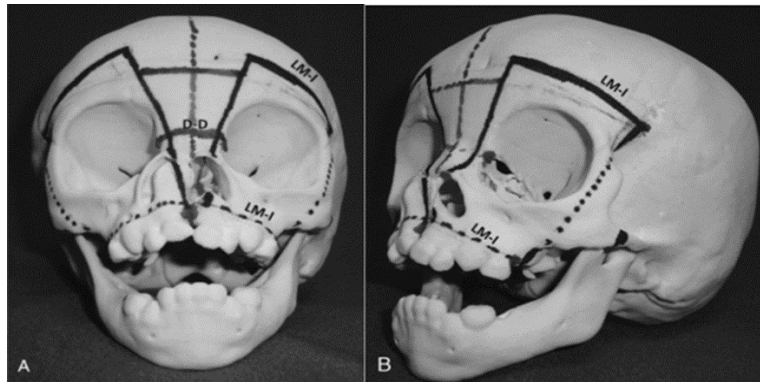
Figure V. Schematic drawing of facial bipartition osteotomy (Cassio Eduardo Raposo-Amaral et al., 2018)



Figure VI. Design sketch of inverted U-shaped osteotomy (Shen et al., 2015)



(a)



(b) Figure VII. (a) A, Frontal view of stereolithographic model with dacryon (D) and interincisal point (I) markings. B, Lateral view with dacryon (D), temporal (T), and lateral maxillary (LM) markings. (b) A, Frontal view of stereolithographic model with dacryon (D) and interincisal point (I) markings. B, Lateral view with dacryon (D), temporal (T), and lateral maxillary (LM) markings (Hidalgo et al., 2009)

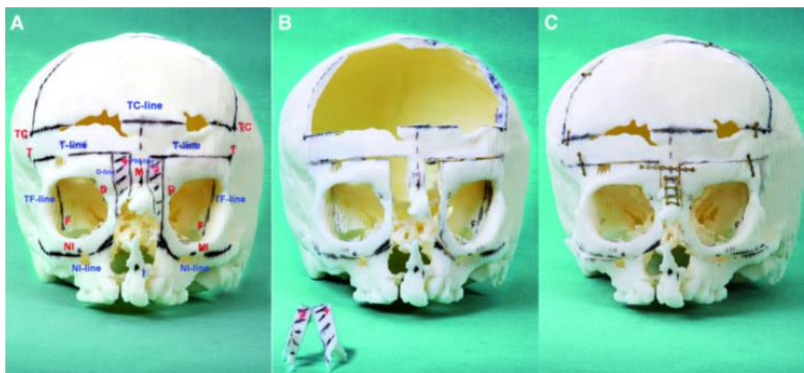


Figure VIII. Preoperative planning using 3D printing modelling technology. A Frontal view with determined anatomic landmarks. b After performing the osteotomies. c after the medial mobilization of the orbits (Hidalgo et al., 2009)

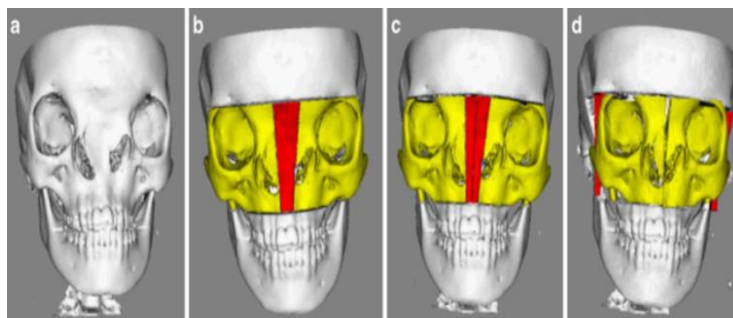


Figure IX. Three-dimensional osteotomy simulation of orbital hypertelorism. a Bone abnormality before surgery. b Orbit is divided into three parts. c Middle bone is lengthways divided into two parts. d Bones of two sides are moved to middle, the removed bone graft in the gap (Xie et al., 2011)

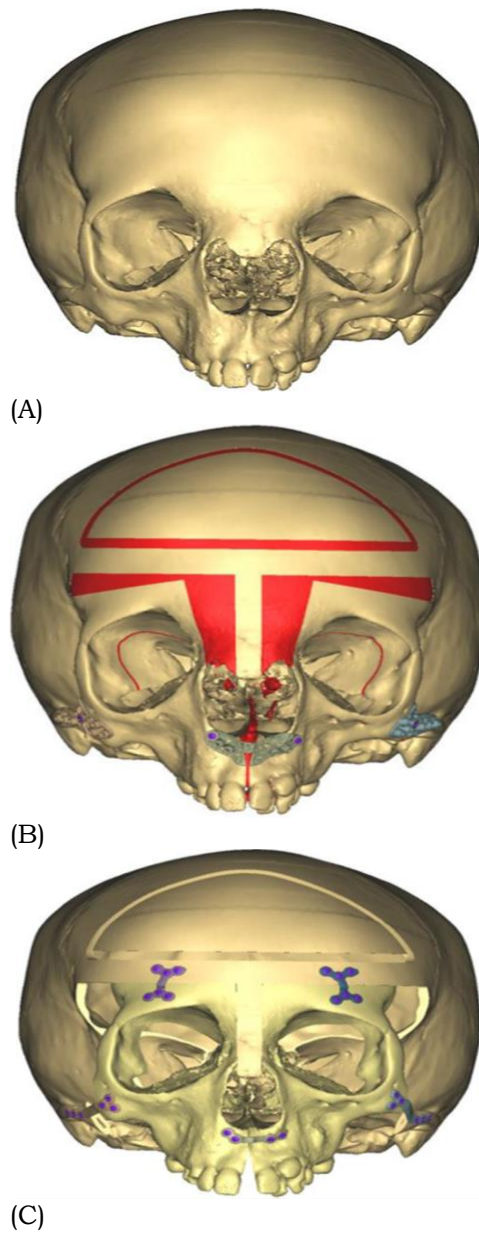


Figure X. (A) Virtual image of the hypertelorism reconstructed via DICOM based on patient's CT-scans. (B) Simulation of facial bipartition osteotomies. (C) Simulation of osteotomies repositioning (Laure et al., 2019)