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Craniosynostosis: State of the Art 2019

Management of isolated and complex craniosynostosis residual deformities: What are the maxillofacial tools?

A. Joly[a,c,d], B. Croise[a,d], N. Travers[b,c], A. Listrat[b,c], A. Pare[a,c,d], B. Laure[a,c,d,*]

[a] Service de Chirurgie maxillo-faciale et Plastique de le Face, CHU Trousseau, Tours, 37044, France
[b] Service de Neurochirurgie Pédiatrique, CHU Clocheville, 37044 Tours, France
[c] Centre comptétence cranioténoses et malformations craniofaciales rares, CHU Clocheville, 37044 Tours, France
[d] Faculté de médecine de Tours, Université de Tours, 2bis, boulevard Tonnellé, 37000 Tours, France

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ABSTRACT

Treatment of craniosynostosis is complex and has greatly progressed in recent decades. From the early stages in the 1950s to today's most recent techniques, surgeons have faced the challenge of overcoming the deformities often caused by such invasive, complex surgeries. In the most recent years, new techniques have been developed that address surgical sequelae, including those of surgery performed in childhood. After a general introduction on craniosynostosis, the present paper describes the various types of deformity that may result from complex surgery and offers an overview of the various tools available to surgeons. An explanation of each indication and procedure is given.

1. Introduction

Craniosynostosis is a rare condition, described by Otto in 1830 and defined as premature closure of one or more cranial sutures. Incidence is 1 per 2,500 births per year [1,2]. In most cases (85%), deformities are isolated (non-syndromic craniosynostosis) but in 15% are associated with other pathologies (congenital heart malformation, maxillofacial deformity, limb abnormality, etc.) or genetic disorders [1].

Premature fusion of cranial sutures may impact skull development and intracranial pressure, especially in syndromic craniosynostosis or synostosis of multiple cranial sutures [2,3]. Moreover, Speltz et al. [4] showed that children with isolated craniosynostosis present mild cognitive deficiency.

Treatment is complex and has steadily progressed since craniofacial surgery was introduced in the 1950s [5] and esthetic and functional issues were recognized. Primary surgery aims to reduce intracranial pressure and treat the resulting craniofacial distortions and asymmetry as well as possible. The most frequent complication following craniosynostosis surgery is incomplete correction of deformities [2]. Long-term assessment is important, to improve our results and develop additional treatments.

Residual deformities include deformities due to craniofacial surgery (bone defect, temporal narrowing, scarring alopecia, restricted mouth opening) [6] or secondary to asymmetric craniofacial skull growth (orbital asymmetry, nasal deviation, upper maxillary retrusion, asymmetric jaw). Despite surgery, bone defects, temporal narrowing and frontal bossing can be observed in patients with scaphocephaly [7]. Also, anterior plagiocephaly causes nasal deviation and increased orbital roof height with enophthalmos, and the chin tip of the mandible is displaced contralaterally [2,6,8]. Patients with trigonocephaly show super lateral depression of the orbital roof, bilateral temporal depression and hypotelorism [2,6]. In syndromic craniosynostosis, complex residual deformities are observe, due to craniofacial disorders, including those forming part of the syndrome. Exophthalmos and jaw retrusion linked to a dental malocclusion is frequently reported [9]. In Saethre–Chotzen syndrome, patients present ptosis, lateral canthus dystopia, lower hair line, and protruding ears [10,11].

These deformities entail severe esthetic and psychological problems, impacting the child's development [12]. It is therefore essential to treat these patients on a multidisciplinary basis and to rehabilitate these asymmetries [13]. The aim of the present

* Corresponding author at: Service de Chirurgie maxillo-faciale et Plastique de le Face, CHU Trousseau, Tours, 37044, France.
E-mail address: b.laure@chu-tours.fr (B. Laure).

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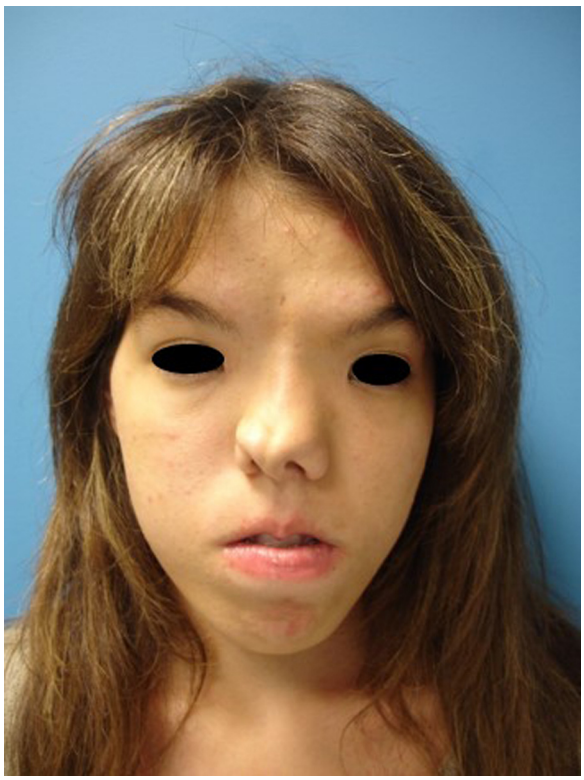

Fig. 1. Fronto-nasal dysplasia sequelae with epicanthus, telecanthus and nasal residual deformity.

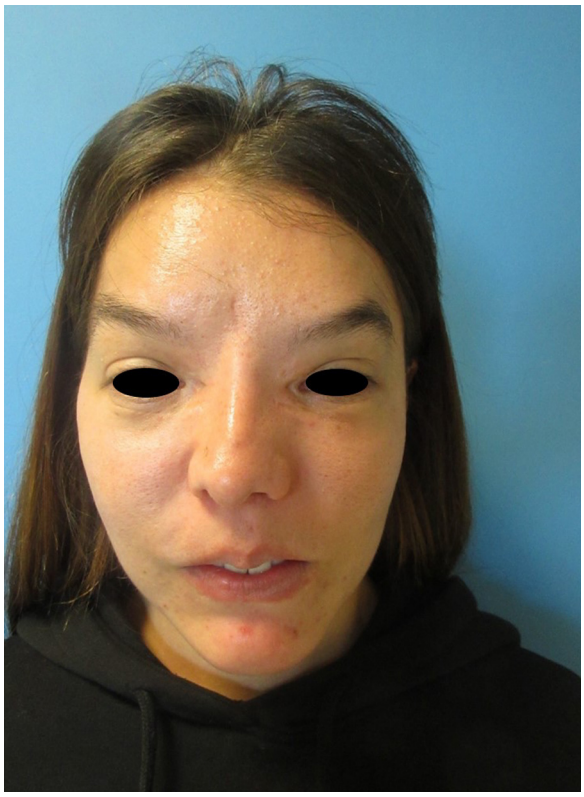

Fig. 2. Outcome of bilateral double Z-Plasty correction, bilateral transnasal canthopexy, rhinoseptoplasty via open approach with parietal bone graft on the nasal dorsum and chin osteotomy with height reduction and advancement.

article is to describe the various maxillofacial tools available to correct these residual deformities.

2. Lipostructure

Lipostructure consists in autologous fat transfer (AFT) to restore volume in a defined area. In the Coleman technique [14], fat is removed through an atraumatic cannula from various areas such as the periumbilical zone, inner side of the knees or thighs, flanks or buttocks [15,16]. The fat is then centrifuged to separate it from the oil layer and from the hematic fraction, which are eliminated. The fat is then reinjected through a cannula into the area to be treated. The technique is simple and minimally invasive, and corrects the congenital defects of the pathology, such as pterional hollows in scaphocephaly and trigonocephaly [17] or receding forehead in oxycephaly [18].

Castro Govea et al. [17] showed that lipoinjection is a good alternative for improving fronto-orbital and temporal appearance in residual craniosynostosis deformities, with 92% successful results. Laurent et al. [19] showed that two operative steps are necessary to get a satisfactory result; in their study, there were no complications.

In craniofacial surgery, lipostructure has to be performed with care, to avoid residual bone defects [16,19].

3. Eyelid correction

Ptosis is due to dehiscence or disinsertion of the muscle raising the upper eyelid (levator muscle) or an abnormality in muscle strength. It can be corrected under local anesthesia, which enables exact symmetric correction of the eyelid. In dehiscence, the levator muscle is fixed to the tarsus via an upper palpebral approach [20]. In levator muscle dysfunction, frontal suspension is required [20], using a temporal or fascia lata strip fixed onto the tarsus via a palpebral approach. The two sides of the strip are inserted in the

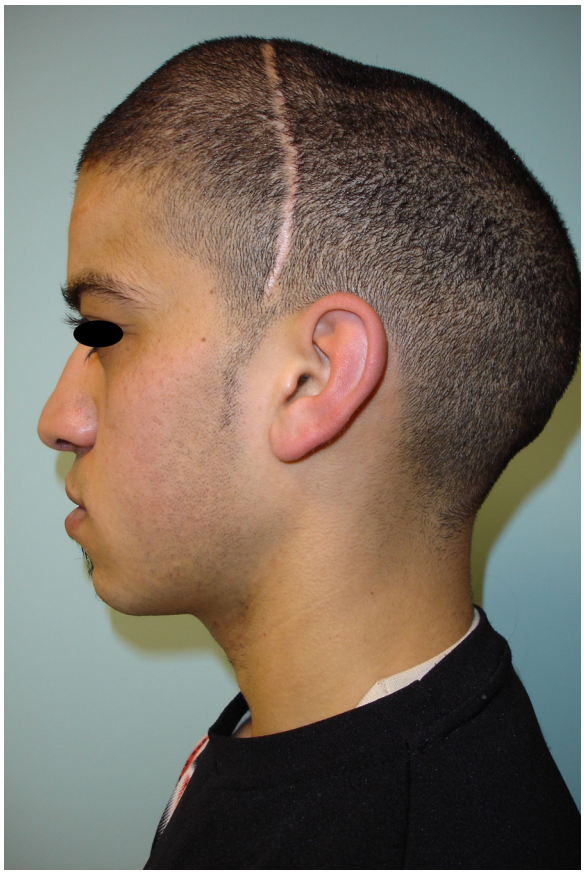

Fig. 3. Scar enlargement in an adult patient.

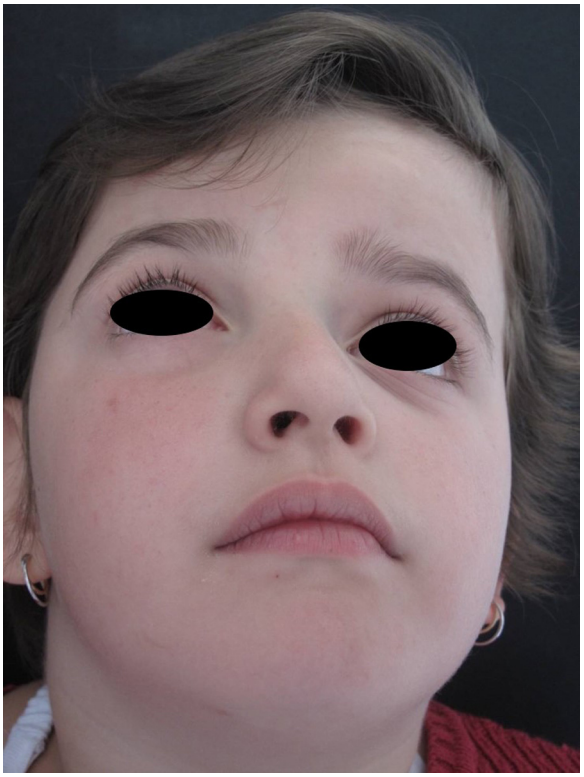

Fig. 4. Left anterior plagiocephaly sequelae with facial asymmetry and left cheekbone retrusion.

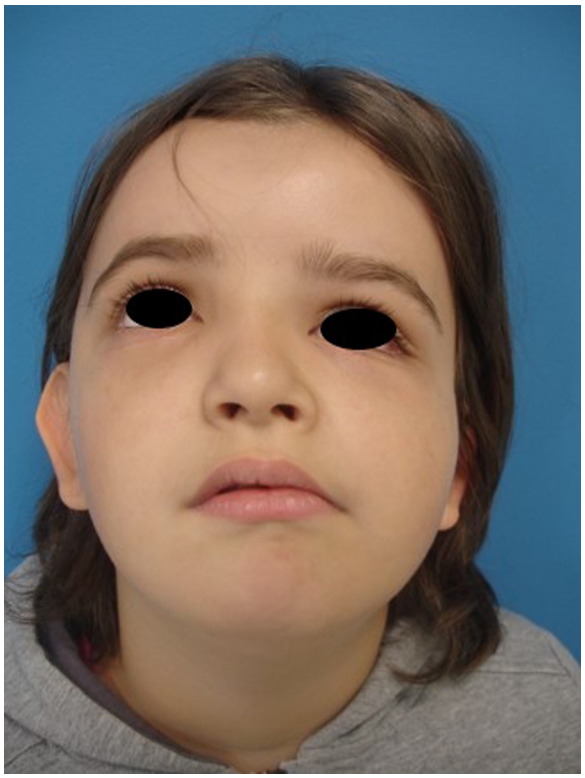

Fig. 5. Parietal apposition graft correction on left zygoma bone with crooked nose rhinoplasty.

retroseptum to the frontal muscle just above the eyebrow. Postoperatively, patients have to raise their eyebrows raise their upper eyelids.

In internal canthal dystopia or telecanthus, transnasal canthopexy enables permanent stable fixation of the medial canthus, using a metallic wire [21].

In external canthal dystopia, lateral canthopexy is performed. A tarsal neo-tendon is fixed by a periosteal suture [22,23]. In craniofacial malformation, a coronal approach is often required; the lateral canthus is fixed to the temporal aponeurosis.

To correct epicanthus, double Z or Y-V plasty is performed [24] (Figs. 1 and 2).

4. Hairline correction

To correct hairline lowering in Saethre-Chotzen syndrome, a mask lift is performed via resection of a scalp strip by bicoronal incision, to raise the scalp line. In the same step, lateral canthopexy can be performed to correct the antimongoloid anatomy observed in these patients [25].

For patients with high hairline, such as patients with scaphocephaly, hairline lowering can be performed during a cranial vault remodeling procedure, which allows broad access to the forehead to decide on the level for the new hairline [26,27].

5. Scarring alopecia

Scarring alopecia is a common presenting symptom in craniofacial surgery. Patients are often operated on in childhood when the skull is growing faster, and scars consequently expand, causing alopecia (Fig. 3).

Depending on the size of the alopecia area, various techniques are available:

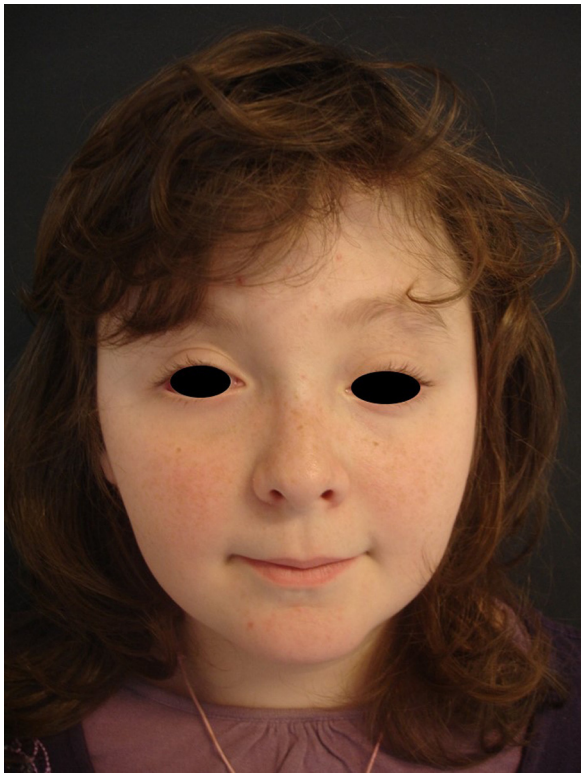

Fig. 6. Sequelae of left anterior plagiocephaly with eye dystopia and cheekbone projection defect.

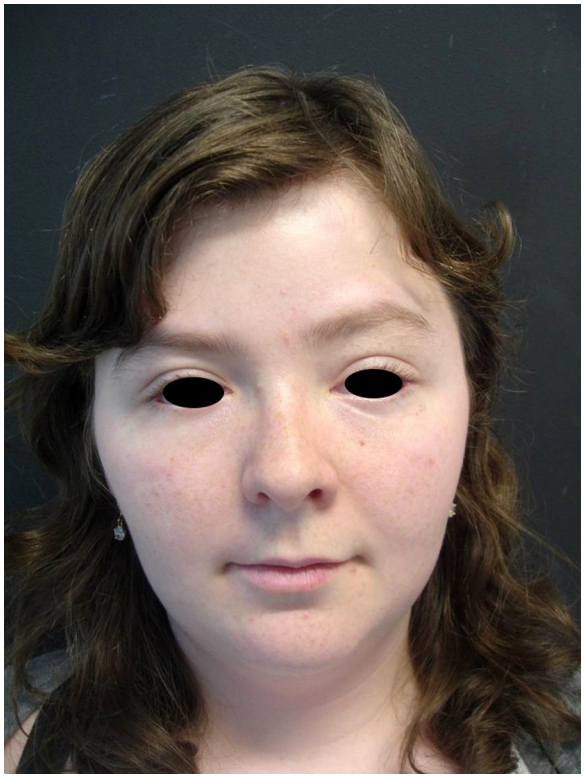

Fig. 7. Correction via a parietal bone graft on left orbital floor and on zygoma.

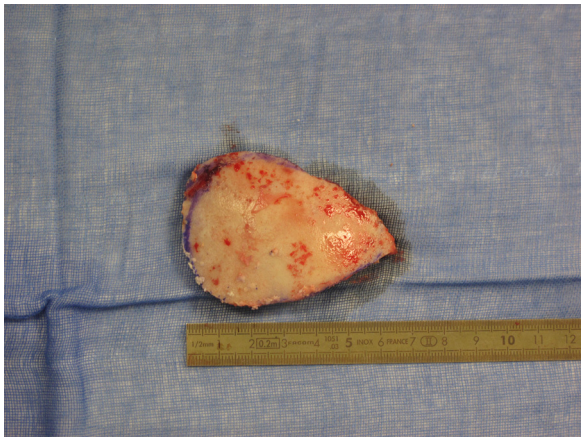

Fig. 8. Parietal bicortical bone harvesting.

- skin resection and direct suturing to soft tissue expanders;
- direct suturing, for limited alopecia with skin laxity;
- for larger alopecia, local scalp flap (transposition, rotation or advancement flap) or skin expansion [28].

For expanders, a subcutaneous inflatable silicon balloon is gradually inflated, producing new skin with hair bulbs [29].

Scalp incisions have to be parallel to the hair follicles, to prevent injury and alopecia [26].

6. Rhinoseptoplasty

Nasal deviation is a classical deformity in craniosynostosis, and can be treated at any age. Several studies reported persistent postoperative nasal angulation in 38% to 49% of patients [30,31].

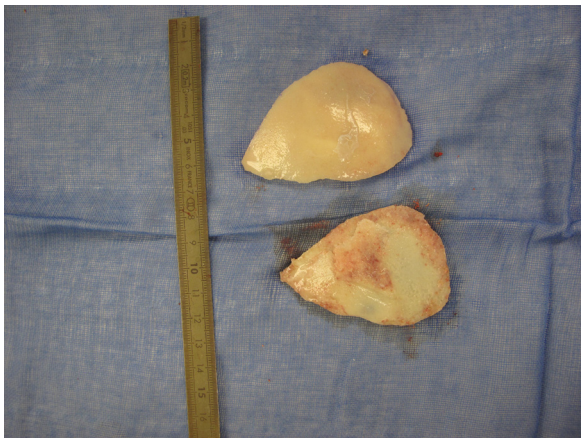

Fig. 9. Outcome of on-table bicortical split by saw: two grafts.

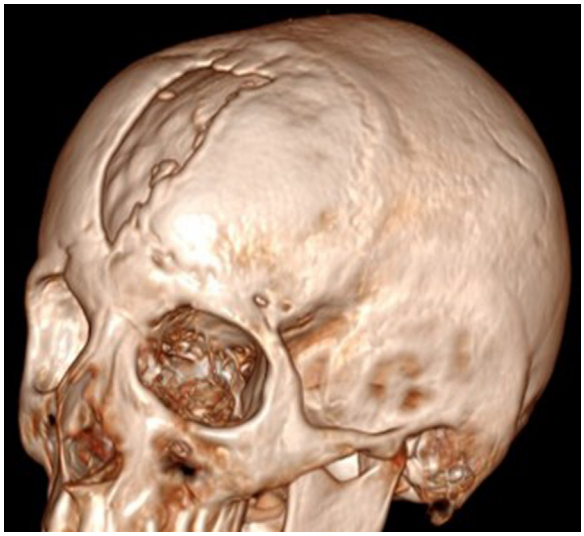

Fig. 10. Pre-operative CT-scan: frontal bone defect, supra-orbital depression after trigonocephaly correction.

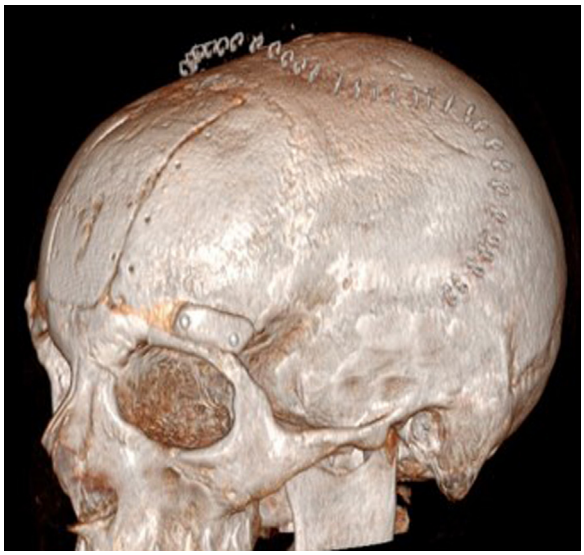

Fig. 11. Result of bone defect correction via bicortical procedure with parietal bone graft and supra-orbital parietal bone grafts.

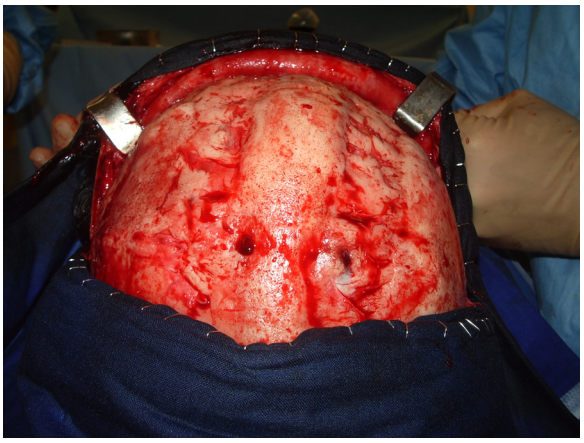

Fig. 12. Operative view of frontoparietal residual deformities after scaphocephaly correction during childhood.

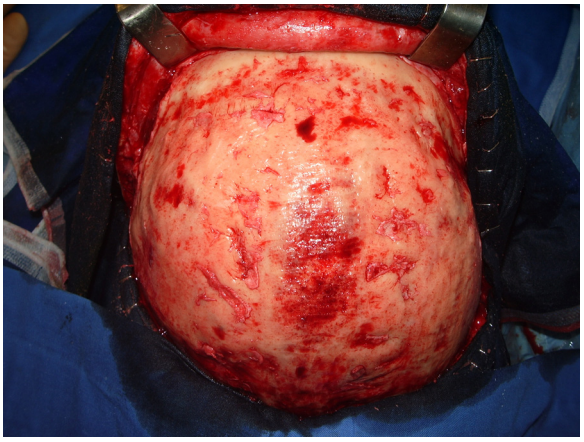

Fig. 13. Correction via external bone remodeling by burr.

Rhinoseptoplasty is most often performed at the end of the growth phase, at 18 years of age, if the deviation persists [32]. Some teams correct frontonasal angulation by nasal osteotomies during the fronto-orbital advancement procedure [33].

In unilateral coronal synostosis (anterior plagiocephaly), the nasal radix is deviated toward the fused suture and the tip of the nose toward the contralateral side. In his book *Plastic Surgery of the Orbit and Eyelids* (Masson 1977), Paul Tessier described a fronto-ethmoido-naso-maxillary osteotomy for a patient with severe nasal deformities. This osteotomy can be performed to correct nasal deviation, associated with bone graft to project the zygoma and supraorbital edge (Figs. 4 and 5).

7. Auricle correction

Ear deformities are frequent in craniosynostosis, as part of craniosynostosis syndrome or simply they are frequent in the general population [34]. Otoplasty is necessary to correct congenital ear deformity and to improve cosmetic results. In Saethre-Chotzen syndrome, patients present small ears with prominent crus ear [35].

Numerous otoplasty techniques have been described to correct prominent ears.

Otoplasty can be performed under general or local anesthesia, depending on the patient's age, via a retro-auricular incision. The aim is to weaken the helix cartilage, so as to be able to plicate it and bury the concha, which is securely attached to the premastoid periosteum [34].

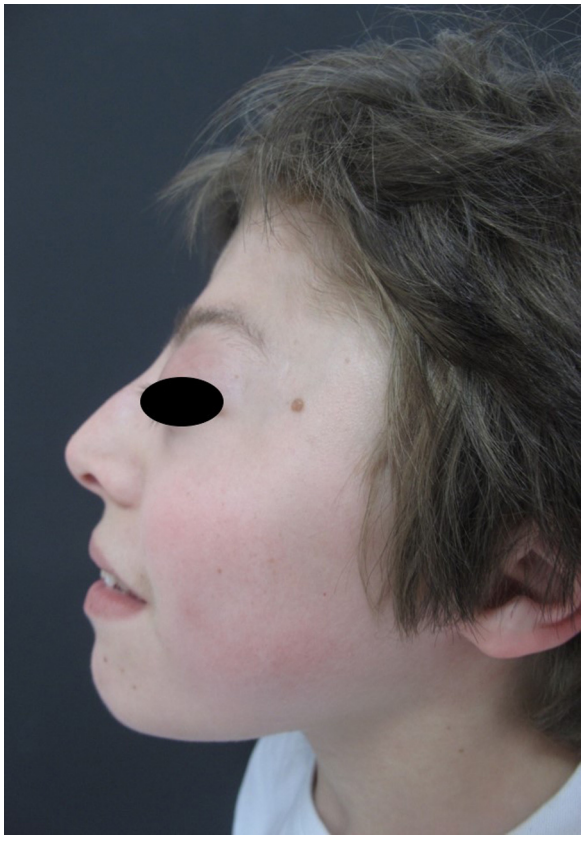

Fig. 14. Saethre-Chotzen syndrome sequelae with frontal retrusion.

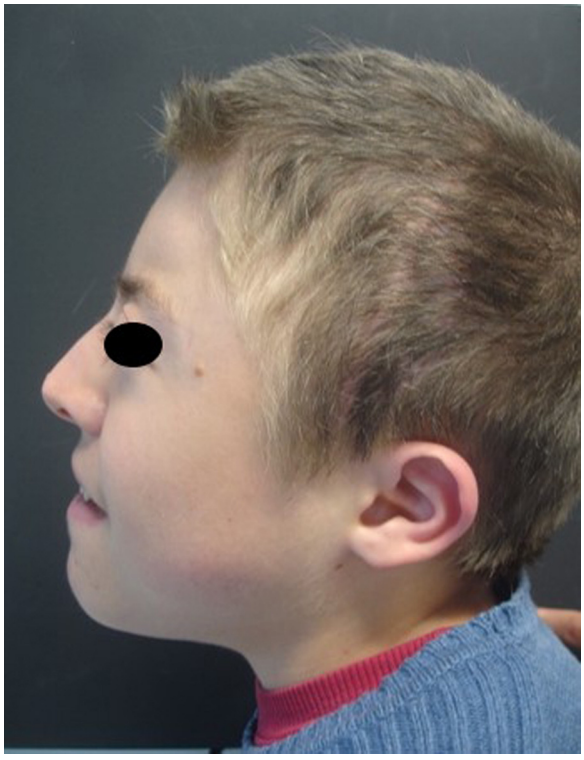

Fig. 15. Correction via reverse frontal cranioplasty.

Surgery	Indications	Age for the surgery
lipostructure	all defect, for example pterional hollows	from 8 years old, when the patient requested
levator muscle surgery	ptosis correction due to dehiscence or disinsertion of the levator muscle	from 3 years old (general anaesthesia), from 15 years old (local anaesthesia)
frontal suspension	ptosis correction due to levator muscle disfunction	from 3 years old
transnasal canthopexy	internal canthal dystopia ou telecanthus	from 10 years old
lateral canthopexy	external canthal dystopia	from 10 years old
hairline correction	lower hairline	from 8 years old, when the patient requested
removal scar	scarring alopecia	when the patient requested
local scalp flap	scarring alopecia	when the patient requested
skin expansion	scarring alopecia	when the patient requested
rhinoseptoplasty	nasal deviation	end of the growth (from 18 years old)
otoplasty	auricle deformities	from 8 years old, when the patient requested
bone graft	facial asymmetry, bone defect, enophtalmos	end of the growth (10 years old for orbits, 18 years old for the rest)
maxillary distraction	more than 1 cm retrusion of the maxillary	11 years old, after canine tooth eruption
Lefort I osteotomy	minor maxillary retrusion or mandibular protrusion	end of the growth, from 18 years old
bilateral mandibular split osteotomy	mandibular retrusion, jaw deviation, open bite	from 15 years old
coronoidectomy	limited mouth opening	when patient have feeding problems

Fig. 16. Summary of all procedures indicated to correct craniosynostosis deformities.

8. Bone defect: cortical bone graft

Bone graft to correct temporal bone defect or restore zygoma projection mainly uses parietal graft, as this is autologous, and harvesting is simple and performed via the same approach. Bone resorption is very low.

For monocortical parietal bone graft, the outer cortical table is removed from the venous sinus, 2 cm lateral to the sagittal suture and 1 cm behind the coronal suture. Using a combination of curved and straight osteotomes, the outer cortical bone graft is separated from the diploe. Figs. 6 and 7 show a hypoglobus and cheekbone projection defect treated by a monocortical procedure after bone graft on the orbital floor and cheekbone.

Larger defects can be addressed by split calvarial bone graft. Monocortical parietal bone graft cannot be performed, because of the difficulty of removing it without breaking it. Bicortical bone graft is therefore performed. The bone is split on the surgical table using a saw in the diploe plane. The external part is used for the bone defect and the internal part replaced in its original position (Figs. 8 and 9).

To optimize symmetry, 3D printed cutting guides are available commercially, calculating the dimensions of the bone sample according to mirroring (Figs. 10 and 11).

It is also possible to produce bone implants to fill bone defects if autologous graft cannot be performed, either by mirroring or by 3D printing [36]. Some complications are reported with these implants: infection, exothermic reaction with burn injury, and artefacts in imaging [37], [38].

Some teams also use costal grafts to correct orbital and zygomatic deformities, considering them thinner and easier to harvest [39].

For frontal bone unevenness, bone remodeling can be performed using a burr (Figs. 12 and 13).

To correct major frontal retrusion, reverse frontal cranioplasty can be performed [40] (Figs. 14 and 15).

9. Orthognathic surgery

Some patients present craniofacial growth disorder of the jaw as well: maxillary retrusion, mandibular protrusion, open bite or jaw deviation. Orthognathic management (orthodontic treatment and orthognathic surgery) is necessary. Follow-up has become of major importance in the last 5–6 years, to prevent occlusion deformity. In case of severe maxillary retrusion, distraction can be scheduled. However, in some cases monobloc advancement or Le Fort III osteotomy are insufficient to correct malocclusion. Le Fort I osteotomy can be performed in such cases to correct maxillary retrusion [41]. In most cases, orthognathic surgery is considered at the end of facial growth, at 15 to 18 years depending on the deformity. The timing of the final orthognathic operation is important, in that maxilla and mandible growth should be complete before surgery [42]. Bi-maxillary surgery is often performed via a Le Fort I osteotomy, with mandibular correction by bilateral sagittal split osteotomy [43].

It is also possible to perform chin osteotomy to medialize the chin, as in plagiocephaly.

10. Correction of limited mouth opening

Craniofacial procedures involve sectioning the temporalis muscle. The resulting fibrosis and myositis give rise to the specific complications in the form of limited mouth opening [44].

In case of limited mouth opening, a coronoidectomy can be performed to restore mouth opening and improve daily life [44]. The approach is intraoral; postoperative physiotherapy is required in order to optimize mouth opening.

To prevent this complication, it is necessary to be meticulous in the reconstruction of the temporalis muscle during the procedure [45], and physiotherapy of mouth opening must be started immediately [46].

11. Conclusion

Management of craniosynostosis involves a multidisciplinary team (neurosurgeons and maxillofacial or plastic surgeons) for global care [13]. Moreover, surgery planning and follow-up need to take account of skull and facial growth for outcome to be satisfactory. Because the face grows until an age of 18 to 20 years, the patient needs to be followed up until this age and the team needs to

analyze functional and esthetic results in order to optimize future procedures.

Craniosynostosis involves numerous craniofacial deformities. To treat residual craniosynostosis deformities, various tools have been described (Fig. 16). Techniques differ in terms of operating time and invasiveness, but surgery is essential for the patient's future comfort. Techniques can be associated in a single step: e.g., combined hypertelorism and epicanthus correction, or rhinoplasty, onlay graft and orthognathic surgery [43].

Because craniofacial deformities can have psychological and social consequences, the child's demands and opinions are important to take into account in follow-up.

Disclosure of interest

The authors declare that they have no competing interest.

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