CASE REPORT

Subcranial Le Fort III Advancement with Equine-Derived Bone Grafts to Correct Syndromic Midfacial Hypoplasia: A Case Report

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Abstract Patients with hypoplasia of the midface normally present a flattening of their facial profile due to insufficient development of the nose and maxilla. Treatment aimed to restore function and an aesthetic appearance calls for a Le Fort III osteotomy and the advancement of the midfacial segment either through distraction or interposition of autogenous bone blocks. However, drawbacks in using autogenous bone suggest that use of alternative graft material may be advisable. The present report describes a Le Fort III advancement using two enzyme-treated equine cancellous bone blocks to correct syndromic midfacial hypoplasia in a 22-year-old patient. A 12-mm surgical advancement was achieved, improving the patient's facial profile. At the 28-month follow-up, the midface advanced position was stable. Equine bone blocks could be a valid alternative to autogenous bone in Le Fort III midface advancement.

Keywords Hypoplasia · Le Fort III · Achondroplasia · Bone substitutes

Introduction

Patients with hypoplasia of the midface present a typical clinical appearance due to missing or partial forward development of the nose and maxilla [1]. The related functional impairments and aesthetic stigma [2] may be corrected with the midface surgical advancement, performed following different techniques (Table 1). Among these, the most common involve advancement on the Le Fort III (LFIII) level, by using either traditional LFIII osteotomy or LFIII distraction osteogenesis (LFIII-DO) (Table 1) [3]. The mean midface advancement these two techniques allow to achieve is 9–12 and 9–16 mm, respectively, with LFIII-DO allowing a maximum advancement up to 22 mm [3]. LFIII-DO is therefore mandatory to treat cases calling for an advancement greater than 12 mm. LFIII-DO is usually applied to treat syndromic patients (e.g., affected by the Crouzon, Apert or Pfeiffer syndromes) presenting with severe obstructive sleep apnea syndrome (OSAS) and proptosis. Both approaches are effective, have good-to-excellent stability and medium rate of relapse [3].

Combined with autologous bone graft harvesting [4], traditional LFIII is generally considered more invasive and has been associated with greater soft-tissue tension and higher complication rates [5]. To overcome autograft-related drawbacks, the use of alternative materials may be advisable.

In the last few years, an enzyme-treated, collagen-preserving, equine-derived bone graft has been widely used in oral surgery [6–8] and orthopedic applications [9]. The native collagen conformation may allow the material to maintain mechanical resistance to compression and shaping. Moreover, the enzymatic treatment preserves the physiological recognition of the graft by osteoclasts, thus

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Table 1 Some surgical procedures to correct the midface deformities

Technique	Short description
Le Fort III (LFIII) osteotomy and the midface advancement	A Le Fort III osteotomy is performed and the Le Fort III segment is advanced <i>en bloc</i> [3]
Le Fort III distraction osteogenesis (LFIII-DO)	A Le Fort III osteotomy is performed and the Le Fort III segment is gradually advanced using external halo or buried fixators [3]
Frontofacial monobloc advancement	The frontal and facial bones are advanced as a monobloc [15]
Le Fort III minus I and Le Fort I midface advancement	A Le Fort III minus I and a Le Fort I segments are created and advanced separately [16]
Modified Le Fort III + I combined with distraction osteogenesis	A Le Fort I osteotomy is performed to advance the lower part of the midfacial segment using internal distractors, while the upper part is advanced <i>en bloc</i> , performing an osteotomy higher than the conventional Le Fort III one [17]
Le Fort II osteotomy and the midface advancement	A Le Fort II osteotomy is performed and the Le Fort II segment is advanced [18]
Monobloc minus Le Fort II midface advancement	A monobloc minus Le Fort II and a Le Fort II segments are created and advanced separately [19]
Le Fort I osteotomy and the midface advancement	A Le Fort I osteotomy is performed and the Le Fort I segment is advanced [20]

favoring bone remodeling [10]. The present report describes a case in which enzyme-treated equine bone blocks were successfully used in an LFIII surgery.

Case Presentation

The 22-year-old male patient was suffering from congenital achondroplasia. Due to the impaired development of the cranial base that characterizes chondrodysplasia, his facial profile was concave, with severe hypoplasia of the midface (Fig. 1a–c). In order to improve his appearance and provide better alignment between the upper and lower jaws, a treatment plan was developed that called for an LFIII osteotomy (Table 1) [3], with 12-mm advancement of the midface. Initial surgical planning envisaged (a) the insertion of two autologous bone blocks collected from the patient's iliac crest into the osteotomy sites at the two pterygomaxillary fissures and lateral orbital walls, and (b) the placement of a third block collected from the patient's calvaria on the glabella and nasal dorsum. The patient refused the collection from the iliac crest because of possible graft donor site morbidity. Therefore, surgeons proposed replacing the two autogenous bone blocks with enzyme-treated equine ones. The patient agreed to this alternative plan and provided his informed consent.

The patient underwent general anesthesia with nasotracheal intubation, while surgery was performed under temporary Altemir's submental intubation. A coronal incision was performed to expose the frontotemporal skull, lateral orbital regions, nasion and zygomatic arches and bodies, after which subcranial osteotomies were performed

according to the Le Fort III—Tessier III design. All osteotomies were performed with a piezoelectric device. A nasal septum U-shaped osteotome was used at the midline to separate the vomer and perpendicular ethmoid plate from the cranial base. The pterygomaxillary junctions were disjointed both through a bicoronal approach and a gingivobuccal access. Rowe forceps were used to mobilize the Le Fort III segment and advance the midface by 12 mm.

Two cancellous enzyme-treated equine bone blocks (Osteopiant, Bioteck, Arcugnano, Italy) (Fig. 2), sized 40 × 30 × 10 mm, were shaped and placed, one for each side, at the gap at the lateral orbital wall. To stabilize the advancement and the two grafts, two titanium plates were placed at the two frontozygomatic pillars. An autologous bone block (25 × 18 mm) was then harvested from the calvaria, molded and covered by a deep temporalis fascia graft and placed and fixed on the glabella and nasal dorsum with a four-hole titanium plate. An additional stabilization of the osteotomized maxillae was achieved using rigid intermaxillary fixation, which was maintained for 7 days, followed by elastic intermaxillary fixation for 3 weeks. The patient was seen every 6 weeks up to 28 months after surgery, during which he did not show any motor functional or sensory impairment.

The 12-mm midface advancement resulted in an improved facial profile with a reduction in concavity and better dental occlusion (Fig. 1d–f). Thanks to the graft at the nasal dorsum, the projection of the area increased. The enzyme-treated equine blocks could easily be visualized in the 3D CT (computed tomographic) scan, and radiograph was taken 9 days after the surgery.

Fig. 1 Preoperative and postoperative profiles of the patient. Picture (a, d), radiograph (b, e) and 3D computed tomography scan (c, f) before (a–c) and 9 days after the surgery (d–f)

Fig. 2 The cancellous equine-derived bone graft used in this study

At 28-month follow-up, the equine bone grafts appeared unchanged and still providing stabilization to the advanced midface. Comparison of the postoperative CT scan with the

one taken 28 months later (Fig. 3a–c) confirmed maintenance of the heterologous graft's shape and thickness over time. Twenty-eight months after surgery, the patient's facial appearance was a cosmetic success. The LFIII surgery resulted in a significant increase in the midface projection that was visible both frontally and laterally (Fig. 4a–d).

Discussion

The present case presents the use of an enzyme-treated, collagen-preserving, equine-derived bone graft to stabilize an LFIII midface advancement for the correction of a midfacial congenital hypoplasia. Traditionally, LFIII midface advancement procedures have been accomplished with autogenous bone blocks harvested from the iliac crest. Lacking any immunoreactivity and immunogenicity, autografts have a better chance of integrating, but are often correlated to higher risk of graft site morbidity [11]. Among possible alternatives, allografts present a residual risk of donor-to-recipient disease transmission, and continuous availability is strictly dependent on efficient tissue

Fig. 3 Computed axial tomography scanning. Scans taken before surgery (**a**), 9 days after surgery (**b**) and at 28-month follow-up (**c**) are shown. Arrowheads indicate the xenograft blocks

Fig. 4 Set of preoperative and postoperative photographs. Frontal (**a**, **b**) and lateral (**c**, **d**) views of the patient before (**a**, **c**) and 28 months after surgery (**b**, **d**)

banking [12]. Synthetic blocks feature insufficient mechanical resistance to compression and limited shaping, and their use in extensive maxillofacial reconstructions is not recommended [13]. Xenografts are good

osteoconductive scaffolds, manufactured by eliminating the antigenic organic bone components. Their resistance to compression and elasticity represents an advantage both during surgery and after implantation, especially in load-

bearing conditions. Different xenografts have been successfully employed in oral and maxillofacial surgery [14]. The one used in the present study displays significant remodeling properties [6, 7]. In the form of blocks, it has optimal mechanical features that allow for effective intra-surgical shaping and fixation [8]. More than 2 years after the LFIII osteotomy described in this case report, the improvements in the patient's appearance and masticatory/breathing functions remained stable. The two equine bone blocks were still integrated at the lateral orbital wall and malar-zygomatic osteotomy, contributing to the facial stability. This suggests that equine-derived bone blocks are a valid alternative to autogenous ones for stabilizing the midface advancement after an LFIII osteotomy. Their use in extensive cranio-maxillofacial surgery should be further investigated.

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Compliance with Ethical Standards

Conflict of interest Mattioli B, Iacoviello P, Aldiano C and Verrina G declare that they have no conflict of interest.

Ethical Approval For this type of study, formal consent is not required.

Informed Consent Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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