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ORIGINAL ARTICLES

Long-Term Outcomes of Free Gracilis Muscle Transfer for Smile Reanimation in Children

Jacqueline J. Greene, MD, Joana Tavares, MD, Suresh Mohan, MD, Nate Jowett, MD, and Tessa Hadlock, MD

Objective To evaluate long-term outcomes of free gracilis muscle transfer (FGMT) for smile reanimation on smile excursion, facial symmetry, and quality of life in a cohort of children with facial palsy.

Study design A retrospective analysis of 40 pediatric patients who underwent FGMT for facial palsy at the Massachusetts Eye and Ear Infirmary Facial Nerve Center was performed. Preoperative and postoperative photography and videography were used to quantify smile excursion and facial symmetry. Preoperative and postoperative quality of life was assessed with the Facial Clinimetric Evaluation (FaCE) survey, a validated, patient-based instrument for evaluating facial impairment and disability.

Results Of the 40 patients who underwent FGMT for facial palsy, 38 patients had complete data including preoperative and postoperative photography and videography from 3 months to 10 years following surgery; 13 cases had >5 years of follow-up. FGMT resulted in significant improvements in smile excursion within several months, with continued improvements in smile excursion and symmetry demonstrated more than 5 years later. Fifteen patients completed preoperative and postoperative FaCE surveys, which demonstrated significant improvement in quality of life scores following FGMT.

Conclusions FGMT significantly improves smile, facial asymmetry, and quality of life for years after this surgery for facial palsy. *(J Pediatr 2018;■■:■■-■■).*

Pediatric facial palsy is a rare[1] but devastating condition that causes physical deformity, functional deficits, including ocular complications, nasal valve collapse, and oral incompetence, and communication difficulties, including inability to express emotion and speech difficulty.[1,2] For patients who undergo facial reanimation surgery, long-term smile excursion outcomes and effects on craniofacial growth have not been comprehensively studied.[3-7]

The etiology of pediatric facial palsy is varied. Congenital causes of facial palsy include birth trauma, Moebius syndrome, unilateral lower lip paralysis, hemifacial macrosomia, Goldenhar-Gorlin syndrome, CHARGE (coloboma, heart defects, atresia choanae, retardation of growth and/or development, genital and/or urinary abnormalities, and ear abnormalities), Chiari malformation, syringobulbia, and Chapple syndrome.[8,9] Most cases of acquired pediatric facial palsy rising from infection resolve (otitis media and viral reactivation, eg, herpes varicella zoster, Epstein-Barr, HIV), whereas intracranial masses, iatrogenic injury, or trauma may lead to chronic residual facial palsy. The most common cause of pediatric facial palsy is debated in the literature, with multiple studies attributing 40%-50% of cases to Bell's palsy[10-13] and other studies attributing the majority of cases to infection and trauma.[14]

Free gracilis muscle transfer (FGMT), introduced in 1976 by Harii et al, is considered the gold standard functional procedure for facial reanimation of longstanding facial paralysis.[3-7,15,16] The gracilis muscle is a long, thin muscle in the medial thigh, where it is the most superficial muscle of the adductor compartment (**Figure 1**; available at www.jpeds.com). The vascular supply to the gracilis muscle flap is typically supplied on inset by the facial artery and vein. Neural innervation derives from either the contralateral facial nerve (cranial nerve VII) via a cross-face nerve graft (CFNG), which is placed in a first-stage procedure 6-9 months earlier, or the masseteric branch of the trigeminal nerve (cranial nerve V). A reliable procedure with low failure rates, FGMT will produce a meaningful smile (at least 3 mm of excursion with smile) in 84% of recipients when driven by cranial nerve VII via a CFNG and in 94% of recipients when driven by cranial nerve V.[16]

Improvements in smile excursion and oral commissure symmetry, as well as quality of life, are well recognized in both the adult and pediatric population following FGMT.[16,18-21] Benefits of FGMT innervated by cranial nerve V include a single-stage procedure and a higher success rate; however, patients are counseled that facial rehabilitation training will be necessary to activate a smile by biting. FGMT innervated by the contralateral cranial nerve VII via a CFNG confers a major advantage of smile spontaneity, but has a slightly lower success rate and requires a 2-stage procedure and harvest of the sural nerve from the lower extremity.

CFNG	Cross-face nerve graft
FaCE	Facial Clinimetric Evaluation
FGMT	Free gracilis muscle transfer
MEEI	Massachusetts Eye and Ear Infirmary

From the Massachusetts Eye and Ear Infirmary, Harvard Medical School, Boston, MA

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The objective of this study was to evaluate the long-term effects of FGMT on smile excursion, facial asymmetry, and quality of life in pediatric patients with facial palsy.

Methods

This study received Institutional Review Board approval from the Massachusetts Eye and Ear Infirmary (MEEI) Human Studies Committee. A retrospective chart review was conducted for all patients seen at the MEEI Facial Nerve Center between June 2003 and January 2018; records of patients with facial paralysis who underwent FGMT on or before the age of 18 were reviewed for 5-year follow-up data.

Data Collection

Demographic data, including age, sex, and etiology of facial paralysis, were recorded. Eyelid weight placement and/or removal was recorded. All FGMT procedures were performed as described previously.[22] Gracilis weight at the time of inset was recorded. The duration of follow-up after free gracilis muscle transfer with complete photography or videography was recorded.

The Facial Clinimetric Evaluation (FaCE) instrument[23] was used to assess quality of life for patients preoperatively and post-FGMT. The FaCE is a validated patient-based instrument for evaluating facial impairment and disability and assesses facial movement, comfort, oral function, eye comfort, lacrimal control and social function. All our patients had been asked to complete the FaCE instrument preoperatively and 5 years after FGMT.

Facial Analysis

Preoperative and postoperative photography and videography were used to measure baseline smile excursion and facial asymmetry using GIMP version 2.8.22 (a free online photography measurement program) and Emotrics version 2.0, a new automated facial analysis software developed at MEEI.[24] Emotrics is a machine-learning computer application that enables rapid automatic facial landmark localization and subsequent computation of facial measurements from standard clinical photographs (**Figure 2**; available at www.jpeds.com). All facial measurements computed in pixels were scaled to distances by conversion using a fixed iris diameter of 11.8 mm, as described previously.[16,25]

A horizontal interpupillary line was automatically plotted through the center of the pupils (**Figure 2**, line 1). This line was bisected by a perpendicular vertical line used to estimate the central axis of the face (**Figure 2**, line 2). Head yaw, pitch, and roll were kept neutral for photographs. Smile excursion was defined as the difference in commissure position relative to the central axis intersection with vermillion border with smile and at rest (line AB), as defined previously.[16,25] Smile asymmetry was defined as the absolute difference in commissure position of the healthy and affected sides at the time of maximum smile (in millimeters).[16,25] Patients with bilateral facial palsy or congenital hemifacial microsomia were excluded from symmetry analysis.

Statistical Analyses

Smile excursion and asymmetry at rest and smile were compared pre-FGMT and post-FGMT using a paired *t* test for the entire cohort (n = 38). For the patients with at least 5 years of follow-up, smile excursion and asymmetry at rest and smile were compared pre-FGMT, 3-18 months post-FGMT, and at least 5 years post-FGMT using a repeated-measures (within subjects) 1-way ANOVA, after the Shapiro-Wilk test to confirm normality and the Mauchly test to confirm sphericity. A post hoc pairwise *t* test was used to assess differences between timepoints. Pre-FGMT and post-FGMT FaCE instrument scores were compared with a paired *t* test. All statistical analyses were performed using R version 3.2.3 (R Foundation for Statistical Computing, Vienna, Austria).

Results

Of the 260 FGMT procedures performed since 2003, 40 were in patients aged ≤18 years at the time of FGMT. In this pediatric group, 38 patients (95%) had complete data photography and videography for their initial presentation and following FGMT. Two of these patients underwent FGMT bilaterally, for a total of 40 FGMTs. Twelve patients had complete data photography and videography at least 5 years following surgery; 1 of these patients underwent bilateral FGMT. Demographic data, including patient sex, laterality of facial paralysis, neural innervation, and etiology of facial paralysis are presented in the **Table**. Within this cohort, 17 patients had an eyelid weight placed, 2 patients had a weight placed and then subsequently

Table. Demographic and operative characteristics and etiology of long-term pediatric FGMT cohort

Variables	Values
Number of patients	38
Sex n (%)	
Male	22 (57)
Female	17 (45)
Age at FGMT, y, mean (SD) [range]	11.4 (3.7) [6-18]
Right-side FGMT, n	16
Left-side FGMT, n	20
Bilateral FGMT, n	2
Motor nerve, n	
Ipsilateral V	8
Contralateral VII	30
Ipsilateral V + contralateral VII	2
Weight of gracilis initial inset, g, mean (SD) [range]	22.8 (12.7) [6-46]
Follow-up after FGMT, y mean (SD) [range]	41 (32) [3.6-125]
Neural reinnervation failure, n (%)	4 (10.5)
Etiology of facial paralysis, n	
Intracranial neoplasm	
Congenital	9
Moebius	1
Iatrogenic	5
Idiopathic	1
Facial nerve schwannoma	2
Neurofibromatosis type II	2
Lyme disease	2
Temporal bone fracture	1
Otomastoiditis	1
Benign parotid mass	2

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removed, 1 patient received a PROSE lens, and 20 patients did not receive an eyelid weight.

Quality of Life Assessment

Fifteen patients had pre-FGMT and post-FGMT FaCE scores. The FaCE scores preoperatively were variable, representing the diverse stages of facial palsy presenting to the MEEI Facial Nerve Center (mean, 52 ± 17; range, 6-71; n = 15). Following FGMT, the FaCE score improved significantly (mean, 67.4 ± 15; range, 41-88; n = 15; $P = .015$).

Facial Analysis following FGMT

The average smile excursion improved significantly following FGMT (from −0.58 ± 3.5 mm to 5.9 ± 3.6 mm; n = 40; $P = 8.2e\text{-}12$) (**Figure 3**, A). The negative smile excursion on the affected side occurs in a flaccidly paralyzed face where the commissure position is displaced to the healthy side owing to unilaterally functioning facial muscles. Four FGMT recipients were deemed failures (<3 mm smile excursion at the latest follow-up appointment); of these, 3 were neurally innervated by cranial nerve VII and 1 was innervated by cranial nerve V. Rest asymmetry did not significantly change following FGMT (**Figure 3**, B). Smile asymmetry improved significantly following FGMT (from 13.8 ± 5.4 mm to 4.7 ± 3.6 mm; n = 34; $P = 2e\text{-}16$). The follow-up time for the post-FGMT group was on average 42 ± 33 months (range, 3.52 months to 10 years).

Long-Term (>5 Years) Smile Excursion

The average smile excursion improved significantly following FGMT in an early postoperative period (from −0.07 ± 3.8 mm to 5.5 ± 4.4 mm; n = 14; $P = .028$) (**Figure 4**, A) and was maintained after 5 years (6.4 ± 3.4 mm; n = 14; $P = .0013$). The average early postoperative period time point was 10.1 ± 5.8 months (range, 3-18 months), and the long-term follow-up time point was 6.6 ± 1.6 years (range, 5-10.3 years). For this long-term cohort, the average patient age was 9.7 ± 2.5 years (range, 6-13.8 years) at the time of FGMT surgery and 16 ± 2.7 years (range 11-21.4 years) at the long-term time point.

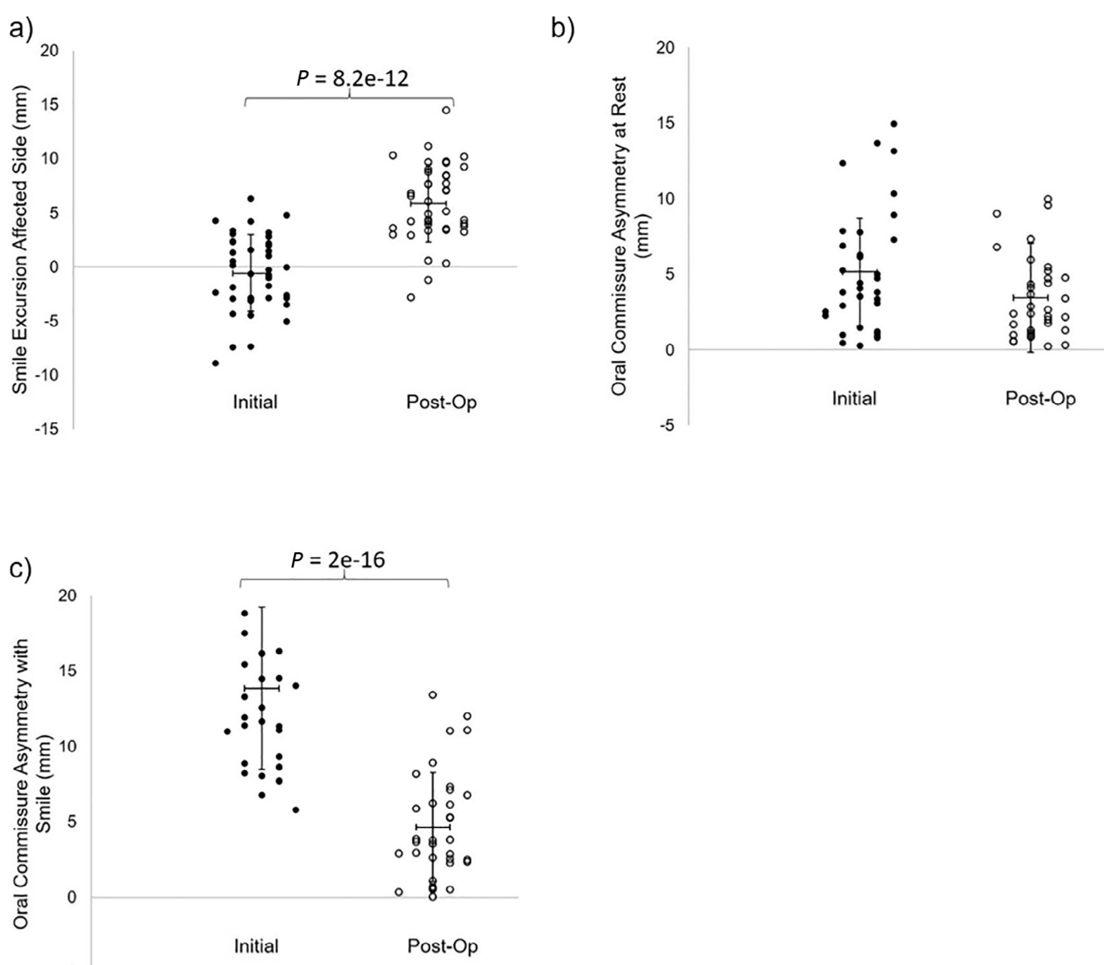

Figure 3. Facial analysis following FGMT. **A,** Smile excursion of the affected side improved significantly following FGMT. **B,** Rest asymmetry did not significantly change. **C,** Smile asymmetry improved significantly following FGMT.

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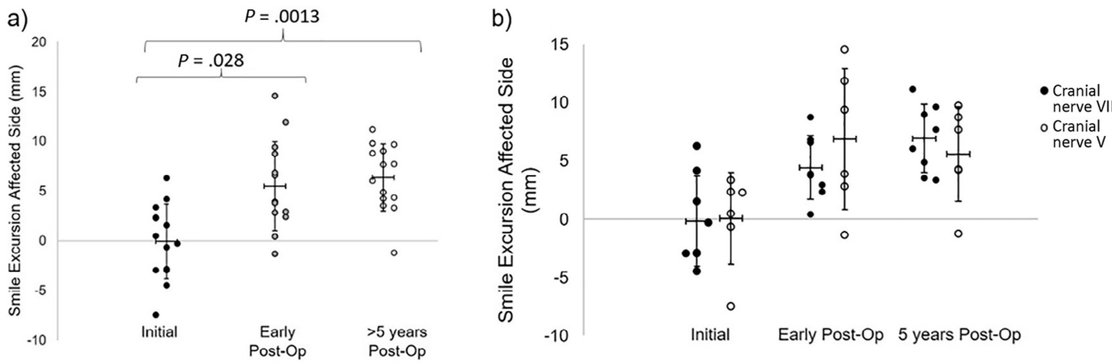

Figure 4. Long-term facial analysis following FGMT. **A,** Smile excursion of the affected side improved significantly following FGMT and more than 5 years later (n = 13). **B,** Smile excursion results represented by neural innervation. Masseteric innervated FGMT demonstrated initial gains in smile excursion followed by a plateau (open dots), whereas contralateral facial nerve innervation via a CFNG demonstrated progressive improvement over 5 years (black dots). The average early postoperative period timepoint was 10.1 ± 5.8 months (range, 3-18 months) and the long-term follow-up timepoint was 6.6 ± 1.6 years (range, 5-10.3 years).

Donor nerve innervation to the FGMT led to different long-term smile recovery trends (Figure 4, B). Innervation by the masseteric branch of the trigeminal nerve resulted in increased smile excursion in the early postoperative period (from 0.06 ± 4.0 mm to 6.9 ± 6.1 mm; n = 6) with a subsequent plateau long-term (5.6 ± 4.1 mm). One patient in this group that received a FGMT innervated by the masseteric nerve did not achieve a successful smile. Innervation by the contralateral facial nerve via a CFNG demonstrated progressive improvement in smile excursion over time (from −0.2 ± 3.9 mm to 4.4 ± 2.7 mm to 6.9 ± 2.9 mm; n = 8).

Long-Term (>5 Years) Oral Commissure Asymmetry at Rest and with Smile

Of the 14 FGMTs performed with ≥5 years of follow-up, 3 were excluded from smile asymmetry analysis due to bilateral facial palsy. Rest asymmetry did not change significantly following FGMT (Figure 5, A; available at www.jpeds.com). Smile symmetry did improve significantly following FGMT (from 13.8 ± 4.8 mm to 5.0 ± 2.3 mm; $P = .0007$) and was maintained for 5 years postoperatively (3.7 ± 2.9 mm; $P = .0001$) (Figure 5, B; available at www.jpeds.com). Smile symmetry was determined by the absolute difference of commissure position of the healthy and affected sides at the time of maximum smile, with a score of 0 indicating perfect symmetry.

Discussion

The goal of this study was to investigate long-term (>5 year) outcomes for pediatric facial reanimation. We analyzed 40 patients who underwent FGMT at age ≤18 years, 38 of whom had complete photography and videography datasets before and after FGMT and 15 of whom had 5-year follow-up data. Smile excursion improved significantly following FGMT and was maintained 5 years later, despite the patients' craniofacial growth and development. Innervation by the masseteric branch of the trigeminal nerve resulted in early smile recovery followed by a plateau, whereas neural innervation from the contralateral facial nerve demonstrated further improvements after 5 years. Smile asymmetry significantly decreased following FGMT and remained stable 5 years later. Failure of neural innervation resulting in smile excursion <3 mm was recorded in 4 patients (10.5%).

Long-term pediatric outcomes for smile reanimation are difficult to obtain owing to the small numbers of cases, the required follow-up, and transition of responsibility of patient care from the parents to the patient. Other large pediatric FGMT cohorts have provided excellent insights into the effect of neural innervation[26] and long-term subjective grading scores and electromyography motor units,[6] but long-term quantitative smile excursion has not been reported previously. Bianchi et al reported 1-year qualitative outcomes (ranked "excellent" to "poor") after FGMT in 17 pediatric patients with Moebius syndrome.[3] Zuker et al reported an average 2-month post-FGMT smile excursion of 1.37 cm in 20 pediatric patients with Moebius syndrome, and Ueda et al reported qualitative grade (1-5) outcomes of 21 cases of pediatric FGMT with follow-up of 2-13 years.[4,5] At the MEEI Facial Nerve Center, 18-month outcomes of a pediatric cohort of 19 FGMTs, including smile excursion and quality of life (FaCE scores), were found to match adult outcomes, albeit with fewer neural failures than in the adult cohort.[19] Bae et al provided quantitative outcomes data for 36 pediatric FGMTs and found greater oral commissure excursion after FGMT innervated by the masseteric nerve than after FGMT innervated by the contralateral facial nerve; however, the time at follow-up was not reported.[26] Terzis et al reported excellent improved subjective grading scores and motor units on electromyography at 5+-year outcomes in 13 cases of pediatric FGMT and 20 cases of pectoralis minor transfer, whereas quantitative assessment of facial displacements was not reported.[6]

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Figure 6. Photographs of an 11-year-old patient who developed slowly progressive right-sided facial paralysis and was found to have a facial nerve schwannoma. **A,** The patient is 8 months postexcision with cable graft repair, which was closely monitored for 12 months postexcision and found to plateau with this level of function (decent facial tone but asymmetric smile). **B,** The patient underwent FGMT by contralateral cranial nerve VII via a CFNG. The CFNG had been previously placed at the time of schwannoma excision in anticipation of possible future FGMT should insufficient recovery occur. In this photograph, she is 6 months post-FGMT and demonstrates improved smile excursion. **C,** The patient is 6 years post-FGMT (age 17) with a spontaneous smile, improved smile excursion, and facial symmetry.

Pediatric facial reanimation surgery presents unique challenges due to confounding craniofacial growth and shorter size of the gracilis muscle, neurovascular pedicle and sural nerves compared with adults. The ability to smile is critical for nonverbal communication, social interaction, and psychological health, particularly for the pediatric population. We have observed that young children with facial palsy often will give a maximum smile with dental show, but as they become older, they learn to moderate or sometimes even flatten their affect to avoid the disfigurement of facial paralysis. Quality of life measures can be difficult to obtain in the pediatric population in addition to the difficulty of obtaining longitudinal postsurgical data. Nonetheless, we were able to obtain FaCE scores preoperatively and 5 years postoperatively in 15 patients, which demonstrated significantly improved scores following FGMT.

Many patients and providers may be unaware of contemporary medical and surgical management options to address the devastating sequelae of facial palsy. At the MEEI Facial Nerve center, patients undergo facial physical therapy, which can greatly aid in reducing the disfigurement of facial paralysis, as well as train patients how to show their "best smile" naturally. As patients age, additional refinements may be performed on an outpatient basis, such as Botox denervation of smile depressors or for facial synkinesis, as well as resection of smile depressor muscles, which can often distort the smile. Although optimal timing for proceeding with FGMT for pediatric patients with chronic facial paralysis is a subject of debate, most surgeons recommend intervention with FGMT at or after age 6 years owing to neurovascular pedicle size as well as a greater social impact from facial palsy by that age (**Figure 6**).

Although sample size is a limitation of our study, our report nevertheless provides a relatively large cohort of pediatric long-term quantitative facial reanimation outcomes (3- to 18-month follow-up in 38 pediatric patients who underwent FGMT, and >5-year long-term follow-up in 13 patients). We found statistically significant improvement in smile excursion and sustained reduction in smile asymmetry 5 years postoperatively. Pediatricians should be aware of the possible surgical options available for sustained improvement of smile excursion and symmetry in children with longstanding facial palsy. ■

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Reprint requests: Jacqueline J. Greene, MD, Massachusetts Eye & Ear Infirmary, Harvard Medical School, 243 Charles St, 9th floor Facial Nerve Center, Boston, MA 02114. E-mail: jacqueline_greene@meei.harvard.edu

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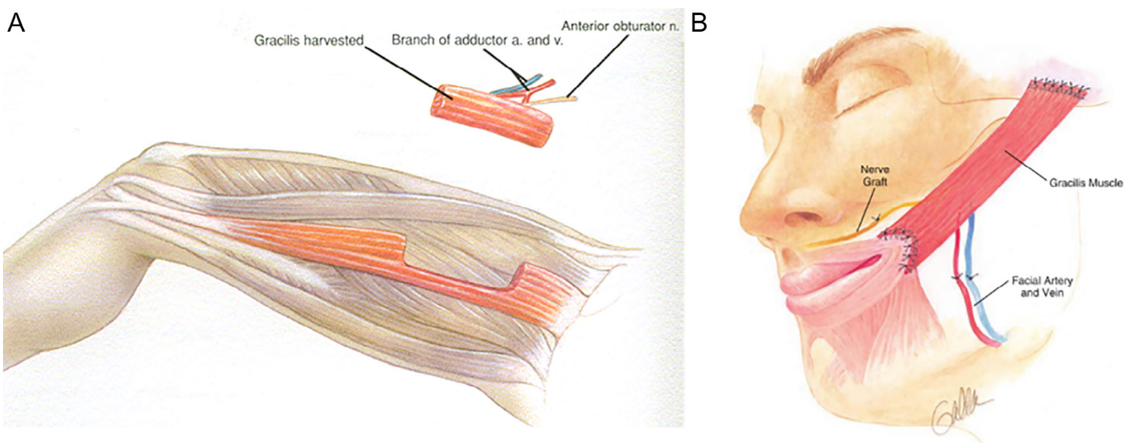

Figure 1. **A,** Gracilis harvest. The anterior aspect of the gracilis muscle has been harvested with its dominant pedicle, branches of the profunda femoris artery and vein, and its nerve supply, the anterior division of the obturator nerve. **B,** Gracilis inset. The gracilis muscle is medially inset to the orbicularis oris muscle and laterally to the temporalis fascia, while the obturator-CFNG neurorrhaphy is tunneled under the upper lip. (Reprinted with permission.[17])

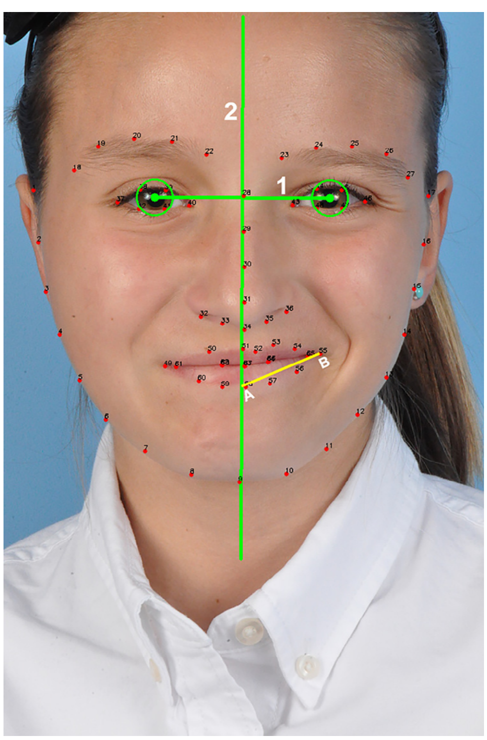

Figure 2. Automated facial analysis: The automated facial analysis program Emotrics[24] was used to automatically trace the contour of the face, interpupillary distance (line 1), and central vertical axis of the face (line 2). These landmarks were then used to calculate smile excursion and smile asymmetry. Smile excursion was defined as the difference in commissure position relative to the central axis intersection with vermillion border (line AB) with smile and at rest. Smile asymmetry was defined as the absolute difference in commissure position of the healthy and affected sides at the time of maximum smile (in millimeters).

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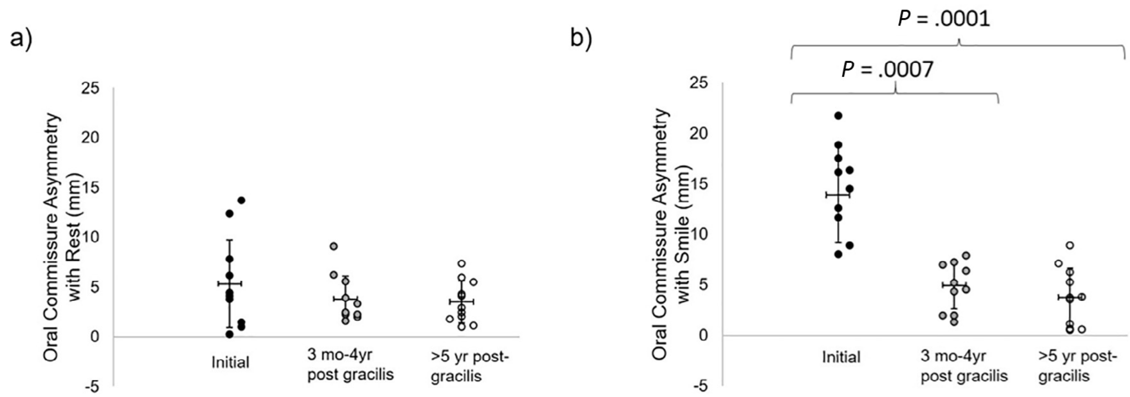

Figure 5. Long-term rest and smile asymmetry. **A,** No significant change in symmetry at rest occurred. **B,** Significant reduction in smile asymmetry was demonstrated at 3-18 months post-FGMT and maintained after 5 years (n = 11).

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