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Facial animation in children with Moebius and Moebius-like syndromes

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Abstract

Background: Moebius syndrome, a rare congenital disorder of varying severity, involves multiple cranial nerves and is characterized predominantly by bilateral or unilateral paralysis of the facial and abducens nerves. The paralysis of the VI and VII cranial nerves leads to a lack of function in the muscles they supply. Facial paralysis often causes bilabial incompetence with speech difficulties, oral incompetence, problems with eating and drinking, including pocketing of food in the cheek and dribbling, as well as severe drooling.

Methods: In this study, we report on pediatric patients with Moebius and Moebius-like syndromes seen and treated surgically from 2003 to September 2007 at the Operative Unit of Maxillofacial Surgery, Head and Neck Department, University of Parma, Italy.

Results: Twelve patients underwent microsurgical reconstruction for restoration of facial movement. The contralateral facial nerve was used as a motor donor nerve in 4 procedures, the motor nerve to the masseter muscle was used in 8 patients, and the gracilis muscle was used in all operations, with a total of 17 free-muscle transplantations. All free-muscle transplantations survived transfer, and no flap was lost. We observed a significant improvement in drooling, drinking, speech, and facial animation with a high degree of patient satisfaction.

Conclusions: The gracilis muscle free transfer is a surgical procedure well tolerated by the young patients and well accepted by their families. We consider it a safe and reliable technique for facial reanimation with good aesthetical and functional results in children with Moebius and Moebius-like syndromes.

Moebius syndrome/sequence is a rare congenital disorder of varying severity characterized predominantly by bilateral or unilateral paralysis of the facial and abducens nerves [1-7].

The paralysis of the VI and VII cranial nerves leads to the consequent lack of function in the muscles they supply. Moreover, lateral gaze and facial animation are absent. When the paralysis is bilateral, it can be asymmetric, and facial movement, when present, is always located in the lower face with platysmal activity or depressor anguli oris activity. However, effective lower-lip support, lower-lip elevation for bilabial speech production and commissure movement, and upper-lip elevation for smiling and emotional expression are absent and the inability of these patients to smile often lead to the mistaken impression that

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they are dull and uninterested. Speech difficulties are complex and multifaceted, and facial paralysis often leads to bilabial incompetence, which causes the characteristic speech pattern of flaccid dysarthria consisting of substitution, distortion, or omission of the bilabial phonemes /p/, /b/, and /m/ and the alveolar phonemes /t/, /d/, and /n/ [1,2,5,6,8]. Paralysis of the lower face often causes problems with eating and drinking, including pocketing of food in the cheek, dribbling, and severe drooling [1,9,10]. Other relevant clinical findings are incomplete eye closure and convergent strabismus. In addition to the abducens and facial nerves, the hypoglossal nerve is the most commonly involved cranial nerve, being affected in 25% of cases [1,9]. This manifests with further problems of articulation, primarily with vowels. Velopharyngeal incompetence is present in about 11% of patients because of glossopharyngeal nerve palsy, which results in a hypernasal voice, glottal substitutions, and increased nasal air emission [9]. Mental disability is overdiagnosed and is only present in 14% of patients [9]. This mistake is often because of the frequent inability of people to understand what these patients are saying or even feeling. When these patients begin school, they are often teased by other children, which can cause them to become withdrawn and reluctant to speak, despite normal intelligence. The inability to show happiness, sadness, or anger frequently results in severe introversion and a reclusive personality with low self-esteem [5].

Impairments of the II, V, X, and XI cranial nerves have also been observed in rare instances [1,5-7].

Patients with Moebius syndrome have been associated with additional deformities that include limb malformation (clubfeet and agenesis, rudimentary fingers or toes, and syndactyly or brachydactyly), malformations of the orofacial structures (bifid uvula, micrognathia, cleft palate, small palpebral fissures, epicanthic folds, ocular hypertelorism, microstomia, external ear deformity with occasional hearing loss, and airway problems with aspiration), musculoskeletal malformations (absence of the sternal head of the pectoralis major muscle, rib defects and arthrogryposis; even cases with dextrocardia have been described), and dysfunction of the cerebrum (mental retardation and epilepsy) [11,12]. Moebius syndrome may also be associated with Poland's syndrome, Klippel-Feil anomaly, Kallmann syndrome, and Hanhart syndrome [5].

The restoration of even a small degree of volitional facial movement can be rewarding for verbal and nonverbal communication. In this study, we report on 12 children with Moebius and Moebius-like syndromes seen and treated surgically from 2003 to September 2007 at the Operative Unit of Maxillofacial Surgery, Head and Neck Department, University of Parma, Italy. We review the surgical techniques used as well as the strategies of reconstruction surrounding their use and the outcomes. We focus on functional issues such as oral competence, speech, and the extent of animation and its impact on these patients. Both early and late complications are discussed.

1. Materials and methods

We reviewed the records of patients with Moebius syndrome evaluated between 2003 and September 2007 at the Operative Unit of Maxillofacial Surgery of the University of Parma, Italy. A standardized neurologic examination was performed in all cases. Facial expression, oral motor function, and speech were evaluated clinically, and most patients during their first office visit underwent electromyographic examinations.

We divided the patients as having Moebius and Moebius-like forms following the classification proposed by Terzis et al [5,13]:

- Moebius: complete bilateral facial and abducens nerve paralysis;
- Moebius incomplete: clinical picture of Moebius with the exception that some residual motor function was noted on one side of the face; and
- Moebius-like: unilateral facial paralysis, but additional cranial nerve palsies were present.

Patients satisfying the following criteria were excluded from "smile surgery":

- age less than 6 years;
- low grade of collaboration because of mental retardation;
- a good functional improvement of the residual motor units after logopedics;
- the involvement of more than one swallowing nerve,
- anomalies in walking; and
- absence of a collaborating family.

1.1. Surgical techniques

To reanimate the impaired side of the face or both sides of the face in patients with classic Moebius syndrome, we transplanted a segment of the gracilis muscle in all cases. Revascularization was via the facial vessels in all patients. For unilateral facial paralysis, the ipsilateral motor nerve to the masseter muscle or the contralateral VII nerve, by means of a cross-facial nerve graft, provided the preferred innervation of the muscle transfer. However, patients who have bilateral facial paralysis require a different nerve for innervation of the transfer. In these situations, we used the motor nerve to the masseter muscle.

1.2. Cross-facial nerve graft

On the normal side through a preauricular incision, the branches of the facial nerve are identified as they exit the anterior portion of the parotid fascia. With the aid of a nerve stimulator, a map of the muscles they innervate can be made, identifying the buccal and zygomatic branches. Segments of

these branches can be removed as their activity is duplicated. Three to 5 fascicles are then selected for division and routed into a sural nerve graft, which is tunneled across the face with the aid of nasal vestibule incisions. On the involved side, the nerve is placed high on the maxilla and zygoma to ease the dissection of this area at the time of muscle transplantation and to keep the nerve graft from being damaged. The regeneration of the graft can be followed by Tinel's sign. After 9 to 12 months, sufficient neural regeneration has occurred in the graft, and the nerve is capable of innervating a muscle transplant. During this second stage, the previously placed cross-facial nerve graft is evaluated histologically, and the best fascicles of the graft are selected to reinnervate the transplant.

1.3. Microvascular muscle transfer

The procedure begins with 2 teams operating simultaneously to elevate the muscle and prepare the face. The ispilateral gracilis is usually used for transfer. The gracilis is approached through a short medial thigh incision, posterior to the line joining the adductor tubercle to the medial condyle. The vascular pedicle is identified on the anterior border of the gracilis at the junction of the upper quarter and lower three quarters. The nerve to the gracilis from the anterior branch of the obturator nerve is traced to the obturator foramen. Only a segment of the muscle is needed. In fact, the use of the entire muscle leads to excess bulk. We have found that approximately one third to half of the muscle is needed to produce the appropriate amount of movement and to avoid excess bulk. The anterior third of the muscle is usually selected, and the fascicle that innervates this segment is identified and labeled.

Facial dissection is performed simultaneously while harvesting the gracilis. The facial incision is begun in the scalp near the upper pole of the ear, courses downward through the preauricular area, and then, after a small posterior curve, courses anteriorly in the neck with submandibular extension. A subcutaneous cheek flap is elevated extending superiorly to the body of the zygoma and the temple and anteriorly to the commissure and upper lip. The facial vessels are identified at the level of the anterior border of the masseter. Three to 5 sutures are placed for secure anchorage and careful positioning of the muscle. The first suture is placed in the oral commissure, the second one is placed in the lower lip, and the third to fifth sutures are placed in the upper lip. They are positioned so that they do not produce eversion or inversion. With traction, they should produce a nasolabial crease that looks as natural as possible. When the motor nerve to the masseter is selected to reinnervate the muscle transplant, it can be identified on the undersurface of the masseter muscle, usually coursing vertically downward at the posterior margin of the muscle just below the zygomatic arch.

The gracilis muscle is transferred to the face with the neurovascular pedicle on the deep surface, enabling subsequent debulking if necessary. The distal end of the muscle is fixed to the lips and oral commissure through the previously positioned sutures. Anastomoses are carried out between the facial vessels and the artery and the larger of the paired vena comitans of the gracilis. In 20% to 25% of patients, the facial vein cannot be found; in these cases, we must isolate the transverse facial vein. Once vascularization is ensured, a fascicular nerve repair unites the recipient nerve in the face to the selected fascicle of the motor nerve in the gracilis. When the donor nerve is the contralateral facial nerve, the nervous repair is performed in the pocket preformed in the superior vestibule of the oral cavity, where we find the previously placed cross-facial nerve.

After the neurovascular repairs, the muscle origin is secured. It is sutured with a slight degree of tension at the corner of the mouth and anchored to the temporal fascia and preauricular fascia. Figs. 1 and 2 show the position of the muscle in the pocket and the sites of neurovascular anastomoses.

Postoperatively, the patients are kept well hydrated, given a graduated diet, and maintained on bed rest for 3 days. Prophylactic antibiotics are used routinely and maintained for the first 72 hours. For early pain control, either a continuous morphine infusion or patient-controlled analgesia is used, depending on the age and capabilities of the patient. Being a microvascular free flap transfer, the patient for the first 2 days has an obligated posture of the head to avoid the flap and pedicle compression. Monitoring of the flap is performed with an eco-Doppler every 2 to 3 hours for the first postoperative 12 hours and every 6 hours for the following 24 hours. Mobilization is done with care on days 4 and 5, and on about days 7 to 8, the patient is discharged from the hospital. Chocolate, tea, coffee, and all drinks with caffeine are not avoided for the first month.

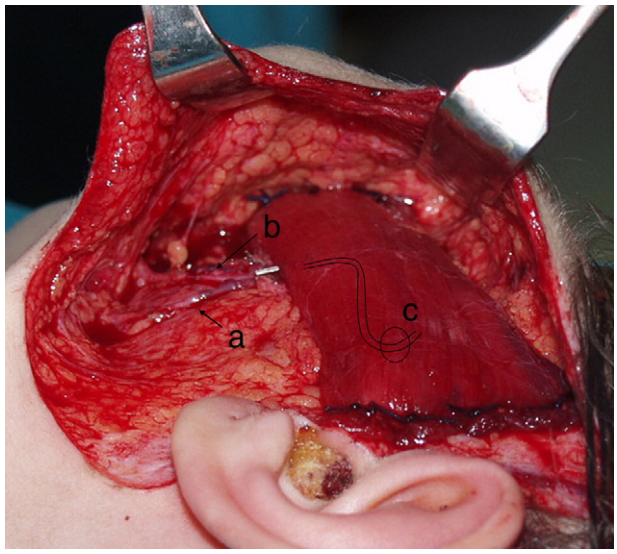

Fig. 1 Muscle position in the subcutaneous pocket with venous (a) and arterious (b) repair; design of the site of anastomosis with the masseteric nerve (c).

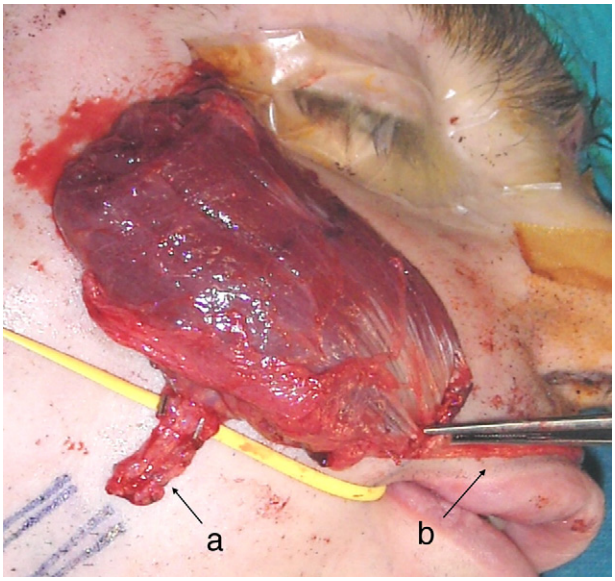

Fig. 2 Position of muscle and of vascular (a) and nervous (b) pedicles when the donor nerve is the contralateral facial nerve through a cross nerve graft.

Reinnervation of the muscle appears 3 to 6 months posttransfer.

To evaluate the functional and aesthetic results of microsurgical reconstruction, we only considered patients with a minimum follow-up of 12 months. The results were analyzed using patient response, clinical examination, and preoperative and postoperative videotaping of patients filmed at rest and performing several standard facial movements to show muscle action, spontaneity, independence, and fine facial movements during speech.

2. Results

Between 2003 and September 2007, 31 pediatric patients with Moebius syndrome were seen at the Department of Maxillofacial Surgery, University of Parma, Italy. They comprised 15 males and 16 females with a mean age, when first seen, of 7.3 years (range, 1-13 years).

The subjects had bilateral Moebius syndrome in 17 cases, and a monolateral form was present in 14 patients with involvement of the right side in 6 and the left side in 8.

We divided the patients into Moebius, Moebius incomplete, and Moebius-like as shown in Table 1 based on the classification proposed by Terzis et al [5,13].

One patient had a previous treatment in a different hospital through a cross-facial graft, without signs of reinnervation.

In those with the Moebius incomplete form, some facial movements were evident, which were always located in the lower face with platysmal activity or depressor anguli oris activity (Fig. 3). The abducens nerve was involved in 72% of the patients. In 11 patients, additional cranial nerve pools were involved, of which the hypoglossal nerve was impaired in 25% and the motor branches of the fifth cranial nerve in 4.2%.

Table 1 Patient classification

	n	%
Moebius	12	38.7
Moebius incomplete	4	12.9
Moebius-like	15	48.4
Total	31	100

We observed alterations of speech patterns with articulation difficulties and substitution or distortion of the bilabial phonemes /p/, /b/, and /m/ in 40% of the patients. Speech was severely altered in 10% of the subjects. Associated findings were observed in 8 patients (25.8%) and are shown in Table 2.

Of the 31 patients evaluated, 12 met the criteria for inclusion in "smile surgery" and underwent microsurgical reconstruction for restoration of facial movement. The contralateral facial nerve was used as a motor donor nerve in 4 procedures, the motor nerve to the masseter muscle was used in 8 patients (5 with bilateral and 3 with monolateral facial paralysis), and the gracilis muscle was used in all operations, with a total of 17 free-muscle transplantations. We performed bilateral free-muscle transplantations in 5 patients. The facial artery and vein were used as recipient vessels in most of the procedures. In one patient, we could not find the facial vein, and we used the transverse facial vein.

In this series, all free-muscle transplantations survived transfer, and no flap was lost. One seroma was seen at the inner thigh after a gracilis muscle harvest, which was treated by aspiration and pressure wrapping. In one patient, who underwent a cross graft, the muscle was well vascularized but did not show signs of reinnervation despite a new operation to revise the nervous anastomosis. In this case, we successfully perfomed, 4 years later, a new gracilis transplantation reinnervated by the masseteric nerve. Three patients developed scars that underwent hypertrophy at the site of the neck incision. Reinnervation of the gracilis muscle took about 3.5 months. Through direct questioning regarding functional problems, all patients with preoperative difficulties regarding oral competence reported a significant improvement with no spontaneous drooling and with adequate capability to control oral fluid. The patients with preoperative abnormalities in speech had an improvement in articulation and in pronunciation of bilabial phonemes (Figs. 4 and 5).

In all, 83.4% of the subjects were happy with the aesthetic and functional results, stated that they would undergo the surgery again, and reported improvement in self-esteem. The long complex surgical procedures were well tolerated by the young patients and well accepted by the families. On the basis of the evaluation criteria previously proposed by O'Brien et al [11], we obtained the results shown in Table 3.

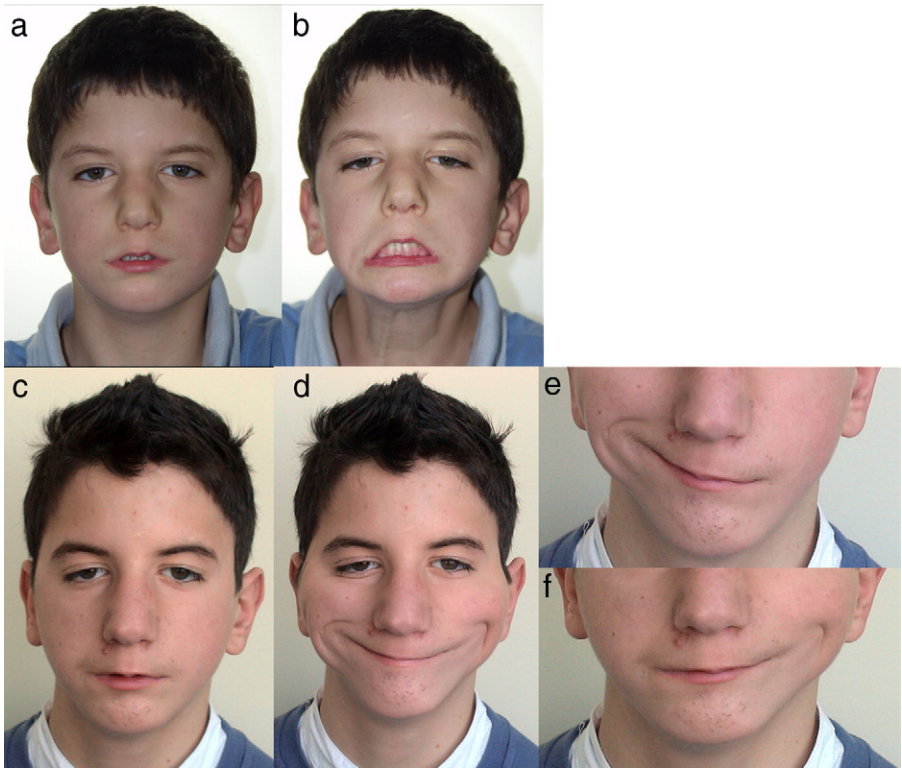

Fig. 3 Seven-year-old male patient with a Moebius incomplete form. Preoperative appearance at rest (a) and while attempting to smile (b) showing some facial movements located in the lower face bilaterally, with depressor anguli oris activity. Three years later, postoperative view at rest and while smiling (c and d) with independent bilateral contraction (e and f). The patient underwent a gracilis muscle transplant both on the right and left sides at 6-month difference one from the other.

Facial symmetry at rest was excellent or good in all patients, and 83.4% maintained good symmetry while smiling. One patient did not have any muscle movement and in one case we observed, involuntary movements and dyskinesias. The overall final results were excellent—good in 83.4% of the patients and fair to poor in 16.6%.

Table 2 Associated findings in young patients with Moebius and Moebius-like syndromes

	n	%
Total of patients with associated findings	8	25,8
Patients operated with associated findings	3	25
Associated findings	n	
Poland's syndrome	2	
Gastroesophageal reflux disease	1	
Cleft palate	1	
Syndactyly	2	
Clubfeet	4	
Micrognathia	7	
Testicle anomalies	1	
Angel wing	1	
Mental retardation	1	
Hearing reduction	1	

3. Discussion

The lack of facial animation in patients with Moebius syndrome poses a major barrier to interpersonal communication and creates severe aesthetic and functional problems. Microneurovascular transfer of a free-muscle transplant is currently the procedure of choice for facial paralysis. The gracilis muscle represents the first choice for facial animation, having the advantages of easy access, dispensability, and appropriate vasculature for free transfer [11,14], and our experience confirms these qualities.

Despite the advances made for the last few years with this surgical approach, problems associated with orientation, bulk, and excursion still remain [15]. Because of the imprecise nature of the muscle excursion, growth, and reinnervation, it is difficult to obtain accurate symmetry of movement and consistently produce a nasolabial crease. This, however, can be overcome through a careful preoperative study of the patient, precise selection of the motor nerve, good positioning of the muscle, and continuous postoperative smile and speech training [15].

Preoperative planning is extremely important, and detailed assessment of the location and direction of desired movement should be made [15]. Before the free-muscle transplant, we usually draw lines on the patient corresponding to the

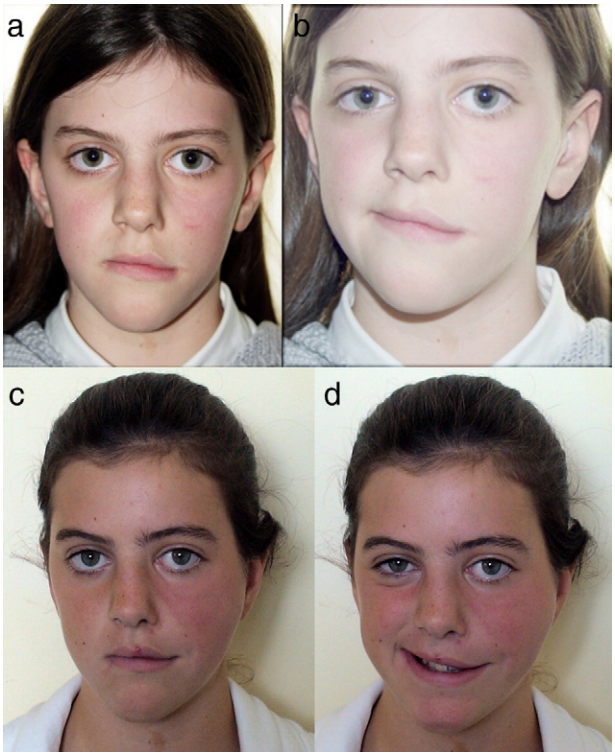

Fig. 4 Eleven-year-old female patient having a Moebius-like form on the left side. Preoperative view at rest (a) and smiling (b). Postoperative view at rest (c) and smiling (d) 2 years later, the reanimation achieved with a the gracilis free-muscle transplant reinnervated by the right facial nerve through a cross graft from the sural nerve.

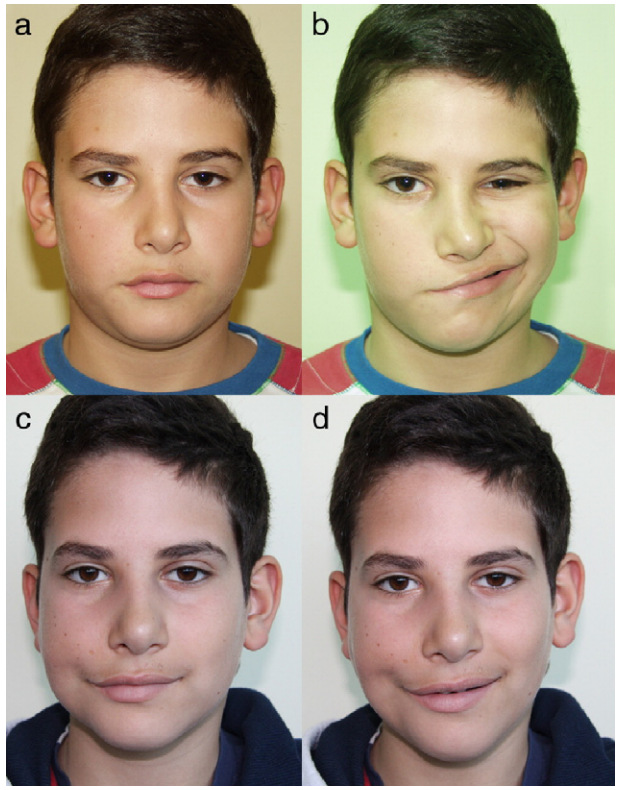

Fig. 5 Eight-year-old male patient with a Moebius-like form on the left side. Preoperative view at rest (a) and smiling (b). Postoperative appearance at rest (c) and smiling (d) 11 months later, the reconstruction achieved with a gracilis free-muscle transplant reinnerveted by the ipsilateral masseteric motor nerve.

nasolabial crease and to the vector of the smile from the oral commissure toward the tragus. These lines direct our positioning sutures on the oral commissure and lips and aid in orienting the gracilis muscle. With traction, the sutures should in fact produce a nasolabial crease that looks as natural as possible. Moreover, it is important to ensure that the direction of contracture of the muscle produces movement that will simulate a normal smile, or in monolateral forms, the contralateral side. Lack of accuracy in muscle positioning will lead to asymmetrical movement, and the most common fault is placing the muscle too transversely.

Excessive bulk at the site of the transplant should also be avoided. According to Zuker et al [16], during dissection, we removed 2 to 6 g of fat from the subcutaneous tissue of the cheeks and Bichat's fat pad. We used approximately one third of the width of the normal gracilis, and we fixed the origin of the muscle on the inferior surface of the zygomatic arch and zygoma, thus, avoiding the problem of muscle protuberance. If the muscle origin is placed on the superficial aspect of the zygomatic arch and zygoma, excess bulk will in fact be apparent. In some patients, we also spread the proximal end of the muscle before anchoring it to the temporal and preauricular fascia.

The selection of a motor nerve is critical to the success of the procedure. Not only must sufficient innervation be present to power the muscle, but also this innervation must be specific to the desired activity to achieve spontaneity, synchronicity, and symmetry. The contralateral seventh nerve, by means of a cross-facial nerve graft, provides the preferred innervation for a muscle transfer in unilateral facial paralysis reconstruction [10,15,17-21]. When the VII nerve is not adequate in a person who has a very powerful smile on their normal side or in patients with bilateral forms, we used the motor nerve to the masseter muscle. As in earlier reports, we noted that in the cross-facial nerve graft group, the extent

Table 3 Clinical results

Parameters	%			
	Excellent	Good	Fair	Poor
Symmetry at rest	83.3	16.7	0.0	0.0
Symmetry smiling	66.7	16.7	8.3	8.3
Muscle action	83.4	0.0	8.3	8.3
Muscle bulk	66.7	33.3	0.0	0.0
Independent action	58.1	25.0	8.3	8.3
Commissure elevation	58.1	25.0	8.3	8.3
Mouth closure	83.4	0.0	8.3	8.3
Involuntary movement	91.7	0.0	0.0	8.3
Overall final results	75.1	8.3	8.3	8.3

of oral commissure movement on the side that was operated on was much less than that of the masseteric group [10,18]. However, the cross-facial nerve graft procedure provides a spontaneous movement that cannot be provided by the motor nerve to the masseter muscle [18,20]. In children, this spontaneity is crucial in producing a normal-appearing smile. Nevertheless, the excursion produced by the motor nerve to the masseter is in the normal range and certainly superior to that provided by a cross-facial graft input from the opposite side [18,21]. Although it does not provide true spontaneity of activity, these children can achieve a good appearance of a smile. In our experience, as in other studies [10,21], most of these patients are able to develop facial movement independent of jaw motion and no longer have to think about moving their face while laughing.

To achieve a spontaneous and symmetrical smile, an important role is played by postoperative smile training by a speech-language pathologist using mirror exercises and biofeedback. When reinnervation of the muscle appears 3 to 6 months posttransfer, an active exercise program must be started; in this way, progress in strength, excursion, and most important, symmetry can be achieved. The age of the patient when operated on did not influence the results from an aesthetic or functional point of view. In children, we observed a shorter time of reinnervation (3.5 months) than in adults (5-6 months).

At the end of the follow-up, we observed a significant improvement in drooling, drinking, speech, and facial animation. In patients with difficulties regarding oral competence, we had adequate capability to control oral fluid and no spontaneous drooling. When preoperative abnormalities in speech were present, we observed an improvement in intelligibility and bilabial competence. Confirming the results obtained by Zuker et al [15,16,21], we obtained a high degree of patient satisfaction; most were happy with the results and reported improvement in self-esteem and social interaction.

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