One-Stage Reconstruction for Bilateral Möbius Syndrome

Simultaneous Use of Bilateral Spinal Accessory Nerves to Innervate 2 Free Muscles for Facial Reanimation

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Background: Möbius syndrome (MS) can present with unilateral or bilateral facial paralysis. In performing 1-stage bilateral MS facial reanimation, we used bilateral spinal accessory (XI) nerves to innervate 2 free functional muscle transfers (FFMTs).

Methods: Of 12 MS patients, 6 had bilateral facial paralysis. Bilateral gracilis were transferred and innervated using bilateral XI nerves. Results were evaluated using smile excursion score, cortical adaptation stage, and patient satisfaction questionnaire.

Results: In all, 13 FFMTs were performed (with 1 gracilis failure). Mean smile excursion score improved from 0.7 to 3.4 (out of 5) postoperatively. Four patients achieved spontaneous smile, 1 achieved independent smile, and 1 achieved dependent smile. Mean satisfaction score was 2.8 (out of 5).

Conclusions: One-stage bilateral FFMTs neurotized by bilateral XI nerves are effective in treating bilateral MS patients. Careful patient selection, adequate neurologic and psychologic examination, and postoperative smile training are all important factors in achieving optimal outcomes.

Key Words: Möbius syndrome, spinal accessory nerve, free functional muscle transfer (FFMT)

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Möbius syndrome (MS) is a rare congenital disorder characterized by unilateral or bilateral facial paralysis and inability for lateral gaze secondary to palsies of the facial (VII) and abducens (VI) cranial nerves.[1,2] The exact cause of this disease is still unknown, and the inclusion of various anomalies to its presentation has rendered it more confusing.[3] Our diagnostic requirements are similar to the Verzijl et al[2] criteria, including facial palsy with impairment of ocular abduction, and other abnormality such as musculoskeletal anomalies as mentioned in Abramson et al classification system.[1] MS patients rarely present with severe neurologic deficiencies and have a normal life span. However, they are often discriminated against because of their inability to convey emotions through facial expression, especially in bilateral MS patients with bilateral facial palsy.

The role of reconstructive surgeons is to restore MS patients' ability to express emotions. Since the advent of nerve transfer[4] and free functioning muscle transfer (FFMT),[5–14] there have been tremendous gains in the treatment of facial palsy. Smile reconstruction using the 2-stage[5–9] or 1-stage FFMT[10–14] approach has been reported. However, in bilateral MS with bilateral facial palsy, the reconstruction poses more challenges and controversies, especially on the timing between the 2 FFMTs and the choice of neurotizers. In this article, we describe our 1-stage operative method of using bilateral spinal accessory (XI) nerves to neurotize 2 FFMTs for smile reconstruction.

MATERIALS AND METHODS

Between January 1986 and December 2010, 305 facial palsy patients were enrolled at Linkou Chang Gung Memorial Hospital and were operated on by the same surgeon (D.C.-C.C.) using FFMTs. Twelve patients (4.0%) were diagnosed with MS, and 6 patients presented with bilateral facial palsy. Detailed physical and neurologic examinations were performed with special emphasis on which cranial nerves were involved. Each patient was classified using the Abramson CLUFT grading system[1] (Table 1). The hypoglossal nerve was involved in 3 patients and the trigeminal nerve in 1 patient. None of these patients showed absence of XI nerve function. They all received 1-stage reconstruction using bilateral XI nerves to innervate 2 FFMTs for smile reconstruction. The mean postoperative follow-up time was 1694 days (range, 383–3727 days, Table 1), with a minimum of 1 year after surgery.

Surgical Techniques

The operation consisted of simultaneous harvesting of both gracilis muscles and dissection of both sides of the face by 2 or 3 teams, 1 for face and 1 or 2 for both gracilis muscles dissection. Adult patients were anesthetized by nasotracheal intubation to minimize interference with lip manipulation, whereas children were intubated by orotracheal intubation because of their nostril size limitations.

In the frog leg position, a 10-cm long incision was made along the upper medial thigh, posterior to the adductor longus muscle to expose the proximal gracilis muscle and its neurovascular pedicles. The proximal fascial aponeurosis of the gracilis was dissected as proximal as possible to its bony insertion. The motor nerve of the gracilis muscle was traced under the adductor longus muscle up to the obturator foramen where the obturator nerve bifurcated into the medial and lateral branches. The vascular pedicle, usually including 1 artery and 2 accompanying veins, was dissected up to the bifurcation. The gracilis muscle was trimmed into a trapezoid shape based on the inside face measurement[9,15] and left in situ until the facial recipient site was ready. The same procedure was repeated on the opposite thigh.

At the facial recipient site, 2 incisions were made: one small incision along the white line of the upper lip, and the other longer incision from the preauricular region down to the submandibular area. Subcutaneous dissection extended from the infraorbital rim superiorly to the mandibular margin inferiorly and to the white line incision of the upper lip medially. Under the facelift skin flap, the facial artery and vein were identified and dissected distally (for a

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TABLE 1. Demographics of Patient Group

Case	Gender	CLUFT Classification		Age of FFMT (Years Old)	Length of Follow-up (Days)
1	M	C: 7 bt, 6 bt, 5 bt	V3, Micrognathia	21	3727
2	F	C: 7 bt, 6 bt		21	2159
3	M	C: 7 bt, 6 bt, 12 bt		20	1434
4	M	C: 7 bt, 6 bt, 12 bt	V3, Micrognathia	15	1105
5	M	C: 7 bt, 6 bt, 12 bt		5	1357
6	F	C: 7bt, 6 bt		52	383
Mean				22.5	1694

M indicates male; F, female; C, cranial nerves; bt, bilateral; FFMT, functioning free muscle transplantation; V3, mandibular nerve of the trigeminal cranial nerve.

distance of about 5 cm), cut, and transposed. A pocket was created for the muscle placement from the infrazygomatic margin down to the lip following soft-tissue removal. A third incision, either transverse or oblique, over the ipsilateral neck was made to identify the XI nerve (Figs. 1A, B). The XI nerve could be found on the deep surface of the lateral trapezius muscle and confirmed by nerve stimulator. Dissection of the XI nerve should be carried as distal as possible until reaching 2 or 3 terminal muscular rami (or when ~10 cm of the nerve was free). The distal end of the XI nerve was transected and transferred to the mandibular angle through a tunnel created deep to the platysma. The same procedure was repeated on the opposite neck.

After both sides of the face and both gracilis muscles have been prepared, the left gracilis was harvested and transferred to the right side of the face and placed in retrograde manner as follows: the proximal thinner cut end (tendinous aponeurosis) was anchored to the posterior wall of the orbicularis oris muscle, and the distal thicker end (muscle belly) was fixed to the periosteum of the zygomatic arch. The facial vessels were anastomosed to the vascular pedicles of the anchored gracilis muscle, and the XI nerve was coapted to the motor nerve of the gracilis. After completing the transplantation, the right face wounds were closed. The right gracilis muscle was then transplanted to the left face in the same manner.

Postoperative Management

After surgery, the neck was immobilized with a prefixed neck splint for 3 weeks. Electric stimulation was initiated 3 weeks after surgery. At approximately 4 months postoperatively, "induction exercise"[9] was begun, during which the patient was instructed to perform shoulder elevation with resistance (trigger movement) to enhance the movement of the transferred muscle. Once the transferred muscle achieved upper lip movement with lateral incisor visible, "smile training" was prescribed to achieve independent movement. The smile training was performed in front of a mirror.

Outcomes Assessment

All patients were followed up for at least 1 year. The patients were asked to smile with maximal excursion, and pictures were taken before and after surgery. Additional videos were also taken. Outcomes were assessed with a new score system (smile excursion score) based on the visibility of teeth during a wide smile (Table 2). The score included 5 categories: score 0, no tooth; score 1, 1 tooth or medial incisor; score 2, 2 teeth or lateral incisor; score 3, 3 teeth or canine; and score 4, 4 teeth or premolar tooth visible. But if the smile is accompanied by either contracture or synkinesis, 0.5 will be deducted for each. A score was given to each side of the face. A postoperative score of 2 or higher was considered successful. Independent reviewers other than the operating surgeon scored these patients based on the pictures and videos taken.

To evaluate the functional progress of the transferred muscle, a new staging system (cortical adaptation stage) was designed to classify the different stages of smile recovery (Table 3). This system included following 5 stages: stage I, no smile or movement of the transferred muscle; stage II, dependent smile with need of trigger movement; stage III, independent smile with a latency; stage IV, spontaneous or immediate smile with present involuntary movement; and stage V, spontaneous smile with absent involuntary movement. Stages I and II were considered poor, whereas stage III and higher were considered satisfactory outcomes.

A questionnaire to evaluate patient satisfaction was performed (Table 4). The questionnaire included the patient's complaint, self-image, use of the newly acquired smile, the possible complications and symptoms associated with the harvest of the XI nerve, and a satisfaction score (5 was maximum). The questionnaire was completed by an independent reviewer via a phone interview, and a written document was mailed to the participants' homes for any additional input. All 6 patients consented to be interviewed for the questionnaire and granted permission for their answers to be used. For patients aged <10 years, their representative guardian gave answers.

RESULTS

A total of 13 FFMTs (12 gracilis muscles, 1 rectus femoris muscle) were performed. One patient (case 3) had undergone an additional surgery because of 1 gracilis failure (Table 5). The subsequent second operation was performed 1 year later. The segmental rectus femoris muscle was reinnervated by the previous XI nerve. Of the original 12 FFMTs, the facial vessels were used in 10 FFMTs, whereas superficial temporal vessels were used in 2 FFMTs for 1 patient (case 4) because the facial vessels were deemed unreliable for anastomoses.

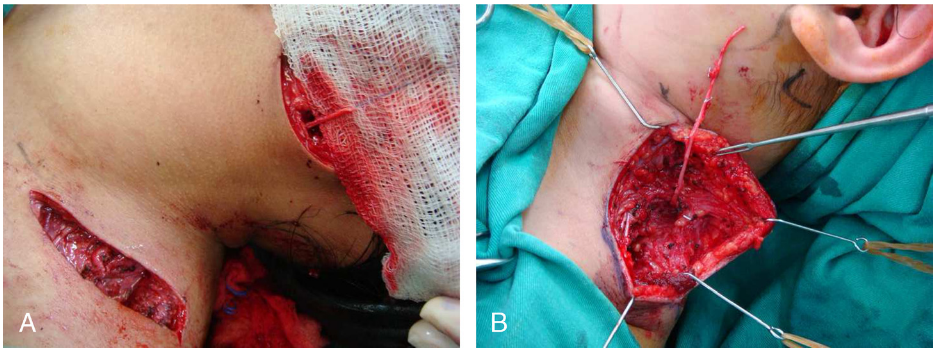

FIGURE 1. The spinal accessory nerve has been dissected through transverse (A) or oblique (B) neck incision, cut distally, and transposed to the mandibular angle.

TABLE 2. Chuang's Smile Excursion Score

Score	Teeth Visible	Contracture	Synkinesis
0	Not visible	−0.5	−0.5
1	First incisor	−0.5	−0.5
2	Second incisor	−0.5	−0.5
3	Canine	−0.5	−0.5
4	Premolar or more	−0.5	−0.5

Acceptable result, total score ≥2 postoperatively.

TABLE 3. Chuang Cortical Adaptation Stage

Stage	Description
I	Inability to smile (no movement of the muscle)
II	Dependent smile (movement) with need of inducing movement to initiate the FFMT
III	Independent smile (movement) of the FFMT with a latency
IV	Spontaneous smile without having to think about it, but still present involuntary movement
V	Spontaneous smile with absent involuntary movement

Acceptable result, stage ≥III postoperatively.
FFMT indicates functioning free muscle transplantation.

All patients achieved at least score 2 on "Smile Excursion Score." The mean score improved from 0.7 preoperatively to 3.4 postoperatively. On the "Cortical Adaptation Stage," cases 4 and 5 (ages, 16 and 5), had excellent results of spontaneous smile with absent involuntary movement (stage V). Cases 1 and 2 (both, 21 years old) had spontaneous smile with involuntary movement of the cheek during abduction of the shoulder (stage IV). Case 3 who underwent second FFMT (rectus femoris) was able to show independent smile without moving the shoulder (stage III). Only case 6 showed smile dependent on shoulder movement (stage II). This may be attributed to her short follow-up. She is still in rehabilitation for smile training. There was a noticeable stepwise progression of the smile excursion score and cortical adaptation stage in all patients during the follow-up periods. In adult patients, it usually took 1 year or longer to achieve independent smile; but in children, it usually occurs spontaneously within a year.

However, the average satisfaction score was 2.8 of 5. The primary concern for dissatisfaction is mostly aesthetic, although 1 patient (case 3) was bothered more by the differential functional recovery of the FFMTs. Five patients (83%) reported symmetry of both transplanted muscles, and all patients were able to perform a controlled and synchronous smile. All reported willingness to use their newly acquired smile in front of family and friends, but not in an unfamiliar environment with strangers. Half of patients complained of initial shoulder pain, but reported alleviation of symptoms over time.

CASE REPORTS

Case 1

A 21-year-old man with bilateral MS presented with bilateral VII, VI, and V cranial nerve palsies. No teeth were visible with attempted smiles (Fig. 2A). He was initially referred to a craniofacial surgeon for jaw correction surgery (midface degloving procedure and Le-Fort I osteotomy). After his recovery from the first surgery, we performed the 1-stage bilateral smile reconstruction. Both FFMTs were inducible by shoulder abduction at 6 months postoperatively. At

TABLE 4. Patient Questionnaire

Facial paralysis recovery
1. Primary complaint
   a. What are you most unsatisfied about the results after the surgery?
   b. Is it a functionally or aesthetically based complaint?
2. Do you think you have a symmetric smile?
   When you smile, do both sides of your smile move synchronously?
3. Do you use your smile in daily activities?
   a. Do you use it in front of family and friends?
   b. Do you use it in front of strangers or public situations?

Spinal accessory nerve and Shoulder function
1. Shoulder abduction
   a. What is the range of motion of the involved shoulder when abducting laterally?
   b. What is the degree of shoulder abduction that can involuntarily initiate the excursion of the functionally free muscle?
2. Shoulder dysfunction
   a. Is there an obvious shoulder drop on the same side which the donor nerve was harvested?
   b. Is there difficulty trying to reach for objects above shoulder level?
   c. Are you unable to keep your shoulder abducted for >30 s?
3. Shoulder discomfort
   a. Is there shoulder pain on the same side which the donor nerve was harvested?
   b. Do you feel tightness of the shoulder?
   c. Do any of these discomforts or dysfunctions detract you from using this shoulder?

Satisfaction score
- 1: Regrets the surgery
- 2: Not acceptable, but does not regret surgery
- 3: Acceptable, but not the smile I was expecting; Needs more improvement
- 4: Satisfied; needs only minor improvements
- 5: Completely satisfied

10-year follow-up, his smile excursion score improved from 0 to 3, and the cortical adaptation stage improved from stage I to IV (Fig. 2B). The patient remained concerned about his facial aesthetic appearance.

Case 2

A 21-year-old woman with bilateral MS presented with bilateral VII and VI cranial nerve palsies. Only first incisors were visible with attempted smiles (Fig. 3A). One-stage bilateral smile reconstruction was performed. The FFMTs showed movement at 3 months and gained independence from the induction exercise at 6 months postoperatively. At 6-year follow-up, her smile excursion score improved from 1 to 3, and the cortical adaptation stage improved from stage I to IV. She remained unsatisfied with her facial aesthetic appearance (Fig. 3B).

Case 3

A 20-year-old man with bilateral MS presented with bilateral VII, VI, and XII cranial nerve palsies. He became increasingly frustrated with having trouble communicating at work and at home. Only first incisors were visible on smiling. One-stage bilateral smile reconstruction was performed. The right gracilis failed and was removed on postoperative day 5. One year later, the second operation was performed using left segmental rectus femoris muscle for right facial reanimation, neurotized using previously dissected XI nerve. Superior thyroid artery with vein graft was used for anastomosis. Three years after the repeated FFMT surgery, the right face showed less excursion than the left. The smile excursion score improved from 1 to 3 on the

TABLE 5. Choice of Muscle, Neurotizer, and Vessels for Free Functioning Muscle Transplantation

Case	Side	Muscle	Neurotizer	Artery	Vein
1	Bilateral	Gracilis × 2	CN XI	FA	FV
2	Bilateral	Gracilis × 2	CN XI	FA	FV
3*	Bilateral	Gracilis × 2	CN XI	FA	FV
		Rectus femoris × 1	CN XI	Neck STA	Neck STV
4	Bilateral	Gracilis × 2	CN XI	Head STA	Head STV
5	Bilateral	Gracilis × 2	CN XI	FA	FV
6	Bilateral	Gracilis × 2	CN XI	FA	FV

CN XI indicates accessory nerve; FA/FV, facial artery/vein; Neck STA/STV, superior thyroid artery/vein; Head STA/STV, superficial temporal artery/vein.

Case 3*, Right FFMT failure → Redo by Rectus femoris, same CN XI.

FIGURE 2. Case 1, A, 21-year-old male with bilateral facial palsies and inability to smile. B, At 10 years after bilateral gracilis FFMTs neurotized by XI nerves, his smile excursion score improved from 0 to 3, the cortical adaptation stage improved from I to IV.

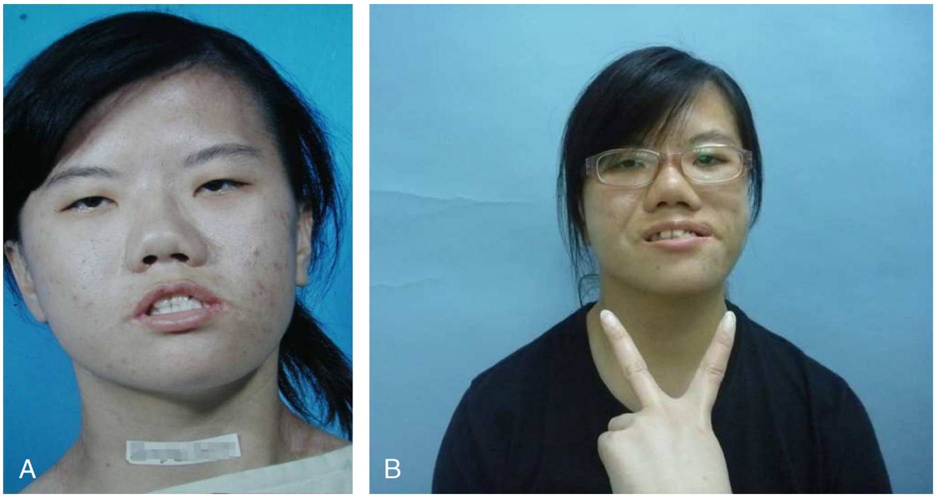

FIGURE 3. Case 2, A, 21-year-old female with bilateral facial palsies and inability to smile. B, At 6 years after bilateral gracilis FFMTs neurotized by XI nerves, her Smile Excursion Score improved from 1 to 3, the cortical adaptation stage improved from I to IV.

left but only from 1 to 2 on the right. The cortical adaptation stage improved from stage I to III.

Case 4

A 15-year-old boy with bilateral MS presented with bilateral VII, VI, XII, and V cranial nerve palsies. He had often been mistaken for having mental retardation (Fig. 4). One-stage bilateral smile reconstruction was performed. Because of poorly developed facial arteries, superficial temporal vessels were used for anastomoses. Postoperatively, the FFMTs showed movement at 2 months and gained independence at 5 months. At 3-year follow-up, he had smile excursion score of 4 and cortical adaptation stage of V (Video 1, online only, supplementary digital content, available at: http://links.lww.com/SAP/A22).

Case 5

A 5-year-old boy with bilateral MS presented with bilateral VII, VI, and XII cranial nerve palsies. No teeth were visible with attempted smiles (Fig. 5). One-stage bilateral smile reconstruction was performed. At 3-year follow-up, he could smile spontaneously (cortical adaptation stage of V), with visibility of both canine teeth (smile excursion score is 4) (Video 2, online only, supplementary digital content, available at: http://links.lww.com/SAP/A23).

Case 6

A 52-year-old woman with bilateral MS presented with bilateral VII and VI cranial nerve palsies (Fig. 6). One-stage bilateral smile reconstruction was performed. One year after surgery, her smile excursion score improved from 1 to 4, and her cortical adaptation stage improved from stage I to II. She remained unsatisfied with her facial aesthetic appearance (Video 3, online only, supplementary digital content, available at: http://links.lww.com/SAP/A24).

DISCUSSION

One Versus Two Versus Multiple Stages for Bilateral MS Facial Reanimation

Smile reconstruction procedures for facial palsy has been discussed extensively, including static procedures (tendon or fascia slings),[16] dynamic procedures (regional muscle transfer),[17,18] and FFMT.[5–14] The goal is to obtain spontaneous and synchronous movements of the face during smiling.

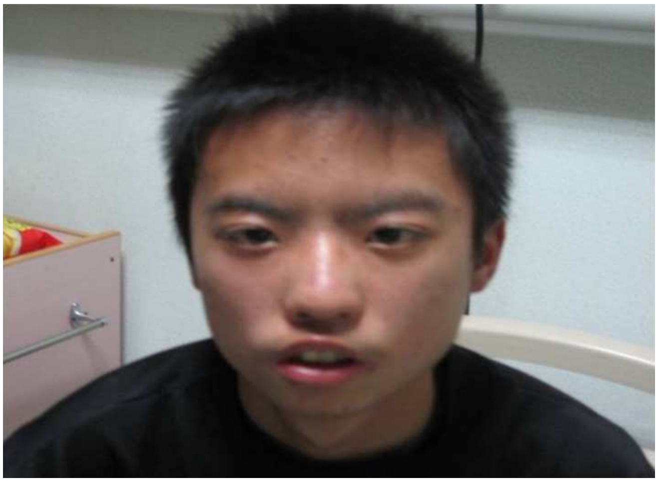

FIGURE 4. Case 4, a 15-year-old male with bilateral facial palsies and inability to smile. Video 1 (online only, supplementary digital content, available at: http://links.lww.com/SAP/A22) demonstrates that he can control and spontaneously smile at 3-year follow-up. His smile excursion score improved from 1 to 4, and cortical adaptation stage improved from I to V.

FIGURE 5. Case 5, a 5-year-old boy with bilateral facial palsies and inability to smile. Video 2 (online only, supplementary digital content, available at: http://links.lww.com/SAP/A23) demonstrates that he can control and spontaneously smile at 3-year follow-up. His smile excursion score improved from 1 to 4, the cortical adaptation stage improved from I to V.

FIGURE 6. Case 6, a 52-year-old woman with bilateral facial palsies and inability to smile. Video 3 (online only, supplementary digital content, available at: http://links.lww.com/SAP/A24) demonstrates that she can smile at 1-year follow-up. Her smile excursion score improved from 1 to 4, and the cortical adaptation stage improved from I to II.

Bilateral MS patients with bilateral facial palsy have masked faces without expression. They may need to undergo numerous surgeries to gain a symmetric smile. Terzis and Noah[19] had performed bilateral FFMTs in 6 patients, applying a 4-stage procedure. They used interpositional nerve grafts to elongate neurotizers, including XI

TABLE 6. Comparison Among Different Neurotizers

	The CFNG	The Masseter Nerve	The XI Nerve
Stage required	2 stages	1 stage	1 stage
Nerve power	1 (standard)	2	2
Technique demanding	1 (standard)	1	2
Muscle movement	1–1.5 y	3–6 mo	3–6 mo
Smile training requirement	Sometimes	Yes	Yes
Independent smile	No	Yes, but not completely	Yes (after training)
Spontaneous smile	Yes	Yes with a latency	Yes with a latency
Persistent trigger movement	Less	Yes	Yes (especially in adult)

CFNG indicates cross-face nerve graft.

nerve, masseter nerve, motor branch of C7, and hypoglossal nerve. The authors suggested a waiting period of 7 months to 2 years between placement of nerve graft and muscle transfer. Zuker et al[14] used the masseter nerve to innervate FFMT technique in 10 patients. They preferred to operate on each side separately with a minimal interval of 3 months. Lischez et al[20] examined 2 patients with bilateral facial reanimation, and the waiting intervals between FFMTs were approximately 1 year. Woollard et al[21] reported 1-stage reconstruction using bilateral masseter nerves to innervate bilateral segmental latissimus dorsi muscles for reconstruction in 20 patients.

Chuang[9,15] has used the 2-stage cross-facial nerve graft followed by FFMT in the past to treat most of his unilateral facial paralysis patients. However, in bilateral MS patients, the facial nerve is not available as a neurotizer. The author decided to use bilateral XI nerves for this purpose because of his experience of performing XI nerve transfer for treating brachial plexus patients.[22] The result was encouraging. Since then, 1 stage bilateral smile reconstruction using bilateral XI nerves to neurotize 2 gracilis muscles has become our first choice to treat bilateral facial palsy. The transferred muscle will regularly start to move at 3 months (stage I) and will gain independent smile (stage III) in 6 months for children and in 1 year for adults.

Comparison Among the Different Neurotizers

A number of donor nerves are available for facial paralysis reconstruction, including contralateral facial nerve, hypoglossal nerve, masseter nerve, C7 motor branch, cervical motor branch, phrenic nerve, and XI nerve.[3,23] The top 3 neurotizers, from the author's point of view, are the contralateral facial nerve, the masseter nerve, and the XI nerve. For bilateral facial paralysis, the masseter nerve seems more popular as the major neurotizer for bilateral facial reanimation.[24,25] However, when performing smile reconstruction using the masseter nerve as the neurotizer, bizarre facial movement is quite frequently seen during eating and biting. In addition, the resulting smile is actually not independent. When activation of the masseter muscle is blocked (by inserting fingers into the patient's mouth as a "bite block" to prevent jaw closure), the patient cannot move the transferred muscle at all.

The use of XI nerve has not been popular for smile reconstruction because of the concerns about inadequate length (thus the need for nerve graft),[4,19] its activity not synergistic with smiling, and its harvest may cause shoulder deficits. In reality, the XI nerve has an adequate length to be transposed to the angle of the mandible without need for nerve graft. In these 6 patients, most are able to reach independence within 1 year, and 4 patients' smile has progressed to be spontaneous at their latest visit. None reported persistent shoulder pain or dropping during follow-up. After 2007, we have extended the use of the XI nerve to neurotize an FFMT for unilateral facial paralysis reconstruction. There have, thus far, been 30 patients (age range, 5–62 years) reconstructed by this technique. Postoperative shoulder complaints consisted of mild pain, and all resolved by time, ranging from 1 to 6 months. The shoulder discomfort came from some trapezius muscle detached from its insertion for ease of dissection of the XI nerve, not from the nerve transection itself. Results after reconstruction have been quite encouraging, showing the XI nerve to be a good alternative to the contralateral facial nerve as a neurotizer. In addition, we have not yet identified any contraindication that would exclude patients from this procedure. Advantages and disadvantages of different neurotizers are shown in Table 6.

Patient Satisfaction

In Bradbury et al[26] survey of 106 hemifacial palsy patients who underwent facial reconstruction, patients with higher scores on the preoperative depression scale were significantly more dissatisfied with surgery. To a bilateral MS patient who has never smiled before, the goal of the surgery is much different than a patient with acquired unilateral facial palsy. Case 6 (Video 3, online only, supplementary digital content, available at: http://links.lww.com/SAP/A24) had significant improvement in smile excursion score and cortical adaptation stage after surgery, but the lowest satisfaction score because of her high expectation. Preoperatively, counseling in the presence of patient's family members is critical in understanding the patient's psychologic condition as well as establishing realistic expectations for perioperative course, postoperative rehabilitation, and final surgical outcomes.

CONCLUSIONS

One-stage bilateral FFMTs neurotized by bilateral XI nerves are effective and safe for treating bilateral facial paralysis in bilateral MS patients. Careful patient selection, adequate neurologic and psychologic examination, thorough preoperative counseling, and postoperative smile training are all important factors in achieving optimal outcomes.

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