Longitudinal Recovery of Speech Motor Function Following Facial Transplantation: A Prospective Observational Study

Bridget J. Perry, PhD, CCC-SLP ^(D); Marziye Eshghi, PhD; Kaila L. Stipancic, PhD, CCC-SLP; Brian Richburg, BA; Hayden Ventresca, BS; Bohdan Pomahac, MD; Jordan R. Green, PhD, CCC-SLP

Objectives: Although facial transplantation is considered effective for restoring facial appearance, research on speech outcomes following surgery is limited. More research is critically needed to inform patients of expected rates and extent of recovery, and to develop interventions aimed at improving speech outcomes.

Methods: Four patients in early recovery (3 weeks–24 months postsurgery) and three patients in late recovery (36–60 months postsurgery) were included. Clinical measures of speech recovery, including speech intelligibility measured using the Sentence Intelligibility Test, a lip strength testing device (Iowa Oral Performance Instrument), and kinematic measures of lip and jaw function measured using high-resolution 3D optical motion capture were used to describe the rate and extent of functional speech and lip recovery, describe and compare the rate of functional speech recovery and kinematic lip and jaw changes in early and late stages of recovery, and explore the association between kinematic measures and functional speech.

Results: Speech intelligibility, speaking rate, and lip strength were below normative values in the first 2 years of postsurgery. Participants in the first 2 years of recovery demonstrated steeper slopes of improvement in clinical and kinematic measures than participants in the later stages of recovery (36–64 months). Gains in jaw range of movement and gains in lip speed and range of movement were significantly correlated with rates of sentence intelligibility improvement. Gains in lip strength were not associated with functional speech improvement.

Conclusions: These findings motivate ongoing work aimed at developing interventions for improving motor speech function in this population.

Key Words: facial transplantation, motor speech, speech kinematics, speech movement, speech recovery. **Level of Evidence:** 3

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INTRODUCTION

To date, upwards of 40 facial transplantation surgeries have been performed worldwide. Although the surgery is widely considered effective for restoring facial appearance, several recent case reports suggest that clinically observable speech deficits persist for years following surgery.^{1,2} At 1-year posttransplant, Van Lierde et al., described improved but still impaired speech deficits in a single patient. The speech impairments noted after

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Send correspondence to Bridget J. Perry, PhD, CCC-SLP, MGH Institute of Health Professions, 96 13th Street, Charlestown, Boston, MA 02129. E-mail: bjperry@mghihp.edu

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1-year posttransplant included moderate hypernasality, mild misarticulations, and lip closure incompetence.² In five patients, including two who were up to 42 months postsurgery, Perry et al. described persistent deficits in lip strength, articulation of bilabial sounds, and speech intelligibility.¹ The enduring deficits found by Perry et al. suggest that recovery of functional speech may continue beyond 3.5 years following surgery.¹

While informative for understanding impairments in patients postsurgery, the findings of previous studies are limited by their relatively short follow-up periods,^{2,3} single case study design,²⁻⁴ and cross-sectional analyses.¹ More longitudinal research on speech recovery is critically needed to (1) better inform patients regarding the expected rate and extent of recovery, (2) evaluate the impact of varying surgical procedures on speech function, and (2) develop evidence-based interventions aimed at improving speech outcomes. In this study, we extended the work of Perry et al.¹ by incorporating a larger sample size, implementing a longitudinal paradigm, and incorporating biomechanical data to further our understanding of the trajectory of motor speech impairment and recovery in this population. To our knowledge, this is the largest facial transplant cohort reported on to date. To better understand the mechanisms of improvement, we also analyzed facial motor recovery via lip strength testing and high-resolution 3D optical facial motion capture. The three aims of the work were (1) to describe the rate and

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From the MGH Institute of Health Professions (B.J.P., M.E., K.L.S., B.R., H.V., J.R.G.), Department of Communication Sciences and Disorders, Boston, Massachusetts, U.S.A.; Brigham and Women's Hospital (B.J.P., B.P.), Department of Plastic and Reconstructive Surgery, Boston, Boston, Massachusetts, U.S.A.; Massachusetts General Hospital (M.E.), Athinoula A. Martinos Center for Biomedical Imaging, MGH, Boston, Massachusetts, U.S.A.; and the University at Buffalo (K.L.S.), Department of Communication Sciences and Disorders, Buffalo, New York, U.S.A.

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| | | | | Participant Characteristics | istics. | | | |
|---|---|--|---|--|--|--|--|--|
| Patient | | P01 | P02 | P03 | P04 | P05 | P06 | P07 |
| Age at transplant | I | 38 | 68 | 60 | 33 | 57 | 30 | 25 |
| Gender | I | Σ | Σ | Σ | Σ | Ľ | Σ | Σ |
| Mechanism of injury | I | Ballistic trauma | Electrical burn | Ballistic trauma | Ballistic trauma | Animal attack | Electrical burn | Electrical burn |
| Functional limitations prior to surgery | 1 | Impaired facial expression, oral incompetence, impaired speech | Impaired facial expression, oral incompetence, impaired speech | Impaired facial expression, oral incompetence, impaired speech | Impaired facial expression, oral incompetence, impaired speech | Absent nasal passage, impaired facial expression, oral incompetence, impaired speech | Impaired facial expression, malocclusion, oral incompetence, impaired speech | Absent nasal passage, impaired facial expression, malocclusion, impaired speech |
| Allograft type | 1 | Osteomyocutaneous, including nose, maxilla, mandible, upper and lower lips | Myocutaneous, including eyelids, nose, upper and lower lips | Osteomyocutaneous, including nose, maxilla, mandible, upper and lower lips | Osteomyocutaneous, including nose, maxilla, mandible, upper and lower lips | Osteomyocutaneous, including bilateral eyelids, bilateral eyes, nose, maxilla, upper and lower lips | Myocutaneous, including eyelids, nose, upper and lower lips | Myocutaneous, including left temporoparietal scalp, bilateral eyelids, left eye, nose, upper and lower lips, teeth |
| Allograft extent | I | Partial | Full | Partial | Partial | Full | Full | Full |
| Facial nerve coaptations | I | Bilateral: buccal and marginal mandibular | Bilateral: buccal and marginal mandibular | Bilateral: buccal and marginal mandibular | Bilateral: buccal and marginal mandibular | Bilateral: frontal, zygomatic, buccal and marginal mandibular | Bilateral: buccal and marginal mandibular | Left: only upper and lower division Right: frontal, zygomatic, buccal and marginal mandibular |
| Months postsurgery | Weeks/months postsurgery at first evaluation | 3 weeks | 2 weeks | 3 weeks | 6 mo | 36 mo | 42 mo | 42 mo |
| | Months postsurgery at last evaluation | 24 | 12 | 17 | 20 | 62 | 62 | 62 |
| Speaking rate | First evaluation | 136.6 | 108.15 | 97.79 | 124.41 | 156.81 | 228.42 | 161.97 |
| minute) | Last evaluation | 174.83 | 131.44 | 116.55 | 129.23 | 164.46 | 200.04 | 143.06 |
| Sentence intelligibility (%) | Sentence-level intelligibilty at first evaluation | 2.00 | 85.45 | 24.55 | 91.8 | 92.7 | 97.2 | 66 |
| | Sentence-level intelligibilty at last evaluation | 86.36 | 95.45 | 58.18 | 96.36 | 94.55 | 97.2 | 66 |
| Word intelligibility (%) | Word-level intelligibilty at first evaluation | 87.14 | 06 | 58.6 | 88.5 | 06 | 95.7 | 98.5 |
| | Word-level intelligibilty at last evaluation | 88.57 | 92.8 | 75.7 | 97.14 | 92.29 | 98.57 | 95.7 |
| Lip strength (kPa [SD]) | Lip strength at first evaluation | 0 (0) | 0 (0) | 0 (0) | 0 (0) | 1.7 (1.6) | 9.7 (2.0) | 5.4 (1.4) |
| | Lip strength at last evaluation | 1.55 (1.0) | 1.33 (0.57) | 2.33 (0.57) | 2.55 (0.40) | | 9.7 (2.0) | 6 (1.5) |

| TABLE II. Functional Speech and Lip Strength Recovery. | | | | | |
|---|----------------------------------|----------------------------------|-----------------------------|-----------------------------|-----------------------------|
| | Year 0–1 (PO1, PO2, PO3, PO4) | Year 1–2 (PO1, PO2, PO3, PO4) | Year 3–4 (PO5, PO6, PO7) | Year 4–5 (PO5, PO6, PO7) | Normal |
| Sentence intelligibility (mean [SD]) | 60% (±43) | 80% (±20) | 96% (±3) | 97% (±2) | >97% ²³ |
| Word intelligibility (mean [SD]) | 83% (±14) | 87% (±11) | 95% (±4) | 96% (±3) | >97% ²³ |
| Speaking rate (mean [SD]) | 120 wpm (±16) | 140 wpm (±31) | 182 wpm (±40) | 170 wpm (±29) | >150 wpm ²⁹ |
| Lip strength (mean [SD]) | 0 kPa (±0) | 2 kPa (±0.5) | 6 kPa (±4) | 8 kPa (±3) | 11.4–14.5 kPa ¹³ |

wpm = word per minute.

extent of functional speech and lip recovery, (2) to describe and compare the rate of functional speech recovery and kinematic lip and jaw changes in early and late stages of recovery, and (3) to explore the association between kinematic measures and functional speech to improve our understanding of underlying mechanisms of motor speech recovery in this population.

METHODS

Participants

The study was approved by the Brigham and Women's Hospital Institutional Review Board. All patients provided written informed consent. Patients who had received either a full or tal between 2011 and 2019 were eligible for the current study, except for one who developed a virus unrelated to surgery causing unilateral facial paralysis. For some, data collection began shortly after surgery, and baseline and follow-up sessions occurred within the first 24 months following surgery (early postsurgical recovery group—P01, P02, P03, P04). At the time of study initiation, other participants (late-postsurgical recovery group—P05, P06, P07) underwent their assessments between 36 and 64 months after facial transplantation surgery (Table I).

partial face transplantation at the Brigham and Women's Hospi-

Clinical Assessment

Sentence-level intelligibility for each patient was obtained using the Sentence Intelligibility Test $\rm (SIT)^5$ and word-level

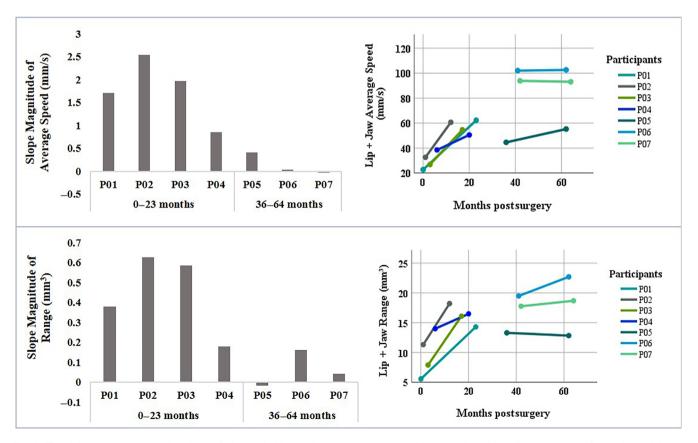


Fig. 1. The right plots represent the slope of change in kinematic measures extracted from the jaw-driven lip movements for each participant. The left plots represent the slope magnitude of observed changes in kinematic measures extracted from jaw-driven lip movement from baseline to follow-up sessions. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

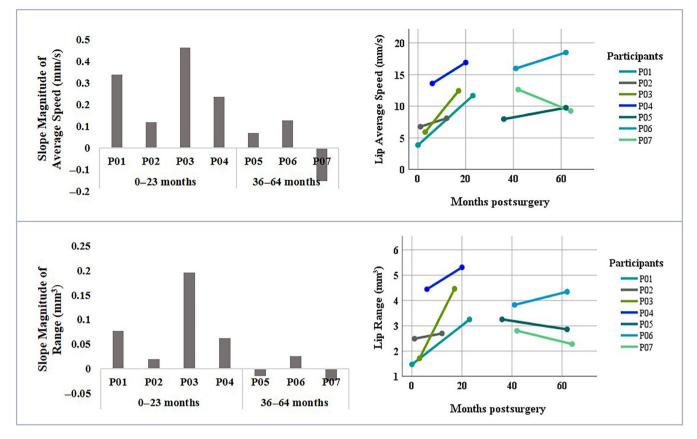


Fig. 2. The right plots represent the slope of change in kinematic measures extracted from lip movement (independent of the jaw) for each participant. The left plots represent the slope magnitude of observed changes in kinematic measures extracted from lip movement (independent of the jaw) from baseline to follow-up sessions. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

intelligibility was obtained using the Word Intelligibility Test (WIT).⁶ Each participant read a unique set of 11 sentences for the SIT and a set of 70 words for the WIT at their habitual speaking rate and volume. Speech samples were recorded digitally (44.1 kHz sampling rate) using a professional quality microphone. Although hearing was not formally tested, it was judged to be adequate during conversational speech for all participants. Trained lab assistants. all of whom were native English speakers reporting no history of language disorders or hearing loss, listened to the recorded samples. For the SIT, sentences were orthographically transcribed, and the total number of words correctly transcribed by the listener was divided by the total number of words spoken by the participant to obtain the percentage of words correctly transcribed (sentence intelligibility percentage). For the WIT, listeners identified each word that the participant said from four options. The total number of words correctly identified by the listener was divided by the total number of words in the set to obtain the percentage of words correctly identified (word intelligibility percentage). The SIT was also used to obtain speaking rate, measured in words per minute (wpm) for each participant. For each sentence on the SIT, the number of words in the sentence was divided by the duration of the sentence in 1 min. This value was averaged across the 11 sentences to yield an overall speaking rate.⁷

Kinematic Assessment

Orofacial movements were recorded using three-dimensional (3-D) motion capture (Motion Analysis, Rohnert Park, CA). Threedimensional motion capture analysis is commonly used to describe motor speech kinematics in both healthy and impaired populations.^{8–10} Procedures used for kinematic data collection, marker placement, movement subtraction, and extraction of kinematic measures from the "nose top center," the "center lower lip," and the "virtual jaw center" markers were the same as those described in detail in Eshghi et al.¹¹ Lower lip movement was represented in two ways: (1) movements of the underlying jaw included (lower lip + jaw) and (2) independent from the movements of the jaw (lower lip – jaw).¹¹

Each participant performed 10 repetitions of the phrase "buy Bobby a puppy" at his or her typical speaking rate and volume. The average speed of movement (mm/s) and range of motion (mm) from lower lip \pm jaw movement time-series were calculated from speech samples using a customized MATLAB algorithm.¹² Each 3-D positional time-series was represented as the 3-D Euclidean distance between the markers. Average speed was calculated as the average value in the first derivative of the 3-D Euclidean distance movement time history. Range of motion was measured as the change in distance between the maximum opening and closing positions during speech. Average speed and range of motion values obtained across the 10 repetitions were averaged for each participant. Because normative values for kinematic speech features are not well established, kinematic speech measures were not used to describe the extent of speech recovery relevant to normative data.

The Iowa Oral Performance Instrument (IOPI Medical Inc) was used to measure lip closure strength. Maximum lip compression strength (kPa) was obtained by asking participants to squeeze a pressurized balloon between their lips with maximal effort. The patient was asked to squeeze the balloon three times

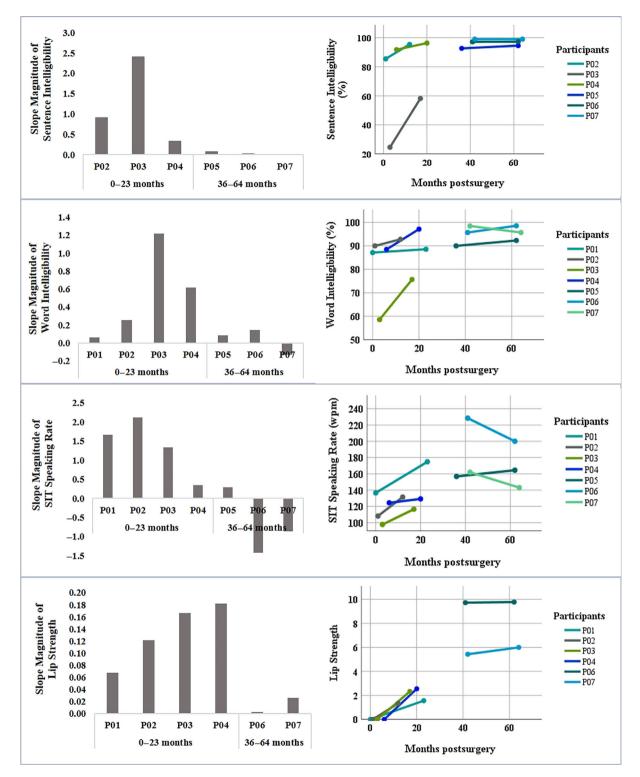


Fig. 3. The right plots represent the slope of change in clinical measures (sentence and word intelligibility scores, speaking rate, and lip strength) for each participant. The left plots represent the slope magnitude of observed changes in clinical measures from baseline to follow-up sessions. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

each at three positions of the mouth (left angle of mouth, right angle of mouth, and center of lips). The average value of these nine trials was considered maximal lip pressure for each participant.

Statistical Analysis

The slopes of change in kinematic (i.e., average speed and range of lip \pm jaw movement) and clinical measures (i.e., word and sentence intelligibility scores, speaking rate, and lip

TABLE III. Mean (SD) of Kinematic Measures Obtained From Each Participant.

| | | | Lip + Ja | aw | Lip – Ja | W |
|---------|-------|---------|--------------------|---------------|--------------------|---------------|
| Subject | Group | Session | Average Speed (SD) | Range (SD) | Average Speed (SD) | Range (SD) |
| P01 | 1 | 1 | 22.781 (3.16) | 5.573 (0.31) | 3.861 (1.38) | 1.473 (1.51) |
| | | 2 | 62.292 (9.32) | 14.298 (0.87) | 11.66 (3.69) | 3.251 (1.12) |
| P02 | 1 | 1 | 32.68 (1.81) | 11.325 (0.68) | 6.776 (0.67) | 2.493 (0.39) |
| | | 2 | 60.685 (46.99) | 18.21 (11.33) | 8.104 (4.83) | 2.702 (1.23) |
| P03 | 1 | 1 | 26.945 (2.11) | 7.91 (0.50) | 5.923 (0.64) | 1.715 (0.131) |
| | | 2 | 54.64 (4.39) | 16.103 (1.52) | 12.436 (1.66) | 4.464 (0.92) |
| P04 | 1 | 1 | 38.53 (7.27) | 14 (1.70) | 13.601 (2.77) | 4.448 (0.72) |
| | | 2 | 50.566 (4.94) | 16.5 (2.01) | 16.913 (1.64) | 5.313 (0.92) |
| P05 | 2 | 1 | 44.6456 (2.02) | 13.31 (1.06) | 7.98 (0.51) | 3.252 (0.67) |
| | | 2 | 55.269 (5.16) | 12.84 (0.83) | 9.751 (1.18) | 2.865 (0.52) |
| P06 | 2 | 1 | 102.025 (8.93) | 19.497 (2.55) | 15.975 (2.01) | 3.826 (0.59) |
| | | 2 | 102.639 (9.41) | 22.699 (1.58) | 18.511 (1.91) | 4.346 (0.47) |
| P07 | 2 | 1 | 93.874 (8.06) | 17.774 (1.90) | 12.628 (0.65) | 2.808 (0.32) |
| | | 2 | 93.125 (3.82) | 18.69 (0.89) | 9.243 (1.63) | 2.28 (0.49) |

strength) were calculated from baseline to follow-up sessions for each participant. Independent-sample *t*-tests were used to statistically compare the rate of improvement in kinematic and clinical measures during the first 23 months and between 36 and 64 months postsurgery. Pearson correlation analyses were performed to examine the association between the slope of change in kinematic measures (average speed and range of lip \pm jaw movement and lip strength) and the slope of change in functional speech measures (sentence intelligibility, word intelligibility, and speaking rate). All statistical analyses were performed in Rstatistical software version 4.0.2 (R development core team, 2013) and an α -level of 0.05 was set as the level of significance.

RESULTS

Participants

Seven patients who had received either a full or partial face transplantation at the Brigham and Women's hospital were included in the study. Each patient received a facial graft from their donor, which included

varying amounts of soft tissue, facial musculature, nerve, and bone as well as facial nerve coaptations leading to the upper and lower lip. The median age of the participants was 38 years (IQ range: 31-58). Demographic information, including age, gender, and details of the transplant are included in Table I. The median time between data collection sessions was 20 months (IQ range: 18.5-24).

Extent of Functional Motor Speech Recovery

Speech intelligibility, speaking rate, and lip strength were well below normative values in the first 2 years following surgery (Table II). Two of the three participants, who had reached the 5-year postsurgery mark, achieved sentence intelligibility within the normal range. Subjectively, occasional misarticulations of bilabial sounds were observed even in participants with intelligibility scores in the normal range. Although all participants were able to

| TABLE IV. |
|--|
| Comparisons of the Slope of Change in Kinematic and Clinical Measures During 0-23 Months Postsurgery and 36-64 Months Postsurgery. |

| | 0–23 Mo Mean (SD) | 36–64 Mo Mean (SD) | <i>t</i> -Tests <i>p</i> -Value | 95% CI Upper Limit, Lower Limit | Cohen's a Effect Size |
|--|----------------------|-----------------------|------------------------------------|------------------------------------|--------------------------|
| Kinematic measures | | | | | |
| Slope of lip + jaw average speed | 1.775 (0.70) | 0.135 (0.24) | 0.013 | 0.579, 2.702 | 2.91 |
| Slope of lip + jaw range of movement | 0.442 (0.21) | 0.061 (0.09) | 0.027 | 0.069, 0.693 | 2.25 |
| Slope of lip – jaw average speed | 0.290 (0.15) | 0.014 (0.15) | 0.064 | -0.024, 0.578 | 1.88 |
| Slope of lip – jaw range of movement | 0.089 (0.08) | -0.004 (0.03) | 0.087 | -0.022, 0.208 | 1.52 |
| Clinical measures | | | | | |
| Slope of % sentence intelligibility | 1.212 (1.07) | 0.025 (0.04) | 0.195 | -1.467, 3.842 | 1.57 |
| Slope of word intelligibility | 0.539 (0.51) | 0.035 (0.14) | 0.141 | -0.273, 1.281 | 1.25 |
| Slope of Sentence Intelligibility Test speaking rate | 1.366 (0.75) | -0.661 (0.87) | 0.032 | 0.283, 3.771 | 2.53 |
| Slope of lip strength | 0.134 (0.05) | 0.014 (0.02) | 0.014 | 0.041, 0.199 | 2.67 |

The number of subjects in the early and late-post surgery groups was 4 and 3, respectively.

| Speech Measures | Biomechanical Measures | Pearson Correlation Coefficient | p-Value |
|--|--------------------------------------|---------------------------------|---------|
| Slope of % sentence intelligibility | Slope of lip + jaw average speed | 0.756 | 0.082 |
| | Slope of lip + jaw range of movement | 0.816 | 0.047 |
| | Slope of lip – jaw average speed | 0.811 | 0.050 |
| | Slope of lip – jaw range of movement | 0.909 | 0.012 |
| | Slope of lip strength | 0.628 | 0.256 |
| Slope of word intelligibility | Slope of lip + jaw average speed | 0.474 | 0.282 |
| | Slope of lip + jaw range of movement | 0.560 | 0.191 |
| | Slope of lip – jaw average speed | 0.768 | 0.044 |
| | Slope of lip – jaw range of movement | 0.891 | 0.007 |
| | Slope of lip strength | 0.789 | 0.062 |
| Slope of Sentence Intelligibility Test speaking rate | Slope of lip + jaw average speed | 0.948 | 0.001 |
| | Slope of lip + jaw range of movement | 0.778 | 0.039 |
| | Slope of lip – jaw average speed | 0.573 | 0.179 |
| | Slope of lip – jaw range of movement | 0.448 | 0.314 |
| | Slope of lip strength | 0.622 | 0.188 |

TABLE V. Correlations Between the Rate of Improvement in Kinematic Measures and Speech Functional Measures

achieve complete lip closure, significant deficits in lip strength continued even in participants at five-five years post-surgery.

Rate of Functional and Kinematic Motor Speech Recovery

All participants with baseline and follow-up data between 0 and 23 months postsurgery demonstrated improvement in all kinematic and clinical measures. The changes in kinematic and clinical measures demonstrated inconsistent trajectories in participants whose baseline and follow-up sessions occurred after 36 months postsurgery (Figs. 1-3).

Table III demonstrates the mean (SD) of kinematic measures obtained from each participant. On average, participants demonstrated steeper slopes of improvement in both kinematic and clinical measures during the first 2 years after facial transplantation and continued to show oromotor recovery, albeit at a slower rate, from 36 to 64 months postsurgery. Improvements in average speed and range of lip + jaw speaking rate and lip strength were significantly faster during the first 23 months following surgery than after the second year (p < 0.05) (Table IV). No significant differences were observed between participants at early and late stages of recovery in terms of the rate of improvement in kinematic measures extracted from the lip movement that was independent from that of the jaw (p > 0.05). There were no differences in the rate of change of word and sentence intelligibility scores between the early and late groups of participants (p > 0.05).

Association Between Kinematic Factors and Functional Motor Speech Recovery

There were significant correlations between the rate of sentence intelligibility improvement and lip + jaw range

of movement improvement, as well as lip-jaw average speed and range of movement improvement (p < 0.05). Similarly, the rate of word intelligibility improvement was significantly correlated with changes in the lip-jaw average speed and range of movement (p < 0.05). The rate of improvement in speaking rate had a significant association with the rate of improvement in lip + jaw speed and range of movement (p < 0.05). The relationships between improvements in functional speech measures and lip strength were not significant (p > 0.05) (Table V).

DISCUSSION

In this article, we report the rate of speech and oromotor recovery following facial transplantation. Our analyses revealed three primary findings: (1) functional speech improvements and kinematic changes of the lip and jaw were greatest during the first 2 years of recovery; (2) lip strength never fully recovered; and (3) increases in lip and jaw movement range of motion and speed were strongly associated with functional speech gains, suggesting a causal link between persistent facial motor impairments and speech deficits. These findings will help inform future surgical procedures and motivate ongoing work aimed at developing therapeutic interventions for improving motor speech function in this population.

Improvements in Speech Movements and Function Occur Primarily in the First 2 Years Following Surgery

In our cohort, large gains in both kinematic and clinical measures of motor speech performance occurred in the first 2 years following surgery. These findings support work by Van Leirde et al., who reported that for a single patient, intelligibility improved most significantly in the first year following transplantation and that intelligibility remained mildly impaired at 21 months postsurgery.² After 3 years postsurgery, changes in both kinematic and clinical speech measures were inconsistent across both patients and tasks, with different patients improving on different measures. This finding may be, partially, the result of heterogeneity in our cohort and the inability to account for all factors impacting speech motor control along the trajectory of recovery (e.g., varying patterns of muscle reinnervation, the emergence of other health conditions, graft rejection episodes, cognitive-mental health status). Although our dataset had a gap between years 2 and 3 postsurgery, most participants in the early group achieved measures within range of the late recovery group by year two following surgery across all measures except for lip strength. Measures of lip strength at year two for the early group are much smaller than measures of lip strength around year three for the late group. The large gap between the early participants' average lip strength at the 2-year time-point and the late participants' lip strength at the 3-year time-point may suggest that lip strength has its fastest recovery rate during this 2-3 year time interval during which we are currently lacking data. An improved understanding of speech motor recovery will require more densely sampled data during the second and third years of recovery.

Lip Strength Deficits are Significant and Persist for Years Following Surgery

At 5 years postsurgery, all participants continued to demonstrate persistent, significant lip weakness compared to that of healthy controls, which range from 11.4 kPa to 14.5.¹³ Similar to the previous findings discussed with functional speech and speech movement recovery, the largest gains in lip strength in our cohort occurred in the first 2 years following surgery. These findings are consistent with Van Leirde et al., who reported that their patient had severe lip weakness until 12 months, at which time lip strength improved but remained decreased relative to healthy controls. Because the muscle activation levels and force generation needed to produce speech are small, and are only a fraction of the maximum muscle activation and force possible,^{14–16} the strength of articulators is typically not associated with speech intelligibility.^{17–19} Accordingly, improvements in lip strength were not associated with improvements in functional speech in our cohort. Although functional speech (i.e., high levels of intelligibility) was grossly attained by the 5-year postsurgery mark in many patients (reported here and in previous work, including Fischer et al.²⁰), ongoing deficits in the control of the articulators may have implications for communicative participation, quality of life, and chewing-swallowing function.²¹⁻²³

Improvements in Speech Function are Driven by Increases in Articulator Speed and Range of Movement

Findings in the current study demonstrate that both speech intelligibility and speaking rate, widely-used clinical measures of speech function,²⁴ were associated with improvements in distinct articulatory movement speeds

and range of motion. These strong associations indicate that recovery of specific articulator movements may underlie improvements in functional speech.

Improvements in Speech Intelligibility are Driven by Gains in Lip Speed and Lip and Jaw Range of Motion

Previous work has shown that the ability to achieve adequate lip closure for producing labial consonants during speech is impaired, particularly during the early stages of recovery postfacial transplantation.¹ While gains in lip strength were not associated with gains in speech intelligibility in our study, interestingly, gains in range of lip movements and lip movement speeds were. The relationship between parameters of lip movement and intelligibility agrees with previous work by Rong et al. who found that among the four speech subsystems, the articulatory system contributed to over half of the variance in intelligibility.²⁵

While speculative, we hypothesize that increased jaw opening early in recovery may be a compensatory mechanism for decreased lip movement. Among the articulators, jaw motor functions appear to be particularly robust to neuromotor damage^{26,27} and individuals with neuromuscular diseases, such as amyotrophic lateral sclerosis, have been shown to use jaw movement to compensate for decreased tongue movement.^{8,28,29} We hypothesize that individuals following facial transplantation may be able to use jaw movements to their advantage in their pursuit of functional speech.

Improvements in Speaking Rate are Driven by Increases in Jaw Speed and Range of Movement

A final finding from this study is that improvements in speaking rate were associated with increases in jaw speed and range of motion. Jaw rotation and translation can be restricted in all dimensions depending on the surgery. Early period postsurgery, jaw movements were relatively small in the vertical directions, which coincided with slow speaking rates (for comparison, speaking rate of healthy individuals has been reported to be from 160 to 200 wpm).^{8,30} Over the first 2 years following surgery, jaw speed and range of movement in the vertical direction significantly increased. These findings are consistent with work by Grigos et al. who described increased jaw displacement during speech in a single patient over 13-months of recovery.³ For some patients who receive osteocutaneous transplants that include the mandible, early limited jaw function that increases over time would be expected as part of recovery. The restrictions were most evident in our findings that jaw range of motion was small early postsurgery, and increased over time. Moreover, this increase was associated with an improved speech outcome-an increase in speaking rate

In our cohort, all patients, regardless of mandible involvement during surgery, presented with increasing jaw movements over the first 2 years of surgery. Increasing jaw movements may be a compensatory strategy used to accommodate compromised lip control, and is in contrast with the more economical strategy used by many healthy speakers, which is to minimize the excursion of the jaw to speak faster.^{10,31,32} Similar findings of an overreliance on the jaw, even at the late stage of recovery postfacial transplantation, were found by Eshghi et al. These findings suggest that surgical techniques and procedures promoting maximal lip and jaw function may improve long-term motor speech outcomes.

Study Limitations

Due to the small number of patients included in this study, the time gap between 23 and 36 months postsurgery, and the large heterogeneity of nonsurgical and surgical factors (i.e., extent of transplant, complications posttransplant, etc.) among patients in this population, it is difficult to extrapolate the present findings beyond the current study. Given the sample size is small, *t*-test may not be an optimal statistical approach. As such, we included the 95% confidence interval and the Cohen's d effect sizes for a more reliable interpretation of the data. Additional longitudinal data spanning from pretransplant between 0 and 5 years is needed to provide a more complete picture of recovery. Lastly, because electromyography was not performed, the contributions of individual facial muscles to speech movements cannot be determined. Although beyond the scope of the current study, additional work is required to account for the impact of factors such as transplant type and extent, level of facial nerve adaptation, revision surgeries, and allograft rejection episodes on recovery.

CONCLUSIONS

The current study highlights both functional speech improvements and kinematic changes of the lip and jaw over the first 5 years of recovery. To our knowledge, this work represents the most complete report of motor speech recovery following facial transplantation surgery to date and sheds light on future areas of exploration. Future work from our laboratory will focus on assessing the efficacy of physiologically based exercise protocols aimed at improving functional speech and orofacial motor recovery following surgery, which is critical for optimizing functional speech in this population. In addition, incorporating surgical data with kinematic data to explore predictors of functional speech recovery would help support patient decision-making and clinical management of speech impairments in this population.

REFERENCES

- Perry BJ, Pomahac B, Bueno E, Su P, Richburg BD & Green JR. Characteristics of speech following facial transplantation. Paper presented at International Conference of Phonetic Sciences; August 2015; Glasgow, Scotland.
- Van Lierde KM, Roche N, De Letter M, et al. Speech characteristics one year after first Belgian facial transplantation. *Laryngoscope*. 2014;124: 2021-2027. doi:10.1002/lary.24585
- Grigos MI, Leblanc É, Hagedorn C, Diaz-Siso JR, Plana N, Rodriguez ED. Changes in articulatory control pre- and post-facial transplant: a case report. J Speech Lang Hear Res. 2019;62:297-306. doi: 10.1044/2018_JSLHR-S-18-0147
- De Letter M, Vanhoutte S, Aerts A, et al. Facial nerve regeneration after facial allotransplantation: a longitudinal clinical and electromyographic follow-up of lip movements during speech. J Plast Reconstr Aesthetic Surg. 2017;70:729-733. doi:10.1016/j.bjps.2017.02.025

- Dorsey M, Yorkston K, Beukelman D, Hakel M. Speech intelligibility test for windows. Institute for Rehabilitation Science and Engineering at Madonna. 2007.
- Kent RD, Weismer G, Kent JF, Rosenbek JC. Toward phonetic intelligibility testing in dysarthria. J Speech Hear Disord. 1989;54:482-499. doi: 10.1044/jshd.5404.482
- Yorkston KM, Beukelman DR. Communication efficiency of dysarthric speakers as measured by sentence intelligibility and speaking rate. J Speech Hear Disord. 1981;46:296-301. doi:10.1044/jshd.4603.296
- Shellikeri S, Green JR, Kulkarni M, et al. Speech movement measures as markers of bulbar disease in amyotrophic lateral sclerosis. J Speech Lang Hear Res. 2016;59:887-899. doi:10.1044/2016_JSLHR-S-15-0238
- 9. Yunusova Y. Articulatory movements during vowels in speakers with dysarthria and healthy controls. J Speech Lang Hear Res. 2008;51:596.
- Mefferd AS, Green JR. Articulatory-to-acoustic relations in response to speaking rate and loudness manipulations. J Speech Lang Hear Res. 2010;53:1206-1219. doi:10.1044/1092-4388(2010/09-0083)
- Eshghi M, Perry BJ, Richburg B, Ventresca HM, Pomahac B, Green JR. Neuromotor speech recovery across different behavioral speech modifications in individuals following facial transplantation. *Front Neurol.* 2021; 11:593153. doi:10.3389/fneur.2020.593153
- Green JR, Wang J & Wilson DL SMASH: A tool for articulatory data processing and analysis. Interspeech. Accessed March 29, 2016. http:// www.mghihp.edu/files/research/sfdl/proc-4.pdf.
- Jeong DM, Shin YJ, Lee NR, et al. Maximal strength and endurance scores of the tongue, lip, and cheek in healthy, normal Koreans. J Korean Assoc Oral Maxillofac Surg. 2017;43:221-228. doi:10.5125/jkaoms.2017.43.4.221
 Kent RD, Kent JF, Rosenbek JC. Maximum performance tests of speech
- Kent RD, Kent JF, Rosenbek JC. Maximum performance tests of speech production. J Speech Hear Disord. 1987;52:367-387.
- Kuehn DP, Moon JB. Induced fatigue effects on velopharyngeal closure force. J Speech Lang Hear Res. 2000;43:486-500. doi:10.1044/jslhr.4302.486
- Mchenry MA, Minton JT, Hartley LL, Calhoun K, Barlow SS. Age-related changes in orofacial force generation in women. *Laryngoscope*. 1999;109: 827-830. doi:10.1097/00005537-199905000-00027
- Neel T, Palmer M, Sprouls G, Morrison L. Muscle weakness and speech in oculopharyngeal muscular dystrophy. J Speech Lang Hear Res. 2015;58:1–12. doi:10.1044/2014_jslhr-s-13-0172
- Solomon NP, Makashay MJ, Helou LB, Clark HM. Neurogenic orofacial weakness and speech in adults with dysarthria. Am J Speech Lang Pathol. 2017;26:951-960. doi:10.1044/2017_AJSLP-16-0144
- McCauley RJ, Strand E, Lof GL, Schooling T, Frymark T. Evidence-based systematic review: effects of nonspeech oral motor exercises on speech. *Am J Speech Lang Pathol.* 2009;18:343-360. doi:10.1044/1058-0360 (2009/09-0006)
- Fischer S, Perry B, Alhefzi M, et al. Follow-up on functional outcomes of face transplantation. *Plast Reconstr Surg.* 2015;136:7-8. doi:10.1097/01. prs.0000472279.22545.96
- Pitts LL, Morales S, Stierwalt JAG. Lingual pressure as a clinical indicator of swallowing function in Parkinson's disease. J Speech Lang Hear Res. 2018;61:257-265. doi:10.1044/2017_JSLHR-S-17-0259
 Pitts LL, Kanadet RM, Hamilton VK, Crimmins SK, Cherney LR. Lingual
- Pitts LL, Kanadet RM, Hamilton VK, Crimmins SK, Cherney LR. Lingual pressure dysfunction contributes to reduced swallowing-related quality of life in Parkinson's disease. J Speech Lang Hear Res. 2019;62:2671-2679. doi:10.1044/2019_JSLHR-S-18-0366
- Hiraoka A, Yoshikawa M, Nakamori M, et al. Maximum tongue pressure is associated with swallowing dysfunction in ALS patients. *Dysphagia*. 2017; 32:542-547. doi:10.1007/s00455-017-9797-z
- Green JR, Yunusova Y, Kuruvilla MS, et al. Bulbar and speech motor assessment in ALS: challenges and future directions. Amyotroph Lateral Scien Frontotemporal Degener, 2013;14:494-500. doi:10.3109/21678421.2013.817585
- Rong P, Yunusova Y, Wang J, et al. Predicting speech intelligibility decline in amyotrophic lateral sclerosis based on the deterioration of individual speech subsystems. *PLoS One.* 2016;11:e0154971. doi:10.1371/journal. pone.0154971
- Lawyer T, Netsky MG. Amyotrophic lateral sclerosis: a clinicoanatomic study of fifty-three cases. AMA Archives of Neurology & Psychiatry. 1953; 69:171-192. doi:10.1001/archneurpsyc.1953.02320260029002
 Lee J, Rodriguez E, Mefferd A. Direction-specific jaw dysfunction and its
- Lee J, Rodriguez E, Mefferd A. Direction-specific jaw dysfunction and its impact on tongue movement in individuals with dysarthria secondary to amyotrophic lateral sclerosis. J Speech Lang Hear Res. 2020;63:499-508. doi:10.1044/2019_JSLHR-19-00174
- Perry BJ, Martino R, Yunusova Y, Plowman EK, Green JR. Lingual and jaw kinematic abnormalities precede speech and swallowing impairments in ALS. *Dysphagia*. 2018;33:840-847. doi:10.1007/s00455-018-9909-4
- Rong P, Green JR. Predicting speech intelligibility based on spatial tonguejaw coupling in persons with amyotrophic lateral sclerosis: the impact of tongue weakness and jaw adaptation. J Speech Lang Hear Res. 2019;62: 3085-3103. doi:10.1044/2018_JSLHR-S-CSMC7-18-0116
- Rong, P, Yunusova, Y, Green, JR. Speech intelligibility decline in individuals with fast and slow rates of ALS progression. Interspeech; 2015. Accessed December 16, 2019. https://www.isca-speech.org/archive/ interspeech_2015/papers/i15_2967.pdf.
- Eshghi M, Stipancic KL, Mefferd A, et al. Assessing oromotor capacity in ALS: the effect of a fixed-target task on lip biomechanics. Front Neurol. 2019;10:1288. doi:10.3389/fneur.2019.01288
- Simione M, Fregni F, Green JR. The effect of transcranial direct current stimulation on jaw motor function is task dependent: speech, syllable repetition and chewing. Front Hum Neurosci. 2018;12:33. doi:10.3389/ fnhum.2018.00033