CHARACTERISTICS OF SPEECH FOLLOWING FACIAL TRANSPLANTATION

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ABSTRACT

To date, over thirty face transplants have been performed worldwide since 2005. Although advances in facial transplantation surgery now offer the possibility of societal reintegration to persons who suffer from severe facial disfiguration, speech deficits can persist. This paper reports on the speech characteristics of five patients following facial transplantation and relates them to labial strength and overall quality of life.

Keywords: Dysarthria, Facial Transplantation, Speech Intelligibility, Speech Errors

1. INTRODUCTION

To date, over 30 facial transplantation surgeries have been completed worldwide. Although the procedure is highly effective for restoring facial appearance, speech outcomes appear to vary across recipients. Brigham and Women's Hospital has performed seven of these surgeries. Although facial mobility has slowly improved in these patients, speech and swallowing abnormalities and deficits persist even in our first patient who is now 3.5 years post transplantation.

The current literature on facial motor and speech outcomes is largely descriptive and sparse in number. Existing descriptions have reported complete facial paralysis immediately following transplantation followed by a prolonged recovery of facial muscle function. Lanteri et al. [7] reported that the first signs of facial musculature contraction occurred within 2-3 months post-surgery. Smeets et al. [15] reported that motor recovery occurred later than sensory recovery, on average at around 7.8 months following surgery. Several studies reported that perceptible gains in facial mobility occurred during the first year [1, 3, 5, 12, 13, 14]. Petruzzo et al. [11] reported that, at five years post-surgery, their patient showed almost a complete restoration of sensation and motion, allowing her to chew, swallow, eat, drink, and speak normally

To our knowledge, the existing research has not described the abnormal speech characteristics following facial transplantation. Persistent weakness of the lip musculature is expected to result in distortions and substitutions of labial sounds [4, 9, 16]. The impact of these errors on speech intelligibility and speaking rate is not well understood but patients are expected to reduce their speaking rate to maximize speech clarity. In this investigation, we profiled the abnormal speech patterns in five patients and examined the association between these errors patterns to labial strength and a measure of quality of life. To our knowledge, this is the first detailed report on speech impairments in multiple patients following face transplantation.

2. METHODS

2.1. Patient History

Patient 1 is a 39 year old male who sustained multiple facial injuries including the shattering of the central part of his face with complete loss of his nose, upper and lower lips including underlying partial maxilla and also central mandible following a self-inflicted gunshot wound to the face in 2000. Attempted repairs were unsuccessful in restoring adequate appearance and function, therefore, he underwent partial facial transplantation during which he received the central lower two-thirds of his face, encompassing the entire nose, the lower orbital rim, the maxilla, the upper and lower lips and the mandible as well as submental tissue down to the level of the hyoid bone.

Patient 2 is a 44 year old female status post physical and sexual assault by her estranged husband in 2007 resulting in 80% TBSA caustic/chemical burns to the head, mouth, neck, chest, bilateral upper and lower extremities, back and the perineum as well as a left minimally displaced humerus fracture, a left distal displaced ulnar fracture, and a right orbital wall fracture. Her injuries left her with significantly disfigured facial features and with multiple painful face and neck contractures. She underwent full facial transplantation, which included the maxilla, extending to zygomatic bones and including nasal bone, the soft tissues including the entire facial skin and underlying muscles, the connection of the supraorbital buccal and mental nerves, and the connection of 5 branches of facial nerve bilaterally with nerve grafts used on the left.

At 1 year status post surgery, patient 2 was enrolled in a case study investigating the effects of an intensive 8-week labial strength training protocol on labial strength and speech [10]. Aside from this trial, therapy targeting speech deficits in all five of these patients has been limited.

Patient 3 is a 57 year-old female who survived a animal attack, which resulted in total vision loss in both eyes, as well as severe trauma to the face and to the bilateral hands. The patient underwent numerous facial reconstruction surgeries prior to undergoing facial transplantation which included all muscles of facial expression, zygoma, maxilla, and hard and soft palates along with a left hand transplant at the level of forearm, and right hand transplant including digits 2-5 at carpal level. Unfortunately, the bilateral hand transplants were removed secondary to infection.

Patient 4 is a 31 year-old male with a history of a severe motor vehicle accident resulting in 45% TBSA electrical burn that left him severely deformed. He sustained burns to his forehead, eyelids, nose, cheeks, upper lip, lower lip, chin, and neck area. He had oral incompetence with full-thickness loss of tissue, with partial reconstruction of in the lower lip region prior to facial transplantation. He underwent full facial transplantation, which included the soft tissues of the entire facial skin and underlying muscles of facial expression, eyelids, the nose, and the connection of the buccal and the marginal mandibular branches of the facial nerve to help innervate the lower third of the face. The upper branches of the facial nerve remained intact, and therefore were not connected to donor nerve during surgery.

Patient 5 is a 26 year-old male who suffered from an electrocution injury, resulting in extensive injury to his face with complete destruction of his left eye and damage to his right eye, leaving him blind. Conventional methods of repair were unsuccessful, leading the patient to undergo full facial transplantation, which included the entire facial skin and all underlying muscles, the left parotid gland, eyelids, nose, and securement of ocular prosthesis on left and right orbit. On the left side, the facial and mental nerves were successfully reconnected. On the right side, the supraorbital and infraorbital nerves were connected along with mental nerve and branches of facial nerve.

2.2. Assessment Methods

2.2.1 Labial Strength

The Iowa Oral Performance Instrument (IOPI Medical Inc.) was used to measure labial strength. Maximum lip compression strength (kPa) was obtained by asking the patient to squeeze a pressurized balloon between the lips with maximal effort three times in three positions: on the left side, the right side and at midline. The average value of the three trials was considered maximal lip pressure in each position.

2.2.1 Sentence Intelligibility

Sentence-level intelligibility was obtained using the Sentence Intelligibility Test (SIT) [18]. This test has been shown to reliably estimate intelligibility within and across judges [18]. Each patient was asked to recite a unique set of 11 sentences, recorded digitally (44.1 kHz sampling rate) using a professional quality microphone. These sentences were then orthographically transcribed by trained lab assistants, all of whom were native English speakers reporting no history of language disorders or hearing loss. The same set of SIT sentences was never scored twice by the same judge.

2.2.3 Word Intelligibility

Word intelligibility was obtained using the Multiple Word Intelligibility Test [6]. Each patient was asked to recite a set of 70 words, recorded digitally (44.1 kHz sampling rate) using a professional quality microphone. Trained lab assistants were then asked to listen to the recording and to identify the words that the listener said from a field of four. To obtain a word intelligibility percentage, the total number of words correctly identified by the listener was divided by the total number of words in the set.

2.2.4 Speech Errors

Phonetic characteristics of speech errors were derived from the words produced during the Multiple Word Intelligibility Test [6]. Trained lab assistants were then asked to listen to the recording and to identify the words that the listener said from a field of four. The field of four words contained the target word as well as three foils that contain specific phonetic contrasts commonly found to be problematic for people with dysarthria. The Multiple Word Intelligibility Test phonetic contrast key was used to determine the phonetic contrast mistakes made by each patient on misidentified words.

2.2.5 Speaking Rate

Speaking rate, measured in words per minute (wpm), was derived from the SIT sentences for each patient. The total word count of the SIT sentence set was divided by the total spoken duration of that set, excluding inter-sentence pauses [19].

2.2.6 Facial Disability Index

Quality of life was assessed using the Facial Disability Index (FDI), a self-report measure of overall disability due to facial neuromuscular impairment. It includes sub-scores for both physical impairment as well as social wellbeing, which are scored on a 100-point scale with 100 representing no disability. It has been shown to be both reliable and valid for the measurement of facial disability [17].

3. RESULTS

3.1 Labial strength.

Labial strength (kPa) was severely impaired in all participants. The average (across participants) lip strength on the left side, right side, and midline was 5.26 kPa, 5.18 kPa, and 4.4 kPa, respectively. At midline, normative values are approximately 33.8 kPa (15.1) for men and 22.4 kPa (7.5) for women [2]. One patient was unable to complete labial strength testing at midline because of her dentition.

Patient	Months	IOPI Left	IOPI Right	IOPI Center
1	6	0	0	0
2	18	6	8	5
3	36	3.3	2	N/A
4	42	7	4.3	5
5	42	10	11.6	7.6

Table 1: Labial strength of patient on the left side, right side. For all tables, scores were ranked based on the number of months post-transplantation.

3.2 Word and sentence intelligibility

On average, following facial transplantation, patients were found to be 91.75% intelligible at the word level and 95.22% at the sentence level. The average speaking rate was 163.36 wpm, which is approximately 30 wpm slower than typical [6].

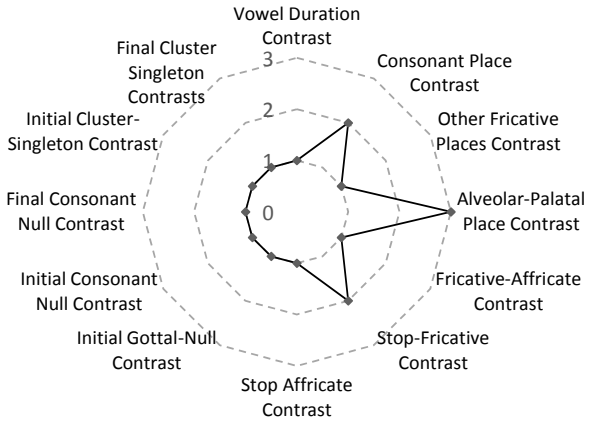
Table 2: Word and sentence intelligibility scores, and mean speaking rates (wpm) for each participant.

Patient	Months	Word Intel %	Sentence Intel %	Speaking Rate
1	6	88.5	91.8	124.4
2	18	90	95.4	145.3
3	36	90	92.7	156.8
4	42	95.7	97.2	228.4
5	42	98.5	99	161.9

3.3 Speech errors

Most of the patients presented with few identifiable speech errors. Alveolar-palatal place contrast errors were the most common and were detected 5 times for patient 1, 2 times for patient 2, and once for patient 3. Consonant place contrast errors and stop-fricative contrast errors were detected in patients 1 and 5, and patients 2 and 3, respectively. Additional error types were observed in single patients.

Figure 1: The number of participants that presented with specific speech errors as derived from the Word Intelligibility Test.



3.4 Quality of life

On average, self-reported levels of social disability as a result of facial impairment were greater than levels of physical disability. Patients, on average, scored 71% on the physical scale of the Facial Disability Index, with 100% indicating no physical disability present. On the social scale of the Facial Disability Index, patients scored a 67.2% on average, with 100% indicating no social disability present. On average, patients obtained a total score of 69.1% on the Facial Disability Index, with 100% indicating no disability as a result of facial impairment.

Table 3: Level of self-reported disability as a result of facial impairment from the Facial Disability Index.

Patient	Months	Physical Scale	Social Scale	Total
1	6	75	84	79.5
2	18	75	76	75.5
3	36	25	48	36.5
4	42	90	60	75
5	42	90	68	79

4. DISCUSSION

This study examined the speech characteristics and their relations to labial strength and quality of life in five participants who have undergone facial transplantation. Our findings demonstrated that patients with greater impairment in labial strength also demonstrated greater impairment in speech intelligibility, suggesting that labial weakness may contribute to reduced speech intelligibility in this population.

Labial strength was greatest in patients who were 42 months status post-surgery, suggesting that improvements in labial strength may continue past 3.5 years. Word intelligibility and sentence intelligibility were also greatest in these patients, a finding that also supports the suggestion that functional improvements continue beyond 3.5 years following surgery. Based on the relative ranking of values with and across variables, labial strength appeared to be predictive of word and sentence intelligibility, and the self-reported physical impairment subscore from the Facial Disability Index.

While only 18 months status post-surgery, patient 2 had better labial strength and speech intelligibility than did patient 3. Patient 2 presented with increased labial strength over patient 3 despite being only 18 months status post-surgery. The advanced status of patient 2 may be because she was the only recipient who received an 8-week intensive labial strengthening therapy [10]. This treatment resulted in significant gains in labial strength and in speed, duration, and range of motion of speech movement [10].

Patients with the lowest word intelligibility scores demonstrated more speech errors across a greater variety of error type. The most frequent speech error type, unexpectedly, was the alveolar-palatal fricative contrasts. A total of 8 alveolar-palatal fricative contrast mistakes were made among patients 1, 2 and 3 (with patient 1 making 5 errors in this category, patient 2 making 2 errors in this category, and patient 3 making 1 error in this category). Patients 4 and 5 did not make any errors of this type, suggesting improvements in this speech error type took place with time since surgery. These errors most commonly involved the substitution of /s/ for /sh/. We suspect that these errors were prevalent in this group because /sh/ requires lip rounding, which is significantly impaired in this population, particularly in the earlier stages of recovery.

Based on patient report and our own clinical observations, we expected to find the greatest impairment to consonants that require bilabial closure. The current findings and our clinical observations, however, suggest that some of these patients were approximating bilabials using lingua-labial closure. Despite its apparent effectiveness, this strategy is visually distracting and it may be effortful for the recipient.

For all but patient 4, speaking rate was found to be well below the norm. Slowing may be a response to oral weakness that is intended to improve the clarity of speech [8]. Impaired labial strength and movement may also limit the rate at which the patient is able to speak. Patient 4 reported that he has always been a “fast-talker”.

Finally, all of the patients reported experiencing reduced quality of life, impacted by their physical function and social wellbeing as a result of their facial impairment. While physical function appeared to improve over time, social wellbeing worsened. Although these results are only based on a small number of participants, the finding suggest that quality of life was not impacted by labial strength, speech intelligibility, or speech error type. Future work should include measures that index impairments in eating and drinking and/or impairments in facial expression.

4. FUTURE WORK AND LIMITATIONS

These data provide new information regarding the speech abnormalities and their potential impact on speech communication and quality of life. The conclusion, however, are only based on a small number of participants and must be viewed as preliminary. Future work is directed toward the continued longitudinal study of these patients while increasing our participant sample size.

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