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Research paper

Longitudinal progress of overall intelligibility, voice, resonance, articulation and oromyofunctional behavior during the first 21 months after Belgian facial transplantation

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ABSTRACT

Purpose: The purpose of this study is to document the longitudinal progress of speech intelligibility, speech acceptability, voice, resonance, articulation and oromyofunctional behavior in a male facial transplant patient 8 days, 15 days, 5 months, 12 months and, finally, 21 months after surgery.
Method: Identical objective (Dysphonia Severity Index, nasometry, acoustic analysis) and subjective (consensus perceptual evaluation, Dutch speech intelligibility test; flexible videolaryngostroboscopy/naso-endoscopy) assessment techniques and questionnaires (speech and voice handicap index, oral health impact profile, facial disability index) were used during each of the five postsurgical assessments.
Results: The pattern of results shows a longitudinal progress of speech intelligibility and acceptability and of the interactive processes underpinning overall speech intelligibility. Vocal quality is normal and resonance is characterized by hypernasality. The phonetic inventory is complete but four phonetic disorders remain. Outcomes pertaining to articulation (formant analysis) show evident progress over time. Lip functions are improving but still decreased.
Conclusions: Transplantation of the face in this patient has largely restored speech. To what extent resonance, articulation, and lip functions can be enhanced by the permanent use of a palatal obturator, by specialized facial and lip movement exercises in combination with motor-oriented speech therapy, is subject for further research.

Learning outcomes Facial transplantation: Readers will be able to (1) describe the relationship between facial transplantation and the impact on speech and oromyofunctional behavior, (2) identify variables that influence the outcome after facial transplantation, (3) define an assessment protocol after facial transplantation, (4) define facial transplantation.

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1. Introduction

Since 2005 twenty-seven facial transplants were performed (Barret et al., 2011; Cavadas, Ibanez, & Thione, 2012; Devauchelle et al., 2006; Dubernard et al., 2007; Gomez-Cia et al., 2011; Guo et al., 2008; Lantieri et al., 2008, 2011; Petruzzo et al., 2012; Pomahac, Nowinski, & Diaz-Siso, 2011, Pomahac et al., 2012; Roche et al., 2014; Shanmugarajah, Hettiaratchy, & Butler, 2011; Shanmugarajah, Hettiaratchy, Clarke, & Butler, 2012; Siemionow et al., 2010; Siemionow, Gharb, & Rampazzo, 2013; Siemionow & Ozturk, 2011, 2012). In most cases the surgical, immunological and functional (i.e. swallowing, eating and lip movements) outcomes are very well described. However, very few authors reported detailed analyses regarding the progress of overall intelligibility, voice, resonance, articulation and oromyofunctional behavior after facial transplantation (FT). Only in Devauchelle et al. (2006) progress of lip function, swallowing, and articulation was mentioned. In this female patient, aged 38 years, a partial myocutaneous facial transplantation was performed in 2005 in Amiens, France. On postoperative day seven, the FT patient was able to eat and drink almost normally, and three months postoperatively she had the ability to move the upper lip and to articulate the bilabials/p//b/with increased lip closure. Six months postoperatively, complete labial contact was observed as well as mastication allowing normal mobilization of the food bolus. Also, phonation continued to improve. Leakage of drinks from the mouth disappeared 12 months postoperatively. After 18 months, facial functional improvements resulted in reflections of emotional expression in the patient's face, allowing her to produce a symmetrical smile. Five years postoperatively blowing, chewing and swallowing was possible and the patient could speak easily and intelligibly. Pouting and kissing were still difficult. Detailed analysis regarding specific voice, resonance and articulation characteristics, although worthwhile knowing, were not provided.

It still needs to be fully determined to what extent the speech characteristics in se can improve after FT. It is known that achieving close relationships with others and developing a positive identity are associated with a subject's ability to communicate well with others (Stinson & Whitmire, 2000). Based on the available reports of the first FT patient, a substantial improvement of speech acceptability, speech intelligibility and the interactive processes (voice, resonance, articulation) behind it, can be expected. The purpose of this study is to document the longitudinal progress of speech intelligibility, speech acceptability, voice, resonance, articulation and oromyofunctional behavior in a male FT patient 8 days, 15 days, 5 months, 12 months and, finally, 21 months after surgery.

2. Methods and materials

This research was approved by the ethical board (2012/809) of the Ghent University Hospital. Identical objective and subjective assessment techniques and questionnaires were used 8 and 16 days, 5, 12 and 21 months after facial transplantation to determine speech characteristics (speech intelligibility, speech acceptability, voice, resonance and articulation) and oromyofunctional behavior. The speech assessments were performed independently by two speech pathologists (KVL, MDL) who were not involved in the daily speech training sessions.

2.1. Subject

The subject was a 54-year-old man, who was admitted to the emergency department in December 2010 due to a ballistic injury to the face. He presented with not only a major soft tissue defect of the lower two thirds of the face, but also an extensive loss of facial bony structures, both maxillae and the left part of the mandible. Vision was lost, as both eyes were involved. The defects were temporarily approximated after debridement and part of the facial fractures stabilized with fixation and reconstruction titanium plates. Five days post trauma, a free anterolateral thigh flap was used to temporarily reconstruct the defect. Plication of the free flap provided coverage of the external skin defect and separation of the oral and nasal cavity in a one-stage procedure. A tracheostomy was required for breathing, as well as a percutaneous gastrostomy tube for feeding. The patient was not able to swallow nor to eat, and speech was very unintelligible. Despite intensive postoperative speech rehabilitation (4–5 sessions/week during six months) swallowing and overall speech intelligibility remained severely disordered.

Clinical evaluation and radiological examination based on CT-scans with three dimensional reconstruction and printing provided an inventory of missing facial structures (see Figs. 1 and 2). Deficient soft tissue included the left side of nose with the nasal cartilages, left lower eyelid, left cheek, including the left upper lip and left oral commissure. The destructed muscles included the left orbicularis oculi, all levators and depressors of the mouth and almost all of the orbicularis oris. The missing bony structures were the medial orbital wall and orbital floor bilaterally, nasal bones, loss of all the hard palate and the horizontal portion of the left mandible from symphysis to ramus ascendens (Roche et al., 2014).

The extensive complexity of the defect and the poor postoperative clinical outcome made it obvious that the conventional reconstructive approach would require multiple extra procedures without guaranteeing an acceptable functional and aesthetic result. Therefore, transplantation of the face was considered as an option to restore swallowing, eating and speech and to re-establish aesthetics in a one-stage procedure. Several multidisciplinary assessments and cadaver dissection training sessions preceded the complex facial transplantation. In a 20-h surgical procedure, a digitally planned FT was performed consisting of a large amount of bone together with the soft tissues of the entire lower 2/3rd of the face. Vascular anastomoses were performed on both facial arteries and veins. A detailed description of both the preparation of the surgery, the surgical procedure of the facial transplant, the immunosuppressive induction protocol and the results of the surgery is

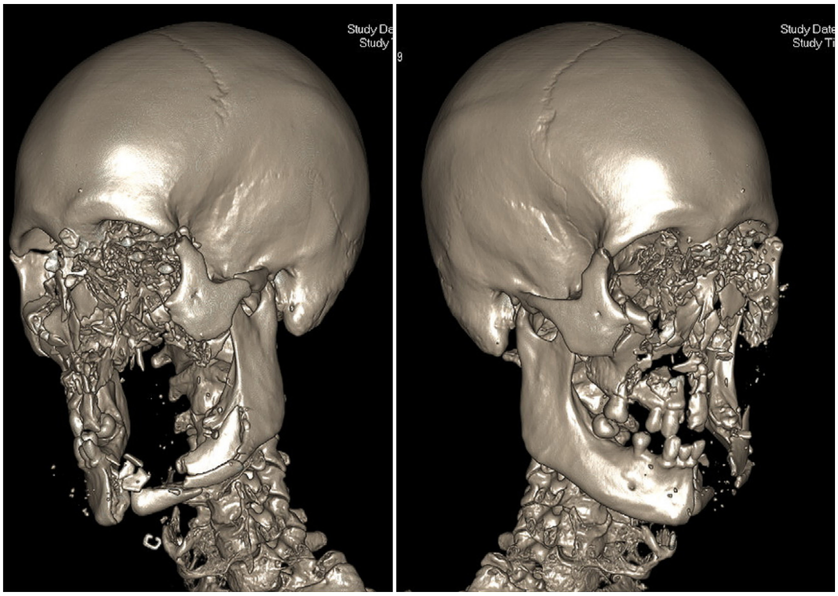

Fig. 1. Defect with extensive loss of facial bony structures.

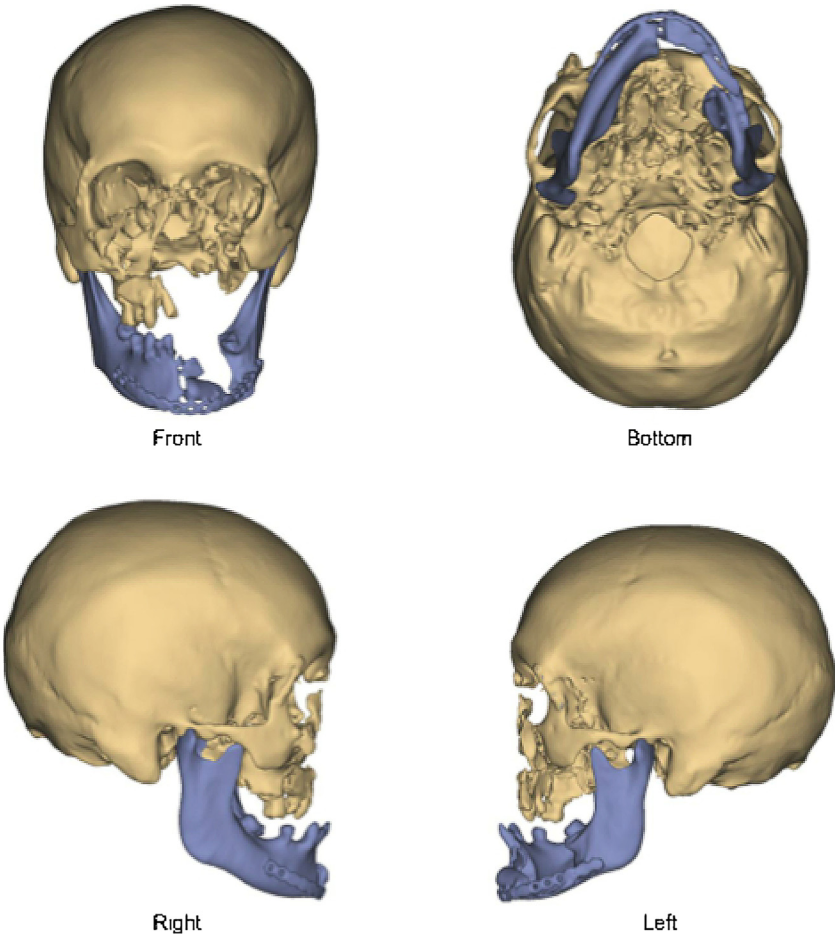

Fig. 2. Pre-operative 3D.

described in Roche et al. (2013). Survival of the graft was complete, the bony structures (both maxillae, part of the left mandible) and mucosal lining of the nasal cavities and hard palate could all be vascularized by connecting only the facial vessels (Fig. 3).

No intraoperative surgical complications occurred and the immediate postoperative course was uneventful. The patient was able to swallow liquids and to produce vowel speech six days after the transplantation. Speech rehabilitation started one week postoperatively, 4–5 times per week during the first three months and focused on breathing, swallowing, oral motor functions, overall speech intelligibility, voice, resonance and articulation of phonemes, syllables, words and short sentences (Van Lierde et al., 2013). After three months the frequency of therapy sessions was reduced to three times per week. The patient was intrinsically motivated to follow speech therapy and his partner functioned as a co-therapist (Lemmens et al., 2014). During logopaedic therapy sessions, the patient was given corrective feedback after every assignment to improve or correct specific productions, using auditory-verbal instructions as well as tactile stimuli. Positive reinforcement was given after every correct attempt and simple yes/no questions were asked, encouraging the patient to comment on his own performance. Also tactile recognition of the facial structures and facial massage were initiated, together with low-vision training. Oropharyngeal endoscopy on postoperative day 26 showed no signs of infection, ischemia or necrosis in the oronasal cavity. Regarding the velopharyngeal mechanism, an adequate lifting of the soft palate with lateral pharyngeal wall constriction during the production of the vowel/a/was observed (100% consensus evaluation, HV, KVL). A fistula located between the hard and the soft palate was responsible for a moderate degree of hypernasality (i.e. excess resonance of vowels and voiced consonants within the nasal cavities). An obturator prosthesis was developed to restore the anatomical structures between the oral and nasal cavity. Pure-tone testing revealed normal hearing sensitivity in both ears. Sensory-motor recovery started between 5 and 8 months postoperatively. Sensory recovery started with restoration of sensations in the oral mucosa and sensations caused by gently touching the chin or one of both cheeks. Recovery of heat and cold sensation started 6 months postoperative. Movements of mouth corners and cheeks and lifting of the nose started 8 months postoperative. Several post-operative complications were observed. At the end of the third month, the patient developed swelling and increasing pain at the left jaw during eating and mime therapy exercises. CT-scan revealed an abscess at an osteosynthesis screw. The abscess was surgically removed. Clinical signs of one acute rejection were diagnosed 6 months postoperatively

Fig. 3. Postoperative 3D.

with total recovery. At twelve months a pulmonary aspergilloma relapsed with clinical symptoms of fever and radiologic progression. The patient had to be hospitalized for 4 weeks. After medical treatment the clinical situation remained stable.

3. Methods and materials

3.1. Speech intelligibility

Speech samples were collected by means of a picture-repeating test (see articulation) in order to judge overall intelligibility of words, sentences and spontaneous speech on a four level ordinal scale (0 = normal speech intelligibility, 1 = slightly, 2 = moderately and 3 = severely impaired speech intelligibility). All analyses were based on a consensus narrow phonetic transcription made by two experienced speech pathologists (KVL, MDL) using the symbols and diacritics of the International Phonetic Alphabet. The speech language pathologists first simultaneously but independently transcribed the samples before comparing transcriptions aiming at a consensus.

Furthermore, the Dutch speech intelligibility test (De Bodt, Guns, & Van Nuffelen, 2006) was applied, requiring the patient to repeat words and sentences. These speech samples were videotaped and subsequently transcribed by the same two speech language pathologists in order to calculate the percentage of sounds produced correctly.

The Speech Handicap Index (SHI) (Van den Steen et al., 2011) is a self-assessment questionnaire reflecting the functional, physical and psychological impact of a speech disorder on the quality of life. The subject has to respond according to the appropriateness of each item (0 = never, 1 = almost never, 2 = sometimes, 3 = almost always, 4 = always). The final result varies between 0 and 60, with the latter representing a maximum perceived impact of the speech disorder on the overall quality of life.

3.2. Speech acceptability

Intelligibility and acceptability of speech are different concepts. Dagenais, Brown, and Moore (2006) described acceptability as the answer to the question, "How would you rate this person's speaking ability?" thus emphasizing the notion that speakers might be able to make themselves intelligible, but at the expense of naturalness, marking their speech as atypical but along different parameters from unintelligible speech. To judge speech acceptability (consensus evaluation by KVL, MDL), a four level ordinal scale was used (0 = normal speech acceptability, 1 = slightly, 2 = moderately and 3 = severely impaired speech acceptability).

3.3. Voice

Subjective assessment: Voice assessment included flexible videolaryngostroboscopy and a consensus perceptual rating during connected speech. Voice ratings were performed by two experienced speech language pathologists (KVL, MDL) on the GRBASI scale (Hirano, 1981). The GRBASI assessment consists of five well-defined parameters: G (overall grade of vocal pathology), R (roughness), B (breathiness), A (asthenicity), and S (strain). A four-point rating scale (0 = normal, 1 = slight, 2 = moderate, and 3 = severe) is used to indicate the grade of each of the six parameters. In addition to GRBASI assessments, vocal pitch and intensity were judged as "normal", "increased" or "decreased". Voice samples of connected speech during reading were audio-recorded for further analysis. Both speech pathologists first independently rated each voice sample. In case of disagreement, the samples were replayed and discussed aiming at a consensus score.

The Voice Handicap Index (VHI) (Jacobson & Johnson, 1997) was used to measure the subject's perceptions of the psychosocial impact of his vocal problem. The VHI is a self-administered questionnaire that consists of 30 questions or statements. The subject has to respond according to the appropriateness of each item (0 = never, 1 = almost never, 2 = sometimes, 3 = almost always, 4 = always). The VHI score varies between 0 and 120, with the latter outcome representing the maximum perceived disability due to vocal difficulties.

Objective assessment: Aerodynamic measurement: The Maximum Phonation Time (MPT) was measured on the basis of two test trials with the vowel/a/, sustained at the subject's habitual loudness and pitch in free field and in sitting position. The duration of the sustained phonation was measured in seconds on a chronometer. Verbal and visual encouragement and coaching was given during the task. The best of both trials was retained for further analysis. Vocal range: Frequency and intensity range were measured using with the Voice Range Profile function of the Computerised Speech Lab (CSL) (Kay Elemetrics, Lincoln Park, HY, 1992). The patient was instructed to inhale in a comfortable way and produce the vowel/a/for at least 2 s, using a habitual pitch and loudness, a minimal pitch (*F*-low), a minimal intensity (*I*-low), a maximal pitch (F-high), an a maximal intensity (*I*-high), respectively. Acoustic analysis: Determination of acoustic voice parameters was performed using the Multi Dimensional Voice Program (MDVP) of the CSL (Kay Elemetrics, 1992). The subject was asked to sustain the vowel/a/ using his habitual tone and without unnecessary effort. A three seconds midvowel segment registered with a sampling rate of 50 kHz was used for analysis. The parameters jitter (%) (perceived as hoarseness) and fundamental frequency (F_0 in Hz) were determined. The overall objective vocal quality was measured by means of the Dysphonia Severity Index (DSI), which is designed to establish an objective and quantitative correlate of perceived vocal quality (Wuyts et al., 2000). The DSI is based on a weighted combination of the following set of voice parameters: MPT (seconds), highest frequency (*F*-high in hertz), lowest intensity (I-low in decibels), and jitter (%). The DSI equation is $(0.13 \times \text{MPT}) + (0.0053 \times F_0\text{-high}) - (0.26 \times I\text{-low}) - (1.18 \times \text{jitter}) + 12.4$. The DSI score ranges from +5 to −5, corresponding with normal and severely dysphonic voices respectively (i.e. the more negative the DSI value, the worse the patient's vocal quality).

3.4. Resonance

Subjective assessment: The velopharyngeal valve mechanism was evaluated by means of flexible naso-endoscopy. It was also perceptually judged by two clinicans (HV, KVL). To evaluate the degree of perceived hypernasality and/or nasal emission, an ordinal scale with five categories was used (1 = normal resonance, 2 = mild hypernasality/nasal emission, 3 = moderate, 4 = severe, 5 = very severe). The hypernasality and nasal emission test designed by Bzoch (1989) was administered. In this procedure, the patient is asked to repeat a series of 10 word pairs, while alternately closing and opening the nares. Rating is done on a scale ranging from 0 to 10, with 0 corresponding to normal resonance (i.e. no perceptual difference between word pairs produced with open vs. closed nares). Words and sentences produced during the picture-naming test were scored by two speech pathologists (KVL, MDL). They first scored the samples independently, and in case of disagreement, the sample was replayed and discussed until a consensus could be reached.

Objective assessment: The Nasometer (model 6300), a Kay Elemetrics microcomputer-based system, was used for measurement of nasalance values. Prior to initiating data collection, the Nasometer was calibrated following the procedure outlined in the manual. The patient was asked to sustain three vowels (/a/,/i/,/u/) and to read two nasometric passages. The "Rainbow passage", an oronasal text with 9.7% (31/318) nasal sounds, and the "Zoo passage", a text containing only oral sounds were read to detect the presence of hypernasality and/or nasal emission.

3.5. Articulation

Subjective assessment: Speech samples for the assessment of articulation were elicited by means of a picture-repeating test. This test requires the speech therapist to name black and white drawings of common objects and actions, the verbal label of which is then to be repeated (by the patient). The speech samples thus collected consisted of 135 different words, containing instances of all Dutch sounds, as well as most consonant clusters in all possible syllable positions. The samples were recorded digitally for further analysis in a sound-treated room of the speech- language and hearing department at the University of Gent. The evaluation included a phonetic inventory and a phonetic analysis. The phonetic inventory reveals which consonants and vowels a patient is capable of producing correctly in his native language. This analysis was conducted without making reference to the intended target sounds. A sound was considered to be present in the inventory when at least two instances of correct productions (i.e. consistent with the standard realization of the sound) were found. In the phonetic analysis, consonant and vowel productions were compared with target productions and analyzed for error types on the segmental level. All analyses were based on a consensus narrow phonetic transcription made by two experienced speech pathologists (KVL, MDL) using the symbols and diacritics of the International Phonetic Alphabet. Both speech language pathologists first simultaneously but independently transcribed the samples before comparing transcriptions and aiming at a consensus.

Objective assessment: To describe the quality of vowel production an objective acoustic analysis of formant frequencies was used. The first two formants (F1 and F2) are considered to be the most important because, based on those two formants, a listener will be able to identify a given vowel. Determination of F1 and F2 frequencies of vowels offers the possibility to describe vowels in terms of high/low and front/back placement of the tongue in the oral cavity and in terms of jaw opening. Also, the effect of lip protrusion is reflected in the frequencies of both formants in that rounded vowels have an overall lower formant structure. The vowels/a/,/i/and/u/represent the extreme articulatory positions of the tongue in English as well as in Dutch. An F1:F2 scatterplot of these vowels yields a so-called 'vowel triangle', i.e. a graphic representation of the articulation space for vowel production with/a/,/i/and/u/as 'corner vowels'. Vowel space, i.e. the surface of the scatterplot shape, is a well-known metric that is often used to objectively describe the degree of articulatory precision. When producing a single vowel, a speaker can concentrate on motor actions per se, to demonstrate maximum motor capabilities. Articulatory maneuvers to produce vowels in a phonetic context, on the other hand, reflect a speaker's ability to deal with neutralization and co-articulation. Neutralization pertains to the decrease of movement amplitudes, proportional to speaking rate. Co-articulation is the inevitable mutual influence of movements needed for neighboring segments in connected speech. Both neutralization and co-articulation are related to intelligibility. In this study, both isolated vowels and vowels in context are studied. Isolated/a/,/i/and/u/vowels were recorded digitally in a sound booth in Ghent University. Midvowel fragments with stable formant patterns were selected (using visual inspection of the oscillogram and the spectrogram) by means of Praat software (Boersma & Weeninck, 2013, version 5.3.52). These fragments had a duration between 500 ms and 800 ms. In addition to isolated vowel samples, [i], [a] and [u] samples were also drawn from digital recordings of monosyllabic nonsense words (NSVO assessment) using the same apparatus and software. As was the case for isolated vowels, mid-vowel segments were selected on graphical displays and were verified auditorily before extracting them using a Gaussian window. The fragments extracted from monosyllabic words had a typical duration of 100 ms, except for some instances of the vowel [a], which is intrinsically longer and therefore often provided stable formant patterns of 200 ms for extraction. To represent each corner vowel, 3 (for isolated vowels) to 6 (for vowels in monosyllable context) fragments were concatenated. These sound files were and subsequently processed using a Praat script involving the Burg algorithm, in order to identify the 50th percentile values of the first two formants. The 50th percentile was chosen as a metric because it is less influenced by artifacts and outliers.

From these formant data, vowel space can be quantified. For vowels in a phonetic context, more specific quantitative indices were derived, including vowel-to-vowel contrasts for the first formant (which is indicative of tongue height and mandibular maneuvers) and the second formant (which is indicative of tongue position in the sagittal plane).

3.6. Oromyofunctional assessment

Five oromyofunctional functions were judged as proposed in the protocol of Lembrachts, Verschueren, Heulens, Valkenburg, and Feenstra (1999), namely lip function (lip position at rest, lip closure, dispersion of the corners of the mouth, lip protrusion, lip strength, lip position during swallowing), tongue function (tongue position at rest, tongue protrusion, tongue retraction, tongue lifting against the upper lip, tongue depression against the lower lip, lateral movements of the tongue, tongue position during swallowing), blowing, sucking and the presence of drooling. Also facial emotional readability (smiling/surprised/sad/angry) was judged on a three-point rating scale (0 = normal function, 1 = decreased function, 2 = function impossible). The above-mentioned speech pathologists (KVL, MDL) first rated independently. In case of disagreement, the samples were replayed and discussed until a consensus was reached.

The Dutch version of the Oral Health Impact profile (OHIP-14) was used (Van der Meulen, John, Naeije, & Lobbezoo, 2008). The OHIP-14 is a self-completion questionnaire that focuses on seven dimensions of oral health impact. The 'functional limitation' domain concerns the loss of function of parts of the body, like difficulty with chewing. The 'physical discomfort' and 'psychological discomfort' domains deal with experiences of pain and discomfort, such as toothache and feeling miserable. The domains labeled 'physical disability', 'psychological disability' and 'social disability' refer to limitations in performing daily life activities, like avoiding certain foods, lack of concentration and feeling irritable with others, respectively. Finally, the 'handicap' domain concerns a sense of disadvantage in functioning, like suffering financial loss because of dental problems. Answers to the 14 questions are scored on a five-point ordinal scale, ranging from 'never' (score 0), 'hardly ever' (score 1), 'occasionally' (score 2), 'fairly often' (score 3) to 'very often (score 4). Thus, higher scores imply a more impaired oral health-related quality of life. The 14 scores are summed yielding a global result (ranging from 0 to 56). Similarly, separate domain scores can be obtained. The patient was also asked to rate overall 'oral health' satisfaction with the transplanted oral cavity on a visual analogue scale with 100% reflecting complete satisfaction and 0% corresponding to completely not satisfied.

The Facial Disability Index (FDB) (VanSwearingen & Brach, 1996) is a reliable and valid self-report questionnaire on physical disability and psychosocial factors related to facial neuromuscular function. The FDI can be used as an initial assessment tool and as a monitoring instrument, providing the clinician with the patient's view of the outcome during interventions. The scores on the physical and psychosocial scale are transformed to a 100-point basis (with 100% reflecting no facial disability).

4. Results

Table 1 offers a synopsis of all outcomes over time, as well as appropriate normative indices to evaluate them.

4.1. Speech intelligibility and acceptability

Speech intelligibility (100% consensus evaluations of words, sentences and spontaneous speech) changed from severely impaired (during the first 15 days post-transplant) towards slightly/moderately impaired (5 months postoperative). Twelve and 21 months after transplantation, intelligibility was evaluated as normal in single words and as slightly impaired in sentences and spontaneous speech. Speech intelligibility scores in words increased from 72% resp. 76% during the first 15 days post-operative to 84% resp. 90% during the following 5 months and, from then on, stayed approximately at the same level, i.e. 80% (12 months post-op) and 88% (21 months post-op). Speech intelligibility scores in sentences were not available during the first 15 days post-operative because the patient was not able to produce sentences. One month post-transplantation the production of sentences became possible yielding scores increasing from 77% (1 month post-op) to 93% (21 months post-operative).

Speech acceptability (consensus evaluation 100%) changed from severely impaired (first 15 days post-transplant) over moderately impaired (1 and 5 months post-surgery) to slightly impaired (12 and 21 months post-transplantation).

4.2. Voice and resonance

At one month at and 5 months post-operative, the VHI showed a psychosocial impact of the vocal problem on the quality of life. However, no psychosocial impact of the vocal problem was perceived immediately post-operative and 1 year after the FT. The perceptual evaluation of the overall grade of vocal pathology changed from moderately impaired (8 days post-operative) over slightly impaired (15 days and 1 month post-operative) to normal (5, 12 and 21 months). During the first month after FT, the voice was rough, breathy, asthenic and strained. One month after FT it was possible to calculate the DSI, yielding values increasing from +1.4 (1 month post-operative) to +3.6 (21 months post FT).

The moderate degree hypernasality that was noticed (consensus evaluation 100%) during the first 5 months after FT decreased to slight hypernasality after 12 month. During the Bzoch hypernasality tests, from the first assessment

Table 1
Results (and reference data as found in the literature) of the assessments of speech intelligibility, speech acceptability, voice, resonance, articulation (vowel triangle data based on vowels in monosyllable words) and oromyofunctional behavior 8 days, 15 days, 5 months and 12 months and 21 months after facial transplantation. NT indicates 'not tested' at that specific post-operative moment. X/X*: nasalance values without obturator/nasalance values with obturator*.The reference data (last column) are mentioned as criteria to evaluate intermediate results obtained during successive postsurgical assessments, e.g. the Dysphonia Severity Index (DSI) reference value for normal voices is +2.5. During the first two post-operative assessments it was impossible to determine DSI values because the patient was not able to perform the nessecary vocal range sound productions. From one month post-operative onwards it became possible to determine the DSI, yielding a values of +1.4. At 5, 12 and 21 months postoperative, DSI values were above the norm/reference and within the normal range.

	Results 8 days post-op	Results 15 days post-op	Results 1 month post-op	Results 5 months post-op	Results 12 months post-op	Results 21 months post-op	Reference
Speech intelligibility							
Consensus perc. evaluation							
Words	3	2	1	1	0	0	0
Sentences	NT	3	2	2	1	1	0
Spontaneous speech	NT	3	3	2	1	1	0
Dutch speech intelligibility score							
Words (%)	72	76	84	90	80	88	100
Sentences (%)	NT	NT	77	78	91	93	100
Speech Handicap Index	23	15	18	18	14	19	5/range: 4–6
Speech acceptability	3	3	2	2	1	1	0
Voice							
Voice Handicap Index	17	15	26	29	18	11	<20/120 no disabilities
Consensus perceptual evaluation							
Vocal quality	G2R2B1A2S1	G1R1B1A1S0	G1R1B0A0S0	G0R0B0A0S0	G0R0B0A0S0	G0R0B0S0	G0R0B0A0S0
Pitch	1	0	0	0	0	0	0
Intensity	1	0	0	0	0	0	0
Aerodynamic measurement Maximum phonation time (s)	10	15	18	17	18	22	22/range: 6.7–37
Vocal range							
Softest intensity (dB)	NT	NT	60	58	55	55	51 (range: 46–57)
Loudest intensity (dB)	NT	NT	80	95	100	101	97 (range: 81–112)
Lowest frequency (Hz)	NT	NT	146	440	135	82	142 (range: 96–188)
Highest frequency (Hz)	NT	NT	493	824	694	670	867 (range: 453–1282)
Acoustic analysis							
Fundamental frequency (Hz)	120	120	122	124	122	119	122 (range: 78–166)
Jitter	1.76	0.52	0.41	0.37	0.29	0.85	0.81 (range: 0–2.1)
Dysphonia Severity Index	NT	NT	+1.4	+3.8	+ 3.9	+3.6	+2.5 (range -5 ± 5)
Resonance							
Consensus perceptual evaluation							
Hypernasality	3	3	3	3	2	2	0
Nasal emission	0	0	0	0	0	0	0
Bzoch hypernasality test	NT	NT	10/10	10/10	10/10	10/10	0/10
Bzoch nasal emission test	NT	NT	0/10	0/10	0/10	0/10	0/10
Nasalance values							
Vowel /a/ (%)	NT	NT	NT	40	25	27/20*	20(range: 0–49)
Vowel /i/ (%)	NT	NT	NT	22	61	54/26*	26(range: 0–55)
Vowel /u/ (%)	NT	NT	NT	14	37	46/9*	9 (range: 0–24)
Oronasal passage (%)	NT	NT	NT	38	47	48/47*	34 (range: 23–45)
Oral passage (%)	NT	NT	NT	37	43	54/39*	11(range: 2.5–20)
Articulation							
Consensus perceptual evaluation							
Phonetic inventory	12/22	21/22	22/22	22/22	22/22	22/22	22/22
Phonetic analysis (amount and specific consonants distorted)	10 /pbmwszfvtd/	10 /pbmwszfvtd/	6 /pbmwsz/	4 /pbmw/	4 /pbmw/	4 /pbmw/	0
Vowel triangle (vowels in monosyllable context)							
F1 frequency (tongue height + mandibular movements) (Hz)							
for /a-i/ F1 contrast	267	354	411	409	361	322	407($\pm$1sd: 294–519)
for /a-u/ F1 contrast	265	342	335	403	371	307	455($\pm$1sd: 356–554)
F2 frequency (sagittal tongue position + lip protrusion) (Hz) for /i-u/ F2 contrast							
Vowel space (kHz^2)	637	647	749	970	814	1058	1194 ($\pm$1sd: 987–1401)
	85	112	131	196	147	165	

Table 1 (*Continued*)

	Results 8 days post-op	Results 15 days post-op	Results 1 month post-op	Results 5 months post-op	Results 12 months post-op	Results 21 months post-op	Reference
Oromyofunctional behavior							
Consensus perceptual evaluation							
Lip function	2	2	2	1	1	1	0
Lip position at rest	2	2	2	2	2	1	0
Lip closure	2	2	2	1	1	1	0
Dispersion of the corners of the mouth	2	2	2	1	1	1	0
Lip protrusion	2	2	2	2	1	1	0
Lip strength	2	2	2	2	1	1	0
Lip position during swallowing	0	0	0	0	0	0	0
Tongue function	1	1	1	0	0	0	0
Tongue position at rest	1	0	0	0	0	0	0
Tongue protrusion	1	1	0	0	0	0	0
Tongue retraction	1	1	0	0	0	0	0
Tongue lifting	2	2	2	1	1	0	0
Tongue depression	2	2	1	0	0	0	0
Lateral tongue movements	2	2	2	2	2	2	0
Tongue position during swallowing	2	2	2	2	2	2	0
Blowing	Present	Present	Present	Absent	Absent	Absent	Absent
Oral Health Impact Profile	2	2	2	2	1	1	0
Total score							
Facial Disability Index	15/56	15/56	8/56	10/56	13/56	8/56	0/56
Physical function (%)	30	55	65	56	85	60	100
Social/well-being function (%)	72	72	72	84	95	90	100

(1 month postoperative) to the last assessment (21 months), all words were produced with hypernasality. No nasal emission was observed (consensus evaluation 100%). Nasalance values changed irregularly during the posttransplant period. The lowest nasalance values for vowels (except for the vowel/a/) as well as for the nasometric passages were obtained 12 months after transplantation. During the last assessment (21 months) a difference in nasalance values with and without the obturator was measured in all vowels and in the nasometric passages.

4.3. Articulation

The phonetic inventory was complete from 1 month postoperative onwards until the last assessment (21 months). An increase of the amount of phonetic distortions was observed (consensus evaluation 100%) in the post-transplant period. Ten phonetic distortions were noticed during the first 15 postoperative days and 6 disorders were observed 1 month after FT. Five, 12 and 21 months after FT 4 phonetic disorders were observed.

Outcomes pertaining to articulation in terms of formant analyses show evident progress over time. In the scatterplots it can be seen that over the course of 21 months, vowel space nearly doubles for both isolated vowels (from 120 to 233 kHz^2 (see Fig. 4) and vowels within a phonetic context (from 85 to 165 kHz^2) (see Fig. 5). Fig. 6 shows that vowel space expansion came gradually, reaching a maximum value at 5 months postoperatively, followed by a limited downfall at 1 year postoperatively. Typical outcomes for vowels produced in a monosyllable context by in male native speakers of Dutch can be derived from data on 50 normal participants in Pols, Tromp, and Plomp (1973). According to these reference data, the last vowel space result registered 21 months postoperatively comes very close to the lower boundary of the ± 1 standard deviation bracket. Since vowel space is a composite index, discrete vowel to-vowel contrasts should also be inspected. These vowel-to-vowel contrasts are illustrated by the intervals along the vertical and horizontal scatterplot axes and precise values are given in Figs. 7 and 8. There is a different progress rate, depending on the type of articulatory maneuver. The F1 [a-i] contrast (governed by differential tongue height and jaw opening for [i] versus [a]) falls within the ± 1 standard deviation bracket from 15 days postoperatively onwards. Vowel-to-vowel contrasts where the rounded vowel [u] is involved grow less. As to the F1 [a-u] contrast, only the 5 month and 12 months post-operative values fall within the ± 1 standard deviation bracket. The only outcome for the F2 [i-u] contrast (governed by differential tongue position in the sagittal plane and by lip protrusion) within the 1 standard deviation bracket normal mean is at 21 months postoperatively.

4.4. Oromyofunctional assessment

The consensus perceptual evaluation (95%) revealed an inability to move the lips during the first postoperative month. Five months after FT lip position at rest, dispersion of the corners of the mouth and lip protrusion became possible, albeit in a reduced manner. Twelve months after FT all lip functions were present but decreased (except for lip closure, which was impossible). During the last assessment at 21 months all lip functions were present, but still decreased.

Perceptual evaluation (95% consensus) revealed a normal tongue position from the first 8 days after FT, but impossible lateral tongue movements and impossible tongue functions during swallowing. Five months after FT all tongue functions

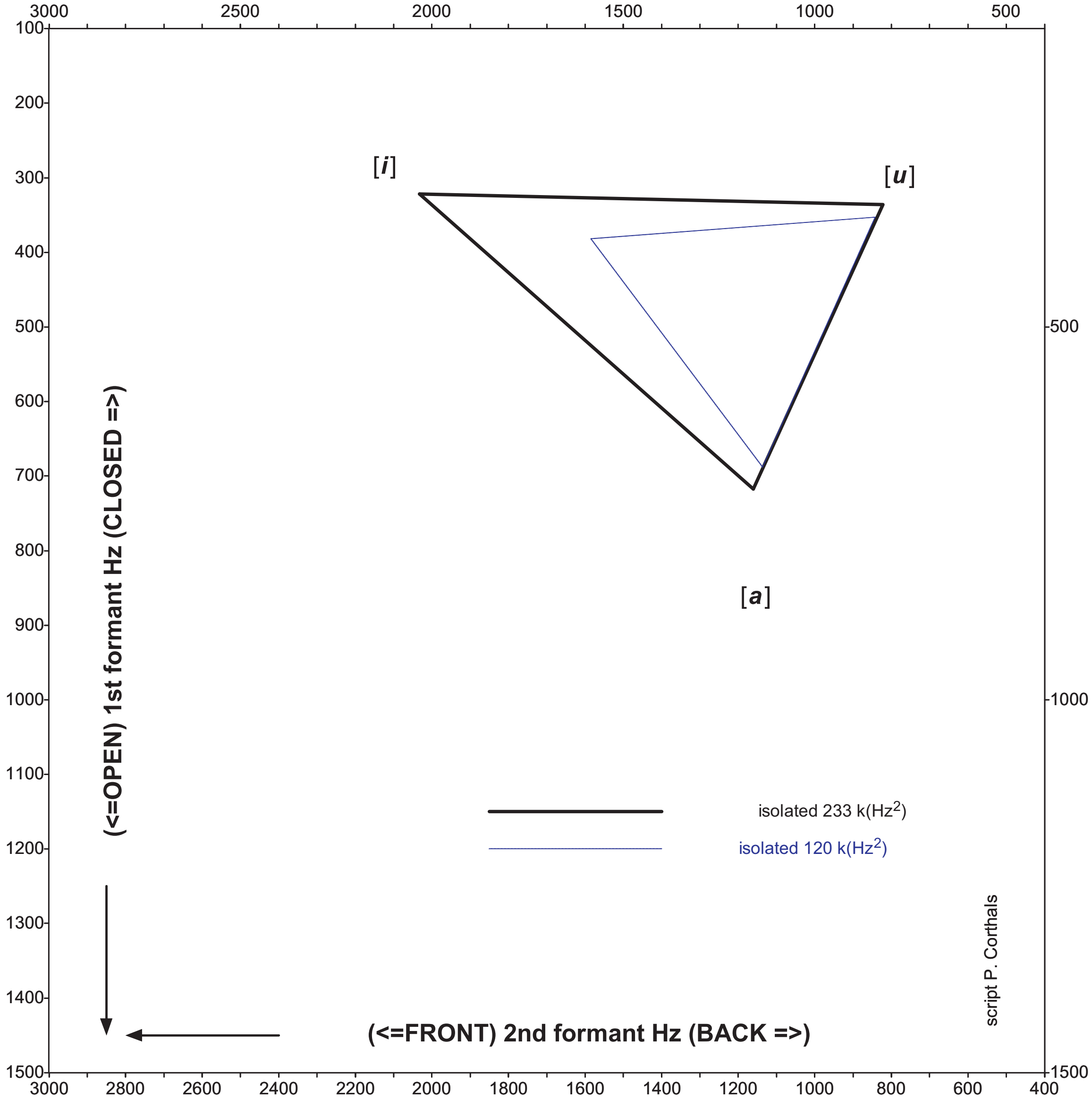

Fig. 4. F1:F2 scatterplot for sustained corner vowels registered at the start (8 days post-operative) and at the end (21 months post-operative) of the study.

were present except for lateral tongue movements, which were decreased in amplitude. During the last two assessments (12 and 21 months after FT) all tongue functions were normal. Blowing and sucking was impossible during all assessments. Drooling was absent from 5 months postoperative onwards. Facial emotional readability was present 12 months after FT. The impact of oral health on the QOL generally decreased from 1 month postoperative until 12 months and 21 months after FT. Only at 12 months after FT a slightly worse result (further from the reference data) on the oral health impact profile was seen. The Facial Disability Index (both the psychological functions and the social well-being functions) showed an improvement at 12 months postoperative (closer to the reference).

5. Discussion

The purpose of this study was to describe the longitudinal progress of several speech characteristics after FT. Transplantation of the face in this patient was expected to restore speech and to re-establish aesthetics in a one-stage procedure. The findings of this study show that, as hypothesized, all speech characteristics in this FT patient improved over time. Very few authors reported detailed analysis of the progress of the different speech characteristics after FT (see Table 2). Comparison of the evolution of speech characteristics in reported facial transplant cases is very difficult, taking into account

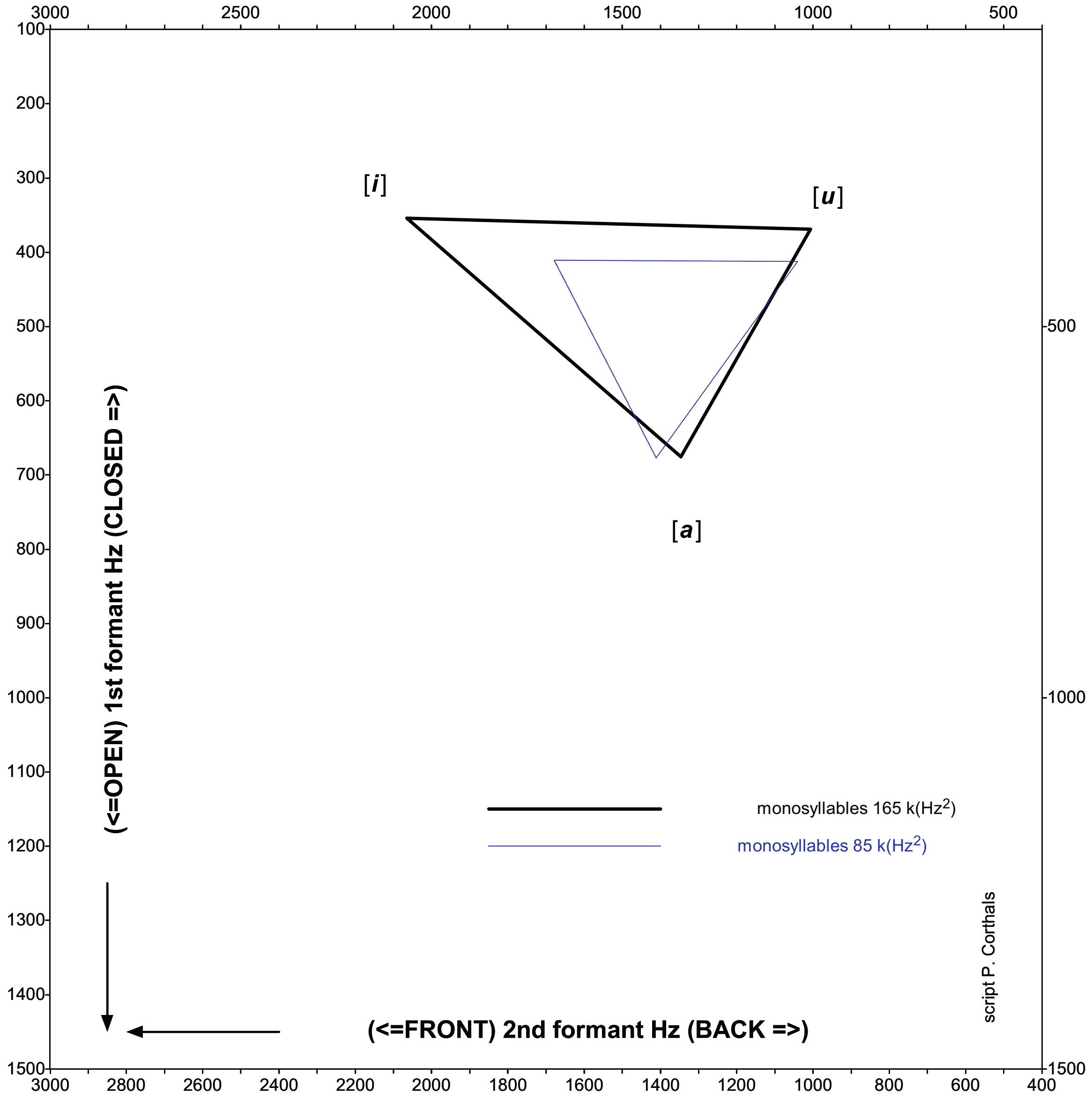

Fig. 5. F1:F2 scatterplot for corner vowels in monosyllable context registered at the start (8 days post-operative) and at the end (21 months post-operative) of the study.

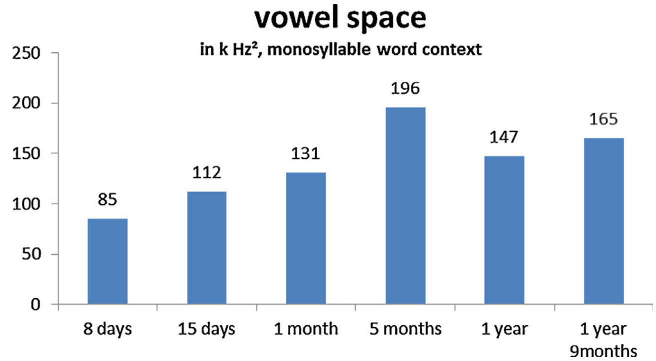

Fig. 6. Vowel space outcomes in monosyllable context registered at all postoperative assessments.

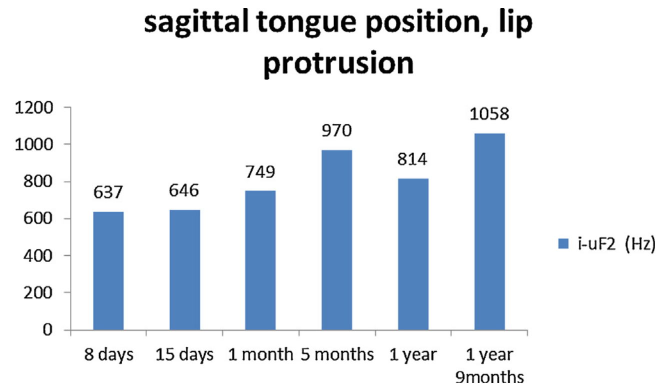

Fig. 7. Vowel-to-vowel 2nd formant contrasts in monosyllable context registered at all the postoperative assessments.

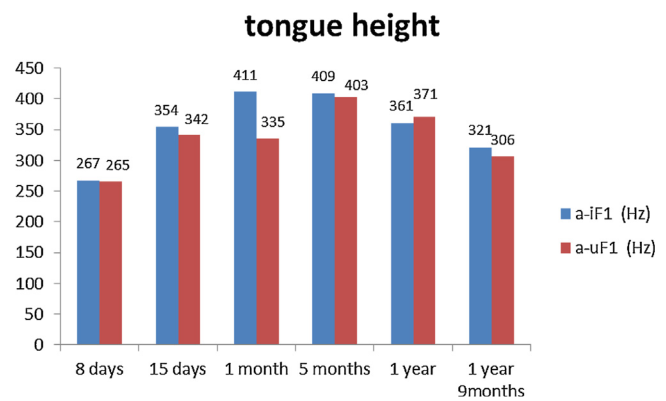

Fig. 8. Vowel-to-vowel 1st formant contrasts in monosyllable context registered at all postoperative assessments.

the uniqueness of each defect. The most detailed description of functional outcome is available for the first FT performed by Devauchelle et al. (2006). Five years after transplantation, this patient could smile, chew, swallow, and blow normally, whereas pouting and kissing remained difficult. Moreover, this patient could talk easily and intelligibly. In the reports of Lantieri et al. (2008, 2011) and Siemionow et al. (2010), the patients were reported to be able to speak and to produce intelligible speech. Our study is unique in that it shows a longitudinal evolution (according to subjective and objective assessment results) of speech intelligibility, speech acceptability, voice, resonance, articulation and oromyofunctional behavior over a period of 21 months after FT. The pattern of results showed progress in acceptability as well as in intelligibility and all the interactive functions underpinning it (voice, resonance, articulation).

In this patient a digitally planned FT was performed, consisting of a large amount of bone together with the soft tissue of the entire lower 2/3rd of the face. As far as speech physiology is concerned, the muscle systems of the vocal tract (supralaryngeal structures) responsible for both resonance, articulation and oromyofunctional behavior were involved. The presence of a moderate vocal problem and the decreased DSI value during the first month post transplantation must be regarded as a temporary sequel of the 20-h surgical procedure. Resonance and articulation are the interactive processes underpinning speech intelligibility and acceptability. Both the resonance and articulation characteristics continued to improve in the postsurgical period until the last assessment. The phonetic inventory was complete 1 month after transplantation and only four phonetic disorders (disordered bilabials/p,b,m,w/) remained after 5 months. Most probably, there is a causal relationship between the persistency of these phonetic disorders on the one hand and on the other hand the improving but still reduced lip functions. Detailed acoustic analysis of vowel articulation revealed that vowel space expansion was more prominent for isolated vowels. Isolated vowel data reflect motor actions per se, illustrating maximum motor capabilities and including compensatory maneuvers. When a speaker produces complete words, co-articulation sets in (i.e. interference by articulatory maneuvers needed for preceding or following speech sounds) as well as neutralization (i.e. the tendency to lessen the amplitudes of all articulatory maneuvers with growing speech rate). In the F1 [a-i] and [a-u] contrasts, where control over jaw opening is a common factor, this neutralization effect is limiting progress, particularly in the most recent recordings. On the other hand, faster speech rate can be seen as a sign of improvement in itself. In this patient, the expansion of both types of vowel space values is due mainly to an upward shift of F2 in [i]. This is the acoustic equivalent of better frontal positioning of the tongue. According to the vowel-to-vowel contrast outcomes where [u] is involved, there is less progress when it comes to dorsal tongue elevation and lip protrusion. In normal speakers, lip

Table 2
Summary of studies on the functional outcome of facial transplantation cases as found in the literature (M: male, F: female, d: days; m: months; y: years).

Surgical team/authors	Date	Location	Recipient age/sex	Allograft	Functional outcome
Devauchelle et al. (2006) Dubernard et al. (2007) Petruzzo et al. (2012)	November 2005	Amiens, France	38,F	Partial myocutaneous	7 d:able to eat and drink almost normally 3 m:ability to move the upper lip 3 m:improvement of lip closure facilitated production of/p,b/ 6 m: complete labial contact 6 m: phonation and mastication continued to improve, with normal mobilization of the food bolus at 6 months 12 m:leakage of drinks from the mouth disappeared 18 m: symmetrical smile; functional improvements are reflected in the emotional expressions of the patient's face (feelings of joy or sadness) 5 y:blowing, chewing and swallowing is possible, pouting and kissing are still difficult, can talk easily and intelligible
Guo et al. (2008)	April 2006	Xi'an, China	30,M	Partial osteomyocutaneous	2 y: able to eat, drink and talk 2 y: no complete and symmetrical smile Died at 27 months
Lantieri et al. (2008)	January 2007	Paris, France	29,M	Partial myocutaneous	10 d: able to speak and eat
Siemionow et al. (2010)	December 2008	Cleveland, USA	45,F	Partial osteomyocutaneous	2 y: regained most of missing facial functions of nasal breathing, sense of smell, drinking from a cup, eating solid foods, and speaking intelligible
Lantieri et al. (2011)	March 2009	Paris, France	27,M	Partial osteomyocutaneous	8 m: complete mouth closure 8 m: intelligible speech
Lantieri et al. (2011)	April 2009	Paris, France	37,M	Partial myocutaneous	NA Died at two months
Pomahac et al. (2011)	April 2009	Boston, USA	59,M	Partial osteomyocutaneous	Ability to breath and speech improved immediately 12 m: unable to pucker lips
Cavadas et al. (2012)	August 2009	Valencia, Spain	42,M	Partial osteomyocutaneous	16 m: swallowing and starting phonation rehabilitation
Lantieri et al. (2011)	August 2009	Paris, France	33,M	Partial osteomyocutaneous	8–12 m: complete mouth closure 10–24 d: recovered intelligible speech
Devauchelle et al. (2006) Shanmugarajah et al. (2011, 2012)	November 2009	Amiens, France	27,M	Partial osteomyocutaneous	NA
Gomez-Cia et al. (2011)	January 2010	Seville, Spain	35,M	Partial osteomyocutaneous	NA
Barret et al. (2011)	Marc 2010	Barcelona,Spain	31,M	Total osteomyocutaneous	4 m: unrestricted masticatory movement
Lantieri et al. (2011) Shanmugarajah et al. (2011, 2012)	June 2010	Paris, France	35,M	Total myocutaneous	NA
Pomahac et al., 2012	Marc, 2011	Boston, USA	25,M	Total myocutaneous	4 m: movement of right-sided muscle groups
Pomahac et al., 2012	April, 2011	Boston, USA	30,M	Total myocutaneous	3 m: return of gross lip motion
Lantieri et al. (2011) Shanmugarajah et al. (2011, 2012)	April 2011	Paris, France	45,M	Partial osteomyocutaneous	NA
Lantieri et al. (2011) Shanmugarajah et al. (2011, 2012)	April 2011	Paris, France	41,M	Partial osteomyocutaneous	NA
Pomahac et al. (2012)	May 2011	Boston, USA	57,F	Total osteomyocutaneous	3 m: no return of motor function
Ozkan et al. (2012) Shanmugarajah et al. (2012)	January 2012	Antalya, Turkey	19,M	Total osteomyocutaneous	NA
Roche et al. (2014)	January 2012	Gent, Belgium	56, M	Partial osteomyocutaneous	Functional outcome is the purpose of this study
Nasir et al. (2012) Shanmugarajah et al., 2012	January 2012	Ankara, Turkey	25,M	NA	NA
Rodriquez et al. (2012) Shanmugarajah et al. (2012)	March 2012	Baltimore, USA	37,M	Total osteomyocutaneous	NA

protrusion elicits lower first and second formants, shifting the top right corner of the vowel triangle to a more extreme position and enhancing all three vowel-to-vowel contrasts. This effect is limited in this patient, due to difficulties with lip functions, particularly with lip protrusion, which were also observed during oromyofunctional assessments. The evolution seen in vowel space and vowel-to-vowel contrasts roughly corresponds with speech intelligibility outcomes, particularly when evaluated at the word level, and with speech acceptability. In fact, this correspondence can be seen as a validation of the techniques and procedures that were used. The downfall of vowel space and all vowel-to-vowel contrasts one year postoperatively should be seen against the background of a general health issue at the time of the assessments, which was not directly related to the transplantation itself (at twelve months a pulmonary aspergilloma relapsed with clinical symptoms of fever and radiologic progression). The degree of hypernasality lessened in the postsurgical period as was reflected in the perceptual evaluations and the nasalance values. During the second year of rehabilitation, the patient was encouraged to wear the palatal obturator permanently to close the palatal fistula. A decrease of the nasalance values while wearing the palatal obturator was seen (reflecting decreased perceived hypernasality) during the last assessment procedure. The longitudinal improvement of the speech characteristics is also reflected in the SHI, VHI, OHIP and the FDB self-assessment questionnaires. The results demonstrate that the FT patient experienced continuous improvements in specific components of speech. These experiences are corroborated by perceptual observations and objective assessments.

This case-report has limitations. First, pre-transplant data regarding speech are not available. Pre-transplant data could have provided a better baseline to evaluate the longitudinal progress of speech performance after FT. Another limitation of this study is the lack of interdisciplinary relationships. Other research efforts may focus on the interaction between the progress in speech and improvement in social and psychological functioning.

In conclusion, the pattern of results showed a longitudinal progress of speech acceptability, speech intelligibility and all the interactive speech production processes underpinning it. To what extent resonance and articulation characteristics and lip functions can be enhanced by the permanent use of the palatal obturator, and by specialized facial exercises and lip movement exercises in combination with the motor-oriented speech therapy is subject for further research

Appendix A. Continuing education

CEU questions

1. The facial transplantation in this subject is the transplantation of:
   a facial skin
   b facial skin, muscles, bones, nerves, arteries and veins
   c facial skin and nerves
   d facial skin, nerves, arteries and veins
2. To describe the quality of vowel production the following method is used:
   a acoustic analysis using Praat software
   b flexible nasoendoscopy
   c nasometry
   d acoustic analysis using the Multi Dimensional Voice Program from the Cumputerized Speech Lab
3. The goal of the facial transplantation in this subject was:
   a to restore swallowing, speech and to re-establish aesthetics in a one-stage procedure
   b to re-establish aesthetics
   c to re-establish aesthetics and psychological quality of life
   d to restore speech and aesthetics
4. The assessment of articulation revealed 21 months post-operative:
   a an incomplete phonetic inventory with distortions of the bilabials
   b an incomplete phonetic inventory with distortions of the bilabials and the alveolars
   c a complete phonetic inventory with distortions of the bilabials
   d a complete phonetic inventory with distortions of the bilabials and the alveolars
5. In this subject two years after facial transplantation the speech is characterized by:

a. the presence of hypernasality
b. the presence of phonetic distortions, hypernasality and decreased lip and lateral tongue functions
c. the presence of a moderately impaired speech intelligibility
d. the presence of a slightly impaired speech intelligibility in sentences with moderate hypernasality and distorted bilabials.

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