



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

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