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## Facial nerve regeneration after facial allotransplantation: A longitudinal clinical and electromyographic follow-up of lip movements during speech

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#### **KEYWORDS**

Facial allotransplant; Facial nerve; Regeneration; Electromyography **Summary** Introduction: Facial allotransplantation constitutes a reconstructive option after extensive damage to facial structures. Functional recovery has been reported but remains an issue.

*Case report* — *Methods:* A patient underwent facial allotransplantation after a ballistic injury with extensive facial tissue damage. Speech motor function was sequentially assessed clinically, along with repeated electromyography of lip movements during a follow-up of 3 years. *Results:* Facial nerve recovery could be demonstrated within the first month, followed by a gradual increase in electromyographic amplitude and decrease in reaction times. These were accompanied by gradual improvement of clinical assessments.

*Conclusions:* Axonal recovery starts early after transplantation. Electromyographic testing is sensitive in demonstrating this early recovery, which ultimately results in clinical improvements.

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#### Introduction

Among the numerous reports on facial transplantation, only three describe the long-term follow-up of reinnervation by the facial nerve.<sup>1-3</sup> Lantieri et al.<sup>1</sup> described a French patient with facial transplantation after resection of a plexiform neurofibroma. Electromyographic examination of the facial musculature was performed 3, 6, 9 and 12 months after surgery, with the first registration of electromyographic activity during voluntary contraction after 6 months. Nine months after surgery, the patient showed spontaneous mimicry, and 12 months post-operatively, electromyography displayed signs of motor reinnervation of both trigeminal and facial areas. Petruzzo et al.,<sup>3</sup> in their first human face-transplanted patient, observed motor recovery of the upper lip by the 12th post-operative week. Dixon et al.<sup>2</sup> suggested a timeframe between 9 and 12 months post-operatively, in which regeneration potentials must be detected to consider the nerve transplant successful.

In the present study, we present a longitudinal follow-up after facial allotransplantation by using clinical assessments of motor speech recovery and electromyographic examination of lip movements during speech. These were tested sequentially during a follow-up period of 3 years. The aim was to determine the time of onset of facial nerve recovery and correlate the electromyographic and clinical findings in this unique case.

#### Case report

#### **Clinical history**

This report describes a 54-year-old man who sustained a ballistic injury to the face, resulting in a major soft tissue defect of the lower two-thirds of his face, loss of both maxillae and nasal bones, destruction of the left half of the mandible and the entire hard palate, and bilateral enucleation due to bilateral damage to the medial orbital wall and orbital floor.<sup>4</sup> Face allotransplantation was considered an option to restore swallowing, feeding and speech and to re-establish aesthetics in a one-stage vascularised composite tissue allotransplantation. The preparation of the surgery, the surgical procedure of the facial transplant (FT), the immunosuppressive induction protocol, and the results of the surgery have been extensively described in earlier publications by Roche et al.<sup>4-6</sup> During follow-up, signs of graft rejection occurred at 4 months, which was successfully treated with immunosuppressants. One episode of Aspergillus infection occurred at 12 months post-operatively.

Speech rehabilitation was started 1 week postoperatively at a rate of five times per week for 3 months and three sessions per week thereafter.<sup>7</sup>

The clinical evolution concerning speech intelligibility, speech acceptability, voice, resonance, articulation and oromyofunctional behaviour in the course of 21 months was reported by Van Lierde et al.<sup>7</sup> In the present study, the authors extended their clinical data to 38 months post-operatively including five evaluations at 1, 5, 12, 22 and

38 months post-operatively. Electromyographic data of lip movements at each evaluation moment were included.

### Methods

The medical and surgical care of the patient has been previously detailed in published work. $^{4-6}$  The reader is referred to these references for the detailed surgical technique employed to optimise the structures directly contributing to speech function. Perceptual evaluations consisted of intelligibility, speech acceptability, articulation and oromyofunctional assessment. Intelligibility was measured by the Dutch Speech Intelligibility Test (NSVO: Nederlandstalig Spraakverstaanbaarheidsonderzoek). To evaluate the speech acceptability, an ordinal scale was used with four levels (0 = normal speech, 1 = slightly)impaired, 2 = moderately impaired and 3 = severelyimpaired speech acceptability), which was scored in a consensus between K.V.L. and M.D.L. Articulation was evaluated by reaching consensus between independent phonetic transcriptions of a picture-repeating test by K.V.L. and M.D.L. Oromyofunctional assessment was performed as suggested by Lembrechts et al.<sup>8</sup> and by means of the selfreporting questionnaire facial disability index (FDI).<sup>9</sup>

To gain insight into the *oral health impact*, the Dutch version of the Oral Health Impact Profile (OHIP-14) was used. The psychosocial impact of vocal and speech changes was assessed with the Dutch versions of the voice handicap index (VHI) and the speech handicap index (SHI), respectively.

*Electromyography* was performed with two electrodes placed over the right upper and lower lip to register bipolar EMG of the orbicularis oris muscle during a sentence completion task. The patient was asked to listen to a sentence and to speak out the most probable word that completed the sentence as rapidly as possible after a click sound, given 1000 ms after the end of the sentence presentation. This task was constructed intentionally with the aim to evoke words that had a bilabial (/m/, /w/, /b/, /p/) or labiodental (/f/, /v/) initial phoneme. Lip movement onset was detected by visual inspection of the EMG data of each trial.<sup>10–12</sup> The EMG data were separately band-pass filtered from 15 to 100 Hz to reduce the contamination by motion artefacts and non-myogenic potentials.<sup>13</sup> Reaction time (RT) was determined as the time between click onset and lip EMG onset. The amplitude of the EMG of the musculus orbicularis oris was also calculated. To overcome variances in amplitude induced by variability of electrode placement between sessions, the ratio between the mean voltage of the EMG during speech and the mean voltage of the EMG during a reference period was calculated. Per word, this reference period was determined as the 500 ms window before the auditory presentation of the sentence.<sup>14,15</sup> Only words that could be included in the RT measurement of all testing sessions were included in the EMG amplitude measure. In this way, 40 words remained available.

To detect motor speech differences between the testing moments, repeated measures ANOVA, with 'test-moment' as an independent 'within-subject' factor, was applied.

For the electrophysiological data, statistical analyses were performed for both RT and amplitude EMG data

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