

# Virtual Reality Augmented Feedback Rehabilitation Associated to Action Observation Therapy in Buccofacial Apraxia: Case Report

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## ABSTRACT

**BACKGROUND:** Buccofacial Apraxia is defined as the inability to perform voluntary movements of the larynx, pharynx, mandible, tongue, lips and cheeks, while automatic or reflexive control of these structures is preserved. Buccofacial Apraxia frequently co-occurs with aphasia and apraxia of speech and it has been reported as almost exclusively resulting from a lesion of the left hemisphere. Recent studies have demonstrated the benefit of treating apraxia using motor training principles such as Augmented Feedback or Action Observation Therapy. In light of this, the study describes the treatment based on immersive Action Observation Therapy and Virtual Reality Augmented Feedback in a case of Buccofacial Apraxia.

**PARTICIPANT AND METHODS:** The participant is a right-handed 58-years-old male. He underwent a neurosurgery intervention of craniotomy and exeresis of infra axial expansive lesion in the frontoparietal convexity compatible with an atypical meningioma. Buccofacial Apraxia was diagnosed by a neurologist and evaluated by the Upper and Lower Face Apraxia Test. Buccofacial Apraxia was quantified also by a specific camera, with an appropriately developed software, able to detect the range of motion of automatic face movements and the range of the same movements on voluntary requests. In order to improve voluntary movements, the participant completed fifteen 1-hour rehabilitation sessions, composed of a 20-minutes immersive Action Observation Therapy followed by a 40-minutes Virtual Reality Augmented Feedback sessions, 5 days a week, for 3 consecutive weeks.

**RESULTS:** After treatment, participant achieved great improvements in quality and range of facial movements, performing most of the facial expressions (eg, kiss, smile, lateral angle of mouth displacement) without unsolicited movement. Furthermore, the Upper and Lower Face Apraxia Test showed an improvement of 118% for the Upper Face movements and of 200% for the Lower Face movements.

**CONCLUSION:** Performing voluntary movement in a Virtual Reality environment with Augmented Feedbacks, in addition to Action Observation Therapy, improved performances of facial gestures and consolidate the activations by the central nervous system based on principles of experience-dependent neural plasticity.

**KEYWORDS:** Virtual reality, augmented feedback, action observation therapy, rehabilitation, buccofacial apraxia

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## Background

Buccofacial Apraxia (BFA) is defined as the inability to perform voluntary movements of the larynx, pharynx, mandible, tongue, lips and cheeks, while automatic or reflexive control of these structures is preserved.<sup>1</sup> BFA frequently co-occurs with aphasia and apraxia of speech (AOS)<sup>2</sup> and has been reported as almost exclusively resulting from lesions of the left hemisphere.<sup>3-10</sup> This latter association is probably due to anatomical contiguity.<sup>11,12</sup>

The first description of the mechanisms of apraxia is credited to Liepmann,<sup>13</sup> reporting that a group of 89 left hemisphere-damaged patients, presenting aphasia, had the highest incidence of apraxia. A lower rate of apraxia was reported in those left hemisphere-damaged patients without aphasia, whereas no evidence of apraxia was observed in patients with right hemisphere damage. A few single case studies suggested

that facial apraxia may appear following lesions in the right hemisphere,<sup>14-19</sup> and results from a group study of oral apraxia indicated that this disorder is extremely rare following right hemisphere lesions.<sup>6</sup>

In some recent studies, in a sample of 22 patients with an acute stroke in the left hemisphere and 19 with a chronic stroke in the left hemisphere, approximately 70% of them performed below cut-off on the Lower Face Apraxia Test.

Moreover, in a sample of 15 patients with an acute right stroke and 19 with a chronic stroke in the right hemisphere, the results showed that approximately half of the acute and a third of the chronic right stroke performed below cut-off in the Lower Face Apraxia Test.<sup>20</sup>

Recent studies of apraxia have reported some success in treating AOS, consisting in a speech motor deficit generally thought to involve motor planning or programming impairments,<sup>21,22</sup>



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**Table 1.** Mouth enlargement index (MEI) and left percentage of mouth displacement (LPMD) parameters during a voluntary smile and while laughing.

TEST	VOLUNTARY SMILE	LAUGHING	P-VALUE
MEI	110.82 (108.39-115.83)	122.96 (117.00-144.34)	.043*
LPMD	47.53 (47.03-48.56)	50.76 (49.05-57.00)	.043*

Data are expressed in Median (first-third quartile).

\* $P < .05$  Wilcoxon Test.

using motor training principles.<sup>2</sup> Based on the findings that speech and non-speech tasks are assumed to use the same motor system,<sup>21</sup> and that AOS and BFA share common characteristics,<sup>23</sup> Katz et al<sup>1</sup> hypothesized that kinematic training techniques will be beneficial for an individual with BFA using an augmented feedback. In addition action observation training (AOT), successfully applied during stroke rehabilitation<sup>24</sup> and Parkinson's disease rehabilitation,<sup>25</sup> could also be a valid approach in motor treatment of BFA. Indeed, research on the relationship between observed and executed actions in apraxia neurorehabilitation has provided insights about the positive effect of a visual-motor training.<sup>26</sup> Also, positive effects of Virtual Reality (VR) in neurorehabilitation are recently investigated, about increasing repetition, engagement and motivation during rehabilitation sessions. VR systems are effective in supporting feedback, have the capability adapt to individual needs, can deliver high intensity and meaningful repetitive exercises to encourage motor control and motor learning.<sup>27</sup>

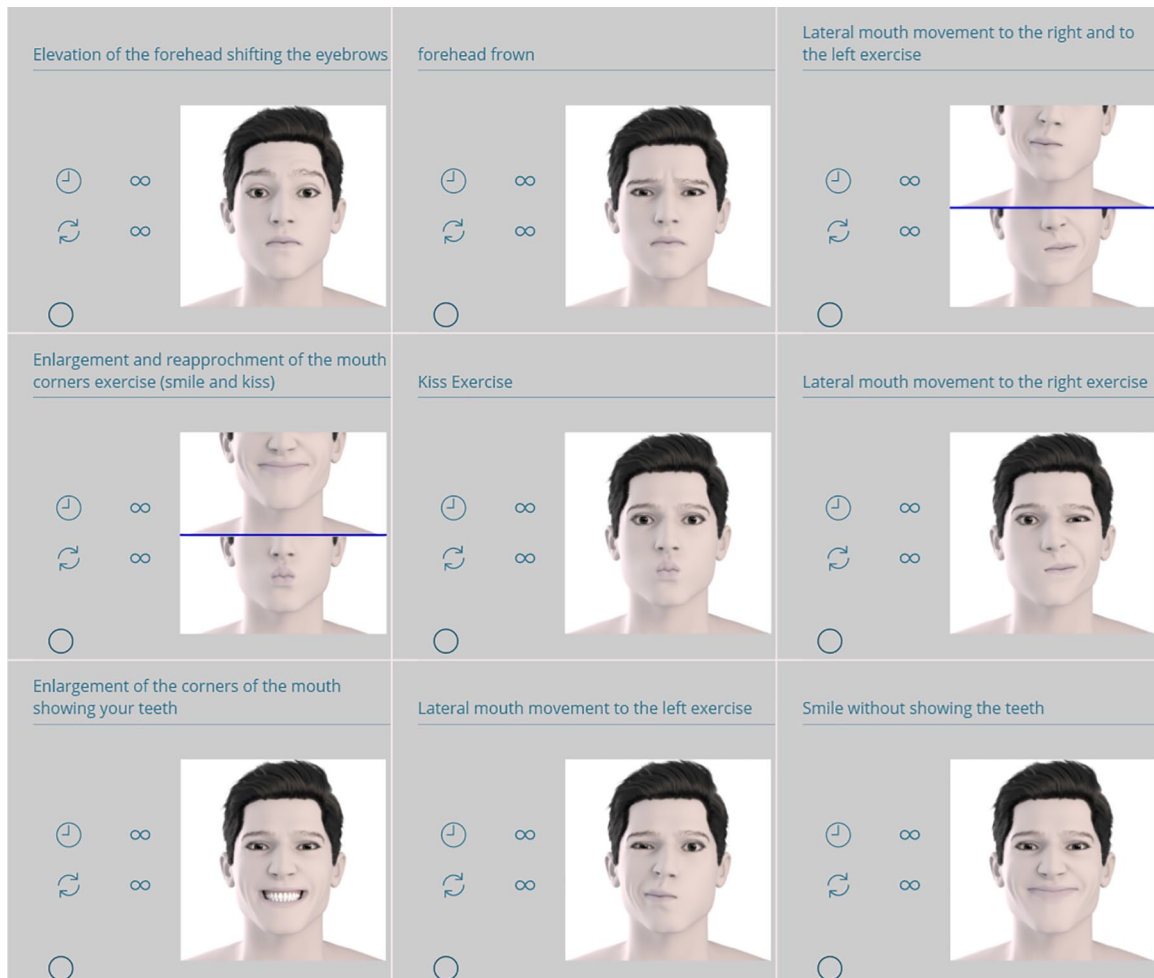
Considering the fact that the role of the right hemisphere in lower face apraxia is far from clear, the goal of our study was to describe the treatment based on immersive AOT and Virtual Reality Augmented Feedback (VRAF) in a patient with Buccofacial Apraxia, without limb apraxia or aphasia, after a neurosurgery resection of a right frontoparietal atypical meningioma.

### Case Description

The participant is a right-handed 58-year-old man, admitted to the Neurosurgery Unit of San Raffaele Hospital. In the past 3 years he suffered from worsening symptoms of paraesthesia and impaired dexterity of left upper limb. On the 10th April 2018 the patient underwent neurosurgery intervention of craniotomy and exeresis of intra axial expansive lesion in the frontoparietal convexity compatible with an atypical meningioma (WHO grade II). The lesion involved the leptomeninges from dura mater and brain parenchyma. No complication occurred after surgery. After 22 days the patient was admitted to the Department of Rehabilitation and Functional Recovery. Neurological assessment showed labial commissure deviation to the left side during both static and dynamic conditions, buccofacial apraxia, face left side hypoaesthesia, left hand pronation at Mingazzini I and impaired dexterity of the same hand. Concerning the upper limb, the left wrist was slightly flexed with metacarpophalangeal joints hyperextended distal and proximal interphalangeal joints flexed, adducted thumb with

flexed interphalangeal joint. Then, most of voluntary wrist movements were performed together with offset activation due to upper limb impairment whilst the patient was not able to perform any isolated hand and finger movements. The level of independence in the activity of daily living was assessed using the Functional Independence Measure (FIM) with a score of 115/126 and Barthel Index (BI) with a score (BI) 95/100. Functional mobility and risk of fall was assessed through the Timed Up and Go test (TUG), performed in 9 seconds. No balance deficits in static conditions were revealed by the Berg Balance Score (BBS) 56/56. Upper limb function was evaluated by the 9 Hole Peg Test (9-HPT) that was impossible to perform with left hand and by the Action Research Arm Test (ARAT) with a score of 6/57 for the left arm and 57/57 for the right one. Finally, Buccofacial Apraxia was assessed using the Upper and Lower Face Apraxia Test. Upper Faces score was 29.96/45.00 points (adjusted for age) and Lower Face score 187.90/435.00 points (adjusted for age and education).

The Upper and Lower Face Apraxia Test (ULFAT) is an assessment tool made up of 2 subscales with specific items for lower and upper face movements. The upper face items include movements subserved by the superior or inferior division of the facial nerve (VII) and by 3 oculomotor nerves (III, IV and VI). The lower face items consist of movements subserved by the inferior division of the facial nerve, the motor component of the 3rd division of the trigeminal (V), glossopharyngeal (IX, X and XII) nerves. Each items are scored as pass or failed (not performed at all, incomplete movement, conduit d'approche, unsolicited movements). Scores are corrected by age and educational level, and range from a worst score of zero to the best score of 45.0 for Upper face and from a worst score of zero to the best score of 435.0 for Lower face.<sup>20</sup> Moreover, lips displacement during a voluntary smile and while laughing was evaluated using anatomical markers captured by a front face camera and processed by a dedicated software (VRRS Khymeia Group). Specifically, patient was instructed by a physiotherapist to repeat 5 voluntary smiles and to laugh for 5 times. Mouth Enlargement Index (MEI) and Left Percentage of Mouth Displacement (LPMD) were then detected applying the marker-based positioning algorithm (Table 1). Specifically, the values of parameters reported in Table 1 were calculated as moving over 5 frames (about 170 ms) in order to reduce noise introduced by the automatic real-time marker-based positioning algorithm. In order to obtain MEI index, the algorithm calculates the initial value of the horizontal distance between the corners of the mouth



**Figure 1.** Figure shows exercises performed during Action Observation Therapy and Virtual Reality Augmented Feedback trainings.

( $D\_Lips\_X$ ) subtracting the horizontal coordinate of the right and left corners of the mouth ( $P\_RightCornerMouth\_X$ ), ( $L\_LeftCornerMouth\_X$ ).

$$D\_Lips\_X = P\_RightCornerMouth\_X - L\_LeftCornerMouth\_X$$

$$MEI = (D\_Lips\_X - D\_Lips\_Initial) * 100$$

Furthermore, LPMD was obtained as follows: algorithm calculates initially the horizontal coordinate of the median point between the left and right eye ( $C\_Eyes\_X$ ) and then the horizontal coordinate of the left corner of the mouth ( $P\_LeftCornerMouth\_X$ ).

$$C\_Eyes\_X = (P\_RightEye\_X + P\_LeftEye\_X) / 2$$

$$LPMD = (C\_Eyes\_X - P\_LeftCornerMouth\_X) / D\_Lips\_X * 100$$

Where  $P\_RightEye\_X$  and  $P\_LeftEye\_X$  are the horizontal position of the eyes, calculated by the average horizontal position of the markers defining a particular eye (Right or Left).

$$LPMD = (C\_Eyes\_X - P\_LeftCornerMouth\_X) / D\_Lips\_X * 100$$

The participant completed fifteen 1-hour sessions of immersive AOT followed by VRAF sessions, 5 days a week, for 3 consecutive weeks. Furthermore, subject performed immersive AOT for 20 minutes and active voluntary movement based on VRAF for 40 minutes every day. During the first 20 minutes the subject paid specific attention on video sequences of face movements (smile, kiss, lateral angle of mouth displacement, raise eyebrows and wrinkle forehead) projected into immersive Oculus Rift in order to do not permit the subject to be distracted. During the other 40 minutes of treatment, the participant performed voluntary facial movements with augmented feedbacks, consisting in the reflection of avatar movements projected on a virtual reality screen. In the first sessions, the execution of motion multiplier on avatar was allowed. Thereby, the subject was able to observe completely his movement on the screen, even when he performed only few and little face movements. During the rehabilitation sessions, the motion multiplier was scaled and reduced taking into account the width of the facial movements of the subject. The sequence of movements provided on virtual reality environment were the same showed during the immersive AOT sessions (Figure 1). Moreover, the subject performed an additional daily hour of conventional physiotherapy, specific to the motor impairment

**Table 2.** Outcome measures at baseline and post treatment.

OUTCOME MEASURE	BASELINE	POST TREATMENT
Functional independence measure (FIM)	115/126	126/126
Barthel index (BI)	95/100	98/100
Timed Up and Go test (TUG)	9 s	7 s
Berg Balance Scale (BBS)	56/56	56/56
9 Hole Peg Test (9-HPT)	N.A.	N.A.
Action Research Arm Test (ARAT)	6/57	30/57
Upper and Lower Face Apraxia Test (ULFAT) – upper face	29.96/45.00	35.45/45.00
Upper and Lower Face Apraxia Test (ULFAT) – lower face	187.90/435.00	376.45/435.00

involving the left upper limb. Firstly, the subject performed repetitive task trainings for fingers and wrist movements. Subsequently he was trained to carry out task-oriented exercises made up of different activities of daily living (eg, cutting, using cutlery). In addition, the treatment included endurance exercises for proximal muscles of the upper limb and aerobic trainings.

## Results

After rehabilitation, the patient achieved full independence in the ADL (FIM score 126/126 and BI score 98/100) and improved his functional mobility (TUG test performed within 7 seconds). In addition, the tests for upper limb showed some improvements in global function through ARAT with a score of 30/57 for the left arm. A total of 9-HPT was still not possible to perform. ARAT score showed great improvement, but there were still difficulties in grasp task with big object and in pinch tasks due to poor hand dexterity during gesture and hand muscle weakness. Patient was able to perform gross movement of shoulder and wrist with good motor control and endurance. Considering facial apraxia, subject achieved great improvements in quality and range of facial movements. He was able to perform most facial movements (kiss, smile, lateral angle of mouth displacement) without unsolicited movements. The ULFAT showed a final score of 35.45/45.00 for Upper Face movements (adjusted for age) and 376.45/435.00 points for Lower Face movements (adjusted for age and education). Outcome measures at baseline and post treatment are shown in Table 2.

## Discussion

This study shows the application of immersive AOT and VRAF for the treatment of BFA in a patient after a neurosurgery resection of a right frontoparietal atypical meningioma. Immersive AOT demonstrated to be a good candidate for rehabilitation due to the fact that the subject could not be distracted during the movements' observation sessions. In

addition, performing voluntary movement in a VRAF provide to the users the opportunity to practice intensive repetition of meaningful task-related activities. That insight should encourage to better corticalize facial gestures and consolidate the activations by the central nervous system based on principles of experience-dependent neural plasticity.<sup>28</sup>

Those findings might suggest the positive effect of the immersive AOT and VRAF rehabilitation in Buccofacial Apraxia recovery. However, additional trials are needed in order to consolidate our hypothesis.

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## Author Contributions

Daniele Emedoli: (i) contributing to the conception, design, analyzing and interpreting data; (ii) drafting the article and revising it critically for important intellectual content; (iii) approving the final version to be published. Maddalena Arosio: (i) drafting the article and revising it critically for important intellectual content; (ii) approving the final version to be published. Andrea Tettamanti: (i) drafting the article and revising it critically for important intellectual content; (ii) approving the final version to be published. Sandro Iannaccone: (i) approving the final version to be published.

## Consent for Publication

Written permission from the patient to participate and to publish results of this report was obtained.

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