Letter to the editor

Restoration of normal swallowing function in Wallenberg syndrome by repetitive transcranial magnetic stimulation and surgery

Dear editor

Oropharyngeal dysphagia is common in patients with Wallenberg syndrome. Although the functional outcome is good for most patients, the risk of inhalation pneumonia is high. Nevertheless, some patients exhibit persistent and chronic oropharyngeal dysphagia with pharyngeal residue and bronchial aspiration. In such cases, the central pattern generator for swallowing may be affected [1]. We report 2 cases for which we proposed a novel strategy based on repetitive transcranial magnetic stimulation (TMS), electrical stimulation and surgery.

Currently, therapeutic possibilities for oropharyngeal dysphagia include speech-language therapy and gastrostomy. New perspectives include cortical modulation involving peripheral electrical stimulation and cortical magnetic stimulation [2]. Several studies have assessed the efficacy of stimulation techniques on brainstem stroke. In one, repetitive TMS (rTMS) was successfully applied to the motor area of both hemispheres in patients with acute lateral medullary infarction [3]. However, patients also often present paralyzed hemi-pharynx with pharyngo-esophageal dys-synergia, for which surgery could be useful. Therefore, we hypothesized that rTMS with electrical stimulation followed by surgery in patients with aphagia after brainstem stroke could help in recovery of the swallowing central pattern generator because the surgery should help in recovery of the upper esophageal sphincter opening.

The 2 patients who volunteered to take part in the treatment (both males, 57 and 44 years old) had aphagia due to a brainstem stroke that occurred 3 years previously. Patients had no other functional incapacity and both were active at home. Nutrition was only possible by gastrostomy because of severe aspirations.

Swallowing was evaluated by standardized videofluoroscopic barium swallow testing (Flexview 8800, General Electric, United Medical Technologies Corp., Fort Myers, FL, USA) associated with high-resolution manometry (MMS, Tubingen, The Netherlands) to characterize aspiration [4], pharyngeal residue, pharyngeal peristalsis and upper esophageal opening [5]. rTMS was previously described by 2 of the authors [6]. Briefly, myohyoid electromyography responses were detected by using 2 pairs of surface electrodes, and cortical stimulations involved a Magstim super-rapid stimulator (Magstim, Whitland, Dyfed, UK) equipped with an air-refreshed, double, 70-mm, figure-of-eight coil (peak magnetic field = 2 T). rTMS was performed on the best point that permitted stimulation of the myohyoid cortical area of each hemisphere. Each hemisphere was stimulated for 10 min (1 Hz at 20% above the threshold value). Stimulation was repeated each day for 5 consecutive days and 5 times at 3-month intervals. During rTMS, surface submental electrical stimulations (TENS Neurostimulator, Schwa-medico GmbH, D 35630 Ehringshausen, Germany) were performed [7]. Surgery was performed after the appearance of the initiation of the swallowing reflex, which was not initially present, after the application of rTMS and TENS.

Pharyngeal resection of the non-contractile pharynx involved the lateral cervical approach with the patient under general anesthesia. The ciropharyngeal muscle was dissected and opened from the upper portion of the thyroid ala to the lower part of the pyriform sinus. A large portion of the inferior constrictor muscles and lateral pharynx mucosa was removed, as was 1 or 2 cm of the lateral thyroid ala. For the pharyngeal constrictor myotomy, a posterior midline muscular incision was made around the upper esophageal sphincter (4 cm high), preserving the mucosal integrity. Finally, a system of laryngeal suspension was created by using non-resorbable material [8]. Swallowing was evaluated by videofluoroscopy and manometry after each rTMS session and at 1, 3 and 6 months after surgery. All treatments proposed are typically used for post-stroke dysphagia.

On initial evaluation, the patients were unable to swallow because of lack of initiation of the swallowing reflex. The consequences were 100% stasis in the paralyzed hemi-pharynx with massive and passive bronchial penetration. Manometry revealed no pharyngeal peristalsis and the upper esophageal sphincter pressure increased during swallowing (Fig. 1). Evaluation of swallowing function demonstrated progressive improvement in pharyngeal peristalsis with rTMS and TENS, with the appearance of a swallowing reflex and pharyngeal peristalsism. Nevertheless, the patient showed continued stasis in the paralyzed hemi-pharynx with aspirations, and the upper esophageal sphincter did not open completely during swallowing. Therefore, we proposed functional surgery to prevent the onset of pharyngeal stasis and to improve swallowing.

After surgical healing, neither patient had any difficulties. Both showed rapid improvement in swallowing after surgery. Both patients had intensive therapy (5 times/week for 2 months) with a speech-language therapist. At 6 months, both patients had normal oral alimentation with food and liquid intake. Swallowing evaluation for both cases demonstrated no aspiration with all food consistencies and slight stasis for pureed food in the operated hemi-pharynx. Thus, the gastostronomy was removed 6 months after surgery.

rTMS over the submental muscle complex of the motor cortex of both hemispheres associated with TENS followed by functional surgery reversed the aphagia in our 2 patients with post-brainstem stroke. This is the first description of complete reversal of aphagia after brainstem stroke. The role of rTMS with TENS and surgery in this recovery and their timing warrants further investigation. The best frequency of rTMS over the motor cortex to induce brainstem plasticity needs to be determined. In

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fact, we chose 1-Hz frequency because we considered that bilateral stimulation of the cortical swallowing motor areas increased the excitability of corticobulbar projections to brainstem swallowing nuclei, which then led to improved swallowing [3]. This excellent response could be related in part to the fact that control of swallowing is usually bilateral, whereas the lesion in lateral medullary infarction is usually unilateral [3]. Thus, the remaining intact ipsilateral premotor neurons and the contralateral center in the medulla oblongata may eventually begin to operate and overcome the severity and long-term persistence of dysphagia. Nevertheless, we cannot exclude other effects on less direct pathways from the cortex to the brainstem that could contribute to the recovery in our patients. Our promising data suggest that rTMS and TENS may be useful as adjuvant therapy along with conventional treatment in brainstem stroke and that surgery combined with rTMS may achieve even better results.

Even if this report concerns only 2 patients, exceptional results were achieved by combined treatment with rTMS, TENS and surgery. Hence, further studies with a larger population are warranted to more clearly define the roles of rTMS and surgery and the natural evolution of dysphagia. Our findings raise the exciting possibility of a wider application of rTMS and TENS combined with surgery to restore normal swallowing function in patients with oropharyngeal dysphagia.

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References


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Disclosure of interest

The authors declare that they have no competing interest.
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