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CASE REPORT

SUCCESSFUL TREATMENT OF LONG-STANDING POST-STROKE DYSPHAGIA WITH BOTULINUM TOXIN AND REHABILITATION

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Cricopharyngeal myotomy is the most common treatment used to restore normal swallowing in patients with persistent (>6 months) cricopharyngeal muscle dysfunction post-stroke. We describe 2 patients whose dysphagia was due to cricopharyngeal muscle over-activity and who significantly improved after a percutaneous botulinum toxin injection in the cricopharyngeal muscle in combination with a rehabilitation treatment (dietary modifications, postural techniques, airflow protection manoeuvres). Swallowing was assessed clinically and by fibreoptic endoscopic evaluation of swallowing and videofluoroscopy; the degree of dysphagia was scored using the penetration-aspiration scale. Two months after the botulinum toxin injection the patients, who were previously fed via percutaneous endoscopic gastrostomy, returned to independent oral feeding and at 6, 12 and 24 month follow-up, both were still able to maintain an adequate oral intake with no signs of aspiration (by videofluoroscopy) or clinical complications. No further botulinum toxin injections or rehabilitation treatments were required. Our findings strongly suggest that even long-standing dysphagia can improve dramatically in selected patients. To the best of our knowledge, there are no other reports with such a long follow-up.

Key words: deglutition disorders, cerebrovascular accident, rehabilitation, botulinum toxins.

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INTRODUCTION

Thirteen percent of patients are still unable to eat due to dysphagia at 6 months following stroke, and feeding alternatives, such as percutaneous endoscopic gastrostomy (PEG), are required (1, 2). These patients often continue indefinitely on non-oral feeding through a PEG (3). Although the nourishment provided may meet nutritional and hydration requirements, a PEG is not devoid of complications, such as reflux aspiration, pneumonia, skin breakdown at the insertion site, digestive difficulties and infections (4). In addition, the individual's inability to feed themselves may have a negative psychological effect, thus delaying proper recovery.

We describe here a multidisciplinary treatment in 2 patients with long-standing (>6 months) post-stroke dysphagia who, despite having relied on a PEG for several months, became capable of independent oral feeding after a combined pharmacological and rehabilitation treatment.

CASE REPORTS

Case one

Twelve months after a spontaneous ponto-cerebellar haemorrhage, a 48-year-old man had tetraparesis, facial nerve palsy and dysphagia. The patient had had a cuffed tracheostomy tube and a PEG positioned since the first month after stroke; two episodes of pneumonia occurred in the 3rd and 6th month after the stroke. Fibreoptic endoscopic evaluation of swallowing (FEES) showed food residues and secretions in pharyngeal recess with massive tracheo-bronchial aspiration. Videofluoroscopy (VFS) showed pharyngeal-oesophageal incoordination, impaired larynx elevation, and aspiration. The degree of dysphagia was scored using the penetration-aspiration scale (PAS) (5) and a score of 7 was assigned. Oesophageal manometry detected a pharyngo-oesophageal motility disorder with an incomplete relaxation of the upper oesophageal sphincter (UES) with a mean pressure of 125 mmHg. Electromyography (EMG) showed a reduced activity of the pharyngeal inferior constrictor muscle and a spasm of the cricopharyngeal muscle. Oesophagogastroduodenoscopy ruled out any gastro-oesophageal reflux or other intrinsic oesophageal lesions.

We performed rehabilitation treatment with a speech therapist that included: (*i*) active exercises to strengthen the swallowing musculature and oral manipulation; (*ii*) procedures to increase oropharyngeal sensory input; (*iii*) postural techniques; (*iv*) dietary modifications. After 20 sessions lasting about one hour, 3 times a week, the patient showed no improvement. Therefore, 25 U of botulinum toxin (BoNT) (Botox®, Allergan Inc., Irvine, CA, USA) reconstituted with 1 ml sterile saline solution to a final concentration of 100 U/ml

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were injected into the cricopharyngeal muscle under EMG guidance and local anaesthesia (6). A progressive clinical improvement occurred from the 2nd day after the procedure onwards, so rehabilitation with a speech therapist was implemented (18 sessions lasting about one hour, 3 times a week), including:

- active exercises to strengthen the swallowing musculature, the swallowing action, and to improve manipulation of material (oral bolus control and propulsion exercises), breathing co-ordination and strength of the respiratory muscles;
- procedures to increase oropharyngeal sensory input;
- postural techniques aimed to redirect food flow and change pharyngeal dimensions and airflow protection manoeuvres to obtain a safer swallowing, improve airway protection, and pharyngeal and laryngeal function in combination with family training.

The above measures were carried out together with dietary modifications using different food consistencies and volumes to make the food more homogeneous and viscous.

Two months later, the patient was able to maintain a regular oral intake of solid and liquid foods with no clinical signs of aspiration. FEES and VFS demonstrated the patients recovery with a spontaneous swallowing action, with a complete UES opening without aspiration; a PAS of 2 was assigned. Enteral feeding could thus be gradually discontinued, and the PEG and tracheostomy removed. At follow-up 6, 12 and 24 months after the BoNT injection, the patient was still able to maintain an adequate oral food intake with no clinical complications or aspiration (as documented by VFG). No further BoNT injections or rehabilitation treatments were required.

Case two

Eight months after a massive right parietal ischaemic lesion a 79-year-old man presented with a left hemiparesis, facial nerve palsy and dysphagia. The patient had had a cuffed tracheostomy tube and a PEG positioned since one month after the stroke and an episode of pneumonia occurred 3 months after the stroke. FEES and VFS showed stasis in the valleculae and pharyngeal recess, pharyngeal-oesophageal incoordination, delayed swallowing reflex, impaired larynx elevation, and aspiration; a PAS of 6 was assigned. Oesophageal manometry showed incomplete UES relaxation (mean UES pressure 112.9 mmHg). EMG detected a bilateral cricopharyngeal myotomy activity impairment during swallowing. Oesophagogastroduodenoscopy revealed no gastro-oesophageal reflux or other intrinsic oesophageal lesions.

We performed rehabilitation treatment, as in the previous case, but there was no clinical improvement. The patient consequently received 15 units of BoNT (Botox®) into each side of the cricopharyngeal muscle. His ability to swallow improved as of the 2nd day after the procedure, so he gradually started a soft diet and the same rehabilitation treatment with the speech therapist as in the previous patient.

Two months after the BoNT injection procedure, the patient was able to ingest solid or liquid foods normally with no clinical signs of aspiration; FEES and VFS confirmed a recovery of the oropharyngeal phase of swallowing with no signs of aspiration, and a complete opening of the cricopharyngeal muscle; PAS score significantly improved up to 2. The tracheostomy tube and PEG were removed over the next month. At follow-up 6, 12 and 24 months after the BoNT treatment, the patient was still able to maintain a normal oral calorie intake with no clinical complications or aspiration (as documented by VFG); no other BoNT injections were required.

DISCUSSION

Our findings strongly suggest that even long-standing dysphagia can improve dramatically in selected patients. Both our patients had abnormal relaxing UES; UES dysfunction is common in patients with post-stroke dysphagia, particularly after brainstem strokes (7). Cricopharyngeal myotomy is the most common treatment used to restore normal swallowing in persistent cricopharyngeal muscle dysfunction, especially when it persists more than 6 months after a stroke (8, 9), but BoNT injections in cricopharyngeal muscle have also been reported to be effective in treating cricopharyngeal dysphagia (10–12). Persistent cricopharyngeal muscle activity (recorded by EMG, which is a useful tool for studying the pathophysiological mechanisms of dysphagia) unrelated to swallowing (spasm) are abnormalities suitable for BoNT treatment (13). The presence of EMG-recorded cricopharyngeal muscle spasm, defined as a permanent electric activity unrelated to swallowing, has been shown to predict a favourable outcome of BoNT therapy (14).

These patients received a BoNT injection in the cricopharyngeal muscle under EMG guidance. This method of BoNT injection was a safe and well-tolerated procedure, and no complications were observed (6). The duration of the effects of BoNT injection is not dose-dependent and probably depends on several factors, such as the injection procedure, the patient's individual response, the careful patient selection and the muscle injected (12). Our patients were still swallowing well, with no clinical signs of dysphagia or pulmonary complications, 24 months after BoNT injection. To the best of our knowledge, there are no other reports with persistent improvement after such a long follow-up. Given the short-lived effect of BoNT (3–6 months), we believe that the durable improvement of our patients' dysphagia is due to the combined use of BoNT and rehabilitation treatment. Previous reports on patients with neurogenic dysphagia did not associate any rehabilitation with the BoNT injection in the cricopharyngeal myotomy; instead, repeated BoNT injection were performed, recording an incomplete and/or short-lived recovery of swallowing (15, 16). In our cases, only one BoNT injection was performed, shortly followed by a specific active rehabilitative program. We believe that our rehabilitation measures soon after the BoNT injection contributed substantially to restoring the swallowing function and assuring a long-term benefit. Based on this preliminary

experience, we propose to add a proper dysphagia rehabilitation program to the BoNT injection in cases of cricopharyngeal muscle spasm in order to achieve a long-term recovery of swallowing function. The combined use of BoNT injection and swallowing rehabilitation would speed up the recovery of dysphagia in selected patients dramatically, reducing long-term side-effects and costs.

REFERENCES

1. Mann G, Hankey GJ, Cameron D. Swallowing function after stroke-prognosis and prognostic factors at 6 months. Stroke 1999; 30: 744–748.
2. Klor BM, Milianti FJ. Rehabilitation of neurogenic dysphagia with percutaneous endoscopic gastrostomy. Dysphagia 1999; 14: 162–164.
3. Ha L, Hauge T. Percutaneous endoscopic gastrostomy (PEG) for enteral nutrition in patients with stroke. Scand J Gastroenterol 2003; 9: 962–966.
4. Nilsson H, Ekberg O, Olsson R, Hindfelt B. Dysphagia in stroke: a prospective study of quantitative aspects of swallowing in dysphagic patients. Dysphagia 1998; 13: 32–38.
5. Rosenbek JC, Robbins JA, Roecker EB, Coyle JL, Wood JL. A penetration-aspiration scale. Dysphagia 1996; 11: 93–98.
6. Restivo DA, Palmeri A, Marchese-Ragona R. Botulinum toxin for cricopharyngeal dysfunction in Parkinson's disease. N Engl J Med 2002; 346: 1174–1175.
7. Palmer ED. Disorders of the cricopharyngeus muscle: a review. Gastroenterology 1976; 71: 510–519.
8. Buchholz DW. Cricopharyngeal myotomy may be effective treatment for selected patients with neurogenic oropharyngeal dysphagia. Dysphagia 1995; 10: 255–258.
9. Bonavina L, Khan NA, DeMeester TR. Pharyngoesophageal dysfunctions. The role of cricopharyngeal myotomy. Arch Surg 1985; 120: 541–549.
10. Dunne J, Hayes M, Cameron D. Botulinum toxin A for cricopharyngeal dystonia. Lancet 1993; 342: 559.
11. American Gastroenterological Association. American Gastroenterological Association medical position on management of oropharyngeal dysphagia. Gastroenterology 1999; 115: 452–454.
12. Haapaniemi JJ, Laurikainen EA, Pulkkinen J, Marttila RJ. Botulinum toxin in the treatment of cricopharyngeal dysphagia. Dysphagia 2001; 16: 171–175.
13. Ertekin C, Aydogdu I. Neurophysiology of swallowing. Clinical Neurophysiology 2003; 114: 2226–2244.
14. Zaninotto G, Marchese-Ragona R, Briani C, Costantini M, Rizzetto C, Portale G, et al. The role of botulinum toxin injection and upper esophageal sphincter myotomy in treating oropharyngeal dysphagia. J Gastrointest Surg 2004; 8: 997–1006.
15. Parameswaran MS, Soliman AM. Endoscopic botulinum toxin injection for cricopharyngeal dysphagia. Ann Otol Rhinol Laryngol 2002; 111: 871–877.
16. Ashan SF, Meleca RJ, Dworkin JP. Botulinum toxin injection of the cricopharyngeus muscle for the treatment of dysphagia. Otolaryngol Head Neck Surg 2000; 122: 691–695.