Impact on Quality of Life of Botulinum Toxin Treatments for Spasmodic Dysphonia and Oromandibular Dystonia

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Objective: To determine the impact on quality of life of botulinum toxin treatments for common dystonias of the head and neck.

Design: Cross-sectional survey study of a patient cohort treated with botulinum toxin injections for spasmodic dysphonia (SD) or oromandibular dystonia (OMD).

Interventions and Outcome Measures: The Glasgow Benefit Inventory was used to quantify the health benefit of treatment. Data were collected for demographics, time intervals relative to diagnosis, treatment duration, and frequency of injections. The groups were compared to determine whether differences existed in benefit from treatment. Correlation analysis was conducted for inventory scores and time intervals.

Results: A total of 23 patients (5 with OMD and 18 with SD) completed the questionnaire. The mean total benefit score was +38.04 (possible range, −100 to +100) for the whole group (P<.001). The OMD group derived a nonsignificantly smaller benefit (+21.67 vs +42.59) (P=.07). The mean subscores for the combined group were +39.67, +26.81, and +42.75 for the general, social support, and physical health subscores, respectively (P≤.001). The difference in mean subscores between the 2 groups was not statistically significant, although patients with OMD had a lower social support subscore (+6.67 vs +32.41). No correlation was found between duration of therapy or frequency of injections and the Glasgow Benefit Inventory score.

Conclusions: Patients with OMD or SD derive considerable benefit when treated with botulinum toxin. The magnitude of benefit is largely independent of the time course of therapy. Treatment with botulinum toxin for these conditions is effective on the basis of quality-of-life criteria.

PATIENTS AND METHODS

This study was approved by our medical center's Committee on Clinical Investigations. The Glasgow Benefit Inventory (GBI) is a well-studied and validated measure of patient benefit developed especially for otolaryngologic interventions. This survey was adapted to examine the quality-of-life impact of repeated botulinum toxin injections for patients with an established diagnosis of SD or OMD. The questionnaire was then administered to a group of 31 patients currently being treated for either SD or OMD with botulinum toxin in a multidisciplinary clinic for movement disorders. Informed consent was obtained from each individual participating in the study.

The data were tabulated and entered into a statistical spreadsheet for analysis. Additional demographic information was gathered from the medical record and confirmed by patient responses on the survey. These data included current age, sex, and dystonia diagnosis. The time interval between onset of dystonic symptoms and diagnosis, average time interval between injections, and duration of therapy with botulinum toxin were also recorded. The data gathered from the GBI were scaled in standard fashion to range from −100 (maximal negative benefit) to +100 (maximal positive benefit). Statistical analysis was conducted with the SPSS statistical package (SPSS Inc, Chicago, Ill). Descriptive statistics were computed for the total score, the general subscore, the social support subscore, and the physical health subscore for the GBI elements. The t test for a population was used to test the hypothesis that the GBI scores would differ from 0 (as a score of 0 implies no positive or negative benefit). The data were further examined to determine whether differences in GBI score existed between patients with a diagnosis of SD and OMD. Finally, the data were examined to determine whether any correlation existed between patient benefit as measured by the GBI and time interval between diagnosis and therapy, injection frequency, or duration of therapy.

The calculated data for the total score and subscores on the GBI for the entire group and each diagnosis subgroup are displayed in Table 1. The combined group derived considerable benefit from recurrent therapy with botulinum toxin for their dystonia, as measured by total score and subscores. Each of these scores demonstrated a statistically significant patient benefit from the injections (Table 1, t test). Combined-group patients reported the least benefit from botulinum toxin injections with respect to their social support subscore (+26.81).

The patients with OMD reported lower derived benefit from the botulinum toxin injections than the SD group. However, these differences were not significant (P > .05, Mann-Whitney statistic for differences in means between groups). Notably, patients with OMD reported a lower social support subscore benefit than patients with SD (+6.67 vs +32.41, respectively; P = .23). All scores were in the positive range, indicating that patients derived no negative benefit from the injections.

Data from the correlation analysis are presented in Table 2. No statistically significant correlation was identified between total GBI score and time interval between symptom onset and diagnosis, duration of therapy, or frequency of injections. Similarly, no correlation was identified between these time intervals and any of the subscores.

The GBI is an outcomes research tool specifically developed to measure patient benefit from otolaryngologic interventions. As a well-studied and validated research tool, it has been found to be sensitive to changes in quality of life and benefit derived from otolaryngologic interventions. It has previously been used to study degree of patient benefit derived from rhinoplasty, acoustic neuroma surgery, and other otolaryngologic interventions, with good success. The standard GBI was modified according to existing recommendations for use in quantifying patient benefit from the intervention of botulinum toxin injections for OMD or SD. The orientation of the questions was kept identical to the basic GBI, so as to prevent response bias. In addition to a total score, the GBI also includes subscales related to general health and well-being (general subscale), social support, and physical health. Each of the subscales measures a change in health status possibly produced by the intervention under consideration. As a postintervention questionnaire, it is maximally sensitive to the change in health status produced by the intervention. This questionnaire method was chosen as our outcomes tool because it is simple, it is not overly burdensome for the patient to complete, and it can be applied to patients who have already initiated their therapy. This is especially important because the standard before-and-after design for an outcomes measure would be very difficult to apply to patients who undergo repeated or recurrent forms of therapy, such as botulinum toxin injections. We believe our high response rate at least partially reflects the ease of completion of the GBI outcomes tool.

Two specific questions contained in the GBI merit some comment. Question 16 (Since your “intervention,” have you taken more or less medicine?) and question 18 (Since your “intervention,” have you taken more or less medicine?) may contribute to lower benefit scores for this group of patients. Generally, with a single intervention, such as a surgical procedure, success after that procedure would be characterized by fewer physician visits and the requirement for less medicine. However, patients receiving botulinum toxin therapy require repeated visits for injections, as well as medicine in the form of the botulinum toxin. Therefore, benefit scores on these 2 questions will tend to be low, as evi-
documented by our data, with mean patient benefit scores of only +13.0 and +17.0, respectively.

The diagnosis and treatment of OMD and SD were brought to the attention of the otolaryngology and neurology communities largely because of the efforts of Blitzer et al. and Ludlow et al. Until the late 1980s, the adductor and abductor forms of SD (laryngeal dystonia) were often attributed to psychogenic causes. Once the efficacy of botulinum toxin was reported and accepted, these diagnoses became better recognized as manifestations of organic dystonia. Before the establishment of botulinum toxin as an effective therapeutic modality, these patients had limited options for long-term control of their dysphonia or dystonia. It was not uncommon for patients to go many years with an inaccurate diagnosis and without opportunity for adequate treatment. Our data highlight this, with an average interval from onset of symptoms to final diagnosis of more than 9 years. With growing awareness of these dystonias among otolaryngologists and neurologists, and the emergence of botulinum toxin as a safe and effective treatment modality, it is likely that more patients will be properly diagnosed and treated.

Proper diagnosis and treatment of these conditions is essential because both OMD and SD may have a substantial impact on quality of life. Patients with OMD often have distracting or disfiguring involuntary jaw and perioral facial movements, which may affect mastication and speech. Patients with adductor SD usually have a “strangled” pattern of voicing that decreases vocal projection and may limit intelligibility of speech. Patients with abductor SD may be perceived as extremely nervous and difficult to understand because of their involuntary voice breaks. Several of our patients have been unable to secure employment because employers perceived their speech or movement patterns as abnormal and even potentially offensive to prospective customers.

Several studies have documented an association between SD and psychological dysfunction. Patients with SD exhibit significantly elevated levels of depression and anxiety, which decrease with botulinum toxin therapy. In a controlled study, psychological and emotional symptoms and an overall poor quality of life in patients with SD were found to be secondary to, rather than the cause of, the voice disorder. Although these studies examined patients with SD, one would expect similar findings for patients with OMD.

Fortunately, botulinum toxin treatment of these dystonias has been successful. Several studies have
Treatment duration, or frequency of injections. Similarly, patient benefit is maintained even years into treatment. Treatment with botulinum toxin for OMD and SD is clearly justified on the basis of patient benefit criteria.

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REFERENCES