ORIGINAL ARTICLE

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 ($P\leq.001$). 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.

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SPASMODIC dysphonia (SD) and oromandibular dystonia (OMD) constitute a group of dystonias relatively frequently encountered by otolaryngologists. The diagnosis is often made by an otolaryngologist or neurologist after an exhaustive search by the patient. Treatment for these conditions has centered on the use of intramuscular injections of botulinum toxin.[1,2] Unfortunately, these dystonias rarely remit spontaneously and, thus, usually require long-term therapy. As the effect of botulinum toxin is temporary, the patient must submit to periodic injections, the frequency of which ranges from several weeks to several months.

Because many patients endure a marked social stigma due to communication difficulties or disfiguring involuntary movements, a diagnosis of SD or OMD carries with it a significant impact on quality of life.[3] Several studies have examined quality-of-life impact of botulinum toxin treatments for cervical dystonia and other dystonic conditions, but relatively little information is available on the impact of such treatment in SD or OMD.[4,5] Furthermore, the recurring nature of these treatments, as well as the cost of botulinum toxin, makes treatment for these conditions expensive on a case-by-case basis. Therefore, measures of quality-of-life impact may aid in counseling patients who undergo botulinum toxin treatments, and may also provide evidence justifying this treatment modality in the face of an ever-shrinking health care dollar.

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RESULTS

Of 31 patients (23 with SD and 8 with OMD), 23 returned a completed survey (response rate, 74%). There were 5 patients with OMD and 18 with SD. The cohort was composed of 17 women and 6 men, with an average age of 55.7 years (range, 26-81

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.[6] 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).[6]

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.

years). The average time interval between the onset of dystonic symptoms and diagnosis was 109.8 months (range, 1 week to 49.7 years). The mean treatment duration was 59.5 months (range, 6-168 months), and the mean time interval between botulinum toxin injections (injection frequency) was 17.8 weeks (standard deviation, 7.3 weeks).

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.

COMMENT

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.[6] It has previously been used to study degree of patient benefit derived from rhinoplasty, acoustic neuroma surgery, and other otolaryngologic interventions, with good success.[7,8] 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.[9] 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.[6] 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 been to a doctor more or less often?) 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-

Table 1. Scores on the Glasgow Benefit Inventory*

Score	OMD Group Mean	OMD Group 95% CI	SD Group Mean	SD Group 95% CI	P†	Combined Group Mean	Combined Group 95% CI	P‡
Total	21.67	10.15-33.19	42.59	35.87-49.31	.07	38.04	29.29-46.79	<.001
General subscore	23.33	12.86-33.80	44.21	37.25-51.17	.08	39.67	31.06-48.28	<.001
Social support subscore	6.67	–9.79-23.13	32.41	21.89-42.93	.23	26.81	13.99-39.63	.001
Physical health subscore	30.00	18.28-41.72	46.30	34.38-58.22	.15	42.75	30.56-54.94	<.001

*OMD indicates oromandibular dystonia; SD, spasmodic dysphonia; and CI, confidence interval.
†Mann-Whitney test for differences between groups.
‡t Test.

denced 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[1,10] and Ludlow et al.[11,12] 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 "strained-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.[3,13] 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.[14] 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

Table 2. Correlation Between Time Intervals and Scores on the Glasgow Benefit Inventory

Score	Time Interval: Symptom Onset to Diagnosis Correlation*	P	Duration of Therapy Correlation	P	Interval Between Injections Correlation	P
Total	0.11	.65	–0.02	.94	0.35	.19
General subscore	0.12	.62	0.08	.73	0.34	.21
Social support subscore	–0.09	.69	–0.26	.29	0.27	.31
Physical health subscore	0.18	.46	–0.09	.73	0.41	.11

*Pearson χ^2 coefficient.

documented high treatment efficacy as well as a low incidence of side effects in the treatment of OMD and SD. In a 12-year review, Blitzer and associates[2] reported an average benefit of 90% for patients with adductor SD, lasting an average of 15.1 weeks. Benefit was measured by means of the universal SD rating scale. A lower average benefit of 66.7%, lasting an average of 10.5 weeks, was noted for patients with abductor SD. Other studies have documented similar success rates with respect to voice outcomes both qualitatively and quantitatively.[15]

Our data confirm that patients with OMD and SD derive meaningful and substantial subjective benefit from therapy with botulinum toxin. This holds true for the social support and physical health domains as well. Our finding that the SD group showed a tendency toward greater benefit in total score and in each of the subscores may result from the fact that patients with SD have a more dramatic communication difficulty and perceive a greater benefit when it improves. Patients with OMD usually experience pain and discomfort, with less impact on social interaction, and therefore may derive less benefit on the social support subscore. Notably, however, none of the treatment groups demonstrated a negative benefit score on any element or subscale (Table 1).

We found no correlation between total GBI score or subscores and time interval from symptom onset to diagnosis, duration of therapy, or interval between injections. This suggests that therapeutic benefit is independent of duration of symptoms before treatment, treat-

ment 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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