Examining Speech Intelligibility and Self-Ratings of Communicative Effectiveness in Speakers With Oromandibular Dystonia Receiving Botulinum Toxin Therapy

Examen de l'intelligibilité de la parole et autoévaluation de l'efficacité de la communication chez les locuteurs affectés de dystonie oromandibulaire qui reçoivent une thérapie à la toxine botulique

KEY WORDS

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Abstract

This brief report presents the results of a pilot study that examined the speech intelligibility and self-ratings of communicative effectiveness of 10 healthy control participants and 10 participants with dysarthria resulting from oromandibular dystonia (OMD). All participants with OMD received botulinum toxin injections to manage the symptoms of OMD, including speech production deficits. Sentence intelligibility was rated pre- and post- BoNT-A injections using the Sentence Intelligibility Test and each participant self-rated communicative effectiveness using the Communicative Effectiveness Survey (CES). Significant differences in speech intelligibility and self-ratings of communicative effectiveness were found between control participants and participants with OMD. No significant differences in listener ratings of speech intelligibility or self-ratings of communicative effectiveness following BoNT-A injections were found. Future research is warranted in a larger scale study to systematically examine communicative participation, speech intelligibility, and response to BoNT-A based on type of dystonia. This may help to ascertain if BoNT-A produces differential effects on intelligibility based on the type of OMD.

Abrégé

Ce bref rapport présente les résultats d'une étude pilote qui examinait l'intelligibilité de la parole et l'autoévaluation de l'efficacité de la communication chez dix participants d'un groupe contrôle et 10 présentant une dysarthrie résultant d'une dystonie oromandibulaire (DOM). Tous les participants ayant une DOM ont reçu des injections de toxine botulique pour gérer les symptômes de la DOM, y compris les déficits en production de la parole. L'intelligibilité des phrases fut notée avant et après les injections de BoNT-A à l'aide du *Sentence Intelligibility Test*, et chaque participant fit une autoévaluation de son efficacité à communiquer à l'aide du *Communicative Effectiveness Survey* (CES). On a découvert des différences significatives pour l'intelligibilité de la parole et pour les autoévaluations de l'efficacité en situations de communication entre les participants du groupe contrôle et ceux ayant une DOM, lors des mesures effectuées avant les injections de BoNT-A. Par contre, il n'y avait aucune différence significative pour ces mesures à la suite des injections de BoNT-A. Une étude à plus grande échelle s'impose afin d'examiner systématiquement la participation dans la communication, l'intelligibilité de la parole et la réaction au BoNT-A selon le type de dystonie. Elle pourrait contribuer à vérifier si le BoNT-A produit des effets sur l'intelligibilité en fonction du type de DOM.

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Oromandibular dystonia (OMD) is a focal dystonia affecting the mouth and face regions (Tan, 2004). In some cases, OMD occurs with blepharospasm or dystonic contractions of the eyelids. The combination of OMD with blepharospasm is called Meige's syndrome. OMD is a slow hyperkinetic movement disorder characterized by forceful involuntary muscular contractions and/or abnormal postures of the lips, tongue, and jaw. As a result, dysarthria can be present which can lead to reductions in speech intelligibility (Blitzer, Brin & Fahn, 1991; Dykstra, Adams, & Jog, 2007). Based on the seminal research of Darley, Aronson, and Brown in 1969, the most deviant speech dimensions of dystonia from most to least severe were identified as imprecise consonant articulation, vowel distortion, harsh voice, irregular articulatory breakdown, strained-strangled voice quality, monopitch, and monoloudness. It should be noted, however, that within this description, spasmodic dysphonia, a laryngeal dystonia, was included with oromandibular dystonia. Therefore, the deviant speech dimensions of dystonia reflecting the phonatory system (i.e., harsh voice, strained-strangled voice quality) and phonatory-prosodic components (i.e., monopitch, monoloudness) are most likely capturing the speech characteristics of spasmodic dysphonia. OMD is likely characterized by more abnormalities in articulatory components such as imprecise consonant articulation, distorted vowels, and irregular articulatory breakdowns.

There is no cure for OMD. Since medical interventions aim to manage symptomatology, the primary goals of treatment focus on reducing dystonic contractions of the orofacial musculature, improving orofacial aesthetics, and ultimately restoring functional speech, masticatory, and swallowing capabilities (Dykstra et al., 2007, Goldman & Comella, 2003).

The underlying site(s) of lesion(s) in dystonia is thought to involve the basal ganglia, cerebellum, and dopaminergic system (Duffy, 2013). Despite an incomplete understanding of the neurological mechanisms underlying oromandibular dystonia, the management of dystonic symptoms has greatly improved since the introduction of botulinum toxin (BoNT-A) therapy. As a result, BoNT-A has become the primary therapy option for treating focal dystonias, including OMD (Goldman & Comella, 2003; Munchau & Bhatia, 2000; Ramachandran & Molloy, 2015).

BoNT is produced by the bacteria *Clostridium botulinum, Clostridium baratii,* and *Clostridium butyricum* (Simpson, 2004) and can occur in seven different serotypes: A, B, C, D, E, F, and G. All of these serotypes act to inhibit the release acetylcholine from nerve terminals; however, they differ in regard to their target proteins and potencies (Dressler & Saberi, 2005). Of the seven existing serotypes, Botulinum toxin type A (BoNT-A) is the most studied for medical use and is considered to be an effective treatment for spasticity, pain, and focal dystonias including blepharospasm, spasmodic dysphonia, and cervical dystonia (Aoki, 2003; Giladi, 1997; Jankovic, 2004; Snow et al., 1990). There are several commercially available preparations of BoNT-A. Some commonly used preparations include Botox® (Allergan, Inc. Irvine, CA, USA), Xeomin® (Merz Pharmaceuticals, Greensboro, NC, USA.), and Dysport® (Ispen Ltd., Slough, Berkshire, UK). Botulinum toxin is injected locally into the symptomatic muscle(s). Dosing is individualized and is based on the mass of the muscle being injected and individual characteristics of the patient such as body mass and any pre-existing weakness (Munchau & Bhatia, 2000). The BoNT-A induced weakness typically appears after one to three days and by two weeks a marked effect is present. The effects of BoNT-A typically lasts approximately three months (Blitzer & Sulica, 2001). Side effects of BoNT-A for OMD can include mild dysarthria, difficulty chewing, and mild dysphagia (Goldman & Comella, 2003; Munchau & Bhatia, 2000).

Despite the prevalence of its use clinically, there appears to be limited empirical literature that has investigated outcome data related to speech intelligibility and communicative participation following BoNT-A injections in OMD population. This is unfortunate, since dysarthria can be a disabling aspect of OMD (Dykstra et al., 2007). It appears that only one study, published in 2007 by Dykstra and her colleagues systematically evaluated the effect of BoNT-A on speech intelligibility and communicative participation in an individual diagnosed with focal lingual dystonia. This case study provided preliminary evidence that BoNT-A had beneficial effects on speech intelligibility and communicative participation.

Purpose

The purpose of the current study is to extend the preliminary research of Dykstra and colleagues by examining speech intelligibility and self-ratings of communicative effectiveness in a larger sample of participants OMD and dysarthria receiving BoNT-A therapy. An additional purpose of the study is to examine if speech intelligibility and self-ratings of communicative effectiveness made by individuals with OMD are significantly different than those of healthy older adults without neurological disease.

Method

Participants

Participants with OMD. This study included 10 participants (6 males, 4 females) with OMD (age range: 44- 80 years, mean age = 66.9 years) with an average OMD onset of 13.8 years. Participants were diagnosed with OMD by a neurologist (M.J.) specializing in movement disorders and participants were judged to demonstrate hyperkinetic dysarthria associated with OMD by a speech-language pathologist (A.D.). These participants were recruited because they were diagnosed with OMD, were receiving therapeutic BoNT-A (Botox® or Xeomin®) injections, demonstrated reduced speech intelligibility resulting from dysarthria, and had no other speech or hearing impairments. Table 1 provides a description of the participants with OMD.

Table 1. Description of participants with OMD

Participant	Age	Sex	Type/location of OMD	OMD Duration (years)	Injection site(s) & Type of BoNT-A	Receiving BoNT-A (years)
1	69	M	Meige's (labial)	4	orbicularis oris: 10u h/s (Xeomin®)	3
2	78	F	jaw opening	2	R&L lateral pterygoid: 30u total, R&L digastric: 40u total, f/s (Botox®)	3 months
3	60	F	lingual	10	Genioglossus: 15u total, R&L digastric: 40u total, f/s (Botox®)	8
4	69	F	lingual, labial, jaw closure	21	R&L pterygoid: 30u total, R&L digastric: 10u total, f/s (Xeomin®)	21
5	78	M	jaw closure, labial	13	Orbicularis oris: 60u total, R&L masseter 40u total, f/s (Botox®)	11
6	56	M	jaw opening, closure, lingual	4	R&L lateral pterygoid: 140u total, R&L digastric: 40u total, tongue: 30u total, f/s (Botox®)	4
7	80	M	Meige's (jaw opening, jaw closure)	23	R&L pterygoid: 120 units total, R&L digastric: 30u total, f/s (Xeomin®)	22
8	68	M	jaw closure	8	R&L masseter: 30u total, medial pterygoid: 30u total, f/s (Botox®)	3
9	67	F	Meige's (labial)	50	R&L digastric: 10u total, R&L pterygoid: 20u total, f/s orbicularis oris: 5u total h/s (Botox®)	4
10	44	M	Meiges's (jaw closure, labial)	3	R&L masseter: 40u total, medial pterygoid: 40u total, f/s (Botox®)	1

Note. R = right; L = left; u = units; f/s = full strength; h/s = half strength

Control participants. Ten healthy control participants (4 males, 6 females) with a mean age of 67.5 years (age range: 59-78 years) also were recruited to participate in this study. Table 2 provides a description of the control participants. All participants had no prior history of speech, language, or hearing problems (other than those resulting from OMD). All participants provided informed consent prior to participation in this study which was approved by the Health Sciences Research Ethics Board at Western University.

Listeners. Five graduate level students aged 22-27 years were recruited to participate in this study as listeners. Listeners were not aware of the purpose of the study and they did not have experience listening to dysarthric speech. Listeners had no history of hearing loss, neurological, or speech impairment and were native English speakers. Listeners passed a 30 dB HL hearing screening at 500, 1000, 2000, and 4000 Hz in both ears. Untrained, younger adults served as listeners since previous studies have suggested that untrained younger adults and untrained older adults rate speech intelligibility of mild to moderately dysarthric speech similarly (Dagenais, Garcia, & Watts, 1998; Dagenais, Watts, Turage, & Kennedy, 1999). Therefore, older adults were not recruited as listeners for this study.

Procedure

Participants with OMD were tested over two sessions: the first experimental session (pre- BoNT-A), occurred immediately before participants received their routinely scheduled BoNT-A injections. This pre-treatment condition occurred approximately three months after participants' last BoNT-A injection to correspond with the wearing off period of BoNT-A. The second experimental session (post-BoNT-A), occurred approximately one month following injection to correspond to peak therapeutic effectiveness of BoNT-A. Control participants were tested during a single experimental visit.

Speech stimuli.

Speech intelligibility. Estimates of speech intelligibility were determined using the Sentence Intelligibility Test (SIT) (Yorkston, Beukelman, & Tice, 2011). Based on psychometric evaluation, the SIT has been found to be reliable and valid measure of speech intelligibility for dysarthric speakers (Yorkston et al., 2011). Each control participant and participant with OMD was seated in a quiet examination room. Speech recordings were obtained with a headset microphone (AKG C520) at 6 cm from the mouth attached to a digital audio recorder (Zoom H4n). The digital audio recorder recorded each participant's speech at a 16 bit and 44 kHz sampling rate. Each participant received a different set of randomly generated SIT sentences during the task. While reading aloud, each participant was audio-recorded for later analysis and to determine sentence intelligibility.

All speech samples were edited using Praat (Boersma & Weenink, 2013) and playlists were created. The order of presentation of speech samples was randomized and counterbalanced across listeners. The speech samples (i.e., SIT sentences) were presented to each of the five naïve listeners separately. Listeners rated each SIT via orthographic transcription. Digital files containing the audio recordings for each participant were numbered, counterbalanced, and randomized to minimize order effects for listeners. Each listener was seated approximately 0.6 meters (24 inches) from two M-Audio speakers (AV40) in a quiet laboratory. Speech samples were transcribed via free-field presentation at a comfortable listening level. There were two listening sessions of approximately 60 minutes in duration. Speech intelligibility was measured as the percentage of words correctly identified by the listeners (expressed as percent intelligibility). For each participant, speech intelligibility (expressed as percent intelligibility) was determined by calculating the mean of the five listener scores.

Communicative effectiveness. Communicative participation can be defined as, "Taking part in life situations where knowledge, information, ideas, or feelings are exchanged. This may take the form of speaking, listening, reading, writing, or nonverbal means of communication" (Eadie et al., 2006; page 309). Communicative effectiveness is a component of communicative participation and it was defined by Hustad as a person's ability to successfully communicate messages in home and community settings to fulfill life roles (Hustad, 1999). Assessing communicative effectiveness can provide important information about self- perceptions of communication in various social contexts and it can facilitate a breadth of outcome measurement. The CES has been used in studies evaluating communication effectiveness following treatment for maxillary cancer (Mahanna, Beukelman, Marshall, Gaebler, & Sullivan, 1998; Sullivan et al., 2002), in individuals with ALS (Ball, Beukelman & Pattee, 2004) and Traumatic Brain Injury with dysarthria (McAuliffe, Carpenter & Moran, 2010). The construct validity of the CES was also evaluated in individuals with dysarthria secondary to Parkinson's disease (PD) (Donovan, Kendall, Young, & Rosenbek, 2008).

The Communicative Effectiveness Survey (CES) (Donovan, Velozo, & Rosenbek, 2007; Hustad, 1999) was administered to participants with OMD in the pre- and

post-BoNT-A conditions and to control participants only once. The CES was administered to obtain self-ratings of communicative effectiveness and to determine if self-perceptions of communicative effectiveness changed as a result of receiving BoNT-A injections, in addition to perceptual ratings of speech intelligibility. The CES is an 8-item questionnaire focusing on communicative participation that is rated on a 4 point, Likert scale. A score of 1 represents communication that is not effective and a score of 4 represents communication that is very effective (Appendix A). Participants with OMD and control participants self-rated how effectively they communicate in a variety of social situations. Verbal instructions were given to the participants prior to completion of the survey. Means of the sums for each individual question was used to designate the ratings of communicative effectiveness for that context.

Results

Reliability

Inter-rater and intra-rater estimates of reliability were calculated for sentence intelligibility measures. Intelligibility scores from each listener were measured against each other to obtain inter-rater reliability values. All five listeners re-measured 10% of data to determine intra-rater reliability.

The values obtained for inter-rater reliability ranged from 0.906 to 0.960, $p<0.001$. These Intra-class correlation coefficients demonstrate overall excellent reliability between listeners for the speech intelligibility measures. Cronbach's alpha revealed an intra-rater reliability estimate of 0.987, $p<0.001$, which demonstrates high intra-rater reliability for all speech intelligibility measurements.

Speech Intelligibility

OMD versus control participants' sentence intelligibility scores. An independent samples t-test ($p < .05$) was conducted to evaluate the sentence intelligibility scores between OMD (pre-BoNT-A) and control participants. The pre-BoNT-A condition was only selected for comparison in order to examine if the OMD group had significantly different speech intelligibility scores than healthy control participants. This analysis revealed a significant difference in sentence intelligibility scores between OMD and control participants (t (18) = 2.54, p = 0.02). More specifically, the mean sentence intelligibility scores for the control group was 99.27% (SD = 0.66) and the OMD group (pre- BoNT-A) was 90.91% (SD = 10.40). This result suggests that the sentence intelligibility (as measured by the Sentence Intelligibility Test) of participants with OMD (pre-BoNT-A) was significantly less (by 8.3%) and had a greater variability than control participants.

Speech intelligibility: pre- versus post-BoNT. The results of this analysis suggest that speech intelligibility did not improve significantly following BoNT-A injections. A paired samples t-test ($p < .05$) demonstrates this non-significant result t (9) = 0.85, p = 0.42. More specifically, the mean sentence intelligibility scores pre- BoNT-A was 90.91% (SD = 10.40) and post- BoNT-A was 89.65% (SD = 12.99).

Communicative Effectiveness

OMD versus control participants' self-ratings of communicative effectiveness. This analysis determined if participants with OMD rated communicative effectiveness differently than control participants. Only the pre-BoNT-A condition was used to compare self-ratings of communicative effectiveness to a healthy control group. A one-factor multivariate analysis of variance, in which the items of the CES served as dependent variables was used to evaluate any differences between groups. The multivariate analysis demonstrated a statistically significant effect for the optimally weighted composite of the CES variables, F(8,11) = 7.40, p = 0.002, η^2= 0.84. Furthermore, 5 of the 8 of the univariate analyses demonstrated a statistically significant effect for the group difference, and all of the differences were in the same direction (i.e., 5/8 scores for individuals with OMD were significantly lower than the healthy control participants). These results are presented in Table 2.

Self-ratings of communicative effectiveness: pre- versus post-BoNT-A. The results of this analysis suggest that self-ratings of communicative effectiveness did not improve significantly following BoNT-A injections. A paired samples t-test demonstrates this non-significant result t (9) = 0.94, p = 0.37. More specifically, the overall mean self-ratings of communicative effectiveness pre-BoNT-A was 2.66 (SD = 0.48) and the mean self-ratings of communicative effectiveness post- BoNT-A was 2.49 (SD = 0.59) (Table 3).

Table 4 shows the mean CES scores pre- and post-BoNT-A injections according to social situation (CES item) and corresponding paired t-test ($p < .05$). No significant differences were identified on any items of the CES. The non-significant paired t-tests suggest relative stability in self-ratings of communicative effectiveness pre- and post-BoNT-A.

Discussion

The purpose of the present study was to examine the potential changes to speech intelligibility and self-

Table 2. Descriptive statistics and results of the univariate analyses of differences between participants with OMD (pre-treatment) and healthy controls based on the CES, ranked in order of largest effect to smallest effect.

CES item	OMD-pre Mean (SD)	Control Mean (SD)	Univariate analyses
Q.4 Conversing with a stranger over the telephone	2.30 (0.67)	3.80 (0.42)	$F(1,18) = 35.53$, $p=0.000$, $\eta^2_p = 0.66$
Q.1 Having a conversation with a family member or friends at home	2.80 (0.63)	3.80 (0.42)	$F(1,18) = 17.31$, $p=0.001$, $\eta^2_p = 0.49$
Q.7 Having a conversation while traveling in the car	2.80 (0.79)	3.70 (0.48)	$F(1,18) = 9.47$, $p=0.006$, $\eta^2_p = 0.35$
Q.2 Participating in conversation with strangers in a quiet place	2.90 (0.57)	3.60 (0.52)	$F(1,18) =8.32$, $p=0.010$, $\eta^2_p = 0.32$
Q.3 Conversing with a familiar person over the telephone	2.90 (0.88)	3.70 (0.67)	$F(1,18) = 5.24$, $p=0.034$, $\eta^2_p = 0.23$
Q.5 Being part of a conversation in a noisy environment (social gathering)	2.40 (0.84)	2.90 (0.74)	$F(1,18) = 1.99$, $p=0.175$, $\eta^2_p = 0.10$ ns
Q.6 Speaking to a friend when you are emotionally upset or angry	2.60 (0.97)	3.00 (0.82)	$F(1,18) = 1.00$, $p=0.331$, $\eta^2_p = 0.05$ ns
Q.8 Having a conversation with someone at a distance (across a room)	2.60 (0.70)	2.90 (0.74)	$F(1,18) = 0.87$, $p=0.363$, $\eta^2_p = 0.05$ ns

Table 3. Mean self-ratings of communicative effectiveness pre- and post- BoNT-A

Pre- BoNT/A	Post- BoNT/A
2.66	2.49
(0.48)	(0.59)

Note. The maximum score of each item on the CES is /4. Standard deviations appear in parentheses below means.

Table 4. Mean CES scores pre- and post- BoNT-A injection according to social situation (CES item) and the corresponding paired t-test and level of significance

CES Item	Pre Mean (SD)	Post Mean (SD)	*t*	*p*
Q.1 Having a conversation with a family member at home	2.80 (0.63)	2.80 (0.79)	0.00	1.00
Q.2 Participating in conversation with strangers in a quiet place	2.90 (0.57)	2.80 (0.63)	0.43	0.68
Q.3 Conversing with a familiar person over the telephone	2.90 (0.88)	2.90 (0.99)	0.00	1.00
Q.4 Conversing with a stranger over the telephone	2.30 (0.68)	2.50 (0.97)	-0.48	0.64
Q.5 Being part of a conversation in a noisy environment (social gathering)	2.40 (0.84)	2.10 (0.88)	1.00	0.34
Q.6 Speaking to a friend when you are emotionally upset or angry	2.60 (0.97)	2.00 (0.94)	1.77	0.11
Q.7 Having a conversation while traveling in the car	2.80 (0.79)	2.70 (0.68)	0.36	0.73
Q.8 Having a conversation with someone at a distance (across a room)	2.60 (0.70)	2.10 (0.99)	1.86	0.10

ratings of communicative effectiveness in participants with OMD receiving BoNT-A injections. All ten participants were receiving BoNT-A therapy in order to manage their symptoms of OMD, including speech production deficits. An additional purpose was to examine potential differences in speech intelligibility scores and self-ratings of communicative effectiveness between participants with OMD and a control group. This analysis was included in order to provide scores that could be compared and interpreted relative to healthy older adults.

Based on the results from the SIT, significant differences emerged in sentence intelligibility between the control group and participants with OMD. This result suggests that the sentence intelligibility of participants with OMD is significantly less and has a greater variability than control participants. Specifically, mean sentence intelligibility scores were approximately 8% less for the participants with OMD than the control participants, and the participants with OMD had a larger variability of intelligibility scores (OMD: 90.91%, *SD* = 10.40 versus control: 99.27%, *SD* = 0.66). The relatively large standard deviation for the PD group suggests a wide range of intelligibility scores that ranged from a severe intelligibility deficit to minimal intelligibility deficits (minimum score: 62.36%, maximum score: 97.82%).

There was also a significant difference between OMD and control participants' overall ratings of communicative effectiveness $F(8,11) = 7.40$, $p = 0.002$, $\eta^2 = 0.84$. More specifically, there were significant differences between OMD and control participants on 5 out of 8 items on the CES. The magnitude of difference between groups suggests that individuals with OMD and dysarthria self-report significant reductions in communicative effectiveness relative to control participants. Upon closer examination of Table 2, the items on the CES with the largest effect size: "Conversing with a stranger on the telephone" and "Having a conversation with a family member at home" accounted for approximately 66.4% and 49% of the variance between OMD and control participants on these items, respectively. Since our participants with OMD presented with dysarthria, but generally had milder intelligibility deficits, it is of interest that the CES items related to a range of communicative situations posed as barriers to perceived communicative

effectiveness. This result may be capturing the everyday consequences of OMD as a pervasive communication disorder that impacts a variety of communicative contexts. Yorkston, Klasner and Swanson (2001) showed that even mild speech impairments resulted in significant restrictions in communicative participation in individuals with multiple sclerosis. This suggests that restrictions in communicative participation cannot necessarily be predicted from the severity of the speech intelligibility deficit. From a clinical standpoint, obtaining information on communicative effectiveness could provide potentially important information for assessment, treatment planning and provision of educational strategies to deal with communication breakdown for individuals with dysarthria.

In general, it appears that our participants with OMD and dysarthria did not show significant improvements in speech intelligibility or self-ratings of communicative effectiveness following BoNT-A injections. In addition, no significant differences were found on item-by-item self-ratings of communicative effectiveness pre- and post- BoNT-A. This suggests relative stability in intelligibility scores and self-perception of communicative effectiveness pre- and post- injection. Even though there were non-significant differences in self-ratings of communicative effectiveness pre- and post-injection, obtaining patient perspectives is an essential component in our understanding of the impact reduced speech intelligibility on successful communicative interactions in this clinical population. This type of information can augment our interpretation of perceptually based measures, such as speech intelligibility.

Despite the non-significant results, on visual inspection of the data, there appear to be two trends that will require further investigation and exploration in a larger scale study. The first trend that emerged was that participants with dystonia predominantly affecting the lingual and, to some extent, the labial musculature appeared to demonstrate a trend for improving speech intelligibility scores and self-ratings of communicative effectiveness following BoNT-A injections (Table 5). That is, there was a tendency for individuals with primarily lingual (and to some extent labial) involvement to demonstrate an improvement in speech intelligibility and communicative effectiveness post-BoNT-A than individuals with primarily jaw involvement. The second trend that emerged was that participants who presented with predominantly jaw involvement had speech intelligibility scores and self-ratings of communicative effectiveness that had a tendency to decrease post-BoNT-A injections (Table 5). Weismer, Yunusova and Bunton (2012) suggest that the tongue is the most influential articulator. Weismer and colleagues also assert that tongue control may be more strongly related to speech intelligibility in individuals with neuromotor pathology than lip/jaw control. Therefore, the trends observed in the current study may be reflective of the differential effects of OMD on speech intelligibility based on the articulator (i.e., lip, tongue, jaw) affected. It may also suggest that different articulators respond differently to BoNT-A which may impact speech intelligibility. This question is worthy of careful consideration in a future study.

Overall, it appears that although BoNT-A injections was associated with isolated beneficial effects for some participants, it did not significantly impair the speech intelligibility or self-perceptions of communicative effectiveness in the majority of the other participants. This study represents preliminary work examining the changes to sentence intelligibility and self-ratings of communicative effectiveness in a modest sample of individuals with OMD receiving BoNT-A injections. The findings of this study should be interpreted with caution due to some study limitations. The primary limitation relates to the sample size which will limit the generalizability of the results.

The second limitation relates to the BoNT-A injection schedule and the relationship to baseline intelligibility scores. Eight out of ten participants received BoNT-A injections on a three month cycle (the remaining two participants were de-novo, but began a three month injection cycle). A three month re-injection schedule is the standard protocol for BoNT-A injections for OMD because the clinical effect has an average duration of three months (Jankovic, Schwartz & Donovan, 1990). The pre-BoNT-A condition corresponded to the final day of each participant's 3-month injection cycle. Based on a wearing off cycle of 3 months (Blitzer & Sulica, 2001) it was expected that participants had experienced the wearing off effects of BoNT-A. However, there is no definitive way to determine with certainty that the effects of the previous BoNT-A injections had diminished completely prior to the next series of injections. Therefore there remains some uncertainty about the baseline intelligibility measurements in the pre-BoNT-A condition. Although the treatment schedule followed by participants in the current study is consistent with previous literature (Blitzer & Sulica, 2001), suggesting that Botox treatments follow a 3 month cycle, a future study may seek to extend the injection cycle to 6 months or more to ensure that BoNT-A had a complete "wearing off" effect before obtaining baseline intelligibility measures, or study only denovo patients. In addition, future studies may seek to measure speech intelligibility across multiple time points within a day to evaluate any dysarthric variability due to fatigue or diurnal fluctuations. For example,

Table 5. A comparison of mean SIT scores and CES scores pre- and post- BoNT-A and the corresponding direction of change post-BoNT-A

Participant	Type of OMD	SIT (%) pre/post		Direction of change (SIT)	CES (mean) pre/post		Direction of change (CES)
1	Meige's (labial)	94.36	94.36	⊘	22	18	-
2	jaw opening	97.82	94.00	-	25	21	-
3	lingual	90.91	98.36	+	28	31	+
4	lingual, labial, jaw closure	91.82	93.82	+	17	21	+
5	jaw closure, labial	95.27	88.55	-	23	20	-
6	jaw opening, closure, lingual	62.36	53.64	-	18	17	-
7	Meige's (jaw opening, jaw closure)	94.00	90.55	-	19	17	-
8	jaw closure	96.73	96.73	⊘	24	17	-
9	Meige's (labial)	89.10	91.27	+	16	23	+
10	Meiges's (jaw closure, labial)	96.54	95.27	-	21	14	-

Note. "⊘" = no change in score, "-"= reduction in score, "+" = increase in score

diurnal fluctuations, which can be present in several types of dystonia, can manifest as little or no involuntary movement in the morning followed by severe disabling dystonia in the afternoon and evening (Evatt, Freeman & Factor, 2011). Although most of our patients were tested at similar time points (usually the afternoon), this variable was not strictly controlled for. The impact of diurnal fluctuations or fatigue on speech production in OMD is worthy of careful future study. Finally, it would be of interest to examine differences in intelligibility scores based on age and familiarity of the listener. Dagenais and colleagues examined ratings of speech intelligibility based on individuals with mild and moderate dysarthria made by unfamiliar younger (19-30 years) and unfamiliar older (61-71 years) adults (Dagenais, Garcia & Watts, 1998; Dagenais, Watts, Turnage, & Kennedy, 1999). These researchers also investigated intelligibility ratings made by speech-language pathologists (S-LP) who were familiar with dysarthric speech production. Across both studies, S-LPs rated speech intelligibility higher than unfamiliar younger and older adults. However, there was general consistency of intelligibility ratings made between younger and older adults for both mild and moderate dysarthria. Despite reported similarity of intelligibility ratings across unfamiliar younger and unfamiliar older adult listeners, a closer examination of listener age and familiarity remains an interesting future direction in the OMD population.

Since BoNT-A is the most contemporary treatment of OMD (Goldman & Comella, 2003; Munchau & Bhatia, 2000) it will be important for future studies to evaluate other aspects of BoNT-A treatment effects such as controlling dystonic contractions, improving comfort,

chewing/swallowing, and orofacial aesthetics. It will be necessary to explore if these other aspects of BoNT-A treatment provide benefit to determine if the apparent minimal benefit to speech intelligibility can be offset by significant improvements in other non-speech domains (e.g., improving comfort, orofacial aesthetics, etc.). It will also be important to explore patient perceptions of treatment (i.e., related to speech and non-speech factors) as well as impact on communicative participation in order to evaluate the effectiveness of BoNT-A from a qualitative perspective. A larger scale study is warranted to systematically examine in greater detail speech intelligibility from both perceptual and acoustic perspectives. Perceptual analyses of single word intelligibility such as the Phonetic Intelligibility Test (PIT) (Kent, Weismer, Kent, & Rosenbek, 1989) would allow for a detailed interpretation and analysis of the specific phonetic errors contributing to the intelligibility deficit (Dykstra, Adams, & Jog, 2005). Acoustic analyses such as F2 slopes, a sensitive measure to the presence of dysarthria, may provide additional information about intelligibility in addition to perceptual analyses (Weismer et al., 2012). Finally, a larger scale study could examine systematically the response to BoNT-A based on the location of dystonia. This may help to ascertain if BoNT-A produces differential effects to intelligibility based on the type of OMD and articulators affected (e.g., lingual, labial, jaw opening, jaw closing, etc.). The results of this preliminary study add new and potentially valuable information regarding changes to speech intelligibility and self-ratings of communicative effectiveness in individuals with OMD receiving BoNT-A therapy.

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Appendix A
Communicative Effectiveness Survey

Communicative Effectiveness Survey

In this survey we ask you to rate how effective your speech is in different communication situations. Please read each statement. Then rate how effectively you communicate in that situation. If you feel your speech is very effective, mark the 4. If your speech does not allow you to communicate at all in a situation, mark the 1. Feel free to use any number on the scale.

1. Having a conversation with a family member or friends at home.

Not at all effective			Very effective
1	2	3	4

2. Participating in conversation with strangers in a quiet place.

Not at all effective			Very effective
1	2	3	4

3. Conversing with a familiar person over the telephone.

Not at all effective			Very effective
1	2	3	4

4. Conversing with a stranger over the telephone.

Not at all effective			Very effective
1	2	3	4

5. Being part of a conversation in a noisy environment (social gathering).

Not at all effective			Very effective
1	2	3	4

6. Speaking to a friend when you are emotionally upset or you are angry.

Not at all effective			Very effective
1	2	3	4

7. Having a conversation while traveling in a car.

Not at all effective			Very effective
1	2	3	4

8. Having a conversation with someone at a distance (across a room).

Not at all effective			Very effective
1	2	3	4

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