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Exploring The Psychosocial Impact Of Botulinum Toxin Type-A Injections For Individuals With Oromandibular Dystonia: A Qualitative Study Of Patients' Experiences

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Abstract

This study explored the psychosocial impact of BoNT-A injections for OMD and explored how participants judge the success of this treatment. Eight participants with OMD participated in a single semi-structured interview. Phenomenological methods were used to analyze the interview data into themes. Two themes and six subthemes emerged from the analysis of the data. The first theme, *Botox has changed me and my experiences*, explored the participants' perspective of receiving BoNT-A injections and its psychosocial impact. The second theme, *What communication is like for me*, explored the psychosocial impact of BoNT-A on speech production and participation. Our results suggest that BoNT-A has a variable impact on domains related to QoL, satisfaction with treatment, speech production, and participation. This study adds novel information related to the psychosocial consequences of BoNT-A treatment in the management of OMD and builds on a literature that studies the consequences and experiences of living with OMD.

Keywords: oromandibular dystonia, BoNT-A, psychosocial outcomes, phenomenology, quality of life, communication

Summary for Lay Audience

Oromandibular dystonia (OMD) is a neurological disorder that may cause difficulties with opening and/or closing of the jaw, and speech production. Furthermore, facial grimacing, lip pursing or retraction, lingual protrusion and/or rotary movements of the tongue may also be present. There is no cure for OMD. Botulinum toxin type-A injections (BoNT-A) is considered to be the most effective treatment for OMD. This study explored the psychosocial impact of BoNT-A for individuals with OMD and explored how participants judge the success of this treatment. Eight participants with OMD participated in a single semi-structured interview. Qualitative phenomenological methods were used to analyze the interview data into themes. Phenomenology is a method of inquiry that allows for the exploration of the experiences of a group of people who share a common phenomenon. In this case, the common phenomenon is the experience of living with OMD and receiving of BoNT-A injections as treatment. Two themes and six subthemes emerged from the analysis of the data. The first theme, *Botox has changed me and my experiences*, explored the participants' perspective of receiving BoNT-A injections and its psychosocial impact. The second theme, *What communication is like for me*, explored the psychosocial impact of BoNT-A on speech production and participation. The results of this study suggest that BoNT-A treatment has variable impact and effect across domains related to quality of life, expectation and satisfaction with treatment, speech production, and participation; with some participants reporting positive effects associated with receiving injections, and others reporting more negative or neutral impact. This research adds novel information relating to the psychosocial consequences of BoNT-A treatment in the management of OMD and builds on a small, but growing literature, which seeks to study the consequences and experiences of living with OMD.

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Chapter 1

1 Introduction

1.1 Dystonia

Dystonia is a neurological movement disorder that is characterized by random, unpredictable movements and abnormal postures that can vary in speed, duration, and amplitude (Duffy, 2013). These unpredictable movements are the outcome of excessive co-contractions of opposing muscles (Duffy, 2013). The movements can be present at rest, during sustained postures, or during activity (Duffy, 2013). Dystonia can be focal (affecting one body region), segmental (affecting more than one adjacent body region), multifocal (affecting more than one non-adjacent body region), hemi-focal (affecting the muscles of only one side of the body), or generalized (affecting multiple muscle regions throughout the body) (Brin, & Comella, 2004).

1.2 Oromandibular Dystonia

When dystonia is focal to the oral and facial regions, it can be characterized as Oromandibular Dystonia (OMD) (Duffy, 2013). OMD is a rare neurological disorder with an estimated annual incidence of 3.3 cases/million and a prevalence of 68.9 cases/million persons (Nutt, Muentert, Aronson, Kurland, & Melton, 1988). The average age of onset is estimated to be 66 years (ranging from 40-86 years of age) with more women affected than men by a ratio of 4:1 (Nutt et al., 1988). There are six types of OMD: jaw-closing dystonia, jaw-opening dystonia, jaw-deviation dystonia, lip dystonia, lingual dystonia (LD), or a combination of any of these. Dystonia can result in forceful involuntary muscular contractions or abnormal postures of the jaw, lower facial, and lingual muscles (Bakke, Larsen, Dalager & Møller, 2013).

OMD may cause difficulties with opening and/or closing of the jaw, speech production, and/or mastication and deglutition. Furthermore, facial grimacing, lip pursing or retraction, lingual protrusion and/or rotary movements of the tongue may also be present (Duffy, 2013). The dystonic contractions associated with OMD are often provoked by voluntary actions, such as eating and speaking. This can result in difficulty managing oral intake of foods and liquids, and can result in reduced speech intelligibility (Blanchet, Rompre, Lavigne, & Lamarche, 2005; Dykstra, Adams, & Jog, 2007a).

1.3 Pathophysiology of OMD

OMD is associated with damage to the basal ganglia, cerebellum, and dopaminergic system (Duffy, 2013). Neuroimaging studies suggest that the abnormalities in somatosensory processing and integration in the sensorimotor cortex may also play a role (Lee, 2007). OMD is often idiopathic in nature, but also can be associated with genetic and environmental etiologies (Lee, 2007). A number of autosomal dominant and recessive and X-linked genes have been associated with dystonia (DYT genes) (Tarsy & Simon, 2006). Genes that are related to isolated dystonia, where dystonia is the only disease apparent, include TOR1A/DYT1, THAP1/DYT6 and GNAL/DYT25 (Klein, 2014). In combined dystonia, where dystonia is accompanied with other movement disorders (i.e., parkinsonism or myoclonus), the linked genes are, GCH1/DYT5a, TH/DYT5b, ATP1A3/DYT12, TAF1/DYT3 and SGCE/DYT11 (Klein, 2014). Recurrent, intensified forms of dystonia may be associated with PRRT2/DYT10, MR-1/DYT8 and SLC2A1/DYT18 genes (Klein, 2014).

The DYT12-ATP1A3 gene is linked to rapid-onset dystonia-parkinsonism. It arises in adolescence or young adulthood in response to physical or mental stress (Klein, 2014). The

DYT12-ATP1A3 gene is inherited in an autosomal dominant manner, with reduced penetrance (where an individual known to carry the dominantly inherited mutated gene may not show clinical evidence of the condition) (Klein, 2014). As a result, dystonic spasms (primarily in the upper limbs), orofacial dystonia, dysarthria, dysphagia, and slow movements may arise as symptoms (Klein, 2014). The DYT6-THAP1 gene is another gene that is linked to adolescent-onset dystonia. This gene has features of focal and generalized primary dystonia and is inherited in an autosomal dominant manner with penetrance (where an individual that is carrying the dominantly inherited gene exhibits symptoms of the condition) (Klein, 2014).

The secondary form of dystonia can develop due to environmental factors, long-term neuroleptic drugs, neurologic diseases, brain injuries, and peripheral traumas (Lee, 2007). It may also arise as the result of surgical complications, and neurodegenerative diseases and disorders (Lee, 2007). Long-term use of a dopamine receptor blocking medication (i.e., levodopa) can decrease the neurotransmission of dopamine, resulting in increased dopamine D2 receptor sensitivity. This increase can lead to a decrease in the inhibitory pathway resulting in dystonia (Lee, 2007). Dystonia may also arise from traumatic brain injuries, affecting the putamen or thalamus (Lee, 2007). Peripheral trauma, such as dental trauma, or trauma to cranial peripheral nerves or nerve roots, can lead to changes in the basal ganglia and may also be associated with dystonia (Jankovic, 2009; Lee, 2007).

1.4 Clinical Characteristics of OMD

The contraction of agonist and antagonist muscles, with overflow of unwanted muscle contractions into the adjacent muscles, results in the dystonic contractions associated with OMD (Lee, 2007). The dystonic movements can either be slow or rapid, and can change during

different activities or postures (Tarsy & Simon, 2006). The abnormal postures can occur during voluntary activities and some task-specific activities; this occurrence is referred to as action dystonia (Tarsy & Simon, 2006).

Alterations of orofacial musculature arise as a result of the contractions and abnormal postures associated with OMD (Sinclair, Gurey, & Blitzer, 2013). A study by Papapetropoulos and Singer (2006), indicated that dystonic-eating dysfunction was a common problem amongst individuals with OMD. Over half of the patients in the study with eating dysfunction reported pain due to the dysfunction and significant weight loss (Papapetropoulos & Singer, 2006). A study by Charles, Davis, Shannon, Hook and Warner (1997), also reported that their participants with OMD presented with reduced and painful masticatory and swallowing capabilities characterized by tongue biting and food being pushed from the oral cavity while chewing. Excessive dental wear, dental fractures, and trauma of the lips, gums, and tongue may also be associated with jaw-closing dystonia, whereas jaw-opening dystonia may be accompanied with temporomandibular joint overload (Bakke et al., 2013).

1.5 Speech Production Deficits

Speech production can be adversely impacted in those with OMD, resulting in dysarthria (Darley, Aronson, & Brown, 1969a). Dysarthria is a collective name for a group of speech disorders resulting from disturbances in muscular control over the speech mechanism, due to damage of the central or peripheral nervous system (Darley et al., 1969a). Dysarthria reflects abnormalities in strength, speed, range, steadiness, tone, or accuracy of movements required for the respiratory, phonatory, resonatory, articulatory, or prosodic aspects of speech production (Duff, 2013). More specifically, OMD can produce a slow hyperkinetic dysarthria (Darley et al., 1969a).

Hyperkinetic dysarthria's are associated with disruption within the basal ganglia control circuit (Duffy, 2013). Imprecise consonant articulation, vowel distortion, and irregular articulation breakdowns are common speech impairments that can arise as a result (Darley, Aronson, & Brown, 1969b). The distinctive speech characteristics associated with hyperkinetic dysarthria can result in a reduction of speech intelligibility (Darley et al., 1969b; Dykstra, Hakel, & Adams, 2007b). Yorkston, Strand and Kennedy (1996), define speech intelligibility as “the degree to which the acoustic signal is understood by the listener” (p. 55). Yorkston, Dowden and Beukelman, (1992) broadly define speech intelligibility as the “understandability of speech” (p. 265).

Dykstra and colleagues (2007a), studied the speech intelligibility of an individual with focal lingual dystonia, and analyzed acoustic and perceptual aspects of intelligibility. The results showed that dystonic movements of the lingual musculature were associated with palatal fricatives being perceived as more fronted alveolar fricatives (e.g. /j/->/s/), which contributed to reductions in speech intelligibility. Another study by Golper, Nutt, Rau and Coleman (1983), examined the speech characteristics of patients with Meige's syndrome, which is OMD with blepharospasm (involuntary closure of the eyelids). The results showed that a patient with severe lingual dystonia, and reduced speech intelligibility, had difficulty with frontal tongue placement (Golper et al., 1983). Dykstra, Adams and Jog (2005) also concluded that abnormal lingual protrusion and elevation could cause speech intelligibility deficits in individuals with focal lingual dystonia, or OMD that affects the tongue.

1.6 Treatment

As there is no cure for OMD, practitioners aim to provide treatments that focus on reducing the dystonic contractions of the orofacial musculature to allow for improvement in functional speech, masticatory capabilities, swallowing, orofacial esthetics, and quality of life (QoL) (Dykstra & Adams, 2009; Dykstra et al., 2007a). These varied treatment approaches can be characterized broadly into the following categories: pharmacological, sensory, prosthetic, behavioural, and medical via chemodenervation.

Pharmacological. Anticholinergics, benzodiazepines, baclofen, and tetrabenazine are drugs that deplete dopamine and block dopamine receptors (Tintner, & Jankovic, 2002). Although these drugs have been found to be beneficial in some patients, they are not a preferred form of treatment for focal dystonias. In treating dystonia, these medications have been shown to provide only modest improvement and are associated with frequent side effects (Tintner, & Jankovic, 2002). These medications are most effective in cases of widespread dystonia (Karp, Goldstein, & Chen, 1999). The most tolerated medication for treating OMD is Clozapine (Karp et al., 1999). Clozapine is different from other drugs in its interactions with neurotransmitter receptors and its range of side effects (Karp et al., 1999). Clozapine acts on the dopaminergic pathways by binding to and increasing dopamine receptor density in the basal ganglia. This reduces the over-activity of the direct pathway, thereby reducing the dystonic contractions (Karp et al., 1999).

Sensory. A sensory trick, or “geste antagoniste”, is a method some patients with OMD use to temporarily relieve dystonic symptoms (Yoshida, 2018). Sensory tricks include motor

tricks (voluntary movement input), imaginary tricks (imagining the performance of their own sensory tricks or other mental imaging), forcible tricks (maneuvers similar to sensory tricks, but require the use of force, and are always antagonistic to the direction of the dystonia) and reverse sensory tricks (sensory trick-like stimuli that result in worsening of dystonia) (Ramos, Karp, & Hallett, 2014; Yoshida, 2018). Sensory tricks can be physical, proprioceptive, visual, auditory, and thermal stimuli (Yoshida, 2018). Sensory tricks are highly individualized, but common examples can include holding an object in the mouth, chewing gum, or touching a certain area of the face, such as the chin, cheek, or eyelid to temporarily relieve dystonic symptoms (Duffy, 2013). The mechanisms behind sensory tricks are unknown (Ramos et al., 2017; Yoshida, 2018).

Yoshida (2018) studied 128 individuals with various presentations of OMD. Participants were fit with a splint designed to elicit a sensory trick in order to reduce dystonic movements. The results of this study revealed that individuals with jaw-closing dystonia, as compared to other dystonia types (i.e., lingual, jaw-opening, labial, jaw deviation), derived the most benefit from the splint when used as an elicitation of a sensory trick. Additionally, participants with intraoral sensory tricks (inserting an object or food into the oral cavity) found the splint more effective than patients with extra-oral sensory tricks (tactile facial stimulation, i.e., touching the chin or teeth with a finger or hand).

Teemul, Patel, Kanatas, and Carter (2016), studied six patients with OMD, and found that their participants reported triggers, such as stress, talking, chewing, and praying resulted in increased dystonic activity. Yoshida (2018), showed that temporary relief, for some, from such symptoms was found by gently touching the lips, chin, teeth, or jaw with a handkerchief or mask; chewing gum; talking; biting on a toothpick or tobacco; or placing a finger underneath the chin. Sensory tricks tend to lose effectiveness over the course of the dystonia, even to the point

of disappearing completely (Ramos et al., 2014).

Prosthetic: bite-block. A bite block is a device that can come in many shapes, sizes and textures and can range from an acrylic block to a block made from dental impression material or putty (Seikel, Drumright, & King, 2016). It is placed between the upper and lower teeth to assist with jaw stability and positioning. Bite blocks can improve facial appearance, articulatory precision, and hyperactive movements associated with hyperkinetic dysarthria (Dworkin, 1996; Goldman & Comella, 2003). Bite blocks can prevent enamel wear, decrease the load on oromandibular joints, and improve chewing function (Blanchet et al., 2005). They may also provide sensory information for the correct placement of other articulators (Lee, 2007). Dworkin (1996) conducted a study where bite-blocks were utilized in two individuals with oromandibular dystonia. Initially, both participants were judged to have moderately reduced intelligibility (Dworkin, 1996). However, once the bite blocks were in place, speech intelligibility was judged as near normal (Dworkin, 1996). It was hypothesized that the bite-block neutralized the dystonic activity by facilitating postural balance and motor stability of the mandible (Dworkin, 1996).

Behavioural. The goal of behavioral therapy is to improve the coordination of articulators for those with OMD (Goldman & Comella, 2003). A study by Netsell and Cleeland (1973) used EMG biofeedback (auditory) with an individual with dystonia affecting the upper lip. Surface electrodes were placed over muscles that were contracting to retract the upper lip, and the individual was presented with a tone whose frequency was equivalent to the voltages recorded from the electrodes (Netsell & Cleeland, 1973). The individual's task was to concentrate on lowering the tone, which served to decrease the dystonic activity in the upper lip

(Netsell & Cleeland, 1973). This behavioural technique helped the participant learn to control and reduce her labial muscle activity (Netsell & Cleeland, 1973).

Speech-language pathologists (SLPs) may also focus on providing patients with management strategies. For example, a speech-language pathologist may teach dyadic strategies or verbal interaction strategies to improve communication. These strategies may include: getting the listener's attention before speaking, making the context and topic clear, maintaining eye contact, using gestures whenever possible, informing the listener that they wish to know immediately when they have not been understood, reiterating an utterance or word when an abnormal movement pattern has potentially interfered with communication and modifying the communicative environment to improve visual and auditory acuity (Duffy, 2013). Similarly, the listener should be attentive to the speaker and inform the speaker immediately of any misunderstandings (Duffy, 2013). Although these techniques are not specific to OMD, they can improve communicative interactions, and these techniques can be used concurrently with other forms of treatment.

Deep Brain Stimulation. Deep brain stimulation (DBS) is a surgical procedure where a stimulator is implanted in the brain. This implant delivers electrical stimulation to the area of the brain affected by dystonia, such as the globus pallidus or the thalamus (Krauss, Yianni, Loher, & Aziz, 2004). The DBS stimulator suppresses the electrical signals causing the dystonia, thereby lessening the dystonic symptoms (Krauss et al., 2004). DBS has been proven to be most beneficial for generalized and segmental dystonia. Further research is required to determine if this treatment option is effective for focal dystonias, such as OMD (Krauss, et al., 2004).

Chemodenervation. To date, chemodenervation is the primary therapeutic treatment for patients with OMD, and this is achieved via botulinum toxin type-A injections (BoNT-A) (Teemul et al., 2016). BoNT-A is considered to be the most effective treatment for OMD (Kazerooni & Broadhead, 2015). BoNT-A causes temporary paralysis by blocking the presynaptic release of acetylcholine at the neuromuscular junction (Simpson, 1981). BoNT-A injections produce temporary chemical denervation, paralysis, muscle atrophy, and local weakness, which help reduce muscle contractions (Giladi, 1997). It takes about one to three days for the weakness induced by the toxin to appear. By two weeks, a marked effect is present (Blitzer & Sulica, 2001). The effects of BoNT are limited in duration, lasting three months on average (Blitzer & Sulica, 2001). At that time the axon terminal begins to sprout and form new synaptic contacts on adjacent muscle fibers. For this reason, the effects of BoNT-A are limited in duration (Simpson, 1989). Depending on the individual and the affected area, the effects of BoNT-A could last for two to three months (Giladi, 1997).

1.7 Clinical Effects of BoNT-A for OMD

Jankovic, Schwartz, and Donovan (1990) reported that injections of BoNT-A into the masseter, temporalis and geniohyoid, resulted in a reduction of dystonic spasms, and an improvement in chewing and speech, in approximately 73% of their patients with OMD. BoNT-A injections have been shown to reduce pain caused by dystonic contractions (Jankovic, 2018; Khan, Anwer, Eliav & Heir, 2015), and can improve orofacial aesthetics by reducing dystonic activity (Merz, Deakin & Hawthorne, 2010). There is some evidence to suggest that bilateral injections of BoNT-A into the intrinsic muscles of the tongue can reduce dystonic activity and improve speech intelligibility. This finding was demonstrated in an individual with focal lingual dystonia (Dykstra et al., 2007a). Although there is an empirical literature demonstrating

improvement following BoNT-A injections, there can be side effects to this treatment. A study by Blitzer, Brin, and Fahn (1991), suggested that injections in the tongue for treatment of lingual dystonia increased the potential for dysphagia and aspiration. Other side effects of BoNT-A include weakness, mild dysarthria, dry mouth, and difficulty chewing and swallowing (Goldman & Comella, 2003; Gn & Ng, 2017). A study by Nastasi, Mostile, Nicoletti, Zappia, Reggio, and Catania (2016), found that no significant associations were discovered between the amount of years of receiving BoNT therapy and the injected muscles or the total dose injected. These results were similar to the findings of Bhattacharyya and Tarsy (2001) and Charous and Comella (2011). This suggests that the therapeutic benefit of BoNT-A is independent of the symptom duration prior to treatment, the treatment duration, or the frequency of the injections. The patient benefits from BoNT-A injections were maintained years into treatment (Bhattacharyya & Tarsy, 2001).

1.8 Psychosocial impact of living with OMD

The term ‘psychosocial’ derives its origin from the World Health Organization’s (WHO) definition of health as ‘a state of complete physical, mental, and social well-being, and not merely the absence of disease and infirmity’ (WHO, 1948). Psychosocial impact is defined as “the effect caused by environmental and/or biological factors on individual’s social and/or psychological aspects” (de Oliveira, Buchain, Vizzotto, Elkis & Cordeiro, 2013).

Psychosocial factors or variables include those that reflect individual responses to disease and even in some cases indicators of the social context itself (Martikainen, Bartley & Lahelma, 2002). They can also be altered based on the social contexts that they exist in (Martikainen et al., 2002). This can include examining an individual’s experience as the result of a health condition and his/her appraisal of QoL (Martikainen et al., 2002). A diagnosis of OMD can result in

alterations to psychosocial functioning. For example, Bakke et al., 2013 found that oral dysfunction arising from OMD resulted in social embarrassment, reduced QoL, and depression. Charles and colleagues reported that their patients with dystonia felt lonely and helpless, leading to social isolation (Charles et al., 1997). Finally, Page and colleagues (2017) reported that 8 participants with OMD self-reported reductions to QoL related to communication, with socialization being an area most affected (Page, Siegel & Jog, 2017).

A study by Papapetropoulos and Singer (2006), also explored how individuals with OMD experience daily living. The results of this study found that eating dysfunction associated with OMD resulted in social embarrassment, especially when accompanied by drooling or choking (Papapetropoulos & Singer, 2006). In another study that examined long-term management of treatment using botulinum toxin in patients with OMD, Sinclair et al., (2013), found that alterations to orofacial aesthetics, resulting from OMD was associated with reductions in perceived QoL. A study by Nastasi et al. (2016) found that individuals with lingual dystonia reported social disabilities that impacted daily activities, such as talking and eating.

Dykstra and colleagues have reported that individuals with OMD have reduced communicative participation, communicative effectiveness, and communication-related QoL relative to control participants (Dykstra, Domingo, Adams & Jog, 2015; Page & Siegel, 2017; Page et al., 2017). Furthermore, several individuals in Bhattacharyya and Tarsy's (2001) study stated that they had difficulty securing employment because employers perceived their speech or movement patterns as abnormal and potentially offensive to customers.

1.9 Psychosocial Impact of BoNT-A for Individuals with OMD

There is a small, but growing literature, which has investigated how BoNT-A injections impact psychosocial functioning in a number of health conditions, yet there still remains a

fundamental gap in our understanding of this construct as it relates to OMD. It has been suggested that the relatively sparse empirical literature exploring psychosocial outcomes could be the result of the rarity of OMD and the resultant difficulties in recruiting research participants (Charous & Comella, 2011). Several studies have examined the impact of BoNT treatment on QoL outcomes for cervical dystonia, but little information is available on the impact of treatment in OMD (Bhattacharyya & Tarsy, 2001).

Bhattacharyya and Tarsy (2001), addressed the gap in the literature and examined how BoNT injections impact QoL in dystonias of the head and neck. The Glasgow Benefit Inventory (GBI; The Glasgow Health Status Questionnaires Manual, 1998) was used to quantify the health benefit of treatment of BoNT in participants with spasmodic dysphonia (SD) and OMD. The GBI is a generic quality of life questionnaire designed for measuring outcomes following otolaryngologic interventions (Robinson, Gatehouse & Browning, 1996). The GBI measures quality of life in three domains: social, general, and physical. Results of the Bhattachartta and Tarsy (2001) study revealed that participants with OMD and SD gained significant benefit within the social support and physical health domains from therapy with BoNT, although patients with OMD had a lower social support sub-score in comparison to patients with SD (Bhattacharyya & Tarsy, 2001).

Dykstra and colleagues (2007a) case study evaluating the impact of BoNT-A injections on speech intelligibility and activity and participation restrictions in an individual with focal lingual dystonia found prominent differences across outcome measures between pre-BoNT-A and post-BoNT-A treatment. For example, results from the Voice Activity and Participation Profile (Ma & Yiu, 2001) showed improvements in the participants self-perception of the severity of her speech problem, the effect on her job, and daily and social communication

following BoNT-A injections. Prior to treatment, the participant reported a loss of employment, reported communicative and social avoidance behaviors, embarrassment, reduced self-esteem, and reduced self-image. Following treatment with BoNT-A, a positive difference was observed in the participants activity and participation limitation scores (Dykstra et al., 2007a). The participant also reported that she felt minimal to no restriction in activity and participation relating to her job and her social communication following BoNT-A injections. Overall, this study demonstrated that BoNT-A treatment significantly improved activity and participation scores, and QoL of the individual studied. This case study highlighted the importance of examining the impact of speech disorders resulting from dystonia on an individual's daily functioning in the context of their personal, social, and environmental settings.

Charous and Comella (2011) conducted a study examining the effects of BoNT injections on QoL for patients with jaw-opening dystonia. Similar to Bhattacharyya and Tarsy's (2001) study, Charous and Comella (2011) used the GBI as their primary outcome measure. The results of the GBI total score and GBI sub-scores following BoNT injections showed improvement and indicated that no patients experienced negative impacts from receiving BoNT injections (Charous & Comella, 2011). Charous and Comella's (2011) study found improvements in QoL for all participants post-injection. Patients reported feeling more optimistic, less embarrassed, more social, and generally felt better about themselves as a result of the injections. Physical disturbances, as a result of dystonia, were also improved due to the BoNT injections. However, the psychosocial gains resulting from BoNT injections were found to be far more significant than the gains to physical well-being (Charous & Comella, 2011).

Merz et al., (2010) was the first research group to develop a disease-specific, patient-reported outcome measure, validated for patients with OMD. The Oromandibular Dystonia

Questionnaire (OMDQ-25) was created to measure health-related quality of life in individuals with OMD. It includes 25 items that are divided into five subscales: general, psychosocial functioning, cosmetics, speech, and eating dysfunction. Each of the 25 items is rated on a five-point scale with different grades of impairment: never, seldom, sometimes, often, and always (Merz et al., 2010). Based on a sample of 30 participants with OMD, Merz and colleagues evaluated the OMDQ-25 at pre-treatment and then at four and eight weeks after BoNT-A treatment. This study found that the cosmetic sub-score demonstrated the greatest difference pre- and post-BoNT-A injections. This result indicated that the patients were highly sensitive to their outer appearance (Merz et al., 2010). Results also demonstrated that following BoNT-A injections, individuals with OMD showed a large benefit in scores related to psychosocial functioning, speech, and eating.

Nastasi et al. (2016) studied the effect of BoNT treatment on QoL in patients with isolated lingual dystonia and OMD affecting the tongue. This study aimed to investigate the impact of BoNT on the QoL of patients with prominent lingual dystonia using the OMDQ-25 to identify clinical and BoNT treatment-related factors that could be associated with better outcomes regarding QoL. The response to BoNT therapy was based on both the OMDQ-25 questionnaire and clinical interviews. Nastasi et al., (2016) found that in both LD and OMD affecting the tongue, QoL was severely affected prior to treatment. However, following BoNT treatment, there was a positive change in total OMDQ-25 scores, demonstrating a significant benefit in overall QoL. These results were seen in patients with LD four weeks after treatment and the benefit to improved QoL persisted after eight weeks. The analysis of the sub-scale scores demonstrated an improvement in many aspects of QoL (Nastasi et al., 2016). The smallest changes were observed in the functional eating category suggesting that the complex and

coordinated process of eating has the least response to the BoNT injections (Nastasi et al., 2016).

Dykstra, Domingo, Adams, and Jog (2015) evaluated sentence intelligibility and self-reported communicative effectiveness of 10 participants with OMD at two time points over the course of a BoNT treatment cycle. Communicative effectiveness was measured using the Communicative Effectiveness Survey (CES) (Donovan, Velozo, & Rosenbek, 2007; Hustad, 1999). Communicative effectiveness is defined as a person's ability to successfully communicate messages in home and community settings (Hustad, 1999). Evaluating communicative effectiveness can provide important information regarding the self-perception of an individual's communication in various social contexts and it can facilitate broader outcome measurements (Dykstra et al., 2015). The results of the Dykstra et al., (2015) study showed significant differences in speech intelligibility and self-ratings of communicative effectiveness between control participants and participants with OMD (Dykstra et al., 2015). However, there were no significant differences in speech intelligibility or self-rated communicative effectiveness observed over the course of the BoNT-A treatment cycle for the participants with dystonia (Dykstra et al., 2015). However, when the group of participants with OMD were looked at based on type of dystonia, there were suggested trends. The 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 (Dykstra et al., 2015). On the other hand, 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 (Dykstra et al., 2015).

Page and colleagues (2017) also conducted a study that examined self-rated

communication-related quality of life (CR-QoL) in 8 control participants and 8 individuals with OMD and dysarthria who were receiving therapeutic BoNT-A injections. The American Speech-Language-Hearing Association's Quality of Communication Life Scale (ASHA QCL) was used to assess CR-QoL (Paul, Frattali, Holland, Thompson, Caperton & Slater, 2004). The ASHA QCL is an 18-item questionnaire rated on a 5-point scale (Paul et al., 2004). The ASHA QCL assesses the impact of a communication disorder on an individual's personal relationships, social life, autonomy, well-being, and participation in educational, work, social, and leisure activities (Paul et al., 2004). The results of the study showed that participants with OMD rated CR-QoL lower, as compared with control participants across all ASHA QCL domains/subdomains. The results also indicated that participants with OMD did not rate CR-QoL significantly different over the course of the BoNT-A treatment cycle.

1.10 Exploring Personal Experiences Through a Qualitative Lens

Qualitative researchers study people in their natural setting, attempting to make sense of, or to interpret, phenomena in terms of the meanings people attribute to them (Denzin & Lincoln, 2011). Qualitative methodology places participant as the "experiencers" and puts them in the position of the expert throughout the research process (Wilding & Whiteford, 2005). Participants in qualitative research are seen as active agents, who are not only describing their life events, but are actively negotiating and reconstructing their story by choosing, interpreting and emphasizing different aspects of their experiences (Sallinen, Kukkurainen, Peltokallio, Mikkelsen & Anderberg, 2012). On the contrary, the researcher in qualitative research is placed in the position of the learner, where the aim is to understand and interpret the phenomena at hand (Wilding & Whiteford, 2005). These roles in the process of qualitative research result in collaborative

dialogue, which produce a genuine representation of participant's experiences (Wilding & Whiteford, 2005). There are varying methodologies and paradigms within qualitative research. Common methodologies include; grounded theory, narrative research, ethnography, and phenomenology, each with varying aims and results. Grounded theory aims to create a theory based on the phenomena at hand (Glaser & Strauss, 1967). Another methodology, Narrative research, aims to represent experiences through individual's stories and portray the complexity of human experiences. The narratives or stories that individuals share can be used as a means to study a research question (Riessman, 2008; Riessman, 1993). Ethnography aims to promote an understanding of why people behave as they do, to reveal culture as shared knowledge and as acquired social behavior (Hammersley, & Atkinson, 1995). Lastly, Phenomenology is a method of inquiry that allows for the exploration of the experiences of a group of people who share a common phenomenon (Dowling, 2007). Each methodology can be underpinned by varying world views called paradigms. Guba and Lincoln (2004) acknowledge four different paradigms. The positivist and post-positivist paradigms, both work within objective, realist approaches and accept one reality. The critical paradigm aims to use naturalistic, subjectivist methodologies and agrees that there are multiple realities, which are structured and shaped by power relations that are historically and socially constructed. Each of these methodologies and paradigms allow for an in-depth understanding of participants personal experiences and perspectives and provide insight that is otherwise difficult to obtain using a quantitative lens. The constructivist and interpretive paradigms, take on more of naturalistic, relativist stance, which recognizes multiple meanings and subjective realities. A constructivist view emphasizes that researchers co-construct data together with the participants. An

interpretivist paradigm aims to describe, understand and interpret participants' experiences (Tuohy, Cooney, Dowling, Murphy & Sixsmith, 2013).

In the current study, interpretive phenomenology was employed to understand the multiple realities of living with OMD and receiving BoNT-A injections. Interpretive phenomenology allows for the exploration of rich, multilayered, intangible, and dynamic phenomena in aspects of an individual's everyday life (Wilding & Whiteford, 2005). Phenomenology takes the participants voice and experience primarily into account to allow for new findings. New findings may also emerge as a result of the phenomenological notion that the data itself should guide the nature and form of the study for the researcher (Dowling, 2007). Phenomenology has been upheld as the approach of choice in areas in which there is little published material, or where areas need to be described in more depth (Wilding & Whiteford, 2005).

1.11 Rationale

There is a relatively robust empirical literature that has investigated the pharmacological benefit of BoNT-A injections in dystonia in general (Bhattacharyya & Tarsy, 2001). In contrast, there is a relatively sparse, but growing empirical literature that has explored BoNT-A therapy in OMD research using patient-reported outcome measures (questionnaires), such as perceived QoL, communicative participation, and effectiveness of treatment from a patient perspective. The problem with questionnaires is that they do not allow an in-depth or individualized exploration of what factors and issues might be underlying the participants' responses. Nevertheless, the emerging research is important because it has provided the rationale to explore more fully the impact of BoNT-A injections on psychosocial outcomes, especially in the context of impaired speech production resulting from OMD. What remains unexplored in the study of

OMD is an understanding of the impact of BoNT-A injections from the perspective of the individual with OMD, in their own words. Each individual will perceive the same phenomenon in different ways and bring forth his or her lived experiences, specific understandings, and historical backgrounds (Finlay, 2002). These individualized experiences are complex to research using quantitative research methods. On the contrary, qualitative research methods acknowledge a subjectivist view of reality based on context and personal experience (Wilding & Whiteford, 2005). Through a subjective lens, qualitative research methods can provide an understanding of each individual's perspective (Creswell, 2014). These experiences and insights can highlight the perspective of individuals living with OMD. Gaining an authentic perspective of the in-depth experience of living with OMD can inform a greater understanding of the depth and detail of individual experiences, determine if patient reported outcome measures are capturing all relevant information or if other patient-reported outcomes should be explored, and finally, qualitative inquiry can aid in the interpretation of questionnaire responses gathered clinically. Acquiring the participants' perspective can also serve to inform meaningful indicators of treatment success and provide much needed insight into living with this diagnosis.

1.11 Purpose

The purpose of this study was to obtain a self-reported account of the psychosocial consequences of BoNT-A injections for individuals living with OMD. Using qualitative, phenomenological methodology, this study explored how participants judge the success of BoNT-A treatment based on the psychosocial issues relevant to them. By understanding the insider's perspective, it is anticipated that this knowledge will help to inform the clinical management of individuals with OMD and associated dysarthria.

Chapter 2

2 Method

2.1 Methodology

Interpretive phenomenological qualitative research methods were used to obtain a self-reported account of the experience of living with OMD and receiving BoNT-A treatment.

Phenomenology is a method of inquiry that allows for the exploration of the experiences of a group of people who share a common phenomenon (Dowling, 2007). In this case, the common phenomenon is the experience of living with OMD and receiving of BoNT-A injections as treatment. Phenomenological research is based on the principle of lived experiences (Dowling, 2007). Lived experiences are the events that naturally occur in the lives of a specific cohort (Dowling, 2007). It is a method of inquiry that offers a way of methodically studying and learning about phenomena that are typically difficult to observe or measure (Wilding & Whiteford, 2005).

The interpretive phenomenological approach does not require researchers to bracket their own preconceptions or theories during the research process (Lowes & Prowse, 2001). The research process instead includes bridling, which was employed in this study. Bridling acknowledges that researchers cannot help but bring their involvement and fore-understandings into the research (Finlay, 2002). The interpretive team acknowledges any assumptions that could both influence the researcher's conduct of interviews and observations, as well as the whole team's interpretations (Crist & Tanner, 2003). It is expected that an interpretive phenomenological approach will allow for the generation of a thorough description of the experience of living with OMD and receiving of BoNT-A injections as treatment, in the words of

the individuals who live it.

2.2 Participant Selection

The inclusion criteria included that all participants could read, speak, and understand English. The recruitment was limited to an age range of 25 to 80 years. This age range was chosen to represent a wide range of the adult population and to properly capture the average age of onset of OMD (66 years; range of 40 – 86 years). All participants were receiving BoNT-A injections to manage symptoms of OMD. There was no exclusion criterion regarding the type of OMD (i.e., lingual, jaw-opening, jaw-closing, mixed) that each individual presented with. In adherence with phenomenological guidelines, the inclusion criteria warranted a sample of participants that all shared the experience of living OMD, in order to reveal the array of lived experiences for this particular clinical population (Patton, 2002).

2.3 Participants

Eight participants (n=8) diagnosed with OMD participated in this study. The current study was part of a larger study that examined speech intelligibility and communicative participation in the same group of participants (see Dykstra et al., 2015; Page et al., 2017). In total there were three females and five males (age range: 44-80 years; mean age: 68 years), with an average OMD onset of 10.4 years. Participants were recruited from a specialized Movement Disorders Clinic, in South Western Ontario. The participants recruited were receiving therapeutic BoNT-A injections (Botox® or Xeomin®) to manage their symptoms of OMD. All participants' demonstrated hyperkinetic dysarthria and reduced speech intelligibility as judged by an experienced speech-language pathologist (A.P.) and had no other speech or hearing impairments.

Table 1 describes the characteristics of each participant.

The investigator explained the nature of the study as well as provided each participant with a letter of information (Appendix A) and a consent form (Appendix B) to sign prior to participating in the study. This study was approved by the Health Sciences Research Ethics Board at Western University in London, Ontario (Appendix C).

Table 1. Characteristics of Participants with OMD

Participant ID*	Sex	Age	OMD Duration (years)	Type/ location Of OMD	Number of Years Receiving BoNT-A	SIT (%)	
						Pre-BoNT-A	Post-BoNT-A
GM	M	69	4	Meige's (labial)	3	94.36	94.36
ST	F	78	2	jaw opening	3 months	97.82	94.00
NF	F	60	10	lingual	8	90.91	98.36
JR	M	44	2	Meige's (pouting, jaw closure)	3 months	96.54	95.27
FI	F	69	21	jaw closure	21	91.82	93.82
SP	M	78	13	lingual, jaw closure	11	95.27	88.55
EP	M	80	23	Meige's (jaw opening, closure)	22	94.00	90.55
BR	M	68	8	jaw closure	3	96.73	96.73

*** Initials are fictitious to protect the identity of participants**

-Pre-BoNT-A refers to a time period of 3 months after the last injection cycle to correspond to a 'wearing off' period of BoNT-A

- Post-BoNT-A refers to a time period of 5 weeks after injection to correspond to 'peak' therapeutic response of BoNT-A

-Please see Dykstra, Domingo, Adams, & Jog (2015) for a description of sentence intelligibility scores (Intelligibility data was collected from the same study participants as part of a larger study)

2.4 Procedure

Interviews. The eight study participants participated individually in one face-to-face interview lasting approximately 60-90 minutes. Interviews were scheduled at five weeks post-BoNT-A injection to correspond with the peak effectiveness of the BoNT-A. Interviews were conducted in a private room by the primary researcher (A.P), who was not involved in the clinical care of the participants. The participants guided the phenomenological interviews to the topics they found relevant (Table 2). The researcher encouraged participants to talk about their experiences, feelings, and to provide stories (Table 2). The questions were expanded upon by each participant and on occasion reiterated by the primary researcher for clarification. The researcher also asked additional questions, when needed, to clarify points or to obtain more information (Table 2). The interviews were audio recorded and transcribed verbatim. If the researcher did not understand a word or sentence spoken by a participant during the interview, she asked for clarification and repetition to ensure correct understanding and meaning. It is anticipated that phenomenological interviews will produce a detailed account of the experience of living with OMD and receiving BoNT-A treatment, in the words of the persons who live it.

Table 2. Interview Questions

The interviews were guided by six general questions:			
The researcher encouraged participants to talk about their experiences, feelings, and provide stories or anecdotes.			
Primary Questions:	The researcher asked additional questions as needed to clarify points or to seek additional information. These questions may have included:		
1) How do you feel your Botox treatment has helped you?			
2) Has using Botox affected your ability to do the things you want to do?	i)	Do you think you would have been able to do exactly the same things, the same way you're doing it now, had you not been using Botox?	
3) Have you noticed changes in your speech since using Botox?	i)	Have you noticed any changes in your speech as far as the clarity or the understandability?	ii) Have you noticed any differences between before you get your Botox and after you get your Botox?
			ii) If yes, are the differences you noticed from using Botox important to you?
4) What is the biggest impact Botox has had on your life?			
5) What has Botox had the least impact on in your life?	i)	Did you have any expectations going into this that you were hoping to have achieved that were not achieved?	
6) What has been the impact of Botox with regard to participating in daily activities?	i)	What are some of the things that you would do now that you wouldn't have done?	
Each interview concluded with the researcher asking the participant if he/she had anything else to add or mention about his/her experience.			

Interview analysis. Interviews were transcribed verbatim from audio recordings by a secondary researcher. The interviews were analyzed using the following qualitative guidelines (Lincoln & Guba, 1985; Tracy, 2010; Finlay, 2002): First, the transcripts alongside the audio recordings were read and listened to multiple times for familiarity. Once a thorough read was completed, the transcripts were then uploaded onto online qualitative analysis software, Dedoose (Dedoose Version 8.0.42, 2017). Next, components of the interviews regarding BoNT-A were highlighted and organized. Through multiple readings of the interviews, sets of codes were created based on the content of the interviews (Appendix D). These codes aided in organizing the content of the transcripts into topic areas. The codes were examined by members of the research team, through an iterative approach and were revised when necessary to obtain consensus. This iterative approach promotes the trustworthiness of the data analysis. Next, a final code dictionary was developed to assign meaningful codes. Lastly, after the interview transcripts were coded and sorted into their content areas, each of the content areas were examined in detail and themes were developed to reflect the most significant patterns within and across the coded topic areas.

The final qualitative analysis focused on to:

- 1) Identify commonalities and differences among participants' experiences;
- 2) Reflect the complexities and multiple realities among participants through descriptive accounts;
- 3) Illustrate the themes through the language of the participants

(Benner, 1994)

2.5 Quality Criteria

Quality criteria in qualitative research aim to provide guidelines to aid in trustworthy research. These are specific criteria and a set of processes that are employed to safeguard the authenticity of the data. These include: reflexivity, credibility, and multivocality.

Reflexivity. Reflexivity can be defined as thoughtful, conscious self-awareness of the researchers (Finlay, 2002). Reflexive analysis includes constant evaluation of subjective responses, inter-subjective dynamics, and the research process itself (Finlay, 2002). Through the application of reflexivity, a phenomenologist begins their research with the data of their experience; their own reflecting, intuiting, and thinking and uses that as primary evidence (Moustakas, 1994). Therefore, reflexive analysis is necessary to examine the impact of the researcher and participants on each other and the research (Finlay, 2002). Reflexivity acknowledges the existence of researcher bias and encourages researchers to provide reflective insights and engage in explicit, self-aware meta-analysis throughout the research process (Finlay, 2002). Reflexivity is seen as an essential component as it improves the quality and validity of research (Guillemin & Gillam, 2004).

Throughout the research process, reflexive notes were recorded alongside an audit trail (a form of reflexivity) to aid in the trustworthiness of the data (Appendix E). Audit trails are an account of all the research decisions and activities throughout the study process, along with records of the participants' reactions or emotions and the researcher's thoughts. Lincoln and Guba (1985) suggest that by implementing an audit trail, an auditor or second party who becomes familiar with the qualitative study and its methodology, findings, and conclusions, can

audit the research decisions and the methodological and analytical processes of the researcher and confirm its findings.

Credibility. Tracy (2010), defined credibility as the ability to create trustworthiness in the data presented. This was ensured during the data collection process when the audio recordings of each interview were transcribed and verified by members of a research team for any errors or discrepancies. Research assistants who were uninvolved in data collection conducted the transcription of the data. A secondary researcher reviewed the transcripts and made notes on where there were discrepancies between what was heard on the recording and what was contained in the transcript. This has been further implemented through the interview analysis, which was conducted through an iterative approach amongst the first author, mentor, and advisory committee. We explored and compared data analysis, and referred to the transcripts to ensure correct interpretation. Any discrepancies were resolved via consensus of the research team. This iterative process relates to an approach called crystallization. Crystallization refers to the practice of using multiple sources, frameworks and methods (Tracy, 2010). Crystallization will be adhered to via the use of multiple researcher viewpoints. Multiple researcher perspectives allow for different aspects of problems to be explored, it helps deepen the understanding of the data, and encourages consistent (re) interpretation (Tracy, 2010). Crystallization accepts that the goal is not to provide a valid singular truth, but to provide a complex and in-depth, understanding of the phenomena at hand (Tracy, 2010).

To further ensure credibility, interpretive phenomenology suggests that the “interpretive team” consists of researchers who are experts in phenomenology or in the content area of inquiry (Crist & Tanner. 2003). In the analyses of the data, the research team consisted of individuals

with clinical and research experience treating and working with individuals with dysarthria and OMD, and research with a background on qualitative research.

Multivocality. Multivocality is implemented in this study to include multiple and varied voices (Tracy, 2010). The heterogeneous nature of the participants recruited will facilitate an understanding of experiences from different genders, ages, occupations, and time since disease onset. As the varied voices of our participants shared their experiences receiving BoNT-A injections, an authentic understanding of his/her lived experiences was revealed and this created a space for a variety of opinions (Tracy, 2010).

Chapter 3

3 Results

Two major themes and six sub-themes were developed through the qualitative analysis of the interview data (Table 3). The first theme, *Botox has changed me and my experiences*, explored the participants' perspective on receiving BoNT-A injections and the psychosocial impact of receiving these injections. The four sub-themes under this category include: *I feel normal again*, *I feel appreciative of what Botox does for me*, *Botox doesn't fix everything*, and *My face looks different*. The second theme, *What communication is like for me* concentrated on the psychosocial impact and changes to speech production and communicative participation that participants experienced as a result of receiving BoNT-A injections. The two sub-themes under this category include: *My speech has changed* and *I can now participate*. All themes and sub-themes will be further expanded on below, alongside direct quotes from the participants' interviews to explain the development of each theme.

Table 3. Themes and Sub-themes

Themes	Sub-themes	Definitions
Botox has changed me and my experiences	I feel normal again	Personal perspective of “disability” and the concept of “normal”
	I feel appreciative of what Botox does for me	Positive emotional reactions, happiness, increased confidence, hopefulness, surprise, improved self-esteem, relief, and impact on QOL
	Botox doesn’t fix everything	Negative emotional reactions, disappointment, fatigue, uncertainty, frustration, fear, and worry
	My face looks different	Alterations to facial appearance/orofacial aesthetics, self-consciousness
What communication is like for me	My speech has changed	Speech production i.e., response rate, speech intelligibility, articulation, and physical effort
	I can now participate	Participating in meaningful activities with family and friends, changes to communicative participation

3.1 Theme 1: Botox has changed me and my experiences

Participants expressed how BoNT-A treatment changed their perspective of themselves, their expectations, and described the psychosocial impact of receiving BoNT-A injections.

Participants also shared their experiences of receiving BoNT-A injections. They discussed how

receiving their injections resulted in positive emotional reactions, such as happiness, increased confidence, hopefulness, surprise, improved self-esteem, and relief. However, many participants also expressed that receiving BoNT-A injections resulted in less positive experiences, such as disappointment, fatigue, uncertainty, frustration, fear, and worry. Participants also discussed the psychosocial impact of BoNT-A injections on their facial appearance.

I feel normal again. Participants discussed how they viewed themselves post-injection, their view of “disability”, and what “normal” means to them. E.P (initials are all fictitious) discussed how the injections helped him from “slipping into the grey area of disability” (EP, M, 80 years). He expressed his concerns about how he would feel about his self-concept without receiving BoNT-A injections by stating:

Yes, yes, I need the shots. Or else, I’m in the area of being disabled... I think I would probably be very limited, definitely. I think the continual eye movement and jaw movement really make you look as a handicap person, whereas [Botox] has prevented me from slipping into that area or suffering any of those thoughts or lower self-esteem (EP, M, 80 years).

Participants also mentioned the idea of feeling “normal” again as a result of the injections. NF explained that she was going to be “hitting the line of permanent disability” (NF, F, 60 years), due to her OMD. However, once she received the injections she described a transformation in her perspective, “It is amazing, it makes me feel normal...I feel like I’ve gone from disabled to abled” (NF, F, 60 years). NF also stated that post-injection “You can develop a normal life and just be aware of a few things, you know common sense things” (NF, F, 60 years). JR also touched on this perception of feeling normal again and how “feeling normal” post-injection

improved his participation in everyday activities. “You don’t feel as embarrassed of uncontrolled movements. I’m able to go outside and be as normal as possible” (JR, M, 44 years).

I feel appreciative of what Botox does for me. Participants discussed the beneficial psychosocial changes they experienced as the result of receiving BoNT-A injections. JR stated: “It improves the quality of your life. Most definitely”(JR, M, 44 years). BR similarly mentioned how he found BoNT-A injections helpful, “Yeah it works. It seems to do its job for me” (BR, M, 68 years). NF discussed that the injections provided her with “inspiration and confidence” (NF, F, 60 years). She also said it “gives me my life back” (NF, F, 60 years). FI discussed how the injections had provided her with confidence, “I feel more confident, I think, a bit more confident because you know, I feel like I try to control [my tongue] when it’s moving a lot. If the Botox is controlling that movement, that’s an important thing that it’s doing really” (FI, F, 69 years). GM mentioned similar changes with the impact BoNT-A treatment had on his life and how it improved his outlook:

It has changed my life I think um, maybe not as far as other people can see, but for me I think it’s changed my life... it gives you, it tends to give you some confidence as well, I think. Um, dystonia can take some confidence away, but Botox gives it some back. And um, I think that’s the big thing (GM, M, 69 years).

NF felt as though she was given a second chance when she regained her speech post-injection. She discussed how the BoNT-A injections gave her the confidence to speak publicly. NF said, “It was almost like a debt I had to pay right because I’m so thrilled to have [my speech] back” (NF, F, 60 years). Participants also mentioned their appreciation for the changes that the BoNT-A treatment has made. GM stated:

I know I've been asked whether I'll be taking Botox my whole life. I don't know if I'll have to or not, but um, with age, things change or not...When people ask, you know, I don't mind taking that the rest of my life. It's a godsend ...it's fixed the thing that I wanted fixed most (GM, M, 69 years).

Participants also expressed their happiness and thankfulness as a result of receiving BoNT-A injections. NF discussed how "the difference between it is survival. I have a job because of Botox. I can talk, I can be happy" (NF, F, 60 years). Similarly to NF, ST expressed "Oh, yes I am happy for any changes, yes, yes" (NF, F, 60 years). EP also shared: "I feel very happy and fortunate they have something for us. I mean without it I wouldn't want to face the consequences without it...I'm very happy about the relief I do get from it" (EP, M, 80 years).

Botox doesn't fix everything. Despite being appreciative of the benefits of BoNT-A treatment, all eight participants also acknowledged that BoNT-A injections didn't fix everything. Participants described failure of expectations and side effects including pain, muscle atrophy and bruising. Although GM experienced improvement with the injections, he recognized that Botox "doesn't make things perfect, but it does make it as near perfect as it can get" (GM, M, 69 years). GM further stated:

I wish I wouldn't still have to have the um, still kind of moving my mouth around. I wish I didn't have that. Botox fixes that a bit...And but, it still hasn't um, gotten my tongue under control. Umm, I would still have to move it around; I'd still have to manage that when I didn't have to manage that before. That might be one thing that it does still leave you with something (GM, M, 69 years).

SP discussed how his expectations of the injections were not met. He had been told once receiving the BoNT-A injections his face would get back to “normal” (SP, M, 78 years), SP stated: “But that didn’t happen. I guess that was a disappointment, but I guess you could live with those things, you know?” (SP, M, 78 years). FI who had been receiving BoNT-A injections for 21 years described her uncertainty of the effect of BoNT-A injections:

I’m not sure now; heaven knows I’m not sure ha-ha... Yeah, um, I think it’s almost a psychological effect now. You know I’ve come to expect to receive months of Botox and uh, something will change anyway, whatever it is ha-ha... Doesn’t sound very positive does that? (FI, F, 69 years).

EP similarly discussed what he wished could be changed: “I wish my muscles didn’t deteriorate and I would get better and get cured” (EP, M, 80 years). Similarly, ST stated, “I thought it might help the jaw a little more. But then, it’s only the first time, so I wasn’t expecting miracles” (ST, F, 78 years).

FI, NF and BR discussed the longevity and variability of symptom relief over the course of the three-month injection life. FI stated: “Um, yeah I mean I think it’s good for my confidence for those first few weeks before it starts to drop off” (FI, F, 69 years). NF and BR discussed the impact of the BoNT-A injection as it wears off. NF stated: “I don’t have as much control when the Botox is wearing off” (NF, F, 60 years). She discussed what she noticed once the injection began wearing off.

Umm I notice that the pain and the tension does start coming back. I can tell within 6 weeks of the injection. The tiredness, the fatigue, the muscle is there, the aches, the pain, the movement, the twitches or you can feel them coming back (NF, F, 60 years).

BR also mentioned:

When I am on the back side or sort of mid-point of the [Botox], when I start to work up to having another injection, that's when it's worse... I'll start to notice little things and then it gradually, it gets very, very pronounced (BR, M, 68 years).

Participants also discussed side effects, such as muscle atrophy, bruising, and pain due to the injections. EP discussed how receiving BoNT-A injections for 22 years affected his muscles. He stated: "The muscles have deteriorated such that they aren't normal ...even the doctor found difficulty when giving me the injections too, especially on the jaw muscles; the eye area is very thin and easy to bruise" (EP, M, 80 years). EP also mentioned how he experienced the injection cycle and the effect on the muscles pre- and post- BoNT-A injections.

Well, all of sudden you're going from unable to control the movement of eyes uhh from closing, your mouth from opening and all of a sudden you're experiencing the extreme opposite, I have to first get used to it the 6 or 7 days and then you feel the Botox just restricting your muscles from the movement that it's hard to shut your eyes, it's the reverse of what's happening and the same way with your mouth is. It's hard to open your jaw muscles it's so restricted. But umm after a month and depending on the strength of the Botox you're never certain how much or how small it is that you're getting. You're loosening up the muscles or at the best of times for at least a month minimum after that the last month is a slow decline, quite often sometimes not as bad as others. But at the time during last two weeks, you really need it (EP, M, 80 years).

Pain, as a result of the injections, was a common experience for four of the eight participants.

NF, although she felt as that the treatments were positive overall, mentioned other experiences post-injection: "There are other symptoms too. I do have problems with the biting of the tongue once in a while. I have to be careful of that. So I have a few nodules from biting the tongue too

hard” (NF, F, 60 years). BR also discussed that “sometimes I’ll bite the inside of my mouth or heaven forbid I bite my tongue and then it sends you right off to the ceiling” (BR, M, 68 years). He goes further to describe more uncomfortable side effects such as “dry mouth especially at night, that’s a problem” (BR, M, 68 years). He stated:

I get up in the morning and sometimes this lower lip is stuck to my teeth. And if you wake up and you don’t recognize that effect, you pull your lip away, you know, just getting up and you open your mouth, that creates a lot of little sort of micro tears in the back of the lip; and then the first thing you know, the thing is swelling and it’s very sore (BR, M, 68 years).

NF also mentioned the pain associated as a result of receiving the injections and the challenge of correct dosing. She explained:

Oh the injection hurts for about 30 seconds, it swells, you have to be careful umm each day it does create more swelling so you have to be more aware, you have to physically be aware of what you’re doing and how much swelling you do have...And being in the tongue you know what are going to be the side effects of that injection, am I going to be back in emergency in 24 hours from now or three days from now with a swollen tongue and not being able to breath (NF, F, 60 years).

SP similarly discussed the pain of the actual injection, the trauma to the muscle and how long it took to heal.

It really was quite sore... I had 25 needles. That’s sore for a while. Any muscle if it’s damaged is going to be. It’s not going to act normal for a while ... It take 2 weeks sometimes it would be a little bit longer, till the healing. Till the muscles healed from the injury, from the trauma (SP, M, 78 years).

SP and JR mentioned their experience of tightness after the injections: SP stated: “Because the Botox releases the muscles. The tightness. There’s more tightness there, especially like I said after the injections, but there’s still some tightness” (SP, M, 78 years). JR also stated: “Just tight, but it wasn’t pain...it still becomes tight when I try to talk (JR, M, 44 years).

My face looks different. Participants described changes related to orofacial aesthetics as a result of receiving injections. They discussed their perceptions of how BoNT-A injections affected their facial appearance, how the injections have impacted the function of their muscles, the trauma resulting from the injections in the form of redness, their level of self-consciousness, and how other people have reacted to their facial appearance.

Participants discussed their perception of positive changes to their facial appearance post-injection. FI discussed that she was less worried about dystonic facial twitches, especially when she was in social situations, during the first couple months after her injections.

When I, the first half anyway [referring to the first half of the injection cycle], I don’t seem to have to worry as much about, you know, if I’m going into a social situation especially if I’m going to be sitting there twitching all the time or am I going to be reasonably personable (FI, F, 69 years).

Similarly, JR discussed how he felt the injections changed his facial appearance and how he is less concerned about dystonic movements in his face. He stated, “I don’t feel like I’m worried about my face being moved so much...It’s calmed my face down a bit” (JR, M, 44 years). NF and SP also reported positive changes to their facial appearance post-injection. NF stated, “There’s your whole appearance issue” (NF, F, 60 years). When asked what Botox had the most impact on, SP stated “facial appearance” (SP, M, 78 years). ST mentioned that prior to her

injections “the kids noticed” changes in her appearance and had said “Mom, you got to do something!” (ST, F, 78 years). She described what her face looked like prior to receiving BoNT-A injections and the resultant changes post-injection. She stated:

My mouth was drawn down. It almost like... have you seen Bell's palsy? Yeah. Almost like that. Only it wasn't Bell's palsy. But that all cleared up [post-injection] because the muscles were relaxed and they could find a way to bring all the facial features back to where they were supposed to be (ST, F, 78 years).

BoNT-A injections did not improve facial aesthetics for all participants. Participants described some perceived negative effects on their facial appearance, or they perceived no noticeable change following injections. GM discussed that post-injection he needed to use his tongue to clear food from sticking to his teeth and he described how he thought this affected his appearance. He stated, “that's an appearance thing, so you don't want to be sticking your tongue out of your mouth” (GM, M, 69 years). GM also discussed how the injections made him self-conscious of his appearance. He explained that as a result of the injections he has a “grimacing” expression, yet the benefit of Botox “outweighs anything it does to appearance” (GM, M, 69 years). He stated:

It does something to my appearance with that... Well I think that the marks here, and you know... it only happened about 6 months ago when me and my wife started to see [Dr.xxx] And so he said we have to adjust [the injections] on the other side. Right now when you... it's kind of hard... it pulls one way so we have to pull it back the other way. Yeah, so um... I think the Botox, has an effect on appearance.... Almost every 3 months we've probably adjusted a little bit. Because I know in the last 6 months they adjusted it. The... my corners of my mouth were falling down I was almost like well, a grimacing

type thing. So we have to counteract that by putting more Botox on the other side so it kind of pulls it up. You can kind of see the red marks in the corner of the mouth up there...I noticed right away, and I think everybody else notices right away. Umm so we kind of do that but I don't want to, like, I said, I didn't want to look like the Joker after (laughs) (GM, M, 69 years).

FI stated that although she is uncertain of how Botox is specifically helping her dystonia, she continues to take the injections for cosmetic reasons.

My mouth was continually moving. So [Dr.xxx] used to try and inject around my mouth, so that would be lessened. Um, but I'm not really sure that that did help.. I mean it has made me feel that the movement is diminished. I know I've noticed that lately I am getting about half of what I used to get fifteen, twenty years ago...we're almost working towards trying to lessen the motion so that they're not moving, you know... in almost distorted ways...So, uh I mean it's becoming almost a cosmetic, I mean I guess it always was a bit of a cosmetic thing, I have to admit that (FI, F, 69 years).

3.2 Theme 2: What communication is like for me

Participants described changes to their speech production and communicative participation as a result of receiving BoNT-A injections and the psychosocial impact associated with receiving the injections. Results were varied, with some participants experiencing noticeable improvements in their speech production, while other participants experienced little to no change in their speech production post-injection. Participants also discussed how BoNT-A injections impacted their communicative participation and psychosocial functioning.

My speech has changed. All participants described changes to their speech production as a result of receiving BoNT-A injections. Participants discussed changes to their speech intelligibility, articulation, response rate, and amount of physical effort required to speak. Participants compared and contrasted their speech production before and after receiving BoNT-A injections.

BR, FI, and EP discussed how their speech intelligibility had improved post-injection, but they also described when their speech production was most improved over the course of the injection cycle. BR stated:

Oh my speech was terribly slurred...I think the clarity and the understandability sort of show up when I'm in this position right now. Like this far from having another shot (6 weeks post-injection). I think [Botox] loosens up these muscles in my face and allows me to pronounce words easier, so I don't have this sort of slurring, if you will (BR, M, 68 years).

Similarly, FI mentioned: "I have to think about the speech thing whether it does help. Uh, I think it does help initially when I have it, for the first you know, few weeks anyways and then there's the, you know, wearing off process" (FI, F, 69 years). EP also stated: "You're much more restricted with formulation of words, and the length of time it takes you to get it out, when you receive the Botox. After Botox is in for a month or so, your speech progressively gets better" (EP, M, 80 years). SP also described that although the injections worked to improve his speech production, it took time for his speech production to be optimized.

I had problems enunciating words. I couldn't enunciate them clearly, uh, and maybe people thought maybe I got marbles in my mouth and wouldn't speak very clearly. And that started to improve after I'll say, for a year. And um, after that, I felt really good about

it because after the first week, or maybe like two weeks I felt like I could speak clearly (SP, M, 78 years).

Participants explained how their articulation improved post-injection, and they also described their reactions to changes in their speech production. For example, NF discussed how Botox provided her with the ability to regain her speech and how it improved her articulation.

I can't say enough about the use of Botox. I mean I can remember, I can speak...hello, you know...I don't think you can imagine going from "wah wah wah" and then 10 seconds after that injection to be able to say something, a word. I'm just flabbergasted. So it's super extreme as far as nothing to 100 percent. I couldn't say a word, it's just phenomenal to me (NF, F, 60 years).

NF continued: "Umm I think my speech is pretty good. Most people don't notice that I ever had a speech problem...I can say t's better, sh's better, but I don't always hit them" (NF, F, 60 years). SP also explained how his speech production improved post-injection. He stated, "The Botox relaxes some muscles, and it's easier for me to form the letters...I have less trouble speaking" (SP, M, 78 years).

Although SP described improvements in his speech, he also described some difficulties that arose post-injections. He stated:

I found that it's either the trauma of the needles in the face, or the Botox itself that would uh, cause me to um, speak... I have a problem with speaking and particularly enunciating because the lips didn't seem to want to make the sounds, you know? You want them to make like o's and a's and you know, r's... or particularly anything that would have... "o" with an "o" in it... and lips, and even as I'm speaking to you right now. Anything with an

“o” in it, the lips don’t seem to one to come together enough to clearly enunciate that “o” (SP, M, 78 years).

Participants explained how the injections increased their ability to respond when speaking. GM discussed how his response rate was “A lot quicker.” He stated:

Well, it has definitely helped me. Umm, see, my tongue and my brain. It seems to be a lot more in sync. Before I took Botox we probably could have measured the time it took after you had asked the question for me to start replying... there is a wire between my brain and my tongue that before was broken. Now it’s fixed that wire (GM, M, 69 years)

SP also mentioned how the injections helped him. He stated:

“You’re not thinking of, how should I answer that question, what words should I use, it’s sort of like an automatic situation. So when I get [Botox], it’s like putting your speech on autopilot. It just seems to work...it makes me think quicker. You think oh, ok” (SP, M, 78).

Participants discussed the amount of physical effort they exerted when speaking. GM discussed how the injections had helped him decrease the amount of physical effort he required to speak.

He stated:

I don’t have to work so hard on the speech things. It’s helped because I always felt I had to work on certain things to make it always the best or the better, but without Botox, I would have a hard time I think doing that (GM, M, 69years).

BR mentioned that post-injection he was able to put in less effort. He explained:

It helps your speech for sort of a month and a half or two months. Then after that, it’s a little more difficult. So you’re not conscious of making an effort. You’re not conscious of

your speech being slurred. So I think that's the biggest thing it has done for me (BR, M, 68 years)

SP also discussed physical effort, but his experience was in contrast to GM and BR. SP stated that after receiving the injections, he needed to “put a special effort into [speaking]” (SP, M, 78). He further explained, “I have to put just a little more effort to making, enunciating certain sounds...I found that it's either the trauma of the needles in the face, or the Botox itself that would uh, cause me to um, speak” (SP, M, 78 years).

Not all participants described a positive change to their speech production post-injection. Some participants discussed their uncertainty about any noticeable changes to their speech production, with some participants stating that they experienced no change to their speech production post-injection, as well as one who previously noted some positive changes. JR explained, “Oh. No, no. My speech. I feel. No, it hasn't increased; it hasn't done my speech at all... It's about the same. It hasn't gotten worse. It hasn't gotten better” (JR, M, 44 years). ST also discussed her uncertainty when asked if there were changes she had noticed. She said, “Uh, not a whole lot, speech, maybe” (ST, F, 78 years). Similarly, when GM was asked about speech intelligibility and any changes that were experienced in his speech he responded, “um, I don't...I don't know...I don't think so” (GM, M, 69 years).

Although EP mentioned improvements in his speech, he also discussed some speech problems he wished BoNT-A injections had helped with. He stated: “Umm, I wish I had less speech problems. I know I'm limited in speech slowness and pronunciation but you just have to accept it and move on” (EP, M, 80 years).

EP who had been receiving BoNT-A injections for 22 years explained that his speech production had changed over the years. He attributed this change to be the result of the deterioration of his

facial muscles due to his long history of receiving BoNT-A injections to manage his OMD. As a result of muscle atrophy, EP mentioned that he received the injections less frequently. He stated:

For the last year or so I find that even at the end of [the injection cycle] now because of the deterioration of the muscles, that period before I get the shots I'm still having trouble, slight limitations than what it used to be. I used to probably speak better before, but it's getting progressively worse so umm I can't get [the injection] every 3 months, for the jaw anyway (EP, M, 80 years).

EP also described changes to his speech intelligibility and speech rate as a result of receiving the injections for 22 years EP stated, "umm speech became thicker and changed, slower, took more time to formulate the jaw, tongue, throat muscles to make pronunciations that would normally just roll of your lips prior to that" (EP, M, 80 years).

I can now participate. Participants described their ability to participate in meaningful activities with their families and friends and how the ability to participate affected their psychosocial functioning. Participants also explained how changes to their speech production post-injection affected their communicative participation, including their ability to have meaningful conversations, communicate in social, public, and private settings and their ability to express their emotions and opinions accurately through their speech.

Participants discussed the meaning of communication and communicative participation and the importance of these in their lives. GM discussed that his "communication ability is a lot better"(GM, M, 69 years), as a result of receiving injections. He explained that the injections had made it easier to talk and increased his confidence to communicate with others. He stated:

I think the big thing is that um, I'm not totally afraid to talk. So if I didn't have the Botox, I would feel very hampered about talking. Uh, but either one-on-one or in a group or anything. I feel um, maybe more confident about that. I think I would always kind of keep trying harder to do something, but this makes it a lot easier. If they're going to ban Botox, I would be very, very afraid (GM, M, 69 years).

NF explained how the ability to communicate changed her life. She stated:

My speech, gave me my job back. Gave me my husband's conversations back, relationships, children and family and work. Isn't that life? That's everything. And you know now I'm able to give back, and I wouldn't be able to before. I feel like I'm contributing to the community (NF, F, 60 years),

NF further discussed the importance of communication and how the injections improved the important relationships in her life, her employment, and her well-being. She stated:

I have a job because of Botox. I can talk, I can be happy. I can laugh and communicate all of that emotionally, socially everything. It makes a difference between living and really not. You're just surviving. When you're not speaking and you don't have that communication, you're surviving. You're self-fulfilling and you can find other ways to study or read and educate yourself but you're not sharing. It's not a sharing thing. It's a very personal, inward type of existence whereas with speech; the difference is night and day. You can communicate, you can share. People can make you laugh you can make them laugh just through speech. You know, you can have a job and you cannot have a job (NF,F, 60 years).

NF also stated:

Well, considering I like to work, umm that's a big impact because then I'll be going back to doing what I like the most in my daily life, I like working umm so that is to me the biggest, and communication with family are the two tops in my book as far as my life. And my life would totally change without it (NF, F, 60 years).

NF and SP both discussed how other people viewed their ability to participate and communicate post-injection. NF mentioned her experience participating in her job post-injection and how people viewed her ability to communicate. She stated:

I think it's affected in so many ways when I went back to work, I became more confident. I became like a mentor kind of. Some of the people and my acquaintances that I knew were so surprised even I think the occupational health and safety person were shocked literally that I was ever able to come back to work from seeing me before I went and then coming back. Matter of fact, I ran into her a week ago and she said "I still can't believe that you were on the disabled road forever and then you're here now many years later" and um she can't believe the difference of my speech like speaking normal. So I know that there's a vast difference there. I had a friend an acquaintance at work who marvelled at how positive (NF, F, 60 years).

SP also mentioned how others viewed his communication skills post-injections. He stated:

Very few people ever asked me to repeat anything - either on the phone or in conversation with uh, an acquaintance or even strangers. The fact is, I had people tell me after about a year, maybe in the second year I think, said "You're speaking well... we never knew you... we never knew you were any different than what you are right now (SP, M, 78 years).

Similar to NF, SP discussed how his communication had changed post-injection and the meaning he attributed to these changes by stating: “I find it easier to speak and to communicate in a way I want to communicate it. That’s a big difference to me. And important to me” (SP, M, 78 years).

SP elaborated:

Well number one, you don’t have to repeat. Number two, you can get your message across. Number three, they can understand it the way you want it communicated. You know I think that... a big thing in communication is, you can say something or write something, but does the hearer, does the reader interpret it the way you want to say... “Well, what do you mean here?” “Well I meant this.” “Well, I’m sorry that’s not what it says.” You know, but it’s... there’s a big skill I think in communicating that everybody can hear you... what you want to say, how you want to say it, and you know, and it’s got the meaning and emotion behind it the way you intended it to be said...being able to, you know, communicate. That people can hear what I want to say, not only the words, but the meaning to the words. I think are important. And that’s important to me. And I said, “Well I didn’t mean it that way, I wasn’t upset.” “How come you’re such an old grouch”?! (SP, M, 78 years).

SP further discussed the importance of regaining effective communication post-injection, and he also described the challenges he faced when he could not communicate effectively. He stated:

Communication is... a two-way communication is easier, because you know, it’s difficult to have good communication, and good relations you know, if one person is saying something, and I don’t know what he’s saying, I have to guess at what he’s saying; I have to guess at what he means. It’s difficult for the other person, but to answer your question specifically, you know. That two-way communication is easier, more relaxed, and less

stressful, and I'm saying the same thing maybe, but you know, if you get that good communication with somebody else, and the most natural situation, you know, it's just, the other person's more fun to be around, and you're more fun to be around the other person, right? But if you have serious impediment, it's difficult for two reasons: they don't understand you and they get frustrated, and they'll feel a certain amount of sorry for you, like you can't communicate any better than what you're doing. I know I always feel that way if someone has a problem that you feel, you feel that you can't communicate with that person so well, but you feel sorry for him that he's... that he or she is in that situation (SP, M, 78 years).

Similarly, NF explained the importance of communication and how it impacts the ability to express one's needs. NF stated, "Communication is a big...I mean with any diagnosis you have to have communication, you go to any doctor and you have to be able to say what's wrong with you"(NF, F, 60 years).

Although JR discussed that his speech stayed "about the same" post-Botox injections, he still felt that the injections had allowed him to become "more social." He stated: "Because I'm not making it affect my job, work-wise, I can be more social" (JR, M, 44 years). Similarly, EP stated, "I find the Botox gives me ability to sit through a movie or just have a conversation with some people, whereas I would be severely restricted" (EP, M, 80 years). ST also described, "Well, I don't hesitate to be out and be talking as much as I was before... it improved my daily activities by going out with people" (ST, F, 78 years).

Chapter 4

4 Discussion

The purpose of this study was to obtain a self-reported account of the psychosocial consequences of BoNT-A injections for individuals living with OMD. Using qualitative phenomenological methodology, this study explored how participants judged the success of BoNT-A treatment based on the psychosocial issues relevant to them. This was achieved by understanding the insider's perspective through an interview process, where participants discussed and shared their treatment experiences. The discussion will further explore the results of the study and compare it to previous literature and research that has explored similar concepts. This section will also outline the study limitations, clinical implications, and future research directions.

4.1 Perspective and Experience

Two major themes emerged from this research. The first theme, *Botox has changed me and my experiences* explored the participants' perspective on receiving BoNT-A injections and the psychosocial impact of receiving these injections. From this first theme, four subthemes emerged. In first subtheme, *I feel normal again*, participants discussed that prior to receiving BoNT-A injections they felt disabled by OMD, but following injections many participants reported feeling less disabled and a return to a pre-disability sense of "normal". A study by Baylor, Yorkston, Eadie, and Maronian (2007), examined the psychosocial consequences of Botox treatment for SD, and reported similar findings to our research. Baylor and colleagues (2007) found that post-BoNT injections participants experienced a relief of SD symptoms, allowing them to feel and act like themselves again. The concept of return to pre-disability

“normal” was examined in a study by Novakovic, Waters, Elia, and Blitzer (2011). Novakovic and colleagues examined the longitudinal effects of BoNT treatment in participants with Adductor Spasmodic Dysphonia (ADSD) on functional outcomes and QoL. Patients completed an evaluation of their voice function after each injection using the percentage of normal function (PNF) scale and the Voice Handicap Index (VHI; Jacobson, Johnson, Grywalski, Silbergleit, Jacobson, Benninger, & Newman, 1997). These results showed that post-injections the average score on the VHI increased by 9.6% and the average PNF score increased by 30.3%.

Participants also discussed the beneficial psychosocial changes they experienced as the result of receiving BoNT-A injections, as captured in the second subtheme, *I feel appreciative of what Botox does for me*. Participants spoke about the perceived improvement in their QoL, increased confidence, and improved happiness post-injection. More specifically, our participants discussed that they experienced increased confidence in their ability to regain control of their orofacial movements and discussed the concept of a ‘second chance’. In this sub-theme, participants also discussed improvements in aspects of OMD they wanted improved following injections such as the ability to communicate more effectively. Finally, participants described how receiving BoNT injections improved their overall happiness.

Our participants’ appraisal of improved QoL as a result of receiving BoNT-A injections is similar to the findings reported by Charous and Comella (2011). This study examined QoL following Botox injections for individuals with jaw-opening dystonia and reported improvements in QoL for all participants post-injection. Their participants reported feeling more optimistic, less embarrassed, and generally felt better about themselves as a result of the injections (Charous & Comella, 2011). Our results are also consistent with the findings of Natasi et al. (2016) who examined the effect of BoNT-A injections on QoL in participants with isolated lingual dystonia

and OMD affecting the tongue. The results of this study revealed that following BoNT-A injections, participants reported a positive change in QoL as measured by a disease-specific questionnaire, the oromandibular dystonia questionnaire-25 (OMDQ-25; Merz et al., 2010). All domains of the OMDQ-25 showed improvement post-injection. Bhattacharyya & Tarsy (2001) also found that participants reported less depression and anxiety related to SD and/or OMD, and improvements in QoL post-BoNT treatment. The GBI was used as a measurement tool, to quantify the health benefit of BoNT injections. The general sub-score, social support sub-score, and physical health sub-score, all demonstrated statistically significant increases, showing that patients benefited from the injections (Bhattacharyya & Tarsy 2001). A study by Hogikyan, Wodchis, Spak and Kileny (2001), explored the longitudinal effects of BoNT injections on voice-related quality of life using the V-RQOL scale (V-RQOL; Hogikyan & Sethuraman, 1999) in 18 patients with ADSD. The results of the study by Hogikyan and colleagues (2001) demonstrated an improvement in V-RQOL, following BoNT injections. Another study by Courey, Garrett, Portell, Billante, Smith, Stone, and Netterville, (2000), studied 38 participants with SD receiving BoNT treatment and found similar improvements in QoL using the Voice Handicap Index scale (VHI; Jacobson et al., 1997) and the Short Form-36 scale (SF-36; Ware, Kosinski & Gandek, 1993), a 36 item patient-reported outcome that measures patient health, related to quality of life. Each outcome measure (i.e., VHI, SF-36) was administered to participants before treatment and one month after treatment. Following BoNT injections, participants reported a significant improvement in scores on both the VHI and SF-36. There were improvements in the participants overall VHI scores, and improvements in their functional, physical, and emotional handicap category scores post-treatment. On the SF-36, patients showed significant improvements in their mental health and social functioning scores.

Finally, a study by Faham, Torabinezhad, Murry, Dabirmoghaddam, Abolghasemi, Kamali, and Asgari, (2018) evaluated QoL and voice changes over multiple time points in 15 participants with ASD. Participants completed the Voice Activity and Participation Profile – Persian edition (VAPPP) (Ma & Yiu, 2001) pre-injection, and at 1, 3, and 6 months post-injection. The results of the Faham et al., study demonstrated that participants reported the greatest gains in QoL (as measured by the VAPPP) at 3 months post-injection, despite voice quality not being judged as normal. This finding of improved QoL, despite remaining voice impairment, relates to the third subtheme, *Botox doesn't fix everything*, which is discussed next.

Although the participants in this study expressed some positive psychosocial benefits of BoNT-A injections, all eight participants also acknowledged that BoNT-A injections did not fix everything. Participants described failure of expectations and side effects as a result of the injections. Participants discussed unpleasant side effects, such as muscle atrophy, bruising, and pain due to the injections. EP discussed how receiving BoNT-A injections for 22 years affected his muscles, causing deterioration and pain. A study by Baylor et al., (2007), examined the psychosocial consequences of Botox treatment for SD. The six participants with SD in the Baylor et al. study described the burden of Botox treatment, such as the fear and pain of receiving the injections, the cyclic and sometimes-inconsistent voice response, and the need for repeated injections. Our participants described dissatisfaction with the cyclic and changing nature of response, over the course of their BoNT-A injection cycle, with respect to speech production (expanded on in the next theme), as well as physical and emotional impacts. Participants in our study reported that their confidence decreased as the BoNT-A injections wore off, as well as loss of control, pain, tension, and fatigue, which developed towards the end of the injection cycle. Dry mouth is another reported side effect of Botox injections (Goldman &

Comella, 2003; Gn & Ng, 2017). Dry mouth was described in our research as an unwanted side effect. One participant, BR described the pain associated with dry mouth post-injection, causing his lower lip to stick to his teeth resulting in tears and swelling.

Failure of expectations regarding the effectiveness of BoNT-A injections in providing relief from a number of OMD-related symptoms was also reported in our study. Participants (SP, GM, FI, SP, and ST) discussed what they had wished BoNT-A injections could have helped or changed. Participants discussed that they wished they could regain control of the continuous orofacial movements, their face would go back to ‘normal’ post-injection, that their muscles would not deteriorate, and that after treatment they would get better and be cured. Baylor et al., (2007) reported similar findings, in regards to the participants’ ‘ideal’ Botox injection. Participants discussed that they wished that the injections were quick and pain-free, that there would be no breathy periods, the injections would last longer, they would have the ability to say sentences correctly, speak without thinking, or feel self-conscious (Baylor et al., 2007). The results of our study support the findings of other studies, demonstrating that BoNT-A injections do not fully meet expectations for every participant and this treatment can be associated with dissatisfaction and unwanted side effects.

Finally, in the fourth subtheme, *My face looks different* participants described changes related to orofacial aesthetics as a result of receiving BoNT-A injections. Many participants discussed experiencing positive changes to their facial appearance post-injection. These participants explained that they felt less self-conscious and less worried about unwanted dystonic movements, such as facial twitches, following injections. In addition, some participants (FI and SP) stated that the most important aspect of receiving Botox injections, were the positive changes to their facial appearance. This finding is consistent with that of Merz and colleagues

who explored and highlighted the importance of facial aesthetics in the participants they studied with OMD. In this study, Merz et al., (2010), evaluated the health-related QoL of 30 participants with OMD using a disease-specific measure, the OMDQ-25, at pre-treatment and then at four and eight weeks after BoNT-A treatment. The results of the Merz study revealed that the *cosmetic* sub-score of the OMDQ-25 demonstrated the greatest positive difference between pre- and post-BoNT-A injections. In our study not all participants experienced positive changes to their facial appearance following BoNT-A injections. Some participants reported feeling self-conscious as a result of the injections. GM described having red marks on his face as the result of the needles, and experienced grimacing as a side effect of the injections. Taken together, these findings indicate that individuals with OMD are highly sensitive to their outer appearance (Merz et al., 2010).

4.2 Speech Production and Participation

The second theme, *What communication is like for me* explored the psychosocial impact related to participants' speech production and communicative participation as a result of receiving BoNT-A injections. Captured in the first subtheme, *My speech has changed* participants reported changes in their voice and changes in the effort to speak as a result of receiving BoNT-A injections. Participants discussed changes to their speech intelligibility, articulation, response rate, and amount of physical effort required to speak. Many of our participants described an improvement to their speech intelligibility and speech production. Dykstra et al., (2007a) explored the effect of BoNT-A on speech intelligibility in an individual with lingual dystonia and reported positive changes to speech production, with a 12.4% increase in single word intelligibility and a 2.7% increase in sentence intelligibility following BoNT-A injections. Participants with SD in the Baylor et al., (2007) study reported a sense of 'freedom'

from not having to worry about their voice post-injection. Baylor and colleagues also found that all participants experienced improved voice quality following Botox injections, with some participants reporting that their voices ‘essentially returned to normal’ when Botox was at its peak effectiveness. Despite some of our participants reporting improved speech production as a result of BoNT-A injection, other participants’ perceived minimal or even no changes to their speech production post-injection. This finding is similar to that of Dykstra et al., (2015), who reported that 7/10 participants with OMD and dysarthria did not show significant improvements in speech intelligibility over the course of a BoNT-A injection cycle. Our participants (BR, FI, EP and SP) also reported dissatisfaction with the cyclic and inconsistent changes to their speech production over the course of their injection cycle. FI mentioned that the injections helped her speech initially, only during the first few weeks. Our findings are consistent with a study by Paniello, Barlow and Serna (2008) administered the V-RQOL to 22 participants with ASD every four weeks over the course of their BoNT-A injections cycles in order to “to quantify the proportion of each cycle during which patients experience meaningful benefit” (Paniello et al., 2008). The results of this study reported that significant voice benefit was derived for only 30% of the injection cycle. Baylor et al., (2007), found that their participants reported that the cyclic and inconsistent voice response after Botox was a burden. Our results suggest that the benefits of BoNT-A on speech production are variable not only over the course of the injection cycle, but across our participants.

Many participants in our study also described that they required less effort to speak post-BoNT-A injection. Baylor et al., (2007) reported a similar finding in their participants with SD. This study reported that their participants ‘sought relief from physical symptoms of effort and fatigue as a result of getting the injections’. However, not all of our participants experienced

reduced effort when speaking. For example, SP stated that, “I have to put just a little more effort to making, enunciating certain sounds”. Baylor et al., (2007), also reported similar results in their participants with SD. More specifically, immediately after the injection, participants felt that their voice was at its weakest and that towards the end of the injection cycle, the amount of physical effort to speak would return.

Another finding of this study related to changes in communicative participation following BoNT-A injections. This finding is captured in the second subtheme, *I can now participate*. Our results align with the results reported by Dykstra et al., (2007a) who found a positive difference in all participation restriction scores (and activity limitation scores) on the VAPP following BoNT-A injections. Importantly, the participant in the Dykstra et al., (2007a) study reported no activity limitations and participation restrictions relating to her job and social communication as a result of receiving BoNT-A injections. Baylor et al., (2007) also discussed that their participants with SD were able to experience new opportunities post-injection, and reported improved participation in social and community activities following injections. Other studies have reported that participants rated themselves as more ‘social’ post-injection (Charous & Comella, 2011), reported improvements in daily communication (Faham et al., 2010), and reported a reduction in social isolation and public avoidance (Epstein, Stygall & Newman, 1997). Similarly, participants (JR, EP, ST, SP, NF and GM) in our study reported the importance and meaningfulness of participating in social activities with family and friends, and that participation was improved post-injection. ST reported that post-injection, she felt an improvement in her daily activities. More specifically, she described that she no longer hesitated to go out and talk with people. The results reported in Baylor et al., (2007), also discussed how Botox injections helped their participants with SD return to activities that they valued and to resume a ‘sense of a

normal life', without their voice 'controlling' their participation in activities. The results presented by Baylor et al., (2007) align with the results of the current study by demonstrating that participants with OMD and SD report improvements to in social engagement and community participation post-injection. Our participants also reported how they were able to return to work following BoNT-A injections. Participants (NF and JR) reported that BoNT-A treatment allowed them to return to work and resume meaningful occupation. Similar to our findings, a study by Meyer, Hu & Hillel (2013), examined employment for individuals with SD who were receiving BoNT-A injections. A total of 145 patients with SD completed the Work Productivity and Activity Impairment (WPAI; Reilly, Zbrozek & Dukes, 1993) instrument, the Voice Handicap Index-10 (VHI-10; Rosen, Lee, Osborne & Murry, 2004) and underwent a perceptual evaluation (Consensus Auditory-Perceptual Evaluation of Voice) (CAPE-V; Kempster, Gerratt, Verdolini Abbott, Barkmeier-Kraemer & Hillman, 2009) in order to compare work outcomes with patient-perceived voice handicap and clinician-perceived voice quality. The results revealed that patients with SD reported that their vocal dysfunction caused a significant negative effect on work productivity and increased their activity limitations, which improved post-injection (Meyer et al., 2013). Across all of the studies, including the present study, it appears that communicative participation is identified as important and is improved as a result of BoNT-A injections.

4.3 Limitations of the Current Study

In this study, qualitative methods were employed to obtain the experience of living with OMD and receiving BoNT-A treatment from a psychosocial perspective. Phenomenology was used in this study because it allowed complex data to be collected through a self-reported account of participants lived experiences. Although this methodology has advantages, there are

also limitations. Data collection, analysis, and interpretation is substantial. Due to the time constraints of a student project and difficulties with participant recruitment resulting from the rare nature of OMD, our study was completed with a sample size of eight participants, despite an original target of 10-12 participants. While common themes emerged with our eight participants studied, a larger sample size would improve the rigor of the data, in terms of multivocality and saturation. A larger sample size would have also allowed for an increase in varied responses from individuals with OMD and their lived experiences of receiving BoNT-A therapy. Furthermore, all participants in this study were recruited from the Movement Disorders Clinic, London Health Sciences Centre in London, Ontario, which may have also led to a uniform demographic.

Member checking or respondent validation is a criterion put in place to improve the rigor of qualitative research to ensure validity and credibility. Member checking is accomplished by having participants validate their responses from the research interviews. It asks that participants verify the themes once the researcher has analyzed the data. (Lincoln & Guba, 1985). This process ensures that the results are in fact capturing what the participant was aiming to say. A component of this practice was completed during the interview process, whereby the researcher clarified participants' responses for correct interpretation and meaning during the actual interview. Unfortunately, the complete implementation of respondent validation was not completed within the time frame of this paper, however, our participants will be contacted in the future to obtain this information.

4.4 Future Directions

The results of this study provide a platform for additional research that examines the psychosocial impact of BoNT-A treatment for the management of OMD. A future study may

wish to employ alternate qualitative methodologies, such as grounded theory (Glaser & Strauss, 1967) in order to provide a generalized theory on the psychosocial impact of receiving BoNT-A injections for patients with OMD. This theory seeks to explain the phenomenon at hand and can be used as a predictor to explain future experiences of the phenomenon (Mills, Bonner & Francis, 2006). This research methodology could allow for the collection of additional data from the participants (i.e., personal diaries), incorporate pre-existing literature and include observations that the researcher has made throughout their interactions with the participants (Mills et al., 2006). This data could provide a broader and deeper understanding of the psychosocial impact of BoNT-A injections in the OMD patient population.

A study consisting of a larger sample size could be collected and compared to the findings in this study to test for theme saturation, or if there are new findings that could be discovered. In addition, a future study may wish to re-interview the same participants with a follow-up study at a five or ten year period to determine and compare if the same issues still present as relevant or if perspectives of treatment have changed or evolved over time. A study comparing traditional objective measures of treatment success could be compared to subjective measures of treatment success in order to determine if the traditional objective measures are capturing the relevant and meaningful outcomes that are important to the individual living with OMD.

4.5 Clinical Implications

The results of this study provide information about the psychosocial consequences of BoNT-A treatment for OMD that can help inform clinical practice. Understanding the participants' perspective on treatment can help improve rehabilitation in this clinical population. The results of this study highlighted the positive and negative consequences of BoNT-A

injections relating to QoL, speech production, and participation from the perspective of the participant. From these results, we were able to distil the following conclusions that have clinical applicability and importance when working with this clinical population.

The first is that BoNT-A injections generally result in positive outcomes and psychosocial consequences, however, chemodenervation should not be viewed as singular or one that addresses all concerns or issues. This is an important clinical issue because chemodenervation via BoNT-A injections is the primary therapeutic treatment for patients with OMD (Teemul et al., 2016) and is considered to be the ‘gold standard’ and the most effective treatment for OMD (Kazerooni & Broadhead, 2015). We advocate that adjunctive therapy, in addition to chemodenervation, should always be considered in order to provide broader and more holistic care for this patient population. This could include counselling, occupational therapy, dentistry, and speech therapy, as examples. Although our participants reported that BoNT-A injections resulted in variable speech production and facial appearance outcomes, these factors were reported as a priority and important for our participants in judging the success of their treatment. Therefore, it is prudent for clinicians to provide additional management such as the provision of a bite block that could improve not only facial appearance, but also articulatory precision, and hyperactive movements associated with hyperkinetic dysarthria (Dworkin, 1996; Goldman & Comella, 2003). Additionally, communicative interaction strategies, such as dyadic strategies to improve communication or modifying the communicative environment to improve visual and auditory acuity, may be appropriate to ensure successful communicative participation (Duffy, 2013). Further, since the experience of having OMD is variable and individualized, any treatment plan should be tailored to the individual based on his/her priorities and personal concerns related to OMD. Finally, assessing psychosocial outcomes of BoNT-A injections

should be ongoing in order to understand and to respond to evolving patient needs, goals, and priorities (Baylor et al., 2007).

Chapter 5

5 Summary and Conclusions

This study explored the lived experiences of eight individuals receiving BoNT-A as a treatment for OMD. The results of this study suggest that BoNT-A treatment has variable impact and effect across domains related to quality of life, expectation and satisfaction with treatment, speech production, and communicative participation; with some participants reporting positive effects associated with receiving injections, and others reporting more negative or neutral effect/impact. This information is extremely important in augmenting our understanding of the impact of BoNT-A treatment, through a psychosocial lens and from the perspective of the individual receiving this treatment.

This research adds novel information relating to the psychosocial consequences of BoNT-A treatment in the management of OMD and builds on a small, but growing literature that seeks to study the consequences and experiences of living with OMD. The results of this study also have practical clinically-based utility because our findings can potentially aid clinicians in targeting relevant and meaningful outcomes of treatment success. With expansion, this line of research inquiry can aid in the eventual development of appropriate patient-reported outcome measures that capture the issues relevant and meaningful to this clinical population.

References

- Bakke, M., Larsen, B. M., Dalager, T., & Møller, E. (2013). Oromandibular dystonia—functional and clinical characteristics: A report on 21 cases. *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology*, *115*(1), 21-26.
- Baylor, C., Yorkston, K.M., Eadie, T.L., and Maronian, N.C. (2007). The psychosocial consequences of BOTOX injections for spasmodic dysphonia: a qualitative study of patients' experiences. *Journal of Voice*, *21*(2), 231-247.
- Benner, P. (1994). The tradition and skill of interpretive phenomenology in studying health, illness, and caring practices. *Interpretive phenomenology: Embodiment, caring, and ethics in health and illness*, 99-127.
- Bhattacharyya, N., & Tarsy, D. (2001). Impact on quality of life of botulinum toxin treatments for spasmodic dysphonia and oromandibular dystonia. *Archives of Otolaryngology–Head & Neck Surgery*, *127*(4), 389-392.
- Blanchet, P. J., Rompre P. H., Lavigne G. J., & Lamarche C. (2005). Oral dyskinesia: a clinical overview. *International Journal of Prosthodontics*, *18*(1), 10-9.
- Blitzer, A., Brin, M. F., & Fahn, S. (1991). Botulinum toxin injections for lingual dystonia. *The Laryngoscope*, *101*(7), 799.
- Blitzer, A. & Sulica, L. (2001). Botulinum toxin: basic science and clinical uses in otolaryngology. *Laryngoscope*, *111*, 218-226.
- Brin, M. F., Comella, C. L., & Jankovic, J. (2004). Pathophysiology of dystonia. *Dystonia: Etiology, Clinical Features, and Treatment*, 5-10.

- Charles, P., Davis, T., Shannon, K., Hook, M., & Warner, J. (1997). Tongue protrusion dystonia: Treatment with botulinum toxin. *Southern Medical Journal*, *90*(5), 522-525.
- Charous, S. J., & Comella, C. L. (2011). Jaw-opening dystonia: Quality of life after botulinum toxin injections. *Ear, Nose and Throat Journal*, *90*(2), 48.
- Courey, M. S., Garrett, C. G., Portell, M. D., Billante, C. R., Smith, T. L., Stone, R. E., & Netterville, J. L. (2000). Outcomes assessment following treatment of spasmodic dysphonia with botulinum toxin. *Annals of Otolaryngology, Rhinology & Laryngology*, *109*(9), 819-822.
- Creswell, J. W. (2014). *Research design: Qualitative, quantitative, and mixed methods approaches* (4th ed.). Thousand Oaks: SAGE.
- Crist, J. D., & Tanner, C. A. (2003). Interpretation/Analysis methods in hermeneutic interpretive phenomenology. *Nursing Research*, *52*(3), 202-205.
- Darley, F. L., Aronson, A. E., & Brown, J. R. (1969a). Differential diagnostic patterns of dysarthria. *Journal of Speech, Language, and Hearing Research*, *12*(2), 246-269.
- Darley, F. L., Aronson, A. E., & Brown, J. R. (1969b). Clusters of deviant speech dimensions in the dysarthrias. *Journal of Speech, Language, and Hearing Research*, *12*, 462-496.
- de Oliveira, A. M., Buchain, P. C., Vizzotto, A. D. B., Elkis, H., & Cordeiro, Q. (2013). Psychosocial impact. *Encyclopedia of Behavioral Medicine*, 1583-1584.
- Denzin, N. K., & Lincoln, Y. S. (Eds.). (2011). *The Sage handbook of qualitative research*. Sage.
- Donovan, N. J., Velozo, C. A., & Rosenbek, J. C. (2007). The communicative effectiveness survey: Investigating its item-level psychometric properties. *Journal of Medical Speech-Language Pathology*, *15*(4), 433-447.

- Dowling, M. (2007). From Husserl to van Manne. A review of different phenomenological approaches. *International Journal of Nursing Studies*, 44, 131-142.
- Duffy, J. R. (2013). *Motor speech disorders: substrates, differential diagnosis, and management* (3rd ed.) St. Louis, MO: Elsevier Health Sciences.
- Dworkin, J.P. (1996). Bite-block therapy for oromandibular dystonia. *Journal of Medical Speech-Language Pathology*, 4(1), 47-56.
- Dykstra, A. D., Adams, S. G., & Jog, M. (2005). Lingual protrusion and elevation in lingual dystonia: A hypothesis. *Canadian Acoustics*, 33(3), 98-99.
- Dykstra, A.D., Adams, S., & Jog, M. (2007a). The effect of botulinum toxin type A on speech intelligibility in lingual dystonia. *Journal of Medical Speech-Language Pathology*, 15(2), 172-186.
- Dykstra, A.D., Domingo, Y., Adams, S. G., Jog, M. (2015). Examining speech intelligibility and self-ratings of communicative effectiveness in speakers with oromandibular dystonia receiving botulinum toxin therapy. *Canadian Journal of Speech, Language Pathology and Audiology*, 39(4), 334-345.
- Dykstra, A.D., Hakel, M.E., & Adams, S.G. (2007b). Application of the ICF in reduced speech intelligibility in dysarthria. *Seminars in Speech and Language*, 28(4), 301-311.
- Dykstra, A.D. & Adams, S. (2009). *Oromandibular dystonia* (pp.359-361). In *Clinical Management of Sensorimotor Speech Disorders, Second Edition*, Malcolm R. McNeil (ed.), New York: Thieme.
- Epstein, R., Stygall, J., & Newman, S. (1997). The short-term impact of Botox injections on speech disability in adductor spasmodic dysphonia. *Disability and rehabilitation*, 19(1), 20-25.

- Faham, M., Torabinezhad, F., Murry, T., Dabirmoghaddam, P., Abolghasemi, J., Kamali, M., & Asgari, M. (2018). Quality of Life and Voice Changes After a Single Injection in Patients With ADSD Over Time. *Journal of Voice; Official Journal of the Voice Foundation*.
- Finlay, L. (2002). "Outing" the researcher: The provenance, process and practice of reflexivity. *Qualitative Health Research, 12*(4), 531-545.
- Giladi, N. (1997). The mechanism of action of botulinum toxin type A in focal dystonia is most probably through its dual effect on efferent (motor) and afferent pathways at the injected site. *Journal of the Neurological Sciences, 152*, 132-135.
- Glaser, B., & Strauss, A. (1967). *The discovery of grounded theory: Strategies for qualitative research*. Chicago: Aldine.
- Gn, S., & Nag, A. (2017). Management of Oromandibular Dystonia: A Case Report and Literature Update. *Case reports in dentistry, 2017*.
- Goldman, J. G., & Comella, C. L. (2003). Treatment of dystonia. *Clinical Neuropharmacology, 26*(2), 102-108.
- Golper, L. A. C., Nutt, J. G., Rau, M. T., & Coleman, R. O. (1983). Focal cranial dystonia. *Journal of Speech and Hearing Disorders, 48*(2), 128-134.
- Guillemin, M., & Gillam, L. (2004). Ethics, reflexivity, and "ethically important moments" in research. *Qualitative inquiry, 10*(2), 261-280.
- Hammersley, M, and Atkinson, P. (1995) *Ethnography: Principles in Praclice*, 2nd ed, London: Routledge.
- Hogikyan, N.D. & Sethuraman, G. (1999). Validation of an instrument to measure voice-related quality of life (V-RQOL). *Journal of Voice, 13*(4), 557-569.

- Hogikyan, N. D., Wodchis, W. P., Spak, C., & Kileny, P. R. (2001). Longitudinal effects of botulinum toxin injections on voice-related quality of life (V-RQOL) for patients with adductory spasmodic dysphonia. *Journal of Voice*, 15(4), 576-586.
- Hustad, K. (1999). Optimizing communicative effectiveness. In K.M. Yorkston, D. R. Beukelman, E. A. Strand, & K. R. Bell (Eds.), *Management of motor speech disorders in children and adults*, 2nd ed. (pp. 483-539). Austin, TX: Pro-Ed.
- Jacobson, B.H., Johnson, A., Grywalski, C., Silbergleit, A., Jacobson, G., Benninger, M.S., and Newman, C.W. (1997). The Voice Handicap Index (VHI): development and validation. *American Journal of Speech-Language Pathology*, 6(3), 66-70.
- Jankovic, J. (2009). Peripherally induced movement disorders. *Neurologic clinics*, 27(3), 821-32.
- Jankovic, J. (2018). An update on new and unique uses of botulinum toxin in movement disorders. *Toxicon*, 147, 84-88.
- Jankovic, J., Schwartz, K., Donovan, D.T. (1990). Botulinum toxin treatment of cranial-cervical dystonia, spasmodic dystonia, other focal dystonias, and hemifacial spasm. *Journal of Neurology, Neurosurgery, and Psychiatry*, 53(8), 633-639.
- Karp, B. I., Goldstein, S. R., & Chen, R. (1999). An open trial of clozapine for dystonia. *Movement Disorders*, 14, 652-657.
- Kazerooni, R. & Broadhead, C. (2015). Cost-utility analysis of botulinum toxin type A products for the treatment of cervical dystonia. *American Journal of Health-System Pharmacy*, 72(4), 301-307.

- Kempster, G. B., Gerratt, B. R., Verdolini Abbott, K., Barkmeier-Kraemer, J., & Hillman, R. E. (2009). Consensus auditory-perceptual evaluation of voice: Development of a standardized clinical protocol. *American Journal of Speech-Language Pathology, 18*(2), 124-132.
- Kent, R. D. (Ed.). (1992). *Intelligibility in speech disorders: Theory, measurement and management* (Vol. 1). John Benjamins Publishing.
- Khan, J., Anwer, H. M. M., Eliav, E., & Heir, G. (2015). Oromandibular dystonia: Differential diagnosis and management. *Journal of the American Dental Association (1939), 146*(9), 690.
- Klein, C. (2014). Genetics in dystonia. *Parkinsonism and Related Disorders, 20*, 137-142.
- Krauss, J. K., Yianni, J., Loher, T. J., & Aziz, T. Z. (2004). Deep brain stimulation for dystonia. *Journal of Clinical Neurophysiology, 21*(1), 18-30.
- Lee, K. H. (2007). Oromandibular dystonia. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology, 104*(4), 491-496.
- Lincoln, Y. S., & Guba, E. G. (1985). *Naturalistic inquiry*. Beverly Hills, CA: Sage Publications.
- Lincoln, Y. S., & Guba, E. G. (2004). The roots of fourth generation evaluation. *Evaluation roots: Tracing theorists' views and influences, 225-241*.
- Lowes, L., & Prowse, M. A. (2001). Standing outside the interview process? The illusion of objectivity in phenomenological data generation. *International Journal of Nursing Studies, 38*(4), 471-480.
- Ma, E. P., & Yiu, E. M. (2001). Voice activity and participation profile: assessing the impact of voice disorders on daily activities. *Journal of speech, language, and hearing research, 44*(3), 511-524.

- Martikainen, P., Bartley, M., & Lahelma, E. (2002). Psychosocial determinants of health in social epidemiology. *International Journal of Epidemiology*, *31*(6), 1091–1093.
- Merz, R. I., Deakin, J., & Hawthorne, M. R. (2010). Oromandibular dystonia questionnaire (OMDQ-25): A valid and reliable instrument for measuring health-related quality of life: Oromandibular dystonia questionnaire (OMDQ-25). *Clinical Otolaryngology*, *35*(5), 390-396.
- Meyer, T. K., Hu, A., & Hillel, A. D. (2013). Voice disorders in the workplace: Productivity in spasmodic dysphonia and the impact of botulinum toxin. *The Laryngoscope*, *123*, S1-S14.
- Mills, J., Bonner, A., & Francis, K. (2006). The development of constructivist grounded theory. *International Journal of Qualitative Methods*, *5*(1), 25-35.
- Moustakas, C. (1994). *Phenomenological research methods*. Thousand Oaks, CA: Sage.
- Nastasi, L., Mostile, G., Nicoletti, A., Zappia, M., Reggio, E., & Catania, S. (2016). Effect of botulinum toxin treatment on quality of life in patients with isolated lingual dystonia and oromandibular dystonia affecting the tongue. *Journal of Neurology*, *263*(9), 1702-1708.
- Netsell, R., & Cleeland, C. S. (1973). Modification of lip hypertonia in dysarthria using EMG feedback. *Journal of Speech and Hearing Disorders*, *38*(1), 131-140.
- Novakovic, D., Waters, H. H., D'Elia, J. B., & Blitzer, A. (2011). Botulinum toxin treatment of adductor spasmodic dysphonia: Longitudinal functional outcomes. *The Laryngoscope*, *121*(3), 606-612.
- Nutt, J. G., Muenter, M. D., Aronson, A., Kurland, L.T., & Melton, L. J. (1988). Epidemiology of focal and generalized dystonia in Rochester, Minnesota. *Movement Disorders*, *3*(3), 188-194.

- Page, A. D., & Siegel, L. (2017). Perspectives on the psychosocial management of oromandibular dystonia. *Seminars in Speech and Language, 38*(3), 173-183.
- Page, A. D., Siegel, L., & Jog, M. (2017). Self-rated communication-related quality of life of individuals with oromandibular dystonia receiving botulinum toxin injections. *American Journal of Speech - Language Pathology, 26*(2S), 674.
- Paniello, R. C., Barlow, J., & Serna, J. S. (2008). Longitudinal follow-up of adductor spasmodic dysphonia patients after botulinum toxin injection: Quality of life results. *The Laryngoscope, 118*(3), 564-568.
- Papapetropoulos, S., & Singer, C. (2006). Eating dysfunction associated with oromandibular dystonia: clinical characteristics and treatment considerations. *Head and Face Medicine, 2*(47), 1-4.
- Patton, M. Q. (2002). *Qualitative evaluation and research methods (3rd ed.)*. Newbury Park, CA: Sage.
- Paul, D., Frattali, C., Holland, A., Thompson, C., Caperton, C., & Slater, S. (2004). *Quality of Communication Life Scale (ASHA QCL)*. Rockville, MD: American Speech-Language-Hearing Association.
- Ramos, VF, Karp, B. I., & Hallett, M. (2014). Tricks in dystonia: Ordering the complexity. *Journal of Neurology, Neurosurgery, and Psychiatry, 85*(9), 987-993.
- Reilly, M. C., Zbrozek, A. S., & Dukes, E. M. (1993). The validity and reproducibility of a work productivity and activity impairment instrument. *Pharmacoeconomics, 4*(5), 353-365.
- Riessman, C. K. (1993). *Narrative analysis (Vol. 30)*. Thousand Oaks, CA: Sage Publications.

- Riessman, C. K. (2008). *Narrative methods for the human sciences*. Thousand Oaks, CA Sage Publications.
- Robinson, K., Gatehouse, S., & Browning, G. G. (1996). Measuring patient benefit from otorhinolaryngological surgery and therapy. *Annals of Otology, Rhinology & Laryngology*, 105(6), 415-422.
- Rosen, C. A., Lee, A. S., Osborne, J., Zullo, T., & Murry, T. (2004). Development and validation of the voice handicap Index-10. *The Laryngoscope*, 114(9), 1549-1556.
- Sallinen, M., Kukkurainen, M. L., Peltokallio, L., Mikkelsen, M., Anderberg, U. M., Medicinska fakulteten, .Socialmedicin. (2012). Fatigue, worry, and fear-life events in the narratives of women with fibromyalgia. *Health Care for Women International*, 33(5), 473-494.
- Seikel, J., Drumright, D., & King, D. (2016). *Anatomy & Physiology for Speech, Language, and Hearing* (5th ed.) Delmar Cengage Learning, (pg. 318).
- Sinclair, C., Gurey, L., & Blitzer, A. (2013). Oromandibular dystonia: Long-term management with botulinum toxin. *The Laryngoscope*, 123(12), 3078-3083.
- Simpson, L. L. (1981). The origin, structure, and pharmacologic activity of botulinum toxin. *Pharmacological Reviews*, 33, 155-188.
- Tarsy, D., & Simon, D. K., (2006). Dystonia. *The New England Journal of Medicine*, 355(8), 818-829.
- Teemul, T. A., Patel, R., Kanatas, A., & Carter, L. M. (2016). Management of oromandibular dystonia with botulinum A toxin: A series of cases. *British Journal of Oral and Maxillofacial Surgery*, 54(10), 1080-1084.

- The Glasgow Health Status Questionnaires Manual*. (1998). Glasgow, Scotland: MRC Institute of Hearing Research, Glasgow Royal Infirmary.
- Tinter, R. & Jankovic, J. (2002). Botulinum toxin type A in the management of oromandibular dystonia and bruxism. *Scientific and Therapeutic Aspects of Botulinum Toxin*, 1-12.
- Tracy, S. J. (2010). Qualitative quality: Eight "big-tent" criteria for excellent qualitative research. *Qualitative Inquiry*, 16(10), 837.
- Tuohy, D., Cooney, A., Dowling, M., Murphy, K., & Sixsmith, J. (2013). An overview of interpretive phenomenology as a research methodology. *Nurse Researcher*, 20(6), 17.
- Ware, J. E., Kosinski, M., & Gandek, B. (1993). *SF-36 health survey: Manual & interpretation guide*. Boston, Mass: The Health Institute, NEMC.
- Wilding, C., Whiteford, G. (2005). Phenomenological research: an exploration of conceptual, theoretical, and practical issues. *OTJR: Occupation, Participation and Health*, 25(3), 98- 104.
- World Health Organization. (1948). Retrieved from http://www.who.int/healthsystems/hss_glossary/en/index5.html
- Yorkston, K., Dowden, P. and Beukelman, D. (1992). Intelligibility measurement as a tool in the clinical management of dysarthric speakers. In: R. Kent, ed., *Intelligibility in Speech Disorders: Theory, Measurement, and Management*. Amsterdam, Philadelphia: John Benjamins Publishing, pp.265-285.
- Yorkston K, Strand E, Kennedy M. (1996). Comprehensibility of dysarthric speech: Implications for assessment and treatment planning. *American Journal of Speech-Language Pathology*, 5, 55 -66.

Yoshida, K. (2018). Sensory trick splint as a multimodal therapy for oromandibular dystonia.
Journal of Prosthodontic Research, 62(2), 239-244.

Appendices

Appendix A: Letter of Information

STUDY TITLE

The effects botulinum toxin A on speech intelligibility, levels of speech usage, communication apprehension, self-perceived communication competence, communicative effectiveness, communication-related quality of life and the lived experiences of individuals with oromandibular dystonia.

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London Health Sciences Centre, University Campus and Western University

INTRODUCTION

This letter of information describes a research study and what you may expect if you decide to participate. You should read the letter carefully and ask the person discussing this with you any questions that you may have before making a decision whether or not to participate. This form contains important information and telephone numbers, so you should keep this copy for future reference. If you decide not to participate in this study, the decision will not be held against you and will not affect your treatment in any way.

You are being asked to participate in this research study because you are an individual with oromandibular dystonia (OMD). The purpose of this study is to investigate the effects of oromandibular dystonia on your speech intelligibility (how understandable your speech is), your level of speech usage, your level of apprehension or concern when you are communicating orally, your self-perceived communicative competence, your effectiveness as a communicator in different social settings and your quality of life as it relates to communication. An additional purpose of this study is to compare how the Botox® injections you are receiving to manage your dystonia affects your speech intelligibility, your communicative apprehension, communication effectiveness and communication-related quality of life. We are also interested in learning about your experience of having oromandibular dystonia.

This study will involve 30 participants with OMD. Information about participants will be collected from patient charts and person-to-person interviews by the principal experimenter or another designated

member of the research team. This will include information about the participant's date of birth, general medical history, neurological history, and speech and hearing history.

This study will be conducted over two sessions, separated by approximately one month and lasting approximately 40 minutes for the first visit and approximately 2 hours for the second visit. Both visits will involve speech recordings of your voice. During this 10 minute recording period you will be asked to read aloud a series of 57 single words and 11 sentences while being recorded with a microphone. Both visits will also involve completing a series of six questionnaires that will look at how you use your speech on a daily basis, your level of concern or apprehension when you are communicating orally, your self-perceived competence when communicating, your effectiveness as a communicator in different social situations and your quality of life as it relates to your communication. It is anticipated that completion of the questionnaires will take approximately 30 minutes. The second visit will involve an additional 60-90 minute one-time in-person interview with the researchers in order to learn more about your experiences of living with oromandibular dystonia. During this interview we will ask you to share stories and information about strategies you have used to help you participate in life activities due to having dystonia. We want to hear about strategies that worked well and those that did not work well. In particular, we want to hear about things that make you more or less confident about your participation in activities. We want to hear your recommendations that you would give to other people in similar circumstances. You do not need to answer any questions you do not want to answer. The interview will be audio-recorded. Only the researchers will have access to the recording of the interview. The audio file would be stored on a secure server at Western University.

The first visit will be completed during your scheduled clinic visit at the Movement Disorders Clinic. The second visit will be scheduled approximately one month later to ensure that your Botox® treatment is working optimally.

If you agree to participate you will be able to complete the first visit of the study directly following your scheduled appointment time at the Movement Disorders Clinic in a separate testing room located within University Hospital. For the second visit of the study you will be asked to come to the Principal Investigator's Lab in Elborn College at the University of Western Ontario for repeat administration of questionnaires, speech recordings and the in-person interview. While at Elborn College, you will be provided with free parking.

The experimental procedures will require very little physical effort, and there is no known discomfort or risk involved in performing them. You will be seated in a comfortable chair throughout the procedures and during the interview and you will be given rest breaks approximately every five minutes or more frequently if required.

The procedures that will be used during this study are experimental in nature and will not provide any direct benefit to the participant's medical condition, however, it is anticipated that results from this study may provide important information about the effect of oromandibular dystonia on speech intelligibility, one's perception of their apprehension when communicating orally, their level of speech usage, their perception of how effective they are as communicators, and their quality of life as it relates to communication. It may also provide important information about the effect of Botox® on speech intelligibility, communication apprehension, communicative effectiveness and communication-related quality of life. Financial compensation will not be provided upon completion of this study. Parking costs over and above your regular clinic visit at the Movement Disorders Clinic will not be reimbursed. While at Elborn College, you will be provided with free parking.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions, or withdraw from the study at any time with no effect on your future care.

All of the information obtained in this study will be held in strict confidence. Your name and any identifying information will be removed from the data. If the results of the study are published, your name will not be used and no information that discloses your identity will be released or published. Representatives of Western University's Health Sciences Research Ethics Board may contact you or require access to your study-related records to monitor the conduct of the research. You do not waive any legal rights by signing the consent form.

Throughout the study, all confidential information and data will be preserved in a locked filing cabinet in the Principal Investigator's laboratory at Elborn College, Western University. All study materials will be destroyed after 25 years.

If requested, you will be provided with a copy of any publication related to the results of this study when it becomes available.

If you have any questions or would like additional information about this study, please contact Professor Allyson Dykstra. If you have any questions about the conduct of this study or your rights as a research subject you may contact Dr. David Hill.

If you agree to participate in this study, please sign the consent form on the next page.

Sincerely,

Allyson Dykstra, PhD Assistant Professor

Appendix B: Consent Form

STUDY TITLE

The effects botulinum toxin A on speech intelligibility, levels of speech usage, communication apprehension, self-perceived communication competence, communicative effectiveness, communication-related quality of life and the lived experiences of individuals with oromandibular dystonia.

PRINCIPAL INVESTIGATOR

Allyson Dykstra, Ph.D.

Assistant Professor

School of Communication Sciences and Disorders, Western University

CO-INVESTIGATOR

Dr. Mandar Jog, MD, FRCPC, Professor

Director, Movement Disorders Program

London Health Sciences Centre, University Campus and Western University

I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction.

_____	_____	_____
Signature of Research Subject	Printed Name	Date

_____	_____	_____
Signature of Person Obtaining Consent	Printed Name	Date

Appendix C: Ethics Approval Notice



Use of Human Participants - Ethics Approval Notice

Principal Investigator: Dr. Allyson Dykstra

Review Number: 17190E

Review Level: Delegated

Approved Local Adult Participants: 30

Approved Local Minor Participants: 0

Protocol Title: The effects of botulinum toxin A on speech intelligibility, levels of speech usage, communication apprehension, self-perceived communication competence, communicative effectiveness and communication-related quality of life in individuals with oromandibular dystonia

Department & Institution: Communication Sciences & Disorders, University of Western Ontario

Sponsor:

Ethics Approval Date: August 19, 2011

Expiry Date: August 31, 2013

Documents Reviewed & Approved & Documents Received for Information:

Document Name	Comments	Version Date
UWO Protocol		
Letter of Information & Consent		2011/08/11

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/CH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced revision(s) or amendment(s) on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

The Chair of the HSREB is Dr. Joseph Gilbert. The UWO HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

The University of Western Ontario
Office of Research Ethics

Support Services Building Room 5150 • London, Ontario • CANADA - N6G 1G9
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Appendix D: Code Dictionary

1. Botox Injection:

- a. **Frequency:** Number of times individuals receive a dosage, number of months in between dosage, number of dosages
- b. **Injection site:** BoNT-A needle injections at specific sites on the facial area
- c. **Physical Effects:**
 - i. **Effect on Muscle:** lack of tension/or extra tense, loosened muscles, tightness, deterioration of muscles
 - ii. **Muscle Movement:** lack of control or improved control, locking muscles, tremor, freedom of movement
 - iii. **BoNT-A Wearing off**

2. Eating/drinking:

- a. **Eating Mechanics:** fear of choking, avoidance of certain foods, food consistency, keeping food/drink in mouth
- b. **Physical Effect on Eating:** loss of taste, dry mouth, saliva and lubrication, pain due to biting inside of mouth/ tongue
- c. **Social Aspect of Eating:** self-consciousness, safety
- d. **Strategies to Improve Eating:** saliva production

3. **Facial Aesthetics:** Facial alteration as a result of the injections, the effect of the needles, the effect of the BoNT-A on the muscles

4. **Good Story:** Personal stories depicting positive or negative interactions/moments post-BoNT-A injections

5. **Job:** Ability to work post- BoNT-A, loss of job as a result of the BoNT-A, type of job, regaining job, personal values attributed to job

6. Pain:

- a. **Pain From Injection:** swollen tongue, not able to breathe, swelling, pain/no pain as a result of the injections, biting inside of mouth/ tongue as result of injection
- b. **Pain Post-Injection:** from the facial movements post- BoNT-A injections, aches, soreness, cuts in mouth, swelling

7. **Personal Perspective:** How participants view themselves post-injection, view of disability and normalcy

8. **Psychological effect:** Emotional reaction as a result of receiving BoNT-A, shock, happiness, disappointment, confidence, hopefulness, embarrassment, surprise, self-

esteem, uncertainty, relief, self-consciousness, feeling intelligent or lacking intelligence, sadness, frustration, fear, excitement, awareness, worry, stress, affect on QOL

9. **Resilience:** Pushing through difficult situations, persevering, tackling physical and emotional barriers, improving outlook, strong character traits, positive outlook

10. Social Aspects:

- a. **Participation in Social Activities/:** Ability to interact in social settings (religious settings, family gatherings)
- b. **Perspective of Others:** Others perception of their treatment process
- c. **Relationships:** Effect on Family and friends relationships post-treatment

11. Speech:

- a. **Communication:** Ability to hold a conversation, to have meaningful conversations and communicate in social and public and private settings, ability to express emotions and opinions through speech
- b. **Fatigue:** exerting effort to speak post-injection, less fatigue as a result of the injections, less effort post-injection, tired from speaking too much, loss of speech due to being tired
- c. **Intelligibility:** Improved or worsened intelligibility, improvement/ or no improvement of slurred speech, individuals understanding of participants speech, and participants' perspective on their speech
- d. **Speech Mechanics:** Response rate, ability to speak, difficulty speaking, pronunciation, Speech rate, strategies and word substitutions, enunciation, formation of letters, loudness, initiating speech, speech production, voice change

Appendix E: Reflexive Notes

Section 1: Qualitative Research: A New Avenue

Once I completed a course in qualitative methods, my social positioning as a researcher became even more evident. What captured me most was how qualitative research had the ability to humanize diverse lived experiences.

After immersing myself in qualitative literature, I became more aware of its multidimensional nature. Qualitative research has the ability to present individual or collective narratives and portray how these experiences shape the complexity of meaningful activities that take place in day-to-day life. As I am studying Oromandibular dystonia, I must consider my outsider status. Even though I do not struggle with the exact disorder the participants may face, qualitative research allows me to share their consciousness at a human level. Qualitative research has completely changed my outlook on my research and made me eager and motivated to understand and empathize with the lived experiences of each participant.

My research was conducted using a phenomenological methodological approach. After learning about phenomenology I realized that it would be the most appropriate methodology to express participants' experiences of receiving BoNT-A injections. I was able to analyze the participants lived experiences and shed light on issues that were relevant to them.

After reading some more papers related to my research topic, I realized that my research would be guided by an interpretivist paradigm. This was due to the fact that I was analyzing previously transcribed interviews. I listened to the audio-recorded interviews to gain a deeper connection with the participants and further immerse myself into their stories and experiences. This paradigm allowed me to combine previous literature findings and the participants' findings,

to share the lived experience of OMD and the psychosocial impact of receiving BoNT-A injections.

Section 2: Shared Consciousness

Prior to my research on OMD and BoNT-A, I began with an assumption that the majority of the participants would describe it as a viable treatment. This assumption was further realized in my neurological motor speech and disorders course. This course provided me with knowledge on OMD, its symptoms and treatment methods. Understanding OMD was my first step in appreciating the impact of BoNT-A on individuals. I gained an appreciation for the vast influence it has on participants, by understanding the regions of the brain affected, the symptoms, dystonic movements and effect on language and speech. I also acquired information on the effect of treatment methods, including BoNT-A injections. This insight provided me with an excellent platform to appreciate the effect of BoNT-A treatment, from a physical perspective.

Alongside the motor speech course, I was enrolled in a course on the International Classification of Functioning (ICF) framework. This course allowed me to shift my understanding of OMD and think about it from a comprehensive perspective, which surpassed its physical impact. Stepping back and trying to understand the impact of OMD from this framework was difficult. I struggled with understanding the impact of OMD from a psychosocial perspective, but this course allowed me to delve into previous qualitative papers and helped me recognize how OMD and BoNT-A could impact an individual at all levels, from participation and QoL to personal and environmental factors, and that this was not a linear or sequential impact, but rather an interactive impact where each component could affect the other in whichever way.

My literature review results produced many papers on OMD and BoNT-A, from a physical aspect, alongside some literature on emotional and social well-being, post-injection. Although the research was sparse, it improved my understanding of OMD and BoNT-A treatment. I had the impression that BoNT-A was a beneficial treatment, in terms of its impact on improving QoL and social and communicative participation.

Section 3: Participant Experiences

Through the audio recorded interviews and transcripts, I gained insight into each participants' personality and experiences, through their own words. I was able to understand and empathize with their struggles and improvements. Inherently, I began to compare the participants' responses to that of previous literature. I appreciated how phenomenology methods allowed for a unique understanding of each participants story, as I compared my participants remarks to former studies, which mainly acquired their results via questionnaires. Hearing the participants' stories made me realize the difficulties that they experienced. That realization further emphasised the importance of filling in the research gap in OMD. Understanding the participants' experiences of OMD, prior to injection, allowed me to gauge the improvements, or lack thereof, post-injections.

I composed a summary for each participant after I had listened to his or her interviews, to gain an overall understanding about each participant's experiences.

EP had very different results than I assumed. EP expressed that his muscles weakened after receiving injections for 22 years. This was not something I anticipated, nor would it have been something I could have understood, had I not been exposed to dysarthria in the motor speech course.

EP has been receiving BoNT-A for 22 years. EP receives BoNT-A in his eyes every 2-3 months, and gets injections in his jaw and neck. As a result of the injections, he experiences deterioration of his eye jaw and neck muscles. EP also reports bruising at the injection sites, and states that they are very painful. EP is uncertain of the effects of BoNT-A and feels as though he can no longer recognize his voice. In recent years EP has begun to have a fear choking, and is very careful when eating. BoNT-A still allows him to travel, as he schedules his injections accordingly. Without BoNT-A, EP would be very limited in his eye and jaw movements and expresses that the limitation would make him feel handicapped. BoNT-A has prevented him from slipping into thoughts aiding in lower self-esteem. BoNT-A injections allow EP to sit through a movie or have a conversation. EP wishes his muscles did not deteriorate as a result of the BoNT-A and that they would get better and be cured. EP is resilient and believes that he needs to accept the situation he is in and move on. He does not allow himself to feel sorry for the situation he is in; he tries to be happy and feels fortunate that at least he has BoNT-A. He does not feel disabled with the BoNT-A injections.

NF seemed very happy with her changes. She expressed a very positive reaction to the BoNT-A injections and often mentioned how it helped her QoL, communication and participation. Her responses were what I anticipated, based on my limited understanding of BoNT-A injections.

NF has been receiving BoNT-A for 8 years. NF receives injections in the tongue and under her chin. Injections in her tongue cause swelling and pain. NF has a tendency to bite her tongue as a result of the BoNT-A. After her first injection, she was shocked after

saying her first words, and felt as though it was a miracle. BoNT-A gave her confidence and allowed her to go back to work 8 months post-injection. She is resilient and makes light of her situation through jokes. 2 weeks Post-injection NF is careful of what to eat, as she fears choking. In that time, NF will try to eat with people around her to make sure she is not alone, in the event that she does choke. NF says that BoNT-A has allowed her to eat more than she used to without contemplating what she can or cannot eat. Her speech improved with the BoNT-A, and she believes that it makes her feel normal and allows her to laugh, express her emotions and be social. BoNT-A has had a huge impact on her communication with her family and she feels that it has taken her from disabled to abled.

JR was less expressive about his journey with BoNT-A; he discussed how the injections helped him in social situations and improved his confidence. Contrary to my assumptions, JR felt that BoNT-A did not assist in his speech production.

JR has been receiving BoNT-A injections for 3 months. JR had twitching eyes and prominent jaw and lip movements that more so affected the left side of his face than the right. These movements have calmed down after injection and as a result, he is less worried and less embarrassed about his facial movements. BoNT-A has also helped JR's driving as the BoNT-A has reduced his eye twitches. The BoNT-A treatment has not helped his speech, but JR feels that his QOL has been improved, and that the injections helped his facial appearance, although his face is now sorer. JR is now more social as a result of the BoNT-A injections. JR feels that BoNT-A has allowed him to be as normal as possible.

SP was also less expressive about his experience with receiving the BoNT-A injections. As everyone mentioned specific things they wished BoNT-A would help, SP had an overall expectation that it would help him get back to normal. Although his expectations were not met, SP did express that BoNT-A helped with communication.

SP has been receiving BoNT-A for 11 years. SP receives injections in between his eyelids, cheek, chin, lips, and jaw. SP injections sites feel sore. After his first injection, SP states that there were no improvements, but his second injection helped with his lip movements and his speech improved. His face used to draw down prior to BoNT-A, and he would have to put in a lot of effort to speak. BoNT-A helped SP with his communication, not only in the sense of stringing words together, but also in conveying meaning. BoNT-A has eliminated barriers to communication for SP. SP expected that BoNT-A would help him get back to normal, but was disappointed that it did not.

BR also seemed very happy with his BoNT-A treatment results. His resilience is one that stood out, as he constantly made reference to pushing through situations. His experiences echoed previous literature on speech intelligibility.

BR has been receiving BoNT-A for 3 years. BR receives injections in his jaw, and collar muscle. BoNT-A has helped his slurred speech and his intelligibility. BR feels that his response rate is faster after the injections and that he does not require the constant effort that he once needed. This has helped BR's phone job. BR no longer overthinks how he should answer a question. BR feels that these improvements are important to him. Following BoNT-A injections, BR notices that he has a dry mouth and has a hard time keeping food in his mouth. BR's perspective on BoNT-A is "if it works it works, if it

doesn't it doesn't". BR is very resilient. He believes that if BoNT-A did not exist he would try to find another solution.

ST had been receiving the injections for the least amount of time. She had not seen any dramatic changes in terms of speech, but did mention her improvements in participation and overall mood. The improvements in participation align with previous research findings.

ST has been receiving BoNT-A for 3 months. ST receives BoNT-A in her mouth and jaw. The BoNT-A has stopped her jaw from locking, and has allowed her to sneeze properly. ST has not found any significant changes to her speech with the injections. BoNT-A injections have allowed ST to want to go out and be with people. She feels as though she no longer has to worry and is happy with the changes the BoNT-A has provided her, it makes her feel good to see improvements. She wishes it helped her jaw a little more, but is not expecting miracles.

FI shared the most varied experiences, as she was unsure of the impact of the injections. FI felt as though the injections were altering her face in a negative manner, this was different than my expectations, as I was assuming the BoNT-A would help improve facial aesthetics. On the other hand, she also explains that facial twitching was improved post-BoNT-A injections.

FI has been receiving BoNT-A for 21 years. She receives injections around her mouth, in her neck and jaw. As a result of BoNT-A, FI experience dry mouth and loses her sense of taste a week post-injection. The injection sometimes makes her mouth open too wide or causes her mouth to be clenched closed. As a result, she finds it difficult to chew and is now eating mashed foods. FI is unsure how BoNT-A has helped and is worried that there

is not an overall plan for her injections. FI feels that there is almost a psychological effect, where she receives BoNT-A and her expectation is that something will change, whatever that change is, she is not sure. FI compares photographs of her face and feels that she looks different post-BoNT-A, that her face looks less relaxed. FI finds that her speech might be better, but her speech sounds weird now and she cannot say precisely how BoNT-A affects her speech. She thought that BoNT-A would be more helpful. FI feels like she is just getting BoNT-A for cosmetics now. FI does not feel that BoNT-A has affected her ADL and she still enjoys travelling. FI finds that BoNT-A helps her confidence for the first weeks before it wears off, she feels more in control. Also, after receiving it she is not worried as much about facial twitching in social settings and she feels she can be more personable before BoNT-A wears off.

GM, similarly to FI, also experienced negative impacts from the BoNT-A, on facial aesthetics. This increased his self-consciousness post-injections. The pain he experienced post-injection also stood out, as he described waking up with his lips stuck to his teeth, due to dry mouth, which further resulted in micro-tears alongside his inner lip. Nevertheless, he was still very positive about the benefits of BoNT-A and expressed resiliency.

GM has been receiving BoNT-A for 3 years. GM receives BoNT-A in corners of his mouth. BoNT-A helped counteract the drooping corners of his lips (making a grimacing face). As a result of the BoNT-A, GM has red marks on the corners of his mouth, and this is something that bugs him, and something that he notices and feels that everyone notices as well. GM does not want to look like the Joker. GM does feel that the BoNT-A makes things better and helps ease the effort it takes to speak. GM is an optimistic person and

does not think BoNT-A has affected his thinking about life or philosophy. GM is resilient and tries not to worry about things if they are not life or death. Although BoNT-A affects his facial appearance, he finds that his response rate is faster and states that this is more important to him. With BoNT-A he is not afraid to talk, and he feels more confident in conversations, whether one-on-one or in a group. He also does not have to exert a lot of effort when speaking. GM does find that the BoNT-A affects his eating and his mouth tends to dry out. He is cautious about his appearance and finds it hard to get food off his teeth when he is eating, as he does not want to have his tongue sticking out of his mouth, so he makes sure he always has water and is eating in a certain way. GM feels that BoNT-A gave him confidence and that it is a “Godsend”. GM wishes that the BoNT-A would help with controlling his mouth from moving around. BoNT-A has changed his life and he would be afraid if it was banned.

Section 4: Code Composition

I have developed codes on Dedoose for each transcript. I highlighted the results related to the treatment of OMD with BoNT-A. I extracted excerpts from the transcripts, and correlated each participants experience into a code. Everything the participants mentioned about the BoNT-A injections was coded. Common concepts were continuously emerging, alongside varying perspectives and experiences.

Participants discussed their communication in private and in public settings and their ability to hold a conversation and adequately express their emotions. I empathized with that concept of struggling to relay emotions and the relief of being able to do so post-injections. This was not something I read about or considered as an effect post-injection. There were a lot of discrepancies between subjects. I noticed that some were even experiencing discrepancies

throughout their own treatment experiences, e.g. a participant may express that at time speech was improved with BoNT-A, and then the same participant would express that speaking was difficult or does not “correct” everything.

Facial alterations were also another topic being raised, again there were varying responses, which I had not considered. I had not read about negative changes to aesthetics throughout my literature review. I had these assumptions that BoNT-A would help dystonic movements, rather than cause facial alterations, such as GM experienced. Feeling tired was another concept that was raised, in terms of effortful speaking or lack of effort post-injections. The number of injections received, also varied between participants and the struggle with perfecting dosages and time of dosage was a problem that was raised, as well as where the participants were receiving the injection. Some were receiving it in the tongue, jaw, neck, etc.

I also included a coding component for good stories. Participants were expressing some positive or negative stories that they have gone through post-injections. The process of phenomenology research allowed me to be able to incorporate these findings in my data collection and this improved my understanding of the patient’s experiences and struggles, through their own words. The nature of phenomenology research allowed me to gain a better understanding of the participants experiences’ with pain. Unlike other research methodologies, phenomenology allowed the participant to elaborate on their experiences. Previous research, used questionnaires as a primary method of data collection, which unfortunately did not provide participants the opportunity to elaborate on their responses.

Understanding the perceptives of each participant was incredibly eye-opening. Although some were taking BoNT-A for a long time, FI in particular seemed uncertain of her improvements post-injections. It was interesting to see how different her reaction was to the

treatments in contrast to NF, who had an overwhelmingly positive experience. Also, how participants viewed themselves post-injections was a new concept. NF and JR both expressed feeling normal post-injections, which was a concept I had not encountered in the literature.

The physical effects and impact of BoNT-A was something I was familiar with. The participants echoed some of what I had learned about BoNT-A and OMD. The psychological effects were also fascinating and echoed some of the previous research on this topic. Each emotional outcome experienced by the participants was very eye-opening. The unfortunate part was learning about the negative emotional experiences, which were a result of expectations that were not met.

The social aspects raised were also ones that I encountered throughout previous papers. They were similar in regards to the improved impact that BoNT-A had on social participation.

Speech improvement was another concept raised. The participants' responses sometimes echoed previous findings, but other times were the complete opposite. I did not expect many negative responses, but with the motor speech course and previous literature, I could appreciate how the participants could have negative responses. The participants positive outlook persisted, even with the negative side effects.

Initially, these concepts produced 15 codes. Through feedback from my supervisor and advisory members, I narrowed the 15 down to 11 codes with sub-codes. 1) Regarding injections, which included the frequency, site and physical effects (effect on muscle, and wearing off), 2) Eating and drinking with sub-codes on mechanics and physical effects of eating, social aspects of eating and strategies that improved eating. 3) Facial aesthetics, 4) Good story, 5) Job, 6) Pain, with sub-codes on pain from injection and pain post-injection. 7) Personal perspective (how participants viewed themselves) 8) Psychological effect, 9) Resilience, 10) Social aspects with

sub-codes on participation in social activities, perceptive of others, and relationships, lastly, 11) Speech with sub-codes, which included communication, fatigue, intelligibility and speech production.

Section 5: Creation of Themes

I began to group the codes into themes that reflected the participants' struggles and improvements post-injections. I struggled with making the themes, as I wanted to make sure I completely captured all the participants' experiences and relayed an accurate depiction of what the participants expressed.

Excerpts that touched on speech and communication were grouped together. The speech aspect tied into the social aspect of communication and participation, which included changes in participants speech, both mechanic changes and intelligibility changes and the social and emotion effect this had on participants. Sub-theme regarding changes in participants speech, such as response rate, intelligibility, articulation and pronunciation, which either improved or stayed the same post-BoNT-A injections, were also included. The second sub-theme was on participation and changes in participation that were experienced. Participants described their ability to interact in social settings (religious settings, friend and family gatherings), and ability to participate in their jobs and daily activities, post-BoNT-A injections.

Another theme was based on perceptive. Participants shared their perspectives on how BoNT-A affected them emotionally, as well as its effect on their perception of themselves and how other perceived them. The first sub-theme was the concept of feeling "normal" and less disabled post-injection, the other sub-themes were on the positive psychological effects, and lastly, the negative psychological effects. The third theme explored how BoNT-A changed the participants' experiences. This included the everyday experiences that changed as a result of the

injections, and how these changes affected participants QOL and day-to-day activities. The sub-themes were eating is not the same, which included topics on choking and food consistency and dry mouth (I tied in a lot of the physical effects here), another sub-theme was face alteration, both positive and negative changes. The next sub-theme was the experience of pain and finally, the last sub-theme was effort and fatigue.

After consulting my supervisor and advisory members, we narrowed down the themes and reorganized them to better represent the psychosocial impact of BoNT-A. The themes were reorganized, the first **Botox has changed me**, The sub-theme *I feel normal again* (tying in their perspective of themselves), then the sub-theme *I feel appreciative of what Botox does for me* (the positive impact), and finally, *Botox doesn't fix everything* (the negative impact). The next theme **What communication is like for me**, the first sub-theme *My speech has changed*, and the second sub-theme, *I can now participate*. The third theme **Botox has changed my experiences**, which incorporated the sub-theme *My eating has changed*, and *My face looks different, I am experiencing pain, I am experiencing fatigue*.

After another review of the themes, pain and fatigue were incorporated into the previous sub-themes, e.g. pain in receiving injections was a negative experience or a positive experience, (pain was relieved post-injections). This sub-theme was included in the theme, **Botox has changed me and my experiences** and sub-theme, *I feel appreciative of what Botox does for me* and the sub-theme *Botox doesn't fix everything*. Also, fatigue, as a result of injection, was always discussed in the context of speech. This was then incorporated into that theme, **What communication is like for me** and the sub-theme *My speech has changed*.

The final consensus for the themes was to remove the sub-theme regarding eating, as it was focused on physical effects, rather than psychosocial effects of BoNT-A injections. Then the

sub-theme regarding facial aesthetics (*My face looks different*) was placed within theme **Botox has changed me and my experiences**, alongside the sub-theme, *I feel appreciative of what Botox does for me*, and *Botox doesn't fix everything*. The second theme was, **What communication is like for me** with the sub-themes *My speech has changed* and *I can now participate*. The excerpts corresponding to each theme were grouped and expanded on and explained in the results section of this paper.

Section 6: Comparison of Findings to Previous Literature

The discussion process was more difficult than I anticipated. I struggled with the organization and logistical flow, of comparing the results. I grouped together all the research articles that discussed each section of the results and compared the findings. There were no research articles that discussed the concept of feeling “normal” post-injection or papers on the negative changes to facial aesthetics. These findings were difficult to find in the research on OMD, as well as research in other types of motor speech disorders. The discussion section of this paper elaborated on other speech disorders to provide a wholesome understanding of the psychosocial impact of BoNT-A. Similar research on speech disorders allowed for a comprehensive understanding of the psychosocial impact of BoNT-A, that previous participants expressed.

The discussion section of this paper verified the importance of conducting further research in the area of OMD and BoNT-A, and the importance of incorporating the participants' experiences, almost as decision-makers, in future research to further improve assessments and treatment measures. I feel extremely lucky to be provided the opportunity to conduct research in the topic of OMD and add to the ever growing research in this field. The participants' stories and lived experiences were a pleasure to listen to.

Curriculum Vitae

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Post-secondary Education and Degrees: Western University of Canada
London, Ontario Canada
2011-2016 BHSc–Honours Specialization in Health Science

Honours and Awards: Western Graduate Research Scholarship
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Dean’s Honour List: Faculty of Health Sciences, *Western University*
2016

The Western Scholarship of Excellence
2011

Aiming for the Top Tuition Scholarship
2011

Work Experience: Research Assistant
Parkwood Institute-Cognitive Clinical Trials
2018-Present

Research Assistant
Western University- Autism and Speech Language Pathology Lab
2016-2016

Research Assistant
Western University- Human Environments Analysis Laboratory (HEAL)
2013-2016

Teaching Assistantship- (CSD 9635)
Western University- Counselling for Speech-Language Pathologists
2017-2018

Teaching Assistantship- (CSD 4411)
Western University- Introduction to Speech and Language Disorders
2017-2018

Teaching Assistantship- (HS 4620)
Western University- Foundations of Mental Health
2017-2017

- Volunteer Experience:**
- Outreach Volunteer
Parkwood Institute-Penny Welch-West (SLP)
2018-Present
 - Outreach Volunteer
Helping Children Talk-Meggan Levson (SLP)
2016-2017
 - Research Assistant
Western University-Janis Cardy (SLP)
2016-2017
 - Outreach Volunteer
Rejuvenate Health Services-Elizabeth Mackinnon (SLP)
2016-2017
 - Outreach Volunteer
Frontier College-Literacy Program
2016-2017
 - Outreach Volunteer
Heart and Stroke Foundation
2013-2016
- Undergraduate Thesis:**
- Honours BHSc.
Spinal Cord and ABI Rehabilitation- Dr. Dalton Wolfe
- Presentations:**
- Smyth, R.E., Elhayek, N., Archibald, L., Johnson, A.M., & Oram Cardy, J. (2017, June). Language development and processing speed in young children: A domain general or domain specific relation? Symposium on Research in Child Language Disorders. Madison, WI, United States of America.
- Elhayek, N., Page, A.D., & Jog, M. (2018, February). Examining the Psychosocial Impacts of Botulinum Toxin Type-A Injections on Individuals with Oromandibular Dystonia: A Qualitative Study of Patients' Experiences. Health and Rehabilitation Science Graduate Research Conference. London, ON, Canada.
- Page, A.D., Elhayek, N., & Adams, S. (2018, November). Examining the Health-Related Quality of Life of Individuals with Oromandibular Dystonia receiving Botulinum Toxin Therapy American Speech-Language-Hearing Association. Boston, MA, United States of America.
- Elhayek, N., Page, A.D., & Jog, M. (2019, February). Examining the Psychosocial Impacts of Botulinum Toxin Type-A Injections on Individuals with Oromandibular Dystonia: A Qualitative Study of Patients' Experiences. Health and Rehabilitation Science Graduate Research Conference. London, ON, Canada.

Borrie, M., Best, S., Fogarty, J., Bassi, N., Di Prospero, C., Elhayek, N., Whitehead, V., Pillon, R., Phillips, N., Chertkow, H. (2019, April). The Canadian Collaboration on Neurodegeneration and Aging – Platform 1 – COMPASS-ND Study-Planning and Implementation. London Health Research Day. London, ON, Canada.

Page, A.D., Elhayek, N., Baylor, C., Adams, S.G., Yorkston, K. (2020, March). Exploring The Psychosocial Impact Of Botulinum Toxin Injections For Individuals With Oromandibular Dystonia: A Qualitative Study Of The Patients' Experiences. Paper submitted for presentation at the International Conference on Motor Speech, Santa Barbara, CA, United States of America.

Borrie, M., Best, S., Fogarty, J., Bassi, N., Di Prospero, C., Elhayek, N., Whitehead, V., Pillon, R., Phillips, N., Chertkow, H. (2019, October). The Comprehensive Assessment of Neurodegeneration and Dementia (COMPASS-ND) study – Recruitment Update. Canadian Conference on Dementia. Québec City, QC, Canada.

Best, S., Wong, T., Elhayek, N., Borrie, M., Dasgupta, M., Rockwood, K., Davis, D., O'Regan, N. (2019, October). The Comprehensive Assessment of Neurodegeneration and Dementia (COMPASS-ND) study – History of Delirium and Prevalence Cases (Case Finding at Parkwood Institute). Canadian Conference on Dementia. Québec City, QC, Canada.