

Electronic Thesis and Dissertation Repository

7-15-2016 12:00 AM

The Consequences of Oromandibular Dystonia on Communicative Participation: A Qualitative Study of the Insider's Experiences

Lauren Siegel

The University of Western Ontario

Supervisor

Dr. Allyson Dykstra

The University of Western Ontario

Graduate Program in Communication Sciences and Disorders

A thesis submitted in partial fulfillment of the requirements for the degree in Master of Science

© Lauren Siegel 2016

Follow this and additional works at: <https://ir.lib.uwo.ca/etd>



Part of the [Communication Sciences and Disorders Commons](#)

Recommended Citation

Siegel, Lauren, "The Consequences of Oromandibular Dystonia on Communicative Participation: A Qualitative Study of the Insider's Experiences" (2016). *Electronic Thesis and Dissertation Repository*. 3925.

<https://ir.lib.uwo.ca/etd/3925>

This Dissertation/Thesis is brought to you for free and open access by Scholarship@Western. It has been accepted for inclusion in Electronic Thesis and Dissertation Repository by an authorized administrator of Scholarship@Western. For more information, please contact wlsadmin@uwo.ca.

Abstract

The purpose of this study was to explore the consequences of oromandibular dystonia (OMD) on communicative participation from the insider's perspective. Qualitative research methods were used to obtain a self-reported account of the experience of living with OMD. Eight individuals with OMD and dysarthria participated in face-to-face phenomenological interviews. Interviews were transcribed from audio recordings and coded using coding software. The codes were then grouped into larger thematic categories based on salience. Results showed that communicative participation is affected by multiple physical, social, and emotional factors caused by OMD. Furthermore, OMD can have significant effects on an individual's job, family, and social life. Lastly, strategies and coping mechanisms used by participants were explored. This study will add to very sparse literature on OMD and will help to reveal the complexity of living with this disorder.

Keywords: oromandibular dystonia, communicative participation, motor speech disorders, phenomenology, dysarthria, hyperkinetic dysarthria

Acknowledgments

First and foremost, I would like to express my gratitude to my supervisor Dr. Allyson Dykstra for introducing me this fascinating area of research, for her constant encouragement, and guidance throughout the duration of this project. This thesis project would not have been possible without her abundant patience and support. I would also like to thank my advisory committee, Dr. Scott Adams and Dr. Carolyn Baylor, for generously providing their mentorship and expertise. Their contribution has been instrumental to this project.

To my fellow Masters/Doctoral candidates in the speech and language sciences field, Carlee Wilson and Cynthia Mancinelli, thank you for their friendship, support, and encouragement. I am proud and grateful to have shared this journey with you both.

A special thank you to my roommates and friends, and to my boyfriend Daniel Martire. This accomplishment would not have been possible without their unconditional love and support.

Lastly, I would like to thank my family, especially my mother, Cheryl Siegel, for always believing in me and constantly encouraging me to pursue my goals.

Table of Contents

| | |
|----------------------------------------------|-----|
| Abstract..... | i |
| Acknowledgments | ii |
| Table of Contents | iii |
| List of Tables | iv |
| List of Appendices..... | iv |
| Chapter 1 | 1 |
| 1 Introduction..... | 1 |
| 1.1 Dystonia | 1 |
| 1.2 Oromandibular Dystonia..... | 2 |
| 1.3 Pathophysiology of OMD | 3 |
| 1.4 Epidemiology of OMD | 4 |
| 1.5 Clinical Features of OMD | 4 |
| 1.6 Speech Intelligibility | 6 |
| 1.7 Treatment of OMD | 7 |
| 1.8 Communicative Participation..... | 11 |
| 1.9 Rationale for Current Study | 19 |
| Chapter 2 | 19 |
| 2 Method | 19 |
| 2.1 Participants..... | 19 |
| 2.2 Research Approach | 21 |
| 2.3 Procedure | 22 |
| 2.4 Credibility | 24 |
| 2.5 Verifiability | 25 |
| Chapter 3 | 26 |
| 3 Results | 26 |
| 3.1 Theme 1: Speaking is Different Now | 27 |
| 3.2 Theme 2: My Roles Have Changed..... | 30 |
| 3.3 Theme 3: I Accept It and Move On | 33 |
| Chapter 4 | 35 |
| 4 Discussion..... | 35 |
| 4.1 Speech Production..... | 36 |
| 4.2 Roles | 37 |
| 4.3 Coping Strategies..... | 39 |
| 4.4 Limitations of the Current Study | 40 |
| 4.5 Future Directions | 41 |
| 4.6 Clinical Implications | 42 |
| Chapter 5 | 44 |
| 5 Summary and Conclusions..... | 44 |
| References | 46 |
| Curriculum Vitae | 61 |

List of Tables

| | |
|--------------------------------------------------------------------------------------------|-----------|
| Table 1: Demographic information of participants with OMD..... | 20 |
| Table 2: Themes and sub-themes describing the consequences of living with OMD | 26 |

List of Appendices

| | |
|------------------------------------------------|-----------|
| Appendix A: Letter of Information..... | 53 |
| Appendix B: Consent Form | 56 |
| Appendix C: Ethics Approval Notice..... | 57 |
| Appendix D: Coding Dictionary | 59 |

Chapter 1

1 Introduction

1.1 Dystonia

Dystonia belongs to a category of neurological movement disorders characterized by random and unpredictable movements that can vary in speed, duration, and amplitude. These movements may be present at rest, during sustained postures, or during activity (Brin & Comella, 2004; Duffy, 2013). These dystonic movements usually stop during sleep and are exacerbated by stress and heightened emotion (Duffy, 2013; Scott, 2000). Dystonic movements range from mild to severe, and may be strong enough to interrupt or alter the direction of intended movement (Duffy, 2013; Tarsy & Simon, 2006). Dystonia results from excessive co-contraction of agonist and antagonist muscles (Tarsy & Simon, 2006). Abnormalities appear to stem from impairment of the basal ganglia, cerebellum, and dopaminergic system (Duffy, 2013; Lee, 2007).

Dystonia may be either primary or secondary. Dystonia is classified as primary if it occurs in the absence of other clinical symptoms, or if it is inherited. Dystonia is classified as secondary when it occurs in association with another known disease (Scott, 2000; Tarsy & Simon, 2006). Dystonia can develop at any point in an individual's lifetime, and can be classified as either early onset or late onset. Early onset dystonia occurs in childhood, before the age of 20. Late onset dystonia occurs in adulthood, after the age of 20 (Defazio, Abbruzzese, Livrea, & Berardelli, 2004). Dystonia is most prevalent among older adults, with an average age of onset of 66 years, and it is more

common in females than males (Lee, 2007). 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

Oromandibular dystonia (OMD) is a type of focal dystonia affecting the muscles of the lips, tongue, and/or jaw, which can impact speech production (Duffy, 2013). It is characterized by involuntary, repetitive, sustained, and sometimes painful muscle contractions (Clark, 2003; Duffy, 2013). There are six types of OMD: jaw closing dystonia, jaw opening dystonia, jaw deviation dystonia, lip dystonia, lingual dystonia, or a combination of any of these. When OMD occurs with blepharospasm (involuntary contractions of the eyelids) it is called Meige's syndrome (Cardoso & Jankovic, 1995). For intended movements, muscle contraction may be disproportionate, or contraction may spread to muscles not normally involved. Secondary contractions may also impede the action of the primary muscles (Cardoso & Jankovic, 1995). OMD may cause difficulties with the opening and/or closing of the jaw, speech, and/or chewing and swallowing. Facial grimacing, lip pursing or retraction, and protrusion or rotary movements of the tongue may be present (Duffy, 2013). The oral mechanism is often normal in size, strength, and symmetry, and pathologic reflexes are usually absent (Duffy, 2013). Patients commonly present with drooling and dysphagia, and complain of food sticking in the throat or difficulty chewing (Duffy, 2013; Tarsy & Simon, 2006).

1.3 Pathophysiology of OMD

As in many neurological conditions, the exact cause of OMD is largely unknown. OMD appears to reflect problems with sensorimotor integration, and so permits inferences about the role of the basal ganglia control circuit, specifically the putamen, cerebellum, and dopaminergic system (Duffy, 2013; Papapetropoulos & Singer, 2006; Tarsy & Simon, 2006). Impulses from the basal ganglia have an inhibitory effect on the thalamus, impeding cortical neuronal firing. Hyperkinetic disorders, such as dystonia, result from the absence of physiological inhibitory control of the basal ganglia over the cortical neuronal firing of the thalamus and brain stem (Duffy, 2013; Papapetropoulos & Singer, 2006). Structural imaging techniques of muscles affected by dystonia have shown a pattern of involuntary rapid firing of fibres, unlike normal firing of muscle electrical activity (Papapetropoulos & Singer, 2006).

Causes of OMD may include trauma to the head and central nervous system, diencephalic stroke, or neurodegenerative disorders such as Parkinson's disease, multiple sclerosis, or Huntington's disease. (Dworkin, 1996; Lai & Jankovic, 1998; Tinter & Jankovic, 2002). Drug-induced dystonia may be caused by levodopa, dopamine agonists, antipsychotic drugs, anticonvulsant agents, serotonin-reuptake inhibitors, and rarely by other miscellaneous drugs (Tarsy & Simon, 2006). Exposure to certain chemicals such as manganese, carbon monoxide, or carbon disulfide may also result in dystonia (Tarsy & Simon, 2006). There is evidence to suggest a genetic component in the appearance of OMD. Genes DYT1 through DYT13 have been associated with dystonia (Klein & Ozelius, 2002; Tarsy & Simon, 2006). These genes include autosomal dominant, autosomal recessive and X-linked causes of primary and secondary dystonia

(Tarsy & Simon, 2006). Specifically, DYT6, DYT7, and DYT13 have been linked to incompletely penetrant, autosomal dominant focal dystonias (Tarsy & Simon, 2006). Occasionally mutations in the torsin A gene at the DYT1 locus have also been identified in patients with adult-onset focal dystonia (Tarsy & Simon, 2006). In some cases, there may be a family history of dystonia (Tarsy & Simon, 2006).

1.4 Epidemiology of OMD

OMD is a rare condition, and the exact number of cases is largely unknown. Many general physicians are unfamiliar with dystonia, and as a result many cases are undiagnosed or misdiagnosed. Patients often have to consult several physicians before their dystonia is correctly identified (Tarsy & Simon, 2006). Common misdiagnoses include temporomandibular joint syndrome, myasthenia gravis, dental malocclusion, and edentulous movements (Tarsy & Simon, 2006). The estimated prevalence of OMD is 68.9 cases/million persons (Nutt, Muentner, Aronson, Kurland, & Melton, 1988). Comparatively, the prevalence of all focal dystonias is approximately 300 cases/million persons (Brin & Comella, 2004). The estimated incidence of OMD is 3.3 cases/million persons (Nutt et al., 1988).

1.5 Clinical Features of OMD

The involuntary spasms associated with OMD can have devastating consequences on the daily functioning of those affected. OMD most commonly causes involuntary clenching, opening, or deviation of the jaw, with muscles of the mouth, tongue, or neck also frequently involved (Tarsy & Simon, 2006). The altered orofacial aesthetics associated with OMD, such as jaw deviation and involuntary tongue protrusions, can lead

to feelings of embarrassment and reduced self-confidence (Lee, 2007). OMD has been linked to depression and a reduction in quality of life (Bakke, Larsen, Dalager, and Moller, 2013). Severe cases may cause jaw pain, difficulty chewing, dysphagia, and dental trauma (Tarsy & Simon, 2006).

A study by Papapetropoulos and Singer (2006) found that dystonic eating dysfunction may complicate OMD leading to weight loss. In a cohort of 53 patients with primary and secondary OMD, there was a 15.6% prevalence of eating dysfunction. Over half of the patients with eating dysfunction also reported significant weight loss. Eating dysfunction associated with OMD may include pain during eating and further social embarrassment when accompanied by drooling or choking (Papapetropoulos & Singer, 2006). OMD may also cause oral trauma. Jaw-closing dystonia most often results in trauma, consisting of persistent grinding of the teeth or biting of the tongue, wearing of the enamel or early loss of teeth, trauma of the lips or gums, or persistent jaw pain (Bakke et al., 2013). Dystonic activity may also cause head and jaw pain, muscular tension or tiredness, and a reduced salivary flow rate (Bakke et al., 2013). Involuntary closing of the jaw may produce inappropriate deviation of the mandible, partial dislocation, intramural soft tissue trauma, and bone resorption (Gandhi, 2010).

In addition to altered orofacial esthetics, orofacial pain, difficulty with chewing, and dental trauma, OMD can also result in a communication disorder called dysarthria. Dysarthria is a collective name for a group of neurologic speech disorders that reflect abnormalities in the strength, speed, range, steadiness, tone or accuracy of movements required for the respiratory, phonatory, resonatory, articulatory, or prosodic aspects of speech production (Darley, Aronson, & Brown, 1969b; Duffy, 2013). According to

Enderby and Emerson (1996), dysarthria is the most commonly acquired disorder of communication. Darley, Aronson, and Brown (1969a, 1969b) created a perceptual classification scheme of dysarthria based on the location of lesion and associated deviant speech dimensions. Following this classification, dystonia is a hyperkinetic dysarthria and an impairment of the extrapyramidal system. Unlike other dysarthrias based on the central nervous system, hyperkinetic dysarthria can result from abnormal movements at only one level of the speech system, for example articulation, or a few muscles at that level, for example the muscles of the lips (Duffy, 2013). Hyperkinetic dysarthria accounts for 19% of motor speech disorders according to evaluations among 8101 people studied at the Mayo clinic from 1993 through 2008 (Duffy, 2013).

Darley et al. (1969b) presumed the neuromuscular deficits associated with the dysarthria of dystonia to cause slow, involuntary movements, with irregular rhythm, reduced range, and excessive tone. Darley et al. (1969b) found the most deviant speech dimensions of dystonia to be: imprecise consonant articulation, vowel distortion, harsh voice quality, irregular articulatory breakdown, strained-strangled voice quality, monopitch, monoloudness, vocal tremor, alternating loudness, and voice stoppages. In this original classification of dystonia, spasmodic dysphonia (hyperadduction of abductor or adductor laryngeal muscles) was included in the description of dystonia. Therefore, the laryngeal impairments described by Darley et al. (1969b) in the description of dystonia such as a harsh voice, strained-strangled vocal quality, voice stoppages, and vocal tremor, should not be included in the perceptual features of the dysarthria associated with OMD.

1.6 Speech Intelligibility

The hyperkinetic dysarthria associated with OMD can result in reductions in speech intelligibility (Dykstra, Adams, & Jog, 2007). Speech intelligibility is defined by Yorkston, Strand and Kennedy (1996) as the “degree to which the acoustic signal is understood by the listener” (p. 55). More broadly, speech intelligibility can be viewed as the “understandability of speech” (Kent, 1992; Yorkston, Dowden, & Beukelman, 1992). For individuals experiencing OMD, involuntary muscle spasms of orofacial musculature may affect the understandability of their speech. Weismer, Yunusova and Bunton (2012) suggest that the tongue is the most influential articulator in speech intelligibility. Weismer and colleagues also assert that tongue control may be more strongly related to speech intelligibility in individuals with neuromotor pathology than lip/jaw control. Assessing speech intelligibility allows researchers and clinicians to categorize dysarthria by severity level, provides a quantitative way to monitor speaker performance during the course of treatment and recovery, and creates a universal way to relay progress amongst other clinicians, to the speaker, or to his/her family members (Yorkston & Beukelman, 1978).

1.7 Treatment of OMD

Since there is no known cure for OMD, clinical focus is placed on various treatments and strategies for reducing dystonic symptoms and improving quality of life.

Pharmacological. Pharmacologic agents used to treat OMD include: anticholinergics (e.g. trihexyphenidyl, benztropine), benzodiazepines (e.g. clonazepam, lorazepam, diazepam), baclofen, and tetrabenazine, which deplete dopamine and block dopamine receptors (Tintner, & Jankovic, 2002). Although these drugs have been found useful in some patients, it is not the preferred form of treatment for focal dystonias since

there is only modest improvement and frequent side effects (Tintner, & Jankovic, 2002). Medications are most effective in cases of widespread dystonia. One of the most well tolerated medications for treating OMD is clozapine, a dibenzodiazepine that differs from conventional neuroleptics in its interactions with neurotransmitter receptors and in its range of side effects (Karp, Goldstein, & Chen, 1999). The pathophysiology of dystonia is thought to involve dysfunction of dopaminergic pathways in the basal ganglia. Clozapine acts on the dopaminergic pathways by binding to, and increasing, D1 dopamine receptor density in the basal ganglia, which reduces the over-activity of the direct pathway (Karp et al., 1999).

Sensory. A sensory trick, or *geste antagoniste*, is a method some patients with OMD use to temporarily relieve their dystonic symptoms (Duffy, 2013). A sensory trick can include such acts as holding an object in the mouth, chewing gum, or touching a certain area of the face, such as the chin, cheek, or eyelid. Sensory tricks may help patients to speak and chew. The mechanism behind sensory tricks is unknown (Duffy, 2013).

Prosthetic. Based on the premise of sensory tricks, various devices have been used in the treatment of OMD. A commonly used device for treating jaw or lingual types of dystonia is a bite-block. A bite-block is a device that is tailored to the individual patient, and placed between the upper and lower teeth to help with jaw stability and positioning. Bite-block therapy has been shown to improve facial appearance, articulatory precision, and hyperactive movements in hyperkinetic dysarthrias (Dworkin, 1996; Goldman & Comella, 2003). Dworkin (1996) found that insertion of a bite-block in two individuals with OMD secondary to Meige's syndrome resulted in immediate conversion

from moderately reduced intelligibility to near normal speech intelligibility in both individuals. It was hypothesized that the bite-block neutralized dystonic activity by facilitating postural balance and motor stability of the mandible. A bite-block may prevent enamel wear, decrease the load on oromandibular joints, and improve chewing function (Blanchet, Rompre, Lavigne, & Lamarche, 2005). In addition to stabilizing the jaw and reducing dystonic movement, a bite-block may provide sensory information for the correct placement of other articulators (Lee, 2007). Although these techniques are not long-lasting, they are noninvasive and may aid other forms of therapy.

Botulinum toxin. The most contemporary and well-tolerated method of treatment for OMD is localized injection of botulinum toxin into the affected muscles (Goldman & Comella, 2003; Munchau & Bhatia, 2000; Ramachandran & Molloy, 2012). Botulinum toxin is a protein produced by the anaerobic bacterium *Clostridium botulinum* (Simpson, 1981). There are seven immunologically distinct toxins, however only types A, B, and E have been used in humans. Botulinum toxin type A (BoNT-A) is the most effective type in the treatment of OMD (Kazerooni & Broadhead, 2015). BoNT-A causes temporary paralysis by blocking the presynaptic release of acetylcholine at the neuromuscular junction (Simpson, 1981). After binding to presynaptic cholinergic nerve terminals, BoNT-A decreases the frequency of acetylcholine release (Avila, Drachman, & Pastronk, 1989). Paralysis occurs within a few hours post-injection (Kao, Drachman, & Price, 1976). Within 2 days after injection, the axon terminal begins to sprout and form new synaptic contacts on the adjacent muscle fibers. For this reason, the effects of BoNT-A are limited in duration. The effect of BoNT-A lasts 3 months on average, after which another injection is required (Simpson, 1989).

Botulinum toxin was introduced as a therapeutic agent for OMD in 1977 (Blitzer & Sulica, 2001). The muscles most commonly implicated in jaw opening or closing dystonia are: the masseters, temporalis or internal pterygoids; the submental muscles or external pterygoids; and the genioglossus and hypoglossus muscles in tongue protrusion dystonia (Tintner & Jankovic, 2002). A variety of methods can be used to determine the proper injection sites. EMG may be used to monitor the muscles that show increased activity during the particular abnormal movement or posture (Tintner & Jankovic, 2002). In the case of OMD, it is difficult to determine all the muscles involved since EMG recordings are not completely accurate, and the pattern of muscle involvement may change over time (Tintner & Jankovic, 2002). For this reason, injection site and dose may vary for each appointment. Injection of BoNT-A into the masseter, temporalis, and lateral pterygoid muscles results in reduction of spasm, and improvement in chewing and speech, in approximately 70% of patients with OMD (Jankovic, Schwartz, & Donovan, 1990). Side effects associated with BoNT-A include: weakness, mild dysarthria, and difficulty chewing and swallowing (Goldman & Comella, 2003).

There are currently three Food and Drug Administration approved BoNT-A products available: onabotulinumtoxinA (Botox[®], Allergan), abobotulinumtoxinA (Dysport[®], Ipsen Biopharmaceuticals), and incobotulinumtoxinA (Xeomin[®], Merz Pharmaceuticals) (Kazerooni & Broadhead, 2015). A cost-utility analysis found that Xeomin[®] was the most cost-effective BoNT-A product compared to Botox[®] and Dysport[®] (Kazerooni & Broadhead, 2015). Botox[®] for non-cosmetic use is covered by most health insurance plans.

Behavioural. Behavioral techniques can be a useful addition to any of the above treatments of OMD. Speech-language pathologists (SLPs) focus on providing behavioral therapy to improve the coordination of articulators (Goldman & Comella, 2003). SLPs may also provide management of masticatory and swallowing capabilities (Goldman & Comella, 2003; Duffy, 2013). An SLP may also suggest dyadic strategies to improve communication between the speaker and his or her communication partners. Strategies can include: getting the listener's attention before speaking, making the context and topic clear, maintaining eye contact, using gestures whenever possible, and modifying the communicative environment to improve visual and auditory acuity (Duffy, 2013). Listeners can also play a supporting role by listening attentively and actively to the speaker, and informing the speaker immediately of any misunderstandings (Duffy, 2013).

Unfortunately, little attention has been given to the impact of these various treatment approaches on speech intelligibility. There is a small empirical literature suggesting improvements to articulatory precision following the use of a bite-block (Dworkin, 1996), and there is preliminary evidence suggesting that, depending on the type and location of OMD, there may be an improvement to speech intelligibility following BoNT-A injections (Dykstra et al., 2007; Dykstra, Domingo, Adams & Jog, 2015). It is unfortunate that there is such a small empirical literature on speech intelligibility in this clinical population since dysarthria is often a disabling feature of OMD and has the potential to result in psychosocial consequences such as reductions in communicative participation.

1.8 Communicative Participation

The characteristics of OMD can be further organized according to the World Health Organization's International Classification of Functioning, Disability, and Health (ICF) (WHO, 2001). The ICF provides a conceptual framework of disability from a biopsychosocial perspective. The ICF asserts "Health is a state of complete physical, mental and social well-being, and not merely the absence of disease or infirmity" (WHO, 2001). The ICF defines 'impairment' as a 'problem in body function or structure,' 'activity' as the 'execution of a task or action by an individual,' and 'participation' as 'involvement in life situations' (WHO, 2001). Eadie, Yorkston, Klasner, Dudgeon, Dietz, Baylor, Miller, and Amtmann (2006) extended the ICF definition of participation to communication by defining 'communicative participation' as 'taking part in life situations where knowledge, information, ideas or feelings are exchanged'. Communicative participation encompasses many life situations including: personal care, household management, leisure, learning, employment, relationships, and community life (Eadie et al., 2006; Yorkston, Baylor, Dietz, Dudgeon, Eadie, Miller, & Amtmann, 2008).

The ICF model also includes environmental (external) and personal (internal) contextual factors that contribute to the consequences of a health condition. Historically, clinical focus has taken an impairment-based perspective to treating communication disorders (Eadie, 2001; Threats, 2000). This is probably because it is easier to measure biomedical aspects, such as physiological functioning, rather than psychosocial aspects of health. Recently, there has been a shift in attention to the psychosocial aspects of health across the field of healthcare in general as well as in the field of communication disorders.

Garcia, Laroche, and Barrette (2002) explored barriers to work integration for individuals with a variety of communication disorders, including those with dysarthria. Findings of this study showed many barriers to employment as a result of these communication disorders, such as the attitudes of communication partners, noise level, phone use, group discussions, and the need for rapid communication. Baylor, Burns, Eadie, Britton, and Yorkston (2011) examined self-reported communicative participation in everyday speaking situations across a wide range of communication disorders including spasmodic dysphonia (SD), stroke, multiple sclerosis (MS), Parkinson's disease (PD), laryngectomy, amyotrophic lateral sclerosis (ALS), and stuttering. Participants described many functional and emotional interferences to communicative participation, including difficulty maintaining adequate loudness, lack of expressiveness, imprecise articulation, difficulty keeping up with conversations, and difficulty speaking in noise and on the phone. Participants also reported using various strategies to improve their intelligibility, including speaking louder, simplifying speech, using different modalities, relying on family and friends, and educating others.

Baylor, Yorkston, and Eadie (2005) took a qualitative approach to explore the biopsychosocial consequences of spasmodic dysphonia (SD). Six adults with SD participated in face-to-face phenomenological interviews. The results of this qualitative study created a model of personal experiences of SD that suggested that communication-related quality of life is shaped by experiences with multiple physiologic, personal, and social factors. For example, participants reported feeling self-conscious, embarrassed, frustrated, less intelligent, less confident, and less competent (Baylor et al., 2005). The attitudes and reactions of other people contributed to these feelings and restricted

participation. Participants stated that comments were often made towards them about their lack of ability based on the sound of their voice. They felt that with the onset of their disorder, they were suddenly viewed as unskilled or unintelligent, when they were actually quite capable and qualified. Participants felt that aside from their close families and friends, people did not understand their disorder, and lacked the patience to communicate with them. Unfamiliar people were quick to make false judgments, and were less accommodating of communication difficulties. Participants adopted strategies such as planning ahead, avoiding difficult situations, and maintaining a good attitude.

Research in the area of neurological speech disorders has also focused on the perception of the self as a communicator. Walshe, Miller, Leahy, and Murray (2008) examined speaker self-perception in a group of 20 people with acquired dysarthria. Speakers self-rated their own speech intelligibility using direct magnitude estimation. These ratings were then compared to intelligibility scores on the Assessment of Intelligibility of Dysarthric Speech (AIDS) (1981). Results showed no correlation between speakers' perceptions of intelligibility and severity ratings on the AIDS. This suggests that speakers view their speech differently than listeners. A proposed explanation is that speakers may rate their intelligibility generally, while listeners rate articulation of specific sentences (Walshe et. al, 2008). Miller, Noble, Jones, Allcock, and Burn (2008) examined how self-perceived communication changes over time in a cross-sectional survey of 104 people with PD. Participants with PD completed a semantic differential questionnaire comprised of bipolar adjectives or statements representing key variables in the domain of communication disorders. Across all participants, there was a statistically significant perception of deterioration in communication after the onset of

PD. People with PD reported a loss of control in communicating, less confidence communicating, difficulties getting their message across, and feelings of frustration, inadequacy, and a sense of lost independence (Miller, Noble, Jones, & Burn, 2006). In a follow-up longitudinal study, Miller, Andrew, Noble, and Walshe (2011) administered a semantic differential questionnaire to 31 people with PD at 3 different time points: in the early stages of PD, and at two later assessment points 3 years apart. The results of this study showed negative changes in perception of self as a communicator in areas involving competence, adequacy, control, and ease of communication.

Measuring communicative participation. Communicative participation is measured in a social context, in which there is an exchange of information and ideas between more than one person (Eadie et al., 2006). Baylor, Yorkston, and colleagues developed a validated tool to measure communicative participation, called the Communicative Participation Item Bank (CPIB) (Baylor, Yorkston, Eadie, Kim, Chung, & Amtmann, 2013; Baylor, Yorkston, Eadie, Miller, & Amtmann, 2009; Yorkston, Baylor, Dietz, Dudgeon, Eadie, Miller, and Amtmann, 2008). The aim of this measure is to better understand how communication disorders impact communication in real life situations. Measuring communicative participation is important because it advances our understanding of both the impact of disablement and the contextual factors that affect it, and helps to develop and revise models of intervention (Eadie et al., 2006).

Unfortunately, the literature regarding the effect OMD has on communicative participation is sparse. Prior to the development of the CPIB, Dykstra, Adams, and Jog (2007) investigated the perception of activity and participation restrictions in an individual with lingual dystonia receiving therapeutic BoNT-A injections. A subjective

evaluation was obtained by administering a modified version of the Voice Activity and Participation Profile (VAPP) pre- and post- BoNT-A treatment. The VAPP is a 28-item self-assessment questionnaire developed to evaluate perceptions of speech problems, activity limitations, and participation restrictions (Ma & Yiu, 2001). Examination of the VAPP revealed dramatic changes on all domains measured by the questionnaire following treatment, such as self-perceived severity of the speech problem, effects on job, and daily social communication and emotion. Results also showed a dramatic change in self-rated activity limitation and participation limitation pre- and post- treatment. Specifically, there was a significant positive difference in the direction of scores following treatment. These results highlight the importance of examining the impact of a speech disorder on an individual's daily functions in the context of their personal, social, and environmental situation (Dykstra et al., 2007; Ma & Yiu, 2001).

One component of communicative participation is communicative effectiveness. Communicative effectiveness is described as a person's ability to successfully communicate their message in home and community settings (Hustad, 1999). Communicative effectiveness can be measured using the Communicative Effectiveness Survey (CES). The CES is comprised of eight items rated on a four-point scale (Hustad, 1999). The CES allows speakers with dysarthria and their communication partners to rate communicative effectiveness in various life situations in order to identify which situations are perceived as most difficult (Hustad, 1999). Ball, Beukelman, and Pattee (2004) used the CES to measure the communicative effectiveness of 25 people with amyotrophic lateral sclerosis (ALS) and resulting dysarthria. The CES was administered to both the participants with ALS and their communication partners, separately. The

results of this study found that speakers and listeners reported similar perceptions of communicative effectiveness, and self-rated communicative effectiveness was positively related to speech intelligibility. Donovan, Kendall, Young, and Rosenbek (2008) examined communicative effectiveness in individuals with PD and their significant others. In contrast, the results of this study found that participants with PD rated themselves with significantly higher ratings of communicative effectiveness than their significant others, and speech intelligibility was not a significant predictor of communicative effectiveness. McAuliffe, Carpenter, and Moran (2010) examined the differences in perceived communicative effectiveness between eight participants with dysarthria following traumatic brain injury (TBI) and their communication partners. Results showed a trend for participants with TBI to rate their communicative effectiveness higher than their communication partners, but this finding was not significant. Furthermore, there was no relationship between communicative effectiveness and intelligibility. Dykstra, Domingo, Adams, and Jog (2015) administered the CES to participants with OMD receiving therapeutic BoNT-A, and to healthy control participants. The results of this study showed a significant difference between OMD and control participants' overall ratings of communicative effectiveness. More specifically, there were significant differences between OMD and control participants on five out of eight items on the CES. The items on the CES with the largest effect size were "conversing with a stranger on the telephone" and "having a conversation with a family member at home". These items accounted for 66.4% and 49% of the variance between OMD and control participants, respectively. These results suggest that individuals with OMD self-report significant reductions in communicative effectiveness relative to control

participants. Furthermore, the results of this study provide insight into the everyday consequences of OMD. This preliminary work by Dykstra et al. (2015) provides a rationale for exploring communicative participation in this clinical population in more depth.

The experience of a speech disorder is highly individualized, and dependent on a wide array of personal, contextual, and environmental factors. Furthermore, the psychosocial impact of impaired communication does not necessarily correlate strongly with any clinical measurement (Walshe & Miller, 2011). This can make it a difficult phenomenon to capture in research. Qualitative research methods are ideally suited for studying the complex nature of speech disorders because they identify the individual with the disability as the “expert,” rather than the researcher or clinician (Dowling, 2007). A qualitative approach provides space for “participant voice,” which allows for a genuine account of the insider’s experience. Participants share with the researcher the information and experiences that are of importance to them (Dowling, 2007). In qualitative research, participants are not restricted by rigid questionnaires and rating scales, nor are they influenced by any presuppositions held by healthcare professionals (Dowling, 2007). Previous research has taken a qualitative approach to study the experience of communication impairments within specific medical conditions (e.g. PD, motor neuron disease, multiple sclerosis, or stroke) revealing changes in relationships, social and emotional effects, and perception of stigmatization (Blaney & Lowe-Strong, 2009; Miller, Noble, Jones, Allcock, & Burn, 2008; Miller, Noble, Jones, & Burn, 2006; Yorkston, Klasner, & Swanson, 2001). However, there is currently limited research investigating the impact of OMD on communicative participation.

1.9 Rationale for Current Study

The purpose of the current study was to obtain a self-reported account of the experience of living with OMD, and to gain a better understanding of both the daily facilitators and interferences to communicative participation specific to this cohort. The goal for this study is that by using qualitative research methods, this research will add novel information to the understanding of the consequences of OMD on communicative participation. By understanding the perspective of the insider, it is further anticipated that this information will help inform the clinical management of individuals with OMD and dysarthria.

Chapter 2

2 Method

2.1 Participants

Eight participants diagnosed with OMD were recruited to participate in this study. In total there were five males and three females (age range: 44-80 years; mean age: 68 years), with an average OMD onset of 10.4 years. Participants' occupations, in no particular order, were: receptionist, ad exec, homemaker, teacher, self-employed, engineer, chief executive officer, and principal. Some participants were retired at the time this study was conducted. Participants were recruited from the Movement Disorders Clinic, London Health Sciences Centre at London, Ontario and were seen by neurologist Dr. Mandar Jog. Participants with OMD were reported to demonstrate hyperkinetic dysarthria by a Speech-Language pathologist (A.D.) and a Neurologist (M.J.) specializing

in movement disorders. The presence of hyperkinetic dysarthria associated with OMD was the primary inclusion criterion of this study. Additional inclusion criteria included: (1) all participants with OMD had no prior history of speech, language, or hearing problems (except those related to OMD); (2) all participants were required to read, speak, and understand English; (3) 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 capture the average age of onset of OMD (66 years; range of 40 – 80 years); (4) all participants were receiving botulinum toxin injections to manage symptoms of OMD; (5) individuals with any type of OMD (i.e., lingual, jaw-opening, jaw-closing, mixed) were eligible to participate in the study. Table 1 contains specific data for each participant. This table includes information about the participants' sex, age, disease duration, type of OMD, and occupation.

Table 1. Demographic information of participants with OMD

| Participant ID | Sex | Age (years) | Years since diagnosis | Years receiving Botox | Type of OMD |
|----------------|-----|-------------|-----------------------|-----------------------|--------------------------------|
| GM | M | 69 | 4 | 3 | Meige's (jaw closure, lingual) |
| ST | F | 78 | 2 | 3 months | Jaw opening |
| NF | F | 60 | 10 | 8 | Lingual |
| JR | M | 44 | 2 | 3 months | Meige's (pouting, jaw closure) |
| FI | F | 69 | 21 | 21 | Jaw closure |

| | | | | | |
|----|---|----|----|----|--------------------------------|
| SP | M | 78 | 13 | 11 | Lingual, jaw closure |
| EP | M | 80 | 23 | 22 | Meige's (jaw opening, closure) |
| BR | M | 68 | 8 | 3 | Jaw closure |

The researcher 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 (Appendix C).

2.2 Research Approach

The current study was conducted using a qualitative phenomenological approach. 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. Phenomenological research is based on the principle of lived experiences, which are the events that naturally occur in the lives of a specific cohort (Dowling, 2007). Phenomenology has become an increasingly popular research method in the health care field, as it takes the patient's voice into primary account allowing for findings to emerge that may have not been previously explored (Dowling, 2007). Qualitative research creates a unique relationship between participant and researcher. Rather than attempting to remove the role of the researcher altogether, as is the case in quantitative research, qualitative researchers

attempt to interpret, to understand, and to describe information in a reflective process (Wilding & Whiteford, 2005). Furthermore, qualitative research adopts a subjectivist paradigm, meaning that reality is a subjective construct based on context and personal experience, rather than an absolute, as suggested in the positivist tradition (Wilding & Whiteford, 2005).

2.3 Procedure

Interviews. Each participant attended one face-to-face interview. Interviews were conducted by the primary researcher in a private room. The primary researcher was not involved in the clinical care of the participants. Interviews lasted between one and one-and-a-half hours, and were audio recorded for later transcription. Since participants had reduced speech intelligibility due to dysarthria, interviews were scheduled at 5 weeks post-BoNT-A injections to correspond to the peak effectiveness of BoNT-A treatment. All participants presented with speech intelligibility that was reduced but understandable to the interviewer. 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.

Phenomenological interviews were guided by the participant to the topics that she/he found relevant. This differs from quantitative research methods, which are guided largely by pre-determined questions. It is expected that phenomenological interviews will produce a detailed account of the experience of living with OMD, in the words of the persons who live it.

Interviews were guided by four general questions:

1) *Tell me about your history with OMD, for example when did your symptoms start and how did that affect you?*

2) *What impact has dystonia had on your life?*

From there, the researcher encouraged participants to talk about their experiences and feelings, and to provide stories or anecdotes. The researcher asked additional questions as needed to clarify points or to seek additional information. These questions included:

- i) Has your job been affected by OMD? If yes, how?
- ii) Has dystonia impacted your role in the family?
- iii) Has dystonia impacted your household duties or responsibilities?
- iv) Has dystonia impacted you emotionally?
- v) Has dystonia impacted your day-to-day communication?
- vi) Has dystonia changed your participation in any social activities?

3) *What is communication like for you?*

Follow-up questions included:

- i) Are there any situations or contexts that you find better or worse?
- ii) Do you do anything that is helpful in improving your speech?
- iii) What worked well? What went wrong?

4) *Are there times when people don't understand you?*

Follow-up questions included:

- i) If yes, what specific contexts/environments/situations were easy versus difficult?

Interview analysis. Interviews were transcribed verbatim from audio recordings by a secondary researcher. Interviews were analysed following qualitative guidelines

(Benner, 1994; Creswell, 1998; Dowling, 2007). First, interviews were read multiple times for familiarity. Then the research team created a set of codes based on the content of the interviews. Codes provide a way of organizing the content of the transcripts into topic areas. Codes were developed in an iterative manner via multiple readings of the interviews and discussions among the research team. Once a final code dictionary had been developed, Dedoose software (Dedoose Version 6.1.18, 2015) was used to assign meaningful codes to excerpts of the transcripts based on subject matter, and then to index and sort these excerpts. An example excerpt was: “I probably felt sorry for myself, and a little bit depressed, and frustrated learning to deal with [OMD]. Working around different scenarios and different life situations each day, it’s not fun.” (N.F.) This excerpt highlights the emotional consequences of OMD, and received the code “emotional reactions”. There were a total of 21 initial codes (Appendix D). After the interview transcripts were coded and sorted into their content areas, each of the content areas were read in detail and summarized for patterns that emerged. Themes were developed to reflect the most salient patterns within and across coded topic areas.

The goals for the final qualitative analysis were 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).

2.4 Credibility

Several steps were taken to promote the credibility of the data. Audio recordings were first transcribed by research assistants who were otherwise uninvolved in data collection and analysis. The secondary researcher then reviewed the transcripts and made any notations where there were discrepancies between what she heard on the recording and what the transcript contained. Discrepancies were resolved via consensus of the research team.

Interview analysis was conducted in an iterative manner by the research team consisting of individuals with varying backgrounds including doctoral training in qualitative methods, years of experience conducting research using the phenomenological approach, and experience treating OMD. Emerging interpretations of the data were reviewed and challenged by each member of the research team.

2.5 Verifiability

In qualitative research, verifiability refers to the extent to which the findings are an authentic representation of the phenomena they are intended to portray (Anderson, 2010). There are multiple techniques that can be used to verify qualitative research findings. This study used constant comparison to ensure rigour in the design. ‘Constant comparison’ means the emerging analyses were compared with previous ideas in an iterative and reciprocal manner ensuring that the data were viewed as a whole rather than in fragments (Anderson, 2010). For example, after each interview was coded, the researcher compared it with all previously coded interviews, and any necessary changes to coding were made. Similarly, after each code group (all quotes that received the code “X”) was analyzed, the researcher compared resulting interpretations to previous interpretations, and again findings were adjusted as needed, so on and so forth. Data

analysis followed in this reciprocal manner until all interviews were coded, analyzed, and interpretations of the data were made. Analyzing data in this manner ensured that findings were representative of the experiences of all the participants involved.

Chapter 3

3 Results

Three major themes and seven sub-themes emerged from the analysis of interview data (Table 2). The first theme, *Speaking is different now*, contained information about the physical effort required to speak with OMD. The three sub-themes under this category included *what my speech is like*, *my environment matters*, and *I use strategies*. The second major theme was *My roles have changed*. This theme addressed changes in participants' everyday lives since their diagnosis and incorporated two sub-themes: *things that are different*, and *why I've made changes*. The third major theme was *I accept it and move on*. This theme focused on how participants were able to deal with living with OMD. The sub-themes under this category were *things that help*, and *OMD has given me a different perspective*. Each of these themes and sub-themes will be described in greater detail below with quotes from participants to demonstrate how these themes were derived from the interviews to describe the consequences of OMD.

Table 2. Themes and sub-themes describing the consequences of living with OMD

| Themes | Sub-themes | Definitions |
|--------|------------|-------------|
|--------|------------|-------------|

| | | |
|---------------------------|------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Speaking is different now | What my speech is like | Speech is effortful, quiet, slow, less intelligible |
| | My environment matters | Situational factors, i.e., eating, fast-moving/group conversations, unfamiliar listeners, unplanned speech, background noise, phone conversations |
| | I use strategies | Strategies to improve ease of communication and intelligibility i.e., using easier words, using shorter, less complex sentences, slowing rate of speech, increasing loudness, have others speak for me |
| My roles have changed | Things that are different | Job status, from care-giver to care-receiver, household management, change in social activities |
| | Why I've made changes | Fatigue, intelligibility deficits, appearance, reactions of others |
| I accept it and move on | Things that help | Educating others, new activities, using humour |
| | OMD has given me a different perspective | Being thankful for other abilities, positive attitude |

3.1 Theme 1: Speaking is Different Now

Participants described both internal and external factors that affected their speech production, including physical aspects of dystonia and environmental factors. Participants then described how they adapted their speech to improve their intelligibility.

What my speech is like. Participants described changes in their speech production such as increased physical effort, slowed rate of speech, and difficulty articulating certain speech sounds.

N.F. (all initials are fictitious) explained that when she first started having symptoms of OMD, her speech slowly became “slurred” and it was more difficult to speak. As her symptoms worsened, she explained, “There was a point of, you could still understand what I was saying but it was an effort. A big effort to be able to make it clear.” E.P. described his speech as: “...thicker and changed and slower; took more time to formulate the jaw, tongue, throat muscles to make the pronunciations that would normally roll off your lips prior to that.” S.P. similarly described trouble speaking because “the lips didn’t seem to want to make the sounds.” These examples highlight authentic patient experiences of effortful and impaired communication resulting from the dysarthria associated with dystonia such as slowed rate of speech, impaired articulation, and reduced speech intelligibility.

My environment matters. Participants highlighted external environmental factors that made speaking more difficult and/or less intelligible. One obstacle common to all participants was eating. Multiple participants expressed difficulty participating in conversations while focusing on eating safely, and some participants reported avoiding situations that involved eating while engaged in conversation. For example, F.I. explained, “Just thinking, oh gosh, you know? You invited people over, you have to talk or eat or both, for a whole evening, it’s a big stress for me.” Since many social situations involve food, this can be a significant barrier for individuals with OMD. In addition to

the difficulties of participating in conversations while eating, participants also described difficulty speaking in group conversations. G.M. stated:

If there were three of us [in conversation],.. how can you just keep on participating in a conversation? I had a hard time to get in there in that conversation. A very hard time getting in. By the time I would try and get ready to say something, the other person's talking. And so I was always kind of behind in getting into that conversation.

Participants also reported unfamiliar listeners, noisy environments, and speaking over the phone as external factors that made speaking more difficult. Four out of eight participants reported difficulty speaking over the phone. For example, S.T. explained:

Well I think there are times [others don't understand me], but they don't want to let on, and then I have to repeat it... But I can tell, and especially on the phone, I notice I have to repeat; that they don't understand.

S.T. explained that speaking in noise exacerbated her dystonic spasms. "I don't talk if we're out and it's a big noise. I just don't talk. It's too hard. You have to talk too loud and my jaw just goes crazy."

I use strategies. Participants described the strategies they used to improve their speech intelligibility. B.R. explained,

I have a little strategy. If I was talking to someone and I had a real problem, I find myself choosing my words. Instead of saying, 'well it's a very overcast day' I might say something like 'it's quite cloudy.' Things that would be easy for me to say.

E.P. also found that choosing his words more carefully helped. "The words I would normally use that are more than three or four syllables, I just can't get it out. So I have to stay with shorter words that I can pronounce, and that sound clearer." Other strategies participants used were increasing loudness and slowing rate of speech. E.P. explained, "If

it's a bad period, then I just try that much more. The alternate ways. Usually I can bring my voice up and speak slower and use fewer words, and it will get more through."

3.2 Theme 2: My Roles Have Changed

The consequences of OMD extend beyond changes in speech production. Participants explained how OMD impacted their roles in the workplace and at home, as well as in social activities. Results varied among loss of roles, role restrictions, and role changes. Interferences extended to many common life situations, and resulted in emotional difficulties.

Things that are different. One area of the participants' lives that that was significantly impacted by OMD was work. Two participants revealed having to leave their job as a result of OMD that they otherwise would have continued.

I had been [working]... I tried to continue [working] until the end of June that year. I had a lot of difficulty, during the speaking part... I found I was losing a lot of confidence trying to do it, you know? I'd find I was doing a lousy job, so I just decided to leave it. (F.I.)

When talking about her job, N.F. said,

My speech had deteriorated and I just finally had to leave [my job]. I'm a pretty tough person and I pushed it to the very end because I loved my job. Considering I like to work, that's a big impact because I'm not doing what I like the most in my daily life.

N.F. stated that she was off work for two years due to symptoms of OMD. She also explained the negative effects of job loss, such as financial worries: "My whole lifestyle was diminished. I mean I'm only getting 60 percent of my income, you know?" The participants with jobs who continued to work stated that OMD significantly interfered

with their productivity. Participants reported decreased confidence at work, taking more time than usual to complete tasks, and needing more help. For example, B.R. stated, “[OMD] was really affecting my job, because I probably spent 80% of my time on the phone, and I even got from my colleagues “pardon?”, “what?” It was kind of embarrassing at times, especially at work.”

In addition to occupational changes, participants explained how OMD affected their roles in the family and in the household. One participant (N.F.) with young children explained having to shift responsibility to other members of her family as a result of her diagnosis.

A lot of those meetings, the bank etc., had to finally be done by my husband or children ... because I’m not communicating what the needs are. I could write a cheque, but if there was any discrepancy over anything in the family household that needed to be dealt with it’s usually through communication.

N.F. further described feelings of hopelessness and guilt for not being able to carry on with her responsibilities, and also disclosed that her children expressed anger at having to take on more work around the household.

Many participants described a change in their social activities. S.T. stated that because of her dystonia she would “prefer to stay at home a lot of times.” She continued, “I avoid going out. As long as I’m at home, well I’m comfortable, there’s nobody around... I’d prefer to just, be alone.” F.I. explained that because of dystonia she preferred solo activities that were less “socially interactive” such as knitting or painting. J.R. explained that he used to go out and socialize with new people at least once a week. Because of dystonia, J.R. said:

I like to watch movies at home. So I just stay home and watch movies in my own space. [Sometimes, but not often, I’ll] invite my friend or my cousin. You know,

watch TV or play video games, stuff like that. They know me and know what happened. So I'm comfortable around them.

OMD can therefore have a significant impact on the individual and their family. Reasons for these changes will be discussed in the next section.

Why I've made changes. Participants provided insight into why their roles changed as a result of OMD. One of the reasons was the inability to be understood by others. E.P. and F.I. provided examples of common life situations where they had trouble being understood by others:

I was in Tim Hortons getting a coffee and I told [the employee], I said two black coffees medium size. And she looks at me and says, "What?" I knew she didn't understand me. So I said, "Two black coffees medium size!" [yells] She jumps up in the air! Same thing happened over the meat counter at Wal-Mart. And I said, "A half pound of roast beef!" [yells]. I felt so bad for her... (E.P.)

In F.I.'s situation, she was unable to successfully complete her task as a result of her speech.

I took in the [roll of film] to be developed in Shoppers Drug Mart... So I am carrying the [roll of film] and I wanted to say, "I need to get this developed." ... I am trying to say this to him, and he says, "Well, I can't help ya if you can't speak, can I!" I thought, well "Okay, you're right!" Actually. I just walked out.

These examples highlight the difficulty some participants' faced while trying to continue on with their regular activities and responsibilities.

Another reason for role changes was the emotional consequences associated with OMD, such as experiencing unsuccessful interactions. Participants identified affective reactions to OMD, including feeling self-conscious, worried, embarrassed, and overly aware. For example, J.R. explained,

I might not approach a woman that I probably would before. Because, I didn't even know what, what to say. Because my voice is going to come out weird, and you'd be like "what is she going to think?" So I might not say anything.

N.F. highlighted how the facial spasms associated with OMD resulted in feelings of worry and fixation. She explained, "I can feel the movements underneath coming through the jaw and through my lips and whatever. I don't know. Are they looking at me? Can they see that it's moving?" Participants explained that they sometimes chose to avoid difficult situations because of how they made them feel.

Lastly, the fatigue felt from prolonged periods of speech restricted the ability of some participants to participate in roles. For example, S.P. explained:

And another thing I did notice was if I'm speaking for a while, like when I go to Bible study and the pastor asked me to read from the Scripture. And the longer I keep reading, the more difficult it is for me to enunciate the words.

N.F. explained that the fatigue from having to speak all day contributed to her decision to leave her job. "...it would be noticeable by the end of the day that it was more difficult for me to speak things clearly, and I was fatigued."

3.3 Theme 3: I Accept It and Move On

The third major theme reflected how participants were able to carry on with their lives after being diagnosed with OMD. Participants explained strategies that they found useful including support from family and friends, educating others, alternative activities, and using humour. Participants also revealed changes in perspective.

Things that help. Participants explained some strategies that were helpful for adapting to life with OMD. For example, N.F. explained that educating others about OMD and alternative modes of communication helped ease communication.

When I was losing a lot of communication, and a lot of friends and people didn't know how to speak [to me], or even figure out that there's an alternative as well as I did. I was actually the one training everyone, "Well you're going to have to do this..." They don't have alternatives in communication really.

Participants also discussed how finding fulfillment in new roles and skills helped them deal with some of their losses. For example, F.I. was able to find new work that made use of her non-speech skills. She explained,

I decided I did enough [educating], and I got into the [computer work]. And I thought, "Oh this is wonderful! Because I don't really have to speak, and I can still [work] and get paid for it."

Three out of eight participants explained that maintaining a sense of humour helped them deal with difficult situations. N.F. stated, "I tried not to lose my sense of humour to some degree. I had a few scenarios there, and you have to be able to laugh at yourself, so I think that helped me get through it as well."

OMD has given me a different perspective. The participants unanimously reported that they came to accept OMD and "move on". Four out of eight participants used the phrase "I just have to deal with it". Participants also expressed that "it could be worse" or "it's not life and death." E.P. disclosed, "I don't feel that I'm a victim... I don't want to sit and think about it, "Oh poor me I can't do this!" and maybe some people maintain that way, but I just don't." Another example of perspective was the participants' new-found appreciation for what they were still able to do. N.F. explained, "I could

physically do things, I could walk, I could breath, I could touch. I can do lots of things just not speak.” N.F. also disclosed that OMD made her a more perceptive person. “Discovering what a good listener is, was remarkable to me. I found a way to gain from that and become a good listener instead of a good speaker.” The ability to focus on the positive aspects of their lives was a meaningful and a useful strategy used by participants to aid in their acceptance of their diagnosis of OMD.

In summary of the results, participants reported changes in their speech production and identified some common environmental factors that interfered with communication. Strategies that improved communication were discussed. In addition to alterations in speech production, participants experienced changes to their work, social, and family lives. Participants also explained some strategies they used to deal with OMD, and expressed the ability to lead meaningful and fulfilling lives despite their diagnosis of OMD.

Chapter 4

4 Discussion

The purpose of this study was to better understand the consequences of living with OMD and dysarthria, and the functional, social, and emotional interferences to communicative participation it may cause. This was accomplished with a focus on patient self-report of experiences related to their OMD. This discussion will further explore the results from this study and how they relate to communicative participation as well as previous literature. Limitations, future directions and clinical implications will be presented.

4.1 Speech Production

From the phenomenological analysis of the data, three overarching themes emerged. The first theme, *Speaking is different now*, dealt with the effects of dysarthria resulting from OMD. Participants in this study reported alterations and reductions to speech intelligibility. This is consistent with the prior literature on dystonia. Darley, Aronson and Brown (1969) studied the effects of hyperkinetic dysarthria associated with dystonia on speech intelligibility and found speech dimensions such as imprecise consonant articulation, vowel distortion, and abnormal direction and rhythm of movement to contribute to a decrease in speech intelligibility. Dykstra, Adams, and Jog (2007) also found reduced speech intelligibility in an individual with lingual dystonia. In the current study, the main concerns reported by the participants were the increase in effort required to produce intelligible speech, and fatigue after speaking for prolonged periods of time. Participants described their speech production as being slurred, slow, and difficult to understand. Furthermore, participants reported external interferences to their speech such as speaking over the phone, fast-paced conversations, and noisy environments.

Participants explained how changes in speech production affected their communicative participation in everyday life. They reported unpredictability of when they may or may not be understood by others which resulted in less frequent communicative participation, especially in high-stress/unfamiliar situations. Baylor et al. (2005) also found “unreliability of the voice” to be a concern for participants with spasmodic dysphonia (SD), contributing to the avoidance of social situations. One of the most difficult real-life situations for participants in the current study was communicating

while eating a meal. Most participants spoke to how it was difficult for them to maintain a conversation while focusing on what they were eating and watching for choking and/or food spillage. This was a difficult adjustment to make for participants who previously enjoyed participating in social situations such as group conversations around the dinner table.

Participants further explained strategies they used to improve their intelligibility. The most commonly reported strategy was being vigilant with speech production and pre-planning the types of words and sounds that were easier than others to produce. Many participants stated that they simplified their speech by using shorter, less complex words and sentences, and speaking slower and louder. Baylor, Burns, Eadie, Britton, and Yorkston (2011) found a similar phenomenon in their qualitative study of communicative participation across different communication disorders. Participants described planning speech carefully, simplifying sentences, and specifically avoiding words that were difficult to say as being a helpful communicative strategy. One participant referred to this strategy as “dumbing down” her speech. This information helps to illustrate the functional restrictions to communicative participation experienced by individuals with OMD and the resulting interferences to communicative participation they may cause.

4.2 Roles

The second major theme, *My roles have changed*, identified participants' occupational, familial, and social roles that were affected by OMD. The most significant role loss identified by participants was occupational. This is problematic as job loss is often associated with economic instability and emotional difficulties (Smith, Taylor,

Mendoza, Barkmeier, Lemke, & Hoffman, 1998). Of the five participants that were working at the time this study was conducted, two had to leave their jobs as a result of OMD. The participants that continued to work after the diagnosis of OMD revealed being less productive and needing to make changes to their job. Previous research also found a significant impact of disordered communication on work life, resulting in job modifications, avoidance of pursuing new job opportunities, or job loss (Baylor et. al., 2005; Smith et. al., 1998). This phenomenon, termed ‘occupational deprivation’, often affects individuals who are born with or have acquired a chronic illness or disability (Christiansen & Townsend, 2004). Occupational deprivation refers to situations and conditions that exist outside of people, depriving them of important occupational opportunities beyond their immediate control (Christiansen & Townsend, 2004, p. 236). Some of these external conditions may include stereotyped perceptions, limited expectations, and physical/functional barriers to occupational environments. Previous narrative accounts from individuals with a disability have highlighted attitudes of others, as well as frustrations with the physical environment, as preventing them from living full and rewarding occupational lives. It is therefore important to change the general public’s stereotyped perceptions of people with disabilities by promoting awareness and education, as well as creating barrier-free environments with unrestricted access and the availability of assistive technology when needed (Christiansen & Townsend, 2004).

Other areas of participants’ lives that were affected by OMD were social and leisure activities. As a result of reduced intelligibility and perceived reactions of others, participants became more hesitant of socialization, and sometimes needed convincing from family and friends to attend social activities. In some cases, participants chose to

avoid social situations altogether. Participants explained a preference for spending more time alone, or with close family and friends, rather than socializing with new people. Baylor et al. (2005) also found changes in the social lives of individuals with SD, with participants stating that they found themselves “sitting in the background” at social activities instead of participating like they normally would.

4.3 Coping Strategies

The third major theme, *I accept it and move on*, reflected how participants managed with the effects of OMD. There is currently limited research on the coping strategies of individuals with communication disorders. Epstein, Hirani, Stygall, and Newman (2009) explored coping mechanisms in individuals with muscle tension dysphonia (MTD) and adductor spasmodic dysphonia (ASD) by administering the Voice Disability Coping Questionnaire. In the Epstein et al. (2009) study, ‘coping’ was defined as ‘the individual’s cognitive and behavioural efforts to manage the stress of illness’. Individuals were found to be either ‘proactive’ by using strategies such as information seeking and social support to eliminate the stressor, or ‘avoidant’ by the use of denial and withdrawal. In the current study, all of the participants described being proactive with managing their disorder, emphasising the acceptance of their diagnosis and the need to “move on”. Coping strategies used by the participants included maintaining a positive attitude, using humour, educating others, and being thankful for their other skills (e.g., being a good listener). Similarly, Baylor et al. (2005) found that for individuals with SD, strategies such as educating family and friends about their disorder helped them to gain support and reduce unpleasant interactions with others. Participants also reported dealing

with their SD by adopting a positive attitude, and keeping their SD in perspective relative to other medical conditions.

Although participants expressed positive reactions of acceptance and resilience, it is important that this not detract from the significance of the negative experiences and themes that emerged throughout the participants' narratives, such as those pertaining to loss, embarrassment, difficulty, and frustration.

4.4 Limitations of the Current Study

This study employed qualitative methods to collect, analyse, and present important findings on the experience of living with OMD. Qualitative research in the field of health sciences allows for the collection of complex data relating to patient experience, and creates a unique space for participant voice. There are, however, accompanying drawbacks. Qualitative research is generally conducted on a smaller scale than quantitative research due to the time-consuming nature of data collection and interpretation. Our research team intended to enrol a larger sample (10-12 participants), but due to feasibility issues such as recruitment difficulties, the rare nature of OMD, and the time constraints and scope of a student project, we completed this study with a sample size of eight participants. Although common themes did emerge from the data, a larger sample may have been required to approach saturation, meaning that the collection of more data would not have led to new information relating to the research question. For these reasons, the current study is intended to be preliminary as opposed to one that is pursuing saturation. A larger sample would have also increased diversity in self-reported experience and would have increased the potential to hear a variety of viewpoints. All

participants in this study were recruited from the Movement Disorders Clinic, London Health Sciences Centre in London, Ontario, which may have led to a uniform demographic.

Lastly, a primary method for verifiability in qualitative research is ‘respondent validation,’ which involves consulting with the participants post-analysis to ensure that the data truly reflect their own interpretations of their experiences. This allows participants to comment on and clarify any points of misinterpretation and verify the authenticity of the researchers’ summaries. Unfortunately, this practice was unable to be completed within the time frame of this paper, however participants will be contacted in the future to participate in brief follow-up visits.

4.5 Future Directions

The results of the present study provide a rationale on which to base future work in the area of OMD. The consequences of OMD are highly complex, having effects on an individual’s social, emotional, and physical functioning. Future studies with a larger sample that rely on qualitative methodologies such as interview analysis may help researchers to further capture the complexities of living with OMD. This study focused on the effects of OMD on communicative participation. An interesting next step may be to use a similar study design to explore other consequences of OMD, such as the experience of various methods of treatment. Currently, the most accepted method of treatment for OMD is BoNT-A injections. Research is currently underway using qualitative methods to examine the effects of botulinum toxin treatment on communicative participation in individuals with OMD.

In some cases, themes relating to role shifts and social participation were dependent on demographic factors specific to the individual. An interesting future direction may be to sample a larger number of participants and explore various demographic factors such as employment status, number of dependents, and age. This information may help to tailor intervention to future patients with similar demographic profiles.

Lastly, the information presented in this study may help adapt questionnaires such as the Communicative Participation Item Bank (Baylor et al., 2009) to include information specific to the effects of OMD on communicative participation.

4.6 Clinical Implications

Understanding the restrictions to communicative participation that affect individuals with OMD is extremely useful in order to improve rehabilitation efforts in the field of speech-language pathology. A benefit of employing qualitative methodologies to collect this information is the ability for participants to speak openly about their disorder without any preconceptions from the healthcare community. The results of this study highlighted specific sources of difficulty, strategies used, and areas of importance to individuals living with OMD, in their own words. By listening to our participants' stories and experiences we were able to draw two main conclusions that have clinical importance.

The first is that the consequences of OMD extend beyond the speech impairment. There has been a shift in focus in healthcare and disability management from simply minimizing symptoms to improving social, emotional, functional, and cognitive aspects

of functioning as well. This concept was solidified by the World Health Organization's (WHO) 2001 definition of health as "a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity." It is now more widely accepted that poor physical or mental health is likely to have many consequences including restrictions on social roles and shifts in social relationships (Dijkers, Whiteneck, & El-Jaroudi, 2000). From the analysis of our data, concepts relating to social interaction, role shifts, and affective responses emerged as salient to the participants. Speech difficulties were rarely presented from only the physical perspective, but more commonly referred to in the context of social communication. For example, some participants expressed worry that because they had difficulty producing speech, others might misinterpret the meaning of their words. Participants also highlighted contextual and environmental factors that restricted their communication such as fatigue, background noise, speaking over the phone, and speaking to unfamiliar listeners.

The second conclusion is that living with OMD is a unique experience that is dependent on demographic factors such as employment status or age. For example, an individual diagnosed with OMD with young children might seek out strategies to aid in household management and child rearing, whereas an older individual might be more worried about choking while eating and maintaining community involvement. This is consistent with the phenomenological framework of inquiry that supports the belief in the existence of not just one reality, but of "multiple realities." According to this belief, there exists no one "truth" to be uncovered; instead each individual constructs his/her own reality based on his/her personal experiences (Denzin & Lincoln, 2000). The results of this study therefore, confirm the idea that the treatment of speech disorders, and OMD

specifically, must take social, emotional, and functional consequences into account as well as physical symptoms. Furthermore, a treatment plan should be tailored to each individual based on his/her personal concerns and goals.

Chapter 5

5 Summary and Conclusions

This study has presented a perspective of the consequences of OMD on communicative participation as experienced by the insider. The results of this study suggest that communicative participation is affected not only by the physical symptoms of OMD, but also by many significant social and emotional components. Changes to communicative participation can affect an individual's job, family, and social life. Lastly, the consequences of OMD are unique to each individual based on their goals and lifestyle.

This study adds to the very minimal literature on OMD, and builds upon the small empirical literature on the consequences of living with OMD. The results of this study make a significant and a novel contribution to the literature due to the phenomenological research methodology used. Participants in this study were able to speak openly about their experience of OMD in the absence of a structured questionnaire or a narrow line of questioning. As a result, novel phenomena emerged that will aid in the understanding of the consequences of OMD for researchers, clinicians, family members, and even newly diagnosed patients.

Although this study included a small number of participants, the overlap in theme content suggests that the main consequences of OMD on communicative participation were explored. Future studies may still benefit from a larger and more varied sample of participants to ensure theme saturation. Future directions may include a more in-depth analysis of the demographic factors involved in the experience of OMD and the effects of various methods of treatment on communicative participation. Once more research is conducted to solidify qualitative themes in this area, quantitative statistical analysis may be beneficial to augment findings.

References

- Anderson, C. (2010). Presenting and evaluating qualitative research. *American Journal of Pharmaceutical Education*, 74(8).
- Avila O. I., Drachman D. B., & Pastronk A. (1989). Neurotransmission regulates stability of acetylcholine receptors at the neuromuscular junction. *Journal of Neuroscience*, 9, 2902-2906.
- Bakke, M., Larsen, B. M., Dalager, T., & Moller, E. (2013). Oromandibular dystonia-functional and clinical characteristics: a report on 21 cases. *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology*, 115(1), e21-6.
- Ball, L. J., Beukelman, D. R., & Pattee, G. L. (2004). Communication effectiveness of individuals with amyotrophic lateral sclerosis. *Journal of Communication Disorders*, 37(3), 197-215.
- Baylor, C., Burns, M., Eadie, T., Britton, D., & Yorkston, K. (2011). A qualitative study of interference with communicative participation across communication disorders in adults. *American Journal of Speech-Language Pathology*, 20(4), 269-287.
- Baylor C. R., Yorkston, K. M., & Eadie, T. L. (2005). The consequences of spasmodic dysphonia on communication-related quality of life: A qualitative study of the insider's experiences. *Journal of Communication Disorders*, 38(5), 395-419.
- Baylor, C.R., Yorkston, K.M., Eadie, T.L., Kim, J., Chung, H., & Amtmann, D. (2013). The Communicative Participation Item Bank (CPIB): Item bank calibration and development of a disorder-generic short form. *Journal of Speech Language and Hearing Research*, 56, 1190-1208.
- Baylor, C., Yorkston, K., Eadie, T., Miller, R. M., & Amtmann, D. (2009). Developing the communicative participation item bank: Rasch analysis results from a spasmodic dysphonia sample. *Journal of Speech, Language, and Hearing Research*, 52, 1302-1320.
- Benner, P. (1994). The Tradition and Skill of Interpretive Phenomenology in Studying Health, Illness, and Caring Practices. In P. Benner (Ed.) *Interpretive phenomenology: Embodiment, caring, and ethics in health and illness* (pp.99-117). Thousand Oaks: Sage.
- 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.

- Blaney, B. E. & Lowe-Strong, A. (2009). The impact of fatigue on communication in multiple sclerosis. The insider's perspective. *Disability and Rehabilitation*, 31(3), 170-80.
- Blitzer, A. & Sulica, L. (2001). Botulinum toxin: basic science and clinical uses in otolaryngology. *Laryngoscope*, 111, 218-226.
- Brin, M.F., & Comella, C.L. (2004). Pathophysiology of dystonia. In M.F. Brin, C.L. Comella, & J. Jankovic (Eds.) *Dystonia: Etiology, Clinical Features, and Treatment* (pp. 5-10). Philadelphia, PA: Lippincott Williams & Wilkins.
- Cardoso, F., & Jankovic, J. (1995). Oromandibular dystonia. In J.K. Tsui & D.B. Calne (Eds.) *Handbook of dystonia* (pp.181–190). New York, NY: Marcel Dekker.
- Christiansen, C. H., & Townsend, E. A. (Eds.). (2004). Disability and Occupational Derivation. *Introduction to Occupation* (236-242). Upper Saddle River, N.J.: Prentice Hall.
- Clark, G. T. (2003). The management of oromandibular motor disorders and facial spasms with injections of botulinum toxin. *Physical Medicine and Rehabilitation Clinics of North America*, 14(4), 727–748.
- Creswell, J. W. (1998). *Qualitative inquiry and research design: Choosing among five traditions* (pp. 80-82). Thousand Oaks: Sage.
- Darley, F. L., Aronson, A. E., & Brown, J. R. (1969a). Differential diagnostic patterns of dysarthria. *Journal of Speech and Hearing Research*, 12, 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.
- Dedoose Version 6.1.18, web application for managing, analyzing, and presenting qualitative and mixed method research data (2015). Los Angeles, CA: SocioCultural Research Consultants, LLC (www.dedoose.com).
- Defazio, G., Abbruzzese, G., Livrea, P., & Berardelli, A. (2004). Epidemiology of primary dystonia. *Lancet Neurology*, 3(11), 673-678.
- Denzin, N.K., & Lincoln, Y.S. (2000). Paradigms and perspectives in transition. In: N. Denzin & Y. Lincoln (Eds.) *Handbook of Qualitative Research* (2nd ed.) (pp 157-162). Thousand Oaks: Sage.

- Dijkers, M. P., Whiteneck, G., & El-Jaroudi, R. (2000). Measures of social outcomes in disability research. *Archives of Physical Medicine and Rehabilitation*, 81(12 Suppl 2), S63–S80.
- Donovan, N. J., Kendall, D. L., Young, M. E., & Rosenbek, J. C. (2008). The communicative effectiveness survey: Preliminary evidence of construct validity. *American Journal of Speech-Language Pathology*, 17(4), 335–347.
- 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.
- 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., & Jog, M. (2007). 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.
- Eadie, T. L. (2001) The ICIDH-2: theoretical and clinical implications for speech-language pathology. *Journal of Speech-Language Pathology and Audiology*, 25, 181-200.
- Eadie, T., Yorkston, K., Klasner, E., Dudgeon, B., Baylor, C., Miller, R., & Amtmann, D. (2006). Measuring communicative participation: A review of self-report instruments in speech-language pathology. *American Journal of Speech Language Pathology*, 15(4), 307-320.
- Enderby, P., & Emerson, J. (2015). Speech and language therapy: does it work? *British Medical Journal*, 312(7047), 1655–1658.
- Epstein, R., Hirani, S. P., Stygall, J., & Newman, S. P. (2009). How do individuals cope with voice disorders? Introducing the voice disability coping questionnaire. *Journal of Voice*, 23(2), 209–217.
- Gandhi, Y. (2010). Oro-mandibular dystonia. *National Journal of Maxillofacial Surgery*, 1(2), 150-152.

- Garcia, L. J., Laroche, C., & Barrette, J. (2002). Work integration issues go beyond the nature of the communication disorder. *Journal of Communication Disorders*, 35(2), 187-211.
- Goldman, J. G., & Comella, C. L. (2003). Treatment of dystonia. *Clinical Neuropharmacology*, 26(2), 102-108.
- Heise, G. J. & Mullen, M. P. (1995). Oromandibular dystonia treated with botulinum toxin: report of case. *Journal of Oral Maxillofacial Surgery*, 53, 332-335.
- Hustad, K. C. (1999). Optimizing communicative effectiveness: bringing it together. In K. Yorkston, D. Beukelman, E. A. Strand, & K. R. Bell (Eds.), *Management of motor speech disorders in children and adults* (pp. 483-537). Austin, TX: Pro-Ed.
- 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.
- Kao, I., Drachman, D. B., & Price, D. L. (1976). Botulinum toxin: mechanism of presynaptic blockade. *Science*, 193(4259), 1256-1258.
- 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, 301-307.
- Kent, R.D. (1992). Introduction. In R.D. Kent (Ed.), *Intelligibility in Speech Disorders* (pp.110). Amsterdam: John Benjamins.
- Klein, C., & Ozelius, L. J. (2002). Dystonia: clinical features, genetics and treatment. *Current Opinion in Neurology*, 15, 491-497.
- Lai, S., & Jankovic, J. (1998). Peripherally induced oromandibular dystonia. *Journal of Neurology, Neurosurgery & Psychiatry*, 65(5), 722-728.
- Langlois, M., Richer, F., & Chouinard, S. (2003). New perspectives on dystonia. *Canadian Journal of Neurological Science*, 1, 34-44.
- Lee, K. H. (2007). Oromandibular dystonia. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*, 104(4), 491-496.

- Ma, E. & Yiu, E. (2001). Voice activity and participation profile: assessing the impact of voice disorders on daily activities. *Journal of Speech, Language, and Hearing Research, 44*, 511-524.
- McAuliffe, M. J., Carpenter, S. & Moran, C. (2010). Speech intelligibility and perceptions of communication effectiveness by speakers with dysarthria following traumatic brain injury and their communication partners. *Brain Injury, 24*(12), 1408-1415.
- Miller, N., Andrew, S., Noble, E., & Walshe, M. (2011). Changing perceptions of self as a communicator in Parkinson's disease: a longitudinal follow-up study. *Disability and Rehabilitation, 33*(3), 204–210.
- Miller, N., Noble E., Jones D., & Burn, D. (2006). Life with communication changes in Parkinson's disease. *Age Aging, 35*, 235–239.
- Miller, N., Noble, E., Jones, D., Allcock, L., & Burn, D. J. (2008). How do I sound to me? Perceived changes in communication in Parkinson's disease. *Clinical Rehabilitation, 22*(1), 14-22.
- Ministry of Health and Long-Term Care. (2003, February). Botulinum toxin injections (Botox ®) for treatment of focal spasticity. Retrieved from <http://www.health.gov.on.ca/en/programsohip/bulletins/4397bul4397fs.aspx>.
- Munchau, A. & Bhatia, K. P. (2000). Uses of botulinum toxin injection in medicine today. *British Medical Journal, 320*(7228), 161-165.
- 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.
- Papapetropoulos, S., & Singer, C. (2006). Eating dysfunction associated with oromandibular dystonia: clinical characteristics and treatment considerations. *Head and Face Medicine, 2*(47), 1-4.
- Ramachandran, T.S., & Molloy, F.M. (2015, April, 27). Dystonia treatment using botulinum toxin (Botox R). [Medscape]. Retrieved from <http://medicine.medscape.com/article/1818592-overview>.
- Scott, B. L. (2000). Evaluation and treatment of dystonia. *Southern Medical Journal, 93*(8), 745-751.
- Simpson, L. L. (1981). The origin, structure, and pharmacologic activity of botulinum toxin. *Pharmacological Reviews, 33*, 155-188.

- Simpson, L. L. (1989). Peripheral actions of the botulinum toxins. In L.L. Simpson (ed). *Botulinum Neurotoxin and Tetanus Toxin* (pp. 153-178). New York, NY: Academic Press.
- Smith, E., Taylor, M., Mendoza, M., Barkmeier, J., Lemke, J., & Hoffman, H. (1998). Spasmodic dysphonia and vocal fold paralysis: Outcomes of voice problems on work-related functioning. *Journal of Voice*, 12, 223–232.
- Tan, E. K., & Jankovic, J. (1999). Botulinum toxin A in patients with oromandibular dystonia: long-term follow-up. *Neurology*, 53, 2102-2107.
- Tarsy, D., & Simon, D. K., (2006). Dystonia. *The New England Journal of Medicine*, 355(8), 818-829.
- Threats, T. T. (2000). The World Health Organization's revised classification: what does it mean for speech-language pathology? *Journal of Medical Speech-Language Pathology*, 8, 13-18.
- Tinter, R. & Jankovic, J. (2002). Botulinum toxin type A in the management of oromandibular dystonia and bruxism. In M.F. Brin, M. Hallet, & J. Jankovic (Eds.) *Scientific and Therapeutic Aspects of Botulinum Toxin* (pp 1-12). Philadelphia, PA: Lippincott Williams & Wilkins.
- Walshe, M. & Miller, N. (2011). Living with acquired dysarthria: The speaker's perspective. *Disability and Rehabilitation*, 33(3), 195-203.
- Walshe, M., Miller, N., Leahy, M., & Murray, A. (2008). Intelligibility of dysarthric speech: perceptions of speakers and listeners. *International Journal of Language & Communication Disorders/ Royal College of Speech & Language Therapists*, 43(6), 633-648.
- Weismer, G., Yunusova, Y., & Bunton, K. (2012). Measures to evaluate the effects of DBS on speech production. *Journal of Neurolinguistics*, 25(4): 74-94.
- 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 (2001). *ICF: International Classification of Functioning, Disability and Health*. Geneva: Author.
- Yorkston, K. M., Baylor, C. R., Dietz, J., Dudgeon, B. J., Eadie, T., Miller, R. M., & Amtmann, D. (2008). Developing a scale of communicative participation: A cognitive interviewing study. *Disability and Rehabilitation*, 30(6), 425-433.

- Yorkston, K. M., & Beukelman, D. R. (1978). A comparison of techniques for measuring intelligibility of dysarthric speech. *Journal of Communication Disorders, 11*, 499-512.
- Yorkston, K., Dowden, P., & 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* (pp 265-286). Amsterdam: John Benjamins.
- Yorkston, K. M., Klasner, E. R., & Swanson, K. M. (2001). Communication in context: A qualitative study of the experiences of individuals with multiple sclerosis. *American Journal of Speech-Language Pathology, 10*(2), 126-137.
- 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.

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.

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

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 for repeat administration of questionnaires, speech recordings and the in-person interview.

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.

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. 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 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/CFR 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 0000040.

Appendix D

Coding Dictionary

| Code | Definition |
|---------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Aesthetics | Change in orofacial aesthetics due to OMD |
| Avoidance | |
| Botox | Effect of botox injections |
| Change of roles | Change of role from caregiver/parent/spouse role to care-receiver, as well as change of role from care-receiver to caregiver (from child's perspective), from able bodied to disabled, loss of independence |
| Communication | ability to communicate, conversations, presentations, communication that serves a social function |
| Diagnosis | Medical appointments, medical procedures, assessments, treatments, symptoms of OMD, side effects |
| Disclosure | |
| Eating | As a result of OMD: difficulty eating, chewing, swallowing, choking, aspirating, drooling, strategies used |
| Emotional reactions | Emotional reactions that result from living with OMD and its treatment: Depression/sadness, frustration, fear, anger, shock, surprise, excitement, loss, confidence, embarrassment, self-consciousness, uncertainty, denial |
| Fatigue | Fatigue due to symptoms related to OMD |
| Good stories | |
| Job | Type of employment, loss of employment, job description, regaining employment, employment choices, personal meaning of employment |
| Pain | Physical pain: due to dystonic symptoms, botox injections |

| | |
|---------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Perceived reactions of others | Reactions of others that are the result of the person's OMD: anger, frustration, sadness, helplessness, pity, honesty about the impact of OMD |
| Perspective | The ability to reflect on experiences as a result of OMD and provide perspective on disability, living with a communication disorder, experiences unique to OMD, being grateful, second chances, giving back |
| Resilience | Ability to work through difficult situations, ability to work through emotional or physical pain, strength of character, outlook, coping mechanisms (eg. humour, trying to cover up the problem) |
| Social outings | Trips, vacations, parties, gatherings, volunteer work, social interactions |
| Speech production | Aspects of speech production, difficulty producing intelligible speech, difficulty being understood by others due to OMD, specific aspects of speech production difficulties |
| Strategies | Strategies used to make speech better, make speech worse, alternate ways of communicating |
| Strategies to improve symptoms of OMD | Sensory tricks, strategies used to help with non-speech aspects of OMD |
| Support | Relationships and individuals who play a supportive role to the individual with OMD. This can take the form of emotional support, physical support, supportive environments |

Curriculum Vitae

| | |
|----------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Name: | Lauren Siegel |
| Post-secondary Education and Degrees: | <p>McGill University Montreal, Quebec, Canada 2010-2014 B.A.</p> <p>Western University London, Ontario, Canada 2014-2016 MSc.</p> |
| Related Work Experience | <p>Research Assistant McGill University 2012-2014</p> <p>Teaching Assistant Western University 2014-2015</p> <p>Research Assistant Western University 2014-2016</p> <p>Research Approval Support Specialist Lawson Research Institute 2016-Present</p> |
| Presentations: | <p>Examining ratings of communication-related quality of life in speakers with oromandibular dystonia receiving botulinum toxin therapy. Poster presentation at the HRS Graduate Research Conference, Western University, London, Ontario, February 4 2015.</p> <p>Examining ratings of communication-related quality of life in speakers with oromandibular dystonia receiving botulinum toxin therapy. Poster presentation at the Faculty of Health Sciences Research Day 2015, Western University, London, Ontario, March 25 2015.</p> |

Examining communication-related quality of life in individuals with oromandibular dystonia. Oral presentation at the 17th Annual Rehabilitation Research Colloquium, Queen's University, Kingston, Ontario, May 1 2015.

Examining speech intelligibility and self-rated quality of life in individuals with oromandibular dystonia receiving botulinum toxin therapy. Poster presentation at 2016 Motor Speech Conference, Hyatt Regency, Newport Beach, CA, March 3 2016.

Examining speech intelligibility and self-rated quality of life in individuals with oromandibular dystonia receiving botulinum toxin therapy. Poster presentation at Faculty of Health Sciences Research Day 2016, Western University, London, Ontario, March 22 2016.