Speech and Language Disorders in Older Canadians: Epidemiologic Primary Care-Based Research Using the Canadian Primary Care Sentinel Surveillance Network (CPCSSN) Database

by

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Abstract

Objectives: The objective of this study is to examine the epidemiology and clinical management in primary care of speech and language disorders in older adults (fifty-five and older) and to assess the utility and feasibility of EMR data to answer such research questions. This study is comprised of a case definition validation study through which prevalence was estimated.

Methods: This study was a combined case definition validation and cross-sectional prevalence study. The study involved development of a case definition through which a chart review was undertaken to serve as the 'gold standard'. Following the chart review, the case definition was converted into a computer algorithm and applied to the same list of patients as the chart review. The results were assessed for sensitivity, specificity, positive predictive value and negative predictive value. Prevalence was assessed using the chart review.

Results: Prevalence within our sample of 1384 patients was 1.2%. The case definition had favourable specificity (99.9%, 95% CI: 99.6% - 100.0%) as well as positive predictive value (75.6%, 95% CI: 25.4% - 96.6%) and negative predictive value (99.0%, 95% CI: 98.8% - 99.2%). Sensitivity was not sufficient for validity (18.8%, 95% CI: 4.05% - 45.6%).

Conclusions: The case definition formulated did not meet an acceptable standard for validity, and thus could not be used to determine national prevalence of speech and language disorders within the CPCSSN database. However, due to the case definition's high positive and negative predictive values, it may be useful for clinical purposes, rather than epidemiologic applications. Due to the compounded issues of 'subjective' diagnosis for speech and language disorders and limitations of EMR-derived data for subjectively diagnosed diseases, speech and language disorders are a set of conditions which may not be easily studied in this context. Finally, while

the case definition did not prove valid, this study has provided a robust estimate of prevalence (1.2%) which is not based on composite data, nor is limited by etiology.

Preface

This thesis is an original work by Rebecca Miyagishima. The research project, of which this thesis is a part, received research ethics approval from the University of Alberta Research Ethics Board, Project Name "Speech and Language Disorders in Older Canadians: Epidemiologic Primary Care-Based Research Using the CPCSSN Database," No. 00049166, August 20, 2014.

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CHAPTER 1: Introduction

I. Background

The populations of Canada and most industrialized countries are aging. In Canada, the number of adults aged sixty-five and over was estimated to be five million as of 2011 (Government of Canada, 2014, par. 2). In 2005, the Government of Canada identified focal points and future "directions" for the Canadian health system to increase productivity and effective service delivery, as well as viability, in the face of these changing population demographics (Health Council of Canada, 2005, 4). As in other countries, including France, Germany, Australia and the United States, there is currently a shift in resource allocation and policy towards primary health care (Schoen et al., 2012). The reason for this shift is related to the unique healthcare needs of older adults. Canadian seniors comprise a significant contingent of primary care service users, with up to 96% of older Canadians indicating that they see a family physician (Public Health Agency of Canada, 2014, par. 98). A 2011 report by the Canadian Institute for Health Information determined that 9.7% of senior Canadians "frequently" (10 visits or more per year) saw a family physician compared to 5.5% of younger adults (CIHI, 2011, 34). The report concluded that "strong primary health care" with a focus on "chronic disease management" was key to improving Canada's health care system in anticipation of health care needs in upcoming years (51). Accomplishing this goal requires expansion of research based in primary care to inform evidence-based primary care practice.

Many of the health issues faced by elderly people are complex and difficult to manage, and some fall outside of family physicians' scope of practice (Wong and Stewart, 2010). While many conditions associated with older age, such as dementia, were formerly the domain of "tertiary care settings, research centres" and other "specialized clinics", shortages of staff and long waiting lists for geriatricians, neurologists and other specialists combined with a "trend of decline" in Canadian family physicians' scope of practice (Wong and Stewart, 2010, e221) will tend to mean that many older Canadians will be diagnosed and treated for their conditions by their family physician and other primary care providers and have limited access to other health care services (Lee et al., 2014, 50). Speech and language disorders provide an important example of conditions that may increasingly be managed in primary care.

II. Types of Speech and Language Disorders Facing Older Adults

Aphasia (also infrequently referred to as 'dysphasia') is characterized by disturbance in the "complex process of interpreting and formulating language symbols" (Zhang et al., 2006, 59). There are several sub-types of aphasia based on both symptoms and etiology. Global aphasia describes the most debilitating sub-type, in which patients lose the ability to produce and understand speech in both spoken and written forms (National Aphasia Association (NAA), 2015). Broca's aphasia (also called non-fluent aphasia) refers to Broca's area in the brain's frontal lobe and indicates a loss of ability to produce fluent speech (Merck Diagnostic Manual; NAA, 2015). Wernicke's aphasia (also known as fluent aphasia) refers to Wernicke's area on the cerebral cortex (Merck Diagnostic Manual; Mayo Clinic, 2014). Patients with Wernicke's aphasia retain the capability to produce speech sounds; however they have difficulty comprehending spoken language (NAA, 2015). Anomic aphasia is a sub-type in which patients lose the ability to "supply" words needed for the phrases they wish to use (NAA, 2015, par.10). Finally, primary progressive aphasia is a "degenerative" form in which language abilities regress without the loss of other mental faculties (NAA, 2015, par.11). Primary progressive aphasia is often associated with frontotemporal dementia (Mesulman, 2001) while the other aphasia sub-types occur most frequently due to stroke (NAA, 2015).

Management for aphasia sometimes includes therapy from a registered speech-language pathologist (Mayo Clinic, 2014). Possible methods include the Schuell-Wepman-Darley Multimodal Stimulation Treatment, and Sentence Level Auditory Comprehension and Language Oriented Treatment (Robey, 1998). In the past, researchers have assessed the efficacy of medications such as pyrithioxine, memantine and piracetem, among others for aphasia treatment, but with little success (Robey, 1998; Tocco et al., 2014; Greener et al., 2001).

Dysarthria is a "motor speech disorder" which may result from stroke, Parkinson's disease, amyotrophic lateral sclerosis (ALS), multiple sclerosis (MS), cerebral palsy, and others (Ashley et al., 2006, 295). There are several sub-types of dysarthria, which vary largely upon the etiology of the speech disorder. Flaccid dysarthria concerns "weakness in the muscles of articulation" resulting from progressive bulbar palsy and stroke (Ashley et al., 2006, 295). Ataxic dysarthria involves "decreased rate of speech, inappropriate stress, fluctuating volume, and poorly controlled coordination of respiration and phonation" most often associated with MS

(Ashley et al., 2006, 295). Spastic dysarthria causes "harsh, strained, or strangled vocal quality; imprecise articulation; and irregular prosody" as a result of stroke (Ashley et al., 2006, 296). Hyperkinetic dysarthria, whose symptoms comprise of "phonotory dysfunction, unpredictable breakdowns of articulation and abnormalities in speech timing and prosody", is most frequently found in Huntington's disease (Ashely et al., 2006; Rusz et al., 2013, 1). Lastly, hypokinetic dysarthria causes the speaker to experience "monotone…loudness and pitch, reduced stress, imprecise articulation, inappropriate silences…and a breathy, harsh vocal quality" and is most commonly found in patients with Parkinson's disease (Ashley et al., 2006, 296).

Treatment for dysarthria is predominantly speech-language therapy (Mayo Clinic, 2014). Management of dysarthria in a speech-language pathology setting will often focus on regulation of speech rate and strengthening muscles involved in speech (RCSLT, 2009, 12). Examples of possible methods include the Lee Silverman Voice Treatment which has been assessed for efficacy in patients suffering from Parkinson's disease and Behavioural Communication Intervention for use in post-stroke dysarthria (RCSLT, 2009, 12).

Apraxia is a "speech programming disorder" (Speech-Language & Audiology Canada (S-L&AC), 2016, par.5). Patients with apraxia lack the ability to properly "sequence" speech sounds in order to create words and phrases (Ashley et al., 2006, 297). Stroke is the most frequent cause of apraxia (Ogar et al., 2005) however; traumatic brain injury and brain tumor are also potential causes (Ashley et al., 2006).

Apraxia does not typically "stand alone", rather it appears alongside other disorders of speech and/or language, particularly aphasia (Davis et al., 1998, 25; Wambaugh et al., 2012). Treatment for apraxia typically focuses on "repeated practice" as in methods such as "traditional articulation therapy" (Wambaugh et al., 2012, S5; Ogar et al., 2005, 431).

Stuttering is a type of fluency disorder in which the "normal flow and rhythm of speech" is upset (S-L&AC, 2016, par. 10). There are a relatively wide variety of symptoms of stuttering, dependent upon the patient themselves, some of which include "repetition of sounds, syllables, words, or phrases, hesitations, prolongations or interjections" (S-L&AC, 2016, par. 10). Stuttering differs from the previously discussed speech and language disorders as it is a developmental condition, as opposed to one which is acquired (American Speech-Language-

Hearing Association (ASHA), 2016). Therapy with a speech-language pathologist is the current best practice for stuttering treatment, which typically involves working to decrease the amount of stuttering in spoken speech through various techniques (NIDCD, 2014). Additionally, some patients use "electronic devices" to regulate the fluency of speech as well as medications, such as anti-depressant and anti-anxiety drugs, as anxiety and depression are often associated as comorbidities with stuttering (NIDCD, 2014, par. 15; Iverach et al., 2010).

III. Speech and Language Disorders Facing Older Adults: Review of the Literature

The literature regarding speech and language disorders in older primary care patients is highly varied and often scarce. While children's speech and language disorders have more often been the focus of research in primary care, speech and language disorder epidemiology lacks a consistent and robust body of literature for the older adult population. Additionally, most research concerns individual speech and language disorders, rather than considering those conditions as a set. However, because there is overlap between speech and language disorders among older adults in terms of etiology, risk factors and patterns of treatment, it is possible to study such disorders together. Thus, this review of the literature will examine aphasia, dysarthria, apraxia and adult stuttering on individual bases as well as collectively.

The epidemiology of speech and language disorders in older adults is a burgeoning field of study, particularly in Canada. As such, current data is limited in scope and to specific population groups, such as patients who have experienced stroke or another related condition (Flowers et al., 2013; Dickey et al., 2010). Therefore, the applicability of this data for epidemiology can be challenging. In 2005, Speech-Language & Audiology Canada (known at the time as the Canadian Association of Speech-Language Pathologists and Audiologists) published a *Speech, Language and Hearing Fact Sheet*, which contained prevalence estimates for speech and language disorders in Canada according to several age ranges. For the 65+ age group, speech disorders, language disorders and stuttering were estimated at 12%, 12% and 1% respectively (S-L&AC, 2005). Thus, for this particular source, one may consider a general estimate of population prevalence for speech and/or language disorders in older Canadians to be approximately 13%. More recently Speech-Language & Audiology Canada have published statistics more specific in focus rather than generalized to the population as a whole. S-L&AC state that 1/6 Canadians (all age categories) have a "speech, language or hearing problem" (2016,

par.11). In *Communication Health and Aging* (2015), the authors note that "over" 100,000 people in Canada suffer from aphasia and 30% of stroke patients experience aphasia (par.9). Issues with communication are also present in 95% of dementia patients (2015, par. 5).

Research on primary care patients suffering from dementia and stroke has been more robust and can provide some insight into the circumstances of patients with related speech and language disorders. Speech and language disorders can occur as a symptom or by-product of stroke or dementia (Borthwick, 2012; Cera et al., 2013; Vidovic et al., 2011); however the epidemiology of speech and language disorders within the dementia spectrum is not well-established in the context of primary care. Survey-based evidence suggests primary care physicians have limited confidence in their ability to diagnose and treat patients suffering from forms of disordered speech (Knopman and Roberts, 2011; Scherz et al., 1991). Knopman and Roberts (2011) found an incidence rate of 3.5 - 4 per 100,000 for speech and language disorders in dementia patients, and many researchers consider current estimates to be highly conservative (Taylor et al., 2009; Cardarelli et al., 2010). In contrast, the National Aphasia Association reported prevalence of speech and language disorder in stroke survivors over the age of 65 to be between 25-40% (2014, par.2). Stroke and dementia are both recognized as being among the top causes of speech and language disorders in elderly adults (S-L&AC, 2015, par.1).

However, much of this research focuses primarily on the conditions themselves (dementia and stroke) rather than the disorders of speech and language, and cannot provide detailed information regarding the experiences of physicians who encounter them in primary care practice. Significant efforts have been made by the speech-language pathology community in countries such as Canada, the United States and Australia, to increase awareness of speech-language therapy as a treatment for elderly patients facing speech and language disorders and engage with physicians and other health-care stakeholders towards accomplishing that goal (Kaf et al., 2011; Finch et al., 2013).

To illustrate why there may be a dearth of more general estimates of prevalence for speech and language disorders, it is helpful to consider the complexity and difficulty of ascertaining reliable prevalence and incidence data on the several individual speech and language disorders included in this study. In his chapter on aphasia in *The Handbook of Language and Speech Disorders*, Chris Code (2010) notes that while a wealth of research has been undertaken

on prevalence and incidence of aphasia in stroke patients, there is a "lack of uniformity" in both methodology of research and clinical definitions of aphasia employed for different studies (Nicholas, 2005, 486). According to Musser et al. (2015), the incidence of aphasia in the United States is 80,000 per year among stroke patients with a population prevalence of one million (1). Speech-Language & Audiology Canada states that "up to 30%" of patients experience aphasia following a stroke (2014, par.8), which both Dickey et al. (2010) and Flowers et al. (2013) confirm, while "over 100,000" people are estimated to be "living with aphasia" in Canada (as stated previously, S-L&AC, 2015, par.9). Studies have demonstrated aphasia in patients post-stroke to be positively associated with age and female gender (Engelter et al., 2006; Ellis et al., 2010). Data concerning prevalence of aphasia caused by disease other than stroke is similarly limited. Norman et al. (2013) examined communication disorders among veterans with traumatic brain injury and determined that within their cohort of 303,716 veterans, 0.2% suffered from aphasia (1625). Among the 14% of the sample who had a traumatic brain injury, 77.1% had been diagnosed with aphasia (1625). Thus, it is possible to gather data based on etiology, however combining such data into a single prevalence statistic is difficult.

Similar issues are common in studies of dysarthria as well (Mayo Clinic, 2014). Flowers et al. (2013) determined dysarthria prevalence to be 42% (95% CI: 35-48) in stroke patients in Ontario (242), while the Royal College of Speech & Language Therapists in the United Kingdom estimate the prevalence to be 50% in stroke patients and 30-80% in "progressive neurological disorders" (2009, 4-5). Dysarthria is also common in patients with MS. Hartelius et al. (2000) estimated a prevalence of 51% (160) while Piacentini et al. (2014) reported 40%-50% (2047). Knuijt et al. (2014) found a prevalence of 56% among a sample of patients with neuromuscular disorders in the Netherlands (1286). Yorkston et al. (2009) states that 65% of patients with acute traumatic brain injury and 22% of patients in OP rehab suffer from dysarthria (3). In patients with Parkinson's disease, Pinto et al. (2004) reported prevalence of dysarthria to be 70% (547). Theodoros (2011) estimates up to 90% of Parkinson's disease patients experience disorder of speech, the most common of which is dysarthria (51). While Flowers et al. (2013) determined that "symptoms of weakness" with a first-time stroke has an odds ratio of 5.3 (95% CI: 2.4-12.0) as a predictor for dysarthria, there was no association between development of dysarthria and gender, age, comorbidity or cause of ischemia (244).

Apraxia, which inhibits a patient's ability to produce and articulate speech sounds, has minimal prevalence data in adults (ASHA, 2016). While childhood apraxia of speech has been studied more thoroughly from both a speech-language pathology perspective and within community-based settings, there has been no such effort made for apraxia in older adults. Duffy (2005) determined a sample prevalence of apraxia to be 7.6% in patients with neurologic speech disorders, making it a less common but nevertheless debilitating condition (12). Additionally, there is little evidence as to predictors of apraxia beyond predictors linked to stroke and other etiologies.

Finally, stuttering is estimated at a prevalence of 0.4% in adults over age fifty years in the United States (Craig, 2007, 1) and approximately 1% for adults over age sixty-five in Canada (Speech Language & Audiology Canada, 2005). Gender is a predictor for stuttering, with stuttering more common in males (Craig et al., 2002). The majority of research pertaining to stuttering in adults focuses on quality of life and is studied using qualitative rather than quantitative methods, creating a difficulty for researchers hoping to generate prevalence data.

Thus, due to the wide variety of causes and associated conditions, a population prevalence estimate for adults with speech and/or language disorders is very difficult to estimate. Research based primarily on qualitative methods has provided valuable insight into the experiences of patients with speech and/or language disorders in primary care settings. Murphy (2006) notes that while those suffering from disordered speech and/or language are likely to have health problems for which health care services are necessary, this population faces considerable challenges in attaining such services. The author illustrates the potentially dangerous consequences: "inadequate communication can result in wrong diagnosis; inappropriate medication and can prevent the client's access to proper assessment necessary for receiving adequate healthcare service" (49-50). Scherz et al. (1991) supports Murphy's assertions in their survey of general practice residents who stated that they lacked knowledge and comfort with speech and language disorders for the purposes of diagnosis and management. While speech-language therapy remains the best practice for treating speech and language disorders in adults (Mayo Clinic, 2014), referral rates have been low in primary care clinics (Murphy, 2006, 55). Murphy (2006) attributes a "limited understanding of the role of the speech and language therapist" as the primary cause for low referral rates (55). Efforts to address this issue have been

bolstered by recent campaigns by speech-language pathologists in Canada and Australia to promote speech-language therapy as a potential treatment for older adults for whom quality of life may improve and rate of recovery accelerated (ASHA, 2014). However, other sources suggest limited resources and access to speech therapy services may also contribute to low numbers of older adults in speech-language pathology clinics (Scherz et al., 1991)

As a final note, the term "speech and language disorders" has been employed here, rather than "communication disorders" as hearing loss and other auditory disorders in addition to voice disorders encompass a branch of research which (though very related) is often considered separately from speech sound and language disorders from an epidemiologic perspective.

IV. Primary Care and the Canadian Primary Care Sentinel Surveillance Network Database: Review of the Literature

Preliminary research of the current prevalence and management patterns of speech and language disorders is necessary before more in-depth epidemiologic analyses may be undertaken. One source of data for this type of study is electronic medical records (EMR) extracted from primary care clinics themselves. EMRs provide patient histories in the form of diagnoses, prescriptions, billing information, referrals, laboratory testing and other information recorded by the attending physician. Thus, EMRs present a comprehensive source of data, through which an accurate and precise understanding of primary care medicine may be gained.

According to Griever et al. (2013), 49% of family medicine practices in Canada used EMR software to record patient visit information as of 2010 (347). The current body of literature regarding EMR-derived data focuses on the validity and availability of such data (Virnig and McBean, 2001). A review of the literature also highlights the differences between countries with regards to clinical data and how health system configuration impacts data.

A recurring finding in the Canadian literature is the potential for inaccuracy in clinical datasets due to the multiplicity of EMR vendors in our primary care system (Tu et al., 2014). In their 2010 retrospective cohort study, Harris et al. determined that a lack of "consult letters [and] visit notes" limited the utility of EMRs for creating a diabetes case definition (351). Birtwhistle et al. (2009) also identified the absence of often critical sources of EMR-derived data, in addition to several other noteworthy challenges such as "dirty data…[including] misspelled words, extra

words, inconsistent word strings…; inconsistent data…[as in] diagnoses stored in several different places…[and an absence of] standardization" (418). Tu et al. (2014), who performed a retrospective comparison of several administrative databases in Ontario, supported Birtwhistle et al. (2009)'s assertion that "lack of standardization" (418) creates significant problems for researchers using EMR-derived data: "the quality of the data captured and the ability to identify discrete data elements in the EMR may not be the same across EMR software packages from different vendors" (Tu et al., 2014, e19-e20).

While Canada's current system of employing different EMR vendors to record patient information creates considerable difficulties at the system-level, there are also strengths and limitations inherent in EMRs as a data source. Virnig and McBean (2001) notes that clinical datasets are a low-cost alternative to study-specific data collection and are more easily acquired by researchers. Additionally, data linkage creates the possibility of using data from various types of administrative sources which is not possible with traditional data collection methods (Virnig and McBean, 2001). However, Harris et al. (2010) argue that such data may not be truly "population-based" and is biased in favour of those who choose to or are able to access health services (351).

In light of these issues in the use of EMR-derived data for research purposes, the Canadian Primary Care Sentinel Surveillance Network (CPCSSN) provides a source of primary care, EMR-derived data. CPCSSN is a network functioning across Canada to survey chronic diseases and improve quality of data for research purposes as well as care for patients (CPCSSN, 2013; Williamson et al., 2014). Primary care clinics participating in CPCSSN provide consent to have health information on patients served by their practices collected and used for academic research (CPCSSN, 2013). CPCSSN currently extracts data from 600 primary care physicians across Canada for 750,000 patients. EMR software products included in the CPCSSN database include Accuro, Bell, DaVinci, Healthscreen, Jonoke, Med Access, Nightingale, Oscar, Practice Solutions, Purkinje, Telin Mediplan and Wolff. In the Southern Alberta Primary Care Research Network, a regional network within CPCSSN specifically, extractions are made from Med Access, Wolff and Telin Mediplan. The CPCSSN database currently tracks the prevalence of eight major conditions: diabetes, osteoarthritis, hypertension, chronic obstructive pulmonary disease (COPD), Parkinson's disease (PD), epilepsy, depression and dementia (CPCSSN, 2013).

A major issue with utilizing EMR-extracted data for research is that the data is used for a purpose other than that for which it was originally recorded (Williamson et al., 2014), however CPCSSN has made significant efforts to extract EMR records and convert them into data which is reliable and useable for epidemiologic research. Data extracted from EMRs from multiple vendors, and primary care clinics is standardized using data mapping (Birtwhistle et al., 2009; Williamson et al., 2014). Data mapping is defined as a "broad technical function" which " 'matches' between a source and a target…[which] enables software and systems to meaningfully exchange patient information, reimbursement claims, outcomes reporting and other data" (McBride et al., 2006, 44). Furthermore data which contains issues such as "misspelled words, extra words in [the] field, inconsistent strings", etc. is cleaned utilizing various algorithms (Birtwhistle et al,. 2009, 418).

Databases such as CPCSSN employ case definitions for conditions of interest in the form of diagnostic algorithms (Williams et al. 2014). These algorithms comprise a "set of criteria" which can indicate 'caseness' when applied to a data set through a combination of free text items, billing codes (ICD-9), medication codes and laboratory test results (Chubak et al., 2012, 344). While diagnostic algorithms have the ability to facilitate data collection easily and accurately, there are issues inherent in their use (Chubak et al., 2012). Chubak et al. (2012) illustrate several ways in which EMR data may be of limited accuracy: firstly, patients may see a physician other than their usual family physician and data linkage between physicians in separate practices (or even health systems) is extremely uncommon; physicians may not record details of the visit factually or with regularity; ICD-9/ICD-10 codes may not effectively convey the patient's diagnosis (particularly lifestyle, risk factors and socioeconomic status) are not typically recorded. Therefore, in order to generate data of sufficient quality for epidemiologic research, case definitions utilized for epidemiology must be evaluated for validity and accuracy.

In a systematic review of validation methods in the United Kingdom, Herrett et al. (2010) determined that the "majority of validations were external [i.e. a questionnaire or record request to GP or comparison of rates of disease incidence or prevalence]" and are used to obtain a measure of positive predictive value (8). Hassey et al. (2001) adds that positive predictive value is a useful measure when used in conjunction with sensitivity and likelihood ratio(s). Nicholson et al. (2011) suggests the main issue from the perspective of the literature is "ambiguity" over the

term ‘validation’ (322). The authors explain that an ‘internal validation’ asks “Did the GP think that the patient had this condition?” whereas an ‘external validation’ questions “[whether] the GP was correct?” (322). Validation studies take both forms, often without clearly differentiating which question is the primary focus of the work (Nicholson et al. 2011). Therefore inappropriateness and incompleteness of methodology and analysis further complicates the current body of literature. In order to address the paucity of validity data for case definitions applied to EMR data (as outlined by Herrett et al. (2010) and Nicholson et al. (2011)), CPCSSN has undertaken two previous validation studies with the goal of increasing the validity of common conditions with the database and increasing the ease with which these conditions may be researched using CPCSSN data. Kadhim-Saleh et al. (2013) was a limited pilot study which validated five case definitions (diabetes, hypertension, osteoarthritis, COPD, depression) using data from a single contributing network (the Kingston-based PBRN-CPCSSN network). The authors noted that while the case definition algorithms that CPCSSN initially employed for osteoarthritis, COPD and depression had high specificity, low sensitivity was a recurring issue. Thus, prevalence may have been higher than reported for those conditions prior to implementing the current (higher sensitivity) algorithms (Kadhim-Saleh et al., 2013). In their study, Kadhim-Saleh et al. (2013) reported high specificities for all five conditions as well as high sensitivities for diabetes (100%), and hypertension (83%). Case definitions for three conditions (osteoarthritis, depression and COPD) continued to report low sensitivities ranging from 39%-45%.

Williamson et al. (2014) followed Kadhim-Saleh et al. (2013) (having found the validation methodology to be appropriate), but included data from 6 CPCSSN networks across Canada and validated case definitions for the original five conditions, in addition to epilepsy, Parkinsonism and dementia. This case definition validation not only included a significantly larger sample size (n=1920) than previous validation studies, it assessed all eight conditions using the same gold standard and included both case positive and case negative patients (Williamson et al. 2014, 371). The validation study resulted in high sensitivity (77.8 – 98.8%), specificity (93.5-99.0%), positive predictive value (72.1-92.9%) and negative predictive value (86.0-99.9%) for all eight conditions

In terms of methodology, Williamson et al. (2014) used an age-stratified approach to assess patients who were overwhelmingly older adults (90%) (368). Chart reviewers were trained to appraise 'caseness' in patients and were blinded to the diagnosis of each case by the algorithm (368). Uncertain cases were reviewed subsequently by an epidemiologist and physician. The case definitions within this study are "specifically developed for use in primary care contexts" (Williamson et al., 2014, 368) and are thus more focused on symptoms, diagnostic patterns, etc. commonly found in primary care than more broad or general case definitions.

Therefore, when undertaking a study employing EMR-derived data, it is necessary to understand and account for the challenges such data may present (particularly for less common diseases in the primary care context). While certain aspects of EMR-derived data are inherent to the source, many of issues explored above can and have been addressed by CPCSSN, resulting in data of high quality and reliability. The methodology developed for case definition validation using CPCSSN data is both rigorous and sufficiently adaptable to a variety of possible conditions.

In order to develop a case definition which could accurately assess prevalence, it was first necessary to determine what factors constitute a case in the EMR context. Compiling a case definition for speech and language disorders is challenging due to the subjectivity of symptoms and variations within disorders themselves. For example, Speech-Language & Audiology Canada (2013) define aphasia as "a language disorder…resulting in difficulty formulating, expressing, and/or understanding language" (par.4) *The Merck Manual of Diagnosis and Therapy* differentiates aphasia into two categories (Wernicke and Broca) and delves further into symptomology: "Wernicke aphasia [patients]…speak normal words fluently…but do not know meaning or relationships [resulting in]…a jumble of words or "word salad"; "Broca aphasia…patients can comprehend and conceptualize relatively well, but their ability to form words is impaired…[which] affects speech production and writing…[and] may include anomia (inability to name objects) and impaired prosody" (par. 9-12). While speech and language disorders are typically diagnosed through observing the communication of a patient and "screening" for abnormalities in speech and comprehension (S-L&AC, 2016, par. 2), there are different manifestations of speech and language problems both within individual speech and language disorders as well as between speech and language disorders. Thus, in order to account

for the symptom-based diagnosis of speech and language disorders and clinical variation, the case definition was intended to be as broad and inclusive as possible.

V. Rationale

Given the number of older patients affected by speech and language disorder in Canada, researchers and physicians must gain a better understanding of the burden of speech and language disorder in Canadian primary care clinics in order to provide effective management. There is currently very little research on speech and language disorders in older adults which focuses on primary care and community-dwelling adults. Thus, there is much uncertainty regarding the number of older adults being treated by primary care physicians for their speech and language disorder, how these disorders are being managed and what the etiology and prognosis is for those patients.

Thus, to help address the considerable gap in epidemiological knowledge regarding speech and language disorders encountered in primary care, this study involves the creation and validation of a case definition for speech and language disorder specific to adult patients and the examination of the prevalence of speech and language disorders in community-based older adult patients. The overarching goal of this study is to provide insight into a condition which poses a significant burden to both patients and caregivers, and increasingly to Canada's primary health care system.

VI. Objectives

The objectives of this study were to examine the epidemiology and clinical management in primary care of speech and language disorders in people aged 55 years and older and to assess the utility and feasibility of using EMR data to answer such research questions. In order to facilitate the objectives of this study, a case definition for speech and language disorders specific to elderly patients was formulated and validated using the CPCSSN database.

VII. Research Questions

1. What is the prevalence in primary care of speech and language disorder in Canadian patients aged 55 years and older?

2. How are speech and language disorders treated in aging populations at the primary care level?

3. Do patient characteristics or type of speech and language disorder lead to differential treatment patterns?

4. Are there differential prevalence rates or treatment patterns across regions?

5. What are the time trends in speech and language disorder prevalence from 2007 through 2014?

6. Can EMR-based data provide an effective means to answer epidemiological questions regarding speech and language disorders at the primary care level in the aging and elderly population?

CHAPTER 2: Study Design and Methods

I. Design

This study employed a retrospective cross-sectional validation design with methodology based on Williamson et al. (201). The case definition was formulated *a priori* and focused on four disorders: three speech disorders (apraxia, dysarthria, and stuttering), and one language disorder (aphasia).

II. Development of the Case Definition

The process of developing a case definition for speech and language disorders in older, community-dwelling adults began by reviewing the literature, including research papers, medical textbooks, and web resources such as *The Merck Manual of Diagnosis and Therapy*, and educational resources published by professional organizations such as the American Speech-Language-Hearing Association and Speech-Language & Audiology Canada. As the case definition was intended to be applied to the CPCSSN database containing EMR-derived data, we supplemented the literature review with an exploratory search of the CPCSSN-SAPCReN database to identify any relevant keywords missed previously. These search terms included

"aphasia," "dysphasia," "dysarthria," "apraxia," "stuttering" as well as more general terms such as "speech disorder" or "speech problems." This exploratory search resulted in the identification of approximately 150 patients, a large number of whom were diagnosed with non-specific or undefined disorder of speech and/or language. To facilitate the inclusion of such patients, thereby widening the parameters of the case definition, an additional section for "undefined" speech and/or language disorders consisting of text phrases identified during the exploratory search was included.

Subsequently, a Speech-Language Pathologist (TH) was consulted to confirm the appropriateness and completeness of the case definition. Commonly related comorbidities and conditions which cause disordered speech and/or language (such as Parkinson's disease or cerebral hemorrhage) were not considered valid evidence of "caseness" without a specific text reference or ICD-9 code. Furthermore, the case definition does not include dysphagia (a swallowing disorder) or congenital anomalies such as cleft palate in the absence of a text or billing code reference specific to an aforementioned speech and/or language disorder. Finally, referral to a speech-language pathologist was not considered sufficient evidence for 'caseness' unless a defined text phrase or ICD-9 code was included. The decision to exclude referrals as evidence for 'caseness' relates to the limited scope of referral data within the CPCSSN database, which results in the user's inability to determine the outcome of a referral visit.

Table 1: Case Definition for Speech and Language Disorders in Adults over 55 years

Text Terms and Phrases	ICD-9 Codes
Aphasia • aphasia; broca; wernicke; "transcortical" "motor"; "transcortical" "sensory"; dysphasia	784.3; 315.31; 438.11
Dysarthria • dysarthria; "slurred speech;" "slurr* speech"	784.5
Apraxia • apraxia	438.81
Stutter/Stammer • "stutter"; "stammer"	307
Undefined speech disorders • "speech" "disturb*"; "speech" "problem"; "speech" "disorder*"; "language" "disorder"; "communication" "disorder"; "nonfluent" "speech"; "speech "deficit"; "speech" "difficult*"; "speech" "challenge*"; "speech – disturb";	438.1*; V40.1

"speech" "disabilit*"; "language" "disabilit*"; "speech" "issue*"; "expressive" "language" "disorder"	

refers to terms for which all related forms of the word was searched and included by the algorithm (e.g. "disorder" would include "disorder" or "disorders")

III. Development of a "Gold Standard"

The "gold standard" for this validation study was a review of patient charts using remote EMR access. Reviewers familiarized themselves with the case definition's terms and billing codes and discussed any questions or concerns pertaining to the definition. A CPCSSN Data Manager (DJ) created an electronic checklist using the case definition, whereby each of the five categories was listed with the components (list of text phrases and ICD9 codes) of each category organized in bullet-point form. Reviewers had the ability to check "Yes" or "No" for presence or absence of each component. In addition to this checklist, reviewers could check "Yes" or "No" for the final decision for each chart as a case or non-case. The gold standard for this study was subsequently employed to estimate prevalence in the study sample.

IV. Population

EMR-derived data were extracted from SAPCReN (Southern Alberta Primary Care Research Network), a regional primary care research network within CPCSSN which includes primary care clinics in Calgary, Lethbridge and rural sites. At the time of sampling, there were 44,590 total patients aged fifty-five and older in the SAPCReN database, which includes patients active in status, inactive and deceased.

V. Validation of the Case Definition

Sample

A sample of 'sentinels' (primary care physicians or nurse practitioners participating in CPCSSN from whom EMR data is gathered) was contacted with the purpose of gaining permission to access patients' charts, at a distribution of approximately 60 charts per clinic. Power analysis using the Wald one sample proportion test at an expected prevalence of 13% for patients aged fifty-five and older indicates at least 80% power for a sample of 1000 with 0.05 significance. This sample was drawn on September 30, 2014, at which time 30, 215 patients

were aged fifty-five and older and had visited a SAPCReN-CPCSSN physician within the previous two years (i.e. were considered 'active' patients).

Patients eligible for inclusion in this study were male and female adults registered as a patient (i.e. active in status) of a SAPCReN – CPCSSN physician who were aged fifty-five and older. Exclusion criteria were defined as age younger than fifty-five years at the time of chart extraction (December 2014). In light of the sample size calculation, a sample of 1000 patients aged fifty-five years and older was selected at random within the records of the participating sentinels. The EMRs of these people were then reviewed directly in order to identify those who met the case definition for speech and language disorders.

Unforeseen technical issues occurred mid-way through the study, which involved loss of access to charts managed by one EMR vendor (Med Access). This vendor reconfigured the search interface, removing certain search fields (including the field which reviewers used to search for patient charts) after the chart review was underway. This resulted in the loss of reviewers' ability to access the charts. The solution for this issue was a partial re-randomization of sample charts from sites using a different EMR vendor (Wolff), which occurred in February 2015. The charts which had been completed from sites using Med Access were kept for analysis and are included in the sample. For this reason, more charts were available for review (and thus included in the sample) than was originally estimated as necessary to demonstrate statistical significance.

Chart Review

The "gold standard" for the case definition validation was a chart review performed by five independent and trained reviewers (RL, SR, HF, MD, DS) using remote EMR access. Reviewers were trained in the identification of speech and/or language disorder cases, as well as how to navigate the EMR and use the electronic case validator tool for data collection. This chart review was undertaken from January to June 2015. The reviewers considered each chart for evidence of 'caseness' and identified the chart as either "Yes" (for cases) or "No" (for non-cases).

All sections of the EMR were reviewed for evidence of 'caseness' with particular emphasis placed on Health Condition (a.k.a. Patient Profile or Problem List), Encounter

Diagnosis (a.k.a. Visits) and Billings (a.k.a. Bills), and evaluated according to whether the attending physician had diagnosed the patient with a speech and/or language disorder within the broad limits of the CPCSSN case definition. The review involved reading through all relevant sections of the EMR, including the physician's "SOAP notes" for any evidence of caseness, whether in text or ICD9 code format. "SOAP" is an acronym for 'Subjective, Objective, Assessment and Plan' which refers to a "problem-oriented tool" and "predominant documentation format" for health care providers wherein "patient's physical history, physical exam, diagnosis, and plan of care" may be easily and systematically recorded (Pearce et al., 2016, 29-30). The final decision made by the reviewer vis á vis caseness was recorded on an electronic case validator tool created by CPCSSN, along with any comments the reviewer may have had about the decision. This tool listed each item (text and billing code) contained within the case definition and allowed reviewers to check "Yes" or "No" for each, along with whether the patient was a speech and/or language disorder case. No personal or identifying information was collected during the review.

Reviewers were blinded to the classification of the chart as a case or non–case according to the CPCSSN algorithm. However, reviewers were able to discuss charts and reach consensus on caseness during the review process as well as after the review had been completed. To assist in reaching consensus for potential cases, a speech-language pathologist provided guidance and evaluated caseness in charts for which there was uncertainty. Cases deemed suspect were initially recorded, with the final decision made by the speech-language pathologist. Additionally, the five reviewers held regular biweekly meetings as well as ad hoc meetings as necessary to discuss any questionable cases, concerns, etc.

Application of the Case Definition Algorithm

Following completion of the chart review, the case definition was translated by a CPCSSN Data Manager into a computer search algorithm. The algorithm was applied to patients in the randomized sample identified using a CPCSSN-assigned identification number within the CPCSSN database and thus identified those it assessed to be cases. Comparison was then made between those identified as cases by the CPCSSN search algorithm and those identified as cases in the 'gold standard' chart review.

VI. Statistical Analysis

Validation analysis consisted of comparing the number of cases identified by the chart review to the number of cases identified by the algorithm in the same sample of patients. The measures used to determine validity included a cross-tabulation to summarize the findings, sensitivity, specificity, positive predictive value (PPV) and negative predictive value (NPV) with 95% confidence intervals. The case definition was set to be considered valid at 70% sensitivity and specificity.

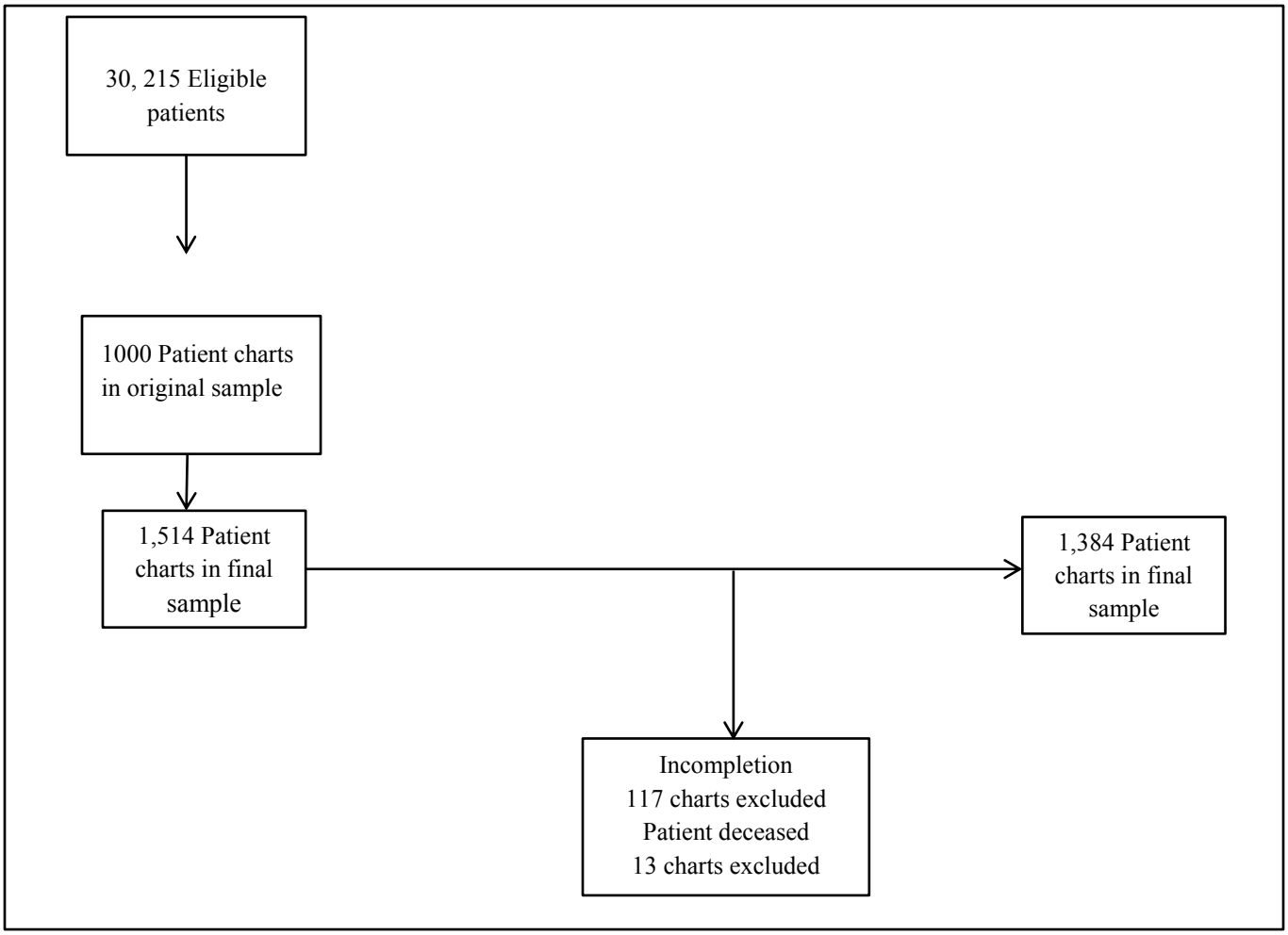
Inter-rater reliability was assessed using a random sub-sample of 10 EMR charts to ensure that reviewers were consistent in their assessment of 'caseness.' Due to the technical issues involving loss of access to Med Access charts, only three of the five reviewers were able to participate in the inter-rater reliability check. Fleiss' kappa coefficient was used to indicate agreement between reviewers.

All analyses were performed using the statistical software Stata IC 13, except for calculation of the Fleiss' kappa, for which SAS was employed.

CHAPTER 3: Results

Of the total SAPCReN population of 44,590, 30, 215 patients met the inclusion criteria of being aged fifty-five and older and had visited a SAPCReN-CPCSSN physician within a two-year timeframe at the time of sampling. While 1000 patients were initially included in the sample, the second sampling resulted in the inclusion of an additional 514 patients. Thus the chart review included 1514 patients in total.

Figure 1: Study Flow Diagram



Following the chart review, 117 charts were excluded due to lack of data. This included charts which were incomplete when access to Med Access charts was lost as well as those found to be unsearchable in the EMR databases due to lack of identifiers. Additionally, 13 deceased patients were excluded from the study sample. Thus statistical analysis included 1384 patients in total.

Table 2 summarizes patient characteristics of the final sample. It consisted of 28.6% more female patients than male patients with a mean age of 67.5 years. Site 8 contributed the highest number of patient charts (22.5%) followed by Sites 4 (19.8%), 5 (19.7%) and 6 (19.4%) respectively.

The case definition and its search algorithm for speech and language disorders identified a prevalence of 1.2% within the sample (95% CI: 0.66%-1.87%). While specificity was favourable at 99.9% (95% CI: 99.6%-100%), sensitivity was considerably lower at 18.8% (95% CI: 4.05%-45.6%). Positive and negative predictive values were determined to be 75.6% (95% CI: 25.4%-96.6%) and 99.0% (95% CI: 98.8%-99.2%) respectively.

Fleiss' kappa statistic for speech and language disorders was -0.034, p = 0.57.

Table 2: Patient Characteristics

Sample	N = 1384	
Sex	Male 35.7% (n=494)	Female 64.3% (n=890
Age (years)	Mean: 67.5 Min.: 55	Std. Dev.: 9.9 Max.: 103
Site	1 2 4 5 6 8	9.5% (n=132) 9.1% (n=126) 19.8% (n=274) 19.7% (n=273) 19.4% (n=268) 22.5% (n=311)

Table 3: 2x2 Table

	Chart Review Case	Chart Review Non-Case	Total
Algorithm Case	3	1	4
Algorithm Non-Case	13	1367	1380
Total	16	1368	1384

Table 4: Validation Analysis

Prevalence	1.2% (n=16)	95% CI: (0.66% – 1.87%)
Sensitivity	18.8%	95% CI: (4.05% - 45.6%)
Specificity	99.9%	95% CI: (99.6% - 100.0%)
Positive Predictive Value (PPV)	75.6%	95% CI: (25.4% - 96.6%)
Negative Predictive Value (NPV)	99.0%	95% CI: (98.8% - 99.2%)

CHAPTER 4: Discussion

I. Case Definition Validation: Discussion of Methods

To our knowledge, there have been no previous studies in which case definitions for speech and language disorders have been validated in the older adult primary care population. Thus it is not currently possible to compare our results to those of other research. However, the results of this study do impart several key findings to researchers concerned with epidemiological methods in the areas of EMR-derived medical database usage and case definition validation.

The case definition validated in this study has very high specificity, which suggests it is effective at ruling out non-cases of speech and language disorders. The low sensitivity however, indicates it is ineffective at ruling in among patients who have a speech and/or language disorder. Thus, as the sensitivity did not meet the acceptable minimum of 70% (Williamson et al., 2014, 368), this case definition cannot be considered valid.

The low sensitivity reported for this case definition may have resulted in several ways. Firstly, there may have been a "misclassification" by the algorithm due to evidence of caseness found in fields of the EMR from which data is not typically extracted (Kadhim-Saleh et al., 2013, 165). For example, physician's SOAP notes are not extracted by CPCSSN due to privacy/confidentiality issues. Referral data is found within the CPCSSN database, however only the type of referral (e.g. neurology, gerontology, etc.) is recorded and reasons for referral are not typically listed. Therefore there may be a possibility for evidence of caseness found by the chart reviewers but not by the algorithm because the data were not extracted into the CPCSSN database. This is however, an unsatisfactory and unlikely explanation for our low sensitivity. It is reasonable to assume that symptoms of speech and/or language disorder or a diagnosed speech and/or language disorder would warrant a mention by the physician in the encounter field (Encounter Diagnoses), particularly if the issue was sufficiently severe as to lead to a referral to a specialist.

A second possible reason for low sensitivity is misclassification in the chart review. After the statistical analysis was completed, the CPCSSN data for the thirteen discrepant cases was analyzed to determine likelihood of error on the part of the algorithm. Of those thirteen

discrepant cases, only one was found to have clear evidence of a speech and/or language disorder and can therefore be considered a misclassification by the algorithm. Among the remaining cases, five showed evidence of a stroke, one case showed evidence of dementia and one was referred to a neurologist for an unknown reason. Two showed no evidence to suggest a speech and/or language disorder nor a related condition or disorder. Two cases showed evidence of voice disorders but nothing else related to speech and/or language disorders and related conditions, such as stroke. These two cases may be the result of misclassification on the part of the chart reviewers as voice disorders were not considered evidence of caseness within our current case definition.

Misclassification may occur at the clinic-level as well. Benchimol et al. (2011) and Kadhim-Saleh et al. (2013) note, the quality and completeness of EMRs rely completely on the discretion of the physician. Physicians may find it more efficient or accurate to favour certain billing codes over others when billing for a patient visit (Kadhim-Saleh et al., 2013). Thus there is a risk of "under-reporting" or differential reporting of certain conditions or billing codes (Kadhim-Saleh et al., 2013. 165). However, as mentioned previously, one would nevertheless expect some indication of caseness to be found in text within the patient encounter, health conditions, etc. in the absence of a billing code.

Unfortunately, the source of low sensitivity for this case definition remains unclear. It appears to be a combination of misclassification on the part of the chart reviewers and the algorithm (though to what extent each occurred cannot be estimated with certainty).

As stated previously, the Fleiss' kappa statistic was too low to provide any indication of the inter-rater reliability of the chart review. This low statistic is likely due to a "paradox" of Cohen's kappa and Fleiss' kappa whereby high inter-rater agreement and low prevalence of the condition in question may result in a low kappa statistic (Feinstein and Cicchetti, 1990, 543).

Falotico and Quatto (2014) suggest the Fleiss' kappa index may "assume a constant and negative value" due to the "paradoxical" (here referring to the "phenomena" of reporting low kappa despite high inter-rater agreement effects on data for low prevalence conditions) (466). Thus, while used often by researchers to demonstrate inter-rater reliability with multiple raters, Fleiss' kappa is insufficient in certain circumstances, such as in this study where prevalence is

very low. Fleiss' kappa was employed due to its usefulness for measuring agreement of more than two raters (Falotico and Quatto, 2014), however prevalence was determined to be significantly lower than expected. Given the limitations of the Fleiss kappa, other statistics were considered to evaluate inter-rater reliability, including Gwet's AC1 and intraclass correlation. However, after consulting with a statistician (TW), the possible alternatives were each deemed to be inappropriate and/or inapplicable to the data.

II. Prevalence of Speech and Language Disorders: Discussion of Findings

At its inception, this study aimed to answer six questions related to the epidemiology of speech and language disorders in older adults in a primary care context. The methodology proposed for this undertaking was a combined case definition validation and cross-sectional prevalence study utilizing EMR-derived data from the CPCSSN database. In this discussion, I will address the six research questions for this study and their implications for future epidemiologic work.

The first and primary objective for this study was to determine prevalence in community-dwelling primary care patients of speech and language disorders in patients aged fifty-five years and older. In this study, we determined the sample prevalence of speech and/or language disorders in older, community-dwelling primary care patients to be approximately 1.2%. This statistic is significantly lower than the 13% statistic published by Speech-Language & Audiology Canada. Thus, it is necessary to examine what may have contributed to this disparity.

Speech-Language & Audiology Canada's 2005 prevalence estimate for Canadians aged 65+ is one of very few statistics available on older Canadian speech and language disorder patients at the population level that is not specific to stroke or other related conditions. Additionally, the authors have not limited the scope of prevalence estimation by location of care (i.e. long-term care, acute care, etc.), thus providing considerable value to researchers pursuing the epidemiologic study of speech and language disorders in Canada as a whole.

Speech-Language & Audiology Canada's 2005 prevalence estimate is based on a "composite" drawn from Statistics Canada, the American Speech-Language-Hearing Association as well as "provincial speech-language pathology and audiology associations" from 1996 (S-L&AC, 2005). In addition to being out-of-date for epidemiologic use, this statistic likely

represents a population which differs from the community-dwelling population of older adults in terms of disease severity and possibly overall health. Researchers in the speech-language pathology community have argued that older and elderly people are under-represented in speech-language pathology services (Hopper et al., 2007) and much of the research focusing on this population is specific to long-term or acute care, rather than community-dwelling older persons. Therefore the population included in Speech-Language & Audiology Canada's 2005 statistic may be comprised of patients with poorer overall health and increased disability than may be found in community-dwelling adults.

Another issue with the 2005 statistic is that it fails to detail the criteria upon which speech and/or language disorders have been evaluated. Primarily, it would be important to note whether episodes of speech and/or language disorder which were short-lived and not recurrent were included. If so, in the case of stroke, there may be greater numbers of speech and language disorders in older patients who are presenting symptoms in acute care, but would not be reported to the family physician following a stroke. It follows then that prevalence of speech and language disorders in primary care-based older adults may be lower due to fewer patients with severe illness, fewer with ongoing disability and higher levels of recovery in adults who have experienced illness. Moreover, while the 2005 statistic published by Speech-Language & Audiology Canada is the best available estimate for prevalence, attempting to extrapolate such a statistic for further epidemiologic research on speech and language disorders in older Canadian adults may be prone to selection bias, rendering comparison between this statistic and others potentially spurious. In light of these issues, Speech Language & Audiology Canada has not published an updated statistic of this kind using recent data, instead presenting a number of population-specific statistics. Neither the older (2005) statistic nor the recently published data (2014-2015) provide sufficient information on speech and language disorders in specific settings of care. The prevalence estimate determined in this study has succeeded in filling a part of this gap in epidemiologic knowledge. Another possibility is that our prevalence estimate, which is not based on survey data or other data sources open to recall and other forms of bias, is a closer representation of speech and/or language disorders in Canadian older adults than the statistic published by Speech-Language & Audiology Canada.

The following four research questions related to the diagnosis and management of speech and language disorders in primary care: how are these disorders treated; do patient characteristics affect the type treatment; does prevalence or treatment patterns differ by region; and have prevalence rates changed between 2007 and 2014? Because this study did not progress to the stage of applying the case definition to a national data set, we are unable to answer these questions.

The final question asked whether EMR-derived data can provide an effective answer to epidemiologic questions regarding speech and language disorders at the primary care level for older adults. To answer this question, it is necessary to consider the issue in two ways: firstly, what is the utility and practicality of using technology in the form of databases and computer algorithms to facilitate research; secondly, what are the strengths and limitations of epidemiologic case definition validation methods? In determining the feasibility of a data source to answer epidemiologic questions, it is necessary to identify the limitations of the data source itself, as well as the appropriateness and constraints of the methods used to study data from a given source. We concluded that low sensitivity may have resulted from "misclassification" of cases by the computer algorithm in which the algorithm misclassified cases as non-cases. The most reasonable explanation for the high degree of apparent misclassification by the computer algorithm is that it is due to the lack of "concrete" or "objective" measures (Kadhim-Saleh et al., 2013, 165) for speech and language disorder diagnoses, resulting in a wide variety of possible text items. In this case, the chart reviewers may have been able to employ other aspects of the chart not readily available to data extraction - in order to infer the physician's intended diagnosis, whereas the algorithm must rely on the specific text phrases listed. Kadhim-Saleh et al. (2013) note that one potential cause for poor sensitivity in their case definition for osteoarthritis was the finding of evidence only in the results of radiographs; similarly it is possible that findings of speech and/or language disorders in referral letters (etc.) led to the determination of some charts as cases in the chart review and non-cases by algorithm where extraction of such information may be less robust. Additionally, as this was a study of primary care patients where population prevalence of speech and language disorders is likely to be lower than other care settings due to lower severity of illness and disability (Williamson et al., 2014), specificity is expected to be higher (Berry et al., 2010, 2). Finally there is a possibility for misclassification by chart reviewers such that errors are found in the 'gold standard' rather than the algorithm. There is

some evidence of this possibility due to findings of evidence for voice disorders in the CPCSSN data (not part of this case definition) in which no further evidence of speech and/or language disorder Thus in this case, the algorithm may have been correct, and the chart review incorrect. While the chart review is considered the gold standard, it is necessary to consider any possibility for human error.

Other studies of diseases with "subjective" diagnoses (Kadhim-Saleh et al., 2013) have reported analogous issues to those illustrated above. Beauchamp et al. (2015) determined that a "lack of a quantitative definition" generated significant challenges in the researchers' attempt to validate a clinical case definition for neuropsychological impairment in pediatric study centers using standardized neurological test batteries (2). The authors discussed how "heterogeneity" in the type and severity of impairment had often deterred the creation of an overarching clinical case definition (2). In Berry et al. (2010), the authors created and validated several configurations of questionnaire items for a case definition for interstitial cystitis/painful bladder syndrome to be validated in patients of urologists and gynecologists. Like speech and language disorders, interstitial cystitis/painful bladder syndrome is diagnosed symptomatically and suffers from lack of awareness among physicians and "wide variability" in diagnosis methods due to a lack of "objective marker[s]" (1848-9) as well as low population prevalence. Thus, while case definition validation studies for diseases including diabetes and hypertension are larger in number and employ definitions which often perform with higher sensitivity and specificity, validation studies for diseases lacking objective or quantitative criteria suffer considerable methodological challenges (Kadhim-Saleh et al., 2013).

While we were not able to fulfill all six of our objectives for this study and provide a more comprehensive contribution to epidemiologic knowledge of speech and language disorders in older persons, this study did succeed in generating a more rigorous estimate of prevalence and validating a case definition which may be effective in clinical settings. Additionally, this study contributes to the epidemiology of speech and language disorders by showing the methodological benefits and issues of validating case definitions, particularly in primary care settings. This is of particular importance, as many definitions for speech and language disorders in research lack sufficient validation using reliable and rigorous methods. Future epidemiologic research into the condition would also benefit from the use of other methods, such as properly

constructed sample surveys including clinical examination. While these expensive, these methods may help alleviate the risk of misclassification by supplementing and bolstering EMR-derived data. As Canadians age in greater numbers and the number of people with conditions related to disordered speech and language increase, it is highly necessary to progress from the current use of composite statistics and wide variations in case definitions for speech and language disorders. Similarly, the epidemiologic methods employed to study such inter-related conditions as speech and language disorders must advance to meet ever-evolving research needs.

In light of our findings with regards to prevalence of speech and language disorders in older primary care patients in Alberta, it is also necessary to consider the implications for planning and policy-making. Our prevalence estimate may suggest that the current estimate for prevalence released by Speech-Language & Audiology Canada over-estimates prevalence in certain care settings – such as primary care.

When discussing policy implications relating to population health, I refer specifically to health outcomes including "length of life and health-related quality and function of those life years)" (Kindig and Stoddart, 2003, 381) - how policymakers and planners can best employ health care resources and direct delivery of health care services to maximize the length and quality of life of older patients. As this study reported speech and language disorders to be of relatively rare occurrence, we must therefore consider where the remaining expected 12% of older Canadian patients with speech and/or language disorders are found. It may be the case that 13% is simply an over-estimation of the burden of speech and/or language disorders in Canadian older adults, at least as a long-term or chronic condition. However, due to the association of speech and/or language disorder with increased severity and disability in stroke (the most common cause of acquired speech and/or language disorder in adults) (Flowers et al., 2013; Dickey et al., 2012), of patients with speech and/or language disorder, more patients may inhabit long-term care facilities rather than reside in the community (and therefore in primary care clinics). Researchers in health economics often differ in their views of whether disease severity should dictate how health care resources are distributed, and to what extent (Gandjour, 2012), particularly when there are shortages of speech-language pathologists to provide needed services (Alberta Health and Wellness, 2007). As speech-language pathology services are the primary (and only effective) treatment for speech and language disorders, it is necessary to consider what

role speech language pathology should adopt in the primary health care "team" (Herbert, 2005, 1) for older adults. In their 2016 report on speech-language pathology scope of practice, the American Speech-Language Hearing Association notes several important aspects of professional practice include "screening[,]assessment..and advocat[ing] for…individuals through…community awareness, prevention activities, health literacy, academic literacy, education, political action, and training programs" (ASHA, 2016, par. 11, 15), which may provide an effective interaction between speech-language pathologists and primary care, in addition to providing speech-language therapy to patients diagnosed with speech and/or language disorders.

Another important implication of this study is the ability of EMR-derived databases to track diseases such as speech and/or language disorders. This study provides evidence for the ability of EMR-derived databases to track diseases such as speech or language disorders, while highlighting the need for increased uniformity and accessibility between and within EMR software. Data linkage to acute care databases is an activity already underway within CPCSSN networks and could add important information to further contextualize the data extracted from EMRs.

Ultimately, implications of policy for this study are difficult to address in a meaningful way without addressing perhaps another key conclusion of this study: the need for further epidemiologic research in Canadian older adults with speech and/or language disorders. Primary care-based research of older adults with speech and/or language disorders could provide important insight into those patients recovering from strokes, coping with dementia, Parkinson's disease or the effects of traumatic brain injury in the community and better inform management and treatment. Future research could include a study of physicians' recording styles and practices in EMRs for patients exhibiting speech or language issues.

III. Strengths and Limitations

This is the first case definition validation study for speech and language disorders in older adults specific to the primary care setting. Though unsuccessful for use in epidemiology in its current state, given the high PPV and NPV values the case definition may be useful in other research or clinical settings or as a starting point from which future case definition development

may proceed. As yet, there are no validated case definitions for speech and language disorders in older adults in use in the primary care setting. However, speech and language issues are important signs of chronic diseases such as dementia as well as markers of recovery after stroke or traumatic brain injury (Mayo Clinic, 2014), which are conditions often managed in primary care (Wong and Stewart, 2010). Without a formal and rigorous method to assess who is included in this population, researchers cannot adequately study it. With this goal in mind, this case definition provides a means of assessing speech and language disorder patients as they may present in primary care and does not rely (but may be modified to include) linkage to other care settings or health practitioners. This case definition may also be further modified to a symptom-based format or further expanded to include voice disorders in future studies.

Another strength of this work is the use of a sample representative of the primary care population. Eligibility for this study did not require patients be referred to a speech-language pathologist, which may result in selection bias based on severity of illness or accessibility to treatment. Our sample included both urban and rural clinics as well as both community and academic-oriented clinics. Finally, as Canada currently lags behind other western countries in studying speech and language disorders in older adults from an epidemiologic standpoint, this study provides an exploratory analysis for future epidemiologic work. Evidence-based medicine is currently promoted as a best practice for patient care by the Canadian Medical Association and Alberta College of Family Physicians (CMA, 2009; CFPC, 2004).

Finally, this study is one of very few to attempt to estimate the prevalence of speech and language disorders in community-dwelling older adults in the primary care context. The prevalence estimate generated here differs from similar research in that it focuses on one specific population in terms of health care service utilization, while combining different populations based on health condition and "clinical variant" of speech and/or language disorder (Dyukova et al., 2010, 593). Additionally, the prevalence we have estimated has been calculated from a single data set and thus does not need to contend with issues of dissimilar methodology or clinical criteria for disease status as other studies and publications which rely on "composite" (S-L&AC, 2005) or aggregate prevalence estimates are required to do.

This study has several limitations. Firstly, technical issues involving one of the EMR systems resulted in a disruption of the chart review and forced us to obtain a sample which was not evenly distributed between sites (and thus EMR systems), as intended.

Secondly, as we were unable to investigate the patient charts following completion of the chart review, we cannot accurately determine whether misclassification occurred for false negative cases. On a related note, speech and language disorders lack quantitative or purely "objective" criteria upon which a diagnosis may be made, thus there are no medications or laboratory tests which lend themselves well to study within EMR databases such as CPCSSN. In the absence of such measures, chart reviewers may have been more likely to designate caseness in error for a speech or language disorder than they might for a disease such as diabetes where definitive medications, laboratory tests exist and billing codes are more frequently employed (Kadhim-Saleh et al., 2014).

CHAPTER 5: Conclusion

This study examined several issues regarding epidemiologic methodology and challenges inherent in administrative data sources. Through the course of the study, the advantages and limitations of EMR-derived data to analyze symptom-based diseases in primary care have been highlighted and the current state of epidemiologic knowledge regarding speech and language disorders in an older, community-based population evaluated. As other researchers have stated, including Code (2010), the body of literature on this topic is in many respects too heterogeneous in both population and methodology to generalize to a variety of populations. Speech-Language & Audiology Canada previously compiled an estimate of prevalence which is broader in scope than most others, but is nevertheless a composite statistic based on discordant methodology, dissimilar populations and somewhat vague diagnostic criteria. In this study, we have generated an estimate of prevalence resulting from a more widely applicable population (i.e. adults in primary care) which is less homogenous and prone to selection biases than studies based in speech-language pathology clinics, acute care or long-term care facilities.

This study has also created a case definition for speech and language disorders in older primary care-based adults which may be effective for clinicians due to its high positive and

negative predictive values. While it did not have sufficient sensitivity for validity in the population studied, the case definition may have greater measures of validity in other populations, such as acute care, where prevalence of speech and/or language disorders is likely to be greater. This case definition is one of very few to be validated for use in epidemiologic research.

Finally, this study has demonstrated some limitations of epidemiologic methodology in populations where the disease studied is of low prevalence. In particular, we have shown how kappa statistics (Fleiss and Cohen's) are inappropriate in such circumstances. As kappa is a widely used and well-known measure of inter-rater reliability, such a limitation is very important for researchers to understand.

While this study did not achieve all of its goals, as defined by our six research questions, the results and conclusions found are important contributions to both the epidemiology of speech and language disorders as well as epidemiologic methodology. This study also establishes the immediate need for research on the population of adult primary care patients with speech and language disorders, about who far too little is currently known.

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