Predictors of Home Care Costs among Persons with Dementia, ALS and MS in Ontario

by

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DECLARATION

I hereby declare that I am the sole author of this thesis. This is a true copy of the thesis, including any required final revisions, as accepted by my examiners.

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ABSTRACT

Purpose: The purpose of this project was to look at the costs of individuals with Alzheimer's disease and related dementias (ADRD), ALS, and MS in long stay home care in Ontario, Canada. The specific goals were to produce estimates of costs for these individuals, as well as identify clinical and personal characteristics associated with these costs. This project also tested the effectiveness of the Resource Utilization Group for home care case-mix system for use in these special populations.

Methods: This project was conducted using a secondary analysis of assessment data from the Canadian Staff Time Resource Intensity Verification Project, a 13-week study of home care costs (N=435 141). The project was guided by the Andersen and Newman (1973) framework for healthcare resource utilization. Descriptive characteristics and mean costs were produced using bivariate frequency and means procedures for each of three conditions. Predictors of costs were identified for each of the three neurological conditions through multivariate regression analysis conducted separately for each condition. In total 41 independent variables were included into the bivariate and multivariate analyses. The dependent variable was the total weekly formal and informal home care costs across all multivariate analyses.

Results: In total, ADRD, ALS, or MS diagnoses were present in 16% of the assessments. The mean costs for the three conditions combined were $594.81. The mean costs for ADRD, ALS, and MS were $593.32, $898.41, and $574.92, respectively. Characteristics that were predictive of cost across all conditions included the Resource Utilization Group for home care case-mix system, ADL functionality, IADL functionality, cognitive performance, unsteady gait, stair use, difficulty swallowing, respiratory challenges, and bowel incontinence. The Resource Utilization

Group for home care case-mix system had the highest level of explained variance of any single item tested in this project across all conditions. However, other clinical characteristics also contributed substantial levels of explained variance to the models for each of the three conditions.

Conclusions: The findings from this project suggest that although diagnosis of ADRD, ALS, and/or MS can describe cost, clinical characteristics are the most important predictors of costs for individuals with these conditions. In addition, the Resource Utilization Group for home care case-mix system can adequately predict costs of individuals with these conditions. The addition of some clinical characteristics would likely improve the predictive abilities of the Resource Utilization Group for home care case-mix system.

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1.0 INTRODUCTION

Neurological conditions are disorders of the central and peripheral nervous systems. In general, few of these conditions are curable, many are both chronic and progressive, and most are clinically complex (Canadian Institute for Health Information [CIHI], 2007; Grima et al., 2000; LePen et al., 1999). A large number of neurological conditions exist, and while conditions such as strokes, migraines, Parkinson's disease and Alzheimer's disease are common, the majority of neurological disorders are quite rare (MacDonald et al., 2000). Overall, the lifetime prevalence of the neurological conditions has been estimated at 625 per 100 000 (MacDonald et al., 2000), but result in substantially greater disease and financial burden because of associated impairments and disabilities (Gustavsson et al., 2011a). According to the World Health Organization (2006), neurological conditions account for six percent of the global burden of disease, and 11 percent in developed countries. In Canada, CIHI estimates that one million Canadians live with some type of neurological disorder, costing roughly $9 billion a year, or 6.7 percent of direct medical expenditures on hospital care, physician care, and drug expenditures alone (CIHI, 2007).

Although estimates of cost for the neurological conditions are typically for hospital and acute-care settings, a substantial amount of care for individuals with these conditions occurs in non-acute care settings. Depending on the region(s) affected, symptoms can include cognitive problems, gait ataxia, muscle spasticity or rigidity, dyspnea, or involuntary movements, resulting in functional limitations. In the past, many of these conditions, their symptoms, and eventual limitations were addressed through hospitalization followed by institutionalization (Lacey, 1999). A combination of health policy shifts and changes in the care for individuals with these conditions has resulted in greater home care use (Coyte, 2000; Lacey, 1999). Due to functional

limitations, services required by these individuals include personal support, occupational therapy, speech language pathology, physiotherapy, nursing, and social work services, all of which are available in home care settings. Overall, these services have been found to be equally effective as services provided within hospitals and long-term care facilities for individuals with neurological conditions (Barnes and Radermacher, 2001). In addition, those with neurological conditions typically prefer to remain in their homes (Barnes and Radermacher, 2001). Finally, the prevalence of neurological conditions is expected to rise (WHO, 2006). Altogether, the demand for home care services by individuals with neurological conditions is expected to grow in coming years.

Despite the importance of home care for individuals with neurological conditions, very little is known about the costs of these conditions, and service needs for individuals within this care setting. With a rising prevalence in the neurological conditions coupled with an increasing reliance on home care services, a better understanding of the resource utilization patterns of these individuals is required in order to adequately plan for the expected rise in home care use by individuals with these conditions. While a small number of studies have attempted to estimate and identify predictors of costs associated with various neurological conditions, constraints in the availability of data have limited the number and types of studies that can be carried out in this research area. In addition, no previous research has attempted to estimate the costs and identify predictors of these costs within the home care setting. With the availability of data from the Resident Assessment Instrument for Home Care, which collect person-level data on over 200 clinical variables, and corresponding cost estimates from the Canadian Staff Time Resource Intensity Verification Project, the aim of this project ws to produce reliable estimates of cost for three common neurological conditions, as well as determine clinical predictors of costs for these

conditions. The three neurological conditions considered include Alzheimer's disease and related dementias (ADRD), amyotrophic lateral sclerosis (ALS), and multiple sclerosis (MS).

2.0 BACKGROUND

2.1 The Neurological Conditions

Neurological conditions are disorders of the central and peripheral nervous system, which include the brain, cranial and peripheral nerves, nerve roots, autonomic nervous system, neuromuscular junction, and muscles (WHO, 2007). Depending on the condition, these neurological disorders can be common, can affect various age groups, and can be costly. The three clinical conditions chosen for this project represent these aspects. ADRD was chosen for this project because of its high prevalence in elderly populations, MS due to its substantial burden on younger populations, and ALS because it is amongst the most costly of neurological conditions. Symptoms associated with each of the three neurological conditions can be found in Table 1. More detailed clinical descriptions for each of the three conditions included in this project can be found below.

Alzheimer's Disease and Related Dementias

ADRD is a progressive and fatal condition primarily afflicting individuals after the age of 60. It is estimated that 24 million individuals worldwide currently have dementia, of which the majority are believed to have Alzheimer's disease (Ballard et al., 2011). Alzheimer's disease is the result of amyloid plaques and neurofibrillary tangles causing cognitive decline and ultimately death (Ballard et al., 2011). Other types of dementia, such as vascular dementia, Pick's disease, frontal lobe dementia, Creutzfeldt-Jacob disease are the result of cerebrovascular disease, nerve cell death, or infectious agents such as prions (Geldmacher and Whitehouse, 1996). Many dementias, including Alzheimer's disease can only be diagnosed definitively post mortem, though probable diagnosis is possible with individuals' detailed history of symptoms in order to

assess the presence of cognitive impairment (Ballard et al., 2011). Accurate diagnosis of ADRD is important in order to manage potentially treatable disorders that contribute to cognitive impairment in individuals with ADRD, including depression, vitamin deficiencies, and hypothyroidism (Ballard et al., 2011). In addition, due to the degenerative nature of ADRD, diagnosis is useful in allowing affected individuals and their families to plan for future life and finances while cognitive functions are still intact (Ballard et al., 2011).

In the mild stage of Alzheimer's disease, symptoms include short-term memory impairment, depression, and anxiety (Gauthier, 2002). During this stage, individuals with Alzheimer's disease are still generally able to live independently (Tyas and Gutmanis, 2008). Recommended care for individuals during this stage includes the management of comorbidities, and consultation with geriatricians and/or neurologists (Hogan et al., 2007). The management of behavioral problems can be achieved through simple environmental modification, task simplification, and redirection at this stage (Tyas and Gutmanis, 2008). As Alzheimer's disease progresses, symptoms include hallucinations and false beliefs, reversal of sleep patterns, motor rigidity and very prominent cognitive decline (Gauthier, 2002). In addition, adverse events including sedation, Parkinsonism, and increased risk of stroke can occur due to the use of pharmacological treatments such as anti-psychotics in controlling symptoms of Alzheimer's disease (Ballard et al., 2011). Individuals with Alzheimer's disease are often institutionalized during the moderate and severe stages of the condition. According to Yaffe et al. (2002), being 80 and above, living alone, and having difficult behaviors such as having psychotic symptoms, being dangerous, and waking up the caregiver were all significantly associated with nursing home admission. The progression for other dementias vary, ranging from rapid decline for

Creutzfeldt-Jacob disease, to a fluctuating course dependent on underlying cerebrovascular disease for vascular dementias (Geldmacher and Whitehouse, 1996).

Amyotrophic Lateral Sclerosis

ALS is one of the major neurodegenerative diseases, causing rapid degeneration of the motor system at all levels due to the death of motor neurons in the anterior horn of the spinal cord (Mitchell and Borasio, 2007). Little is known about the epidemiology of the condition, although estimates have placed the incidence at 3 per 100 000 person years for males, and 2.4 per 100 000 person years for females (Kiernan et al., 2011). The condition is primarily sporadic, but is inherited in about five to 10 percent of cases (Kiernan et al., 2011; Rowland and Shneider, 2001). At this time, the cause of ALS is unclear. Onset typically occurs after the age of 45, but rarely after 80 (Kiernan et al., 2011). Approximately half of all individuals with ALS die within 30 months of symptom onset, although the survival can range from one to 20 years (Kiernan et al., 2011; Ropper and Samuels, 2009; Rowland and Shneider; 2001).

Initial symptoms of ALS vary depending on the ALS type, but are marked by the presence of upper and lower motor neuron features that involve the brainstem and areas of innervation along the spinal cord (Kiernan et al., 2011). The progression of ALS is contingent on the disease type and whether it is familial or sporadic, and also has implications on appropriate therapies (Kiernan et al., 2011). For individuals with bulbar-onset ALS, initial symptoms include dysarthria and/or dysphagia (Mitchell and Borasio, 2007). This type of onset can be caused by upper or lower motor neuron dysfunction (Rowland and Shneider, 2001). Similarly, cervical-onset ALS can be indicative of upper or lower motor neuron damage; however, early symptoms for cervical-onset ALS include proximal and distal weakness of the arms (Mitchell and Borasio, 2007). Finally, lumbar-onset ALS presents proximal weakness in the legs (Mitchell and Borasio,

2007). Roughly a quarter of individuals present with bulbar-onset, 70 percent present with cervical or lumbar onset, and five percent present with both bulbar and limb-onset (Kiernan et al., 2011). Regardless of initial onset, fasciculations, weakness, and degeneration of muscle eventually spread to other regions (Kiernan et al., 2011). Additionally, fatigue, dysphagia, and respiratory challenges are present across all ALS types (Kiernan et al., 2011). For some individuals with ALS, associated frontotemporal dementia is also present (Mitchell and Borasio, 2007).

Together, these symptoms lead to the need for assistance with ADLs, as well as therapies and treatments for weight loss and malnutrition, shortness of breath, and depression associated with initial diagnosis of ALS (Kiernan et al., 2011). Almost all individuals with ALS prefer to remain in their homes, with care from a spouse (Krivickas et al., 1997). Home care services are generally required as the condition progresses, and the caregiver can no longer provide care alone (Krivickas et al., 1997). For individuals without caregivers, institutionalization is usually required (Krivickas et al., 1997). Although still not used by all individuals with ALS, palliative care near the end of life provides individuals with ALS and their families with comfort (Borasio and Voltz, 1997). Death for individuals with ALS is typically the result of respiratory problems, in many cases leading to pneumonia (Kiernan et al., 2011).

Multiple Sclerosis

MS is a central nervous system disorder that is characterized by inflammatory demyelination and axonal loss, followed by some remyelination (Compston and Coles, 2002). Ultimately, this process results in lesions in the brain, the optic nerve, brainstem or spinal cord (Compston and Coles, 2002). The condition is caused by a combination of genetic susceptibility and environmental factors that are currently unknown (Compston and Coles, 2002). The typical

age of onset is in the third or fourth decades of life (Compston and Coles, 2002). The worldwide incidence is about seven cases per 100 000 person years, but primarily affects northern Europeans (Compston and Coles, 2002). The incidence of MS is higher in females than in males at a ratio of roughly 2 to 1 (Ropper and Samuels, 2009).

The progression and symptoms of MS are dependent on the type of MS, the severity of the condition, as well as the location of the lesions. Individuals with relapsing-remitting MS accumulate disability over time as relapses cause gradual damage (Compston and Coles, 2002). Approximately 80 percent of individuals have this type of MS (Compston and Coles, 2002). On the other hand, primary progressive MS does not remit, and presents in about 20 percent of individuals (Compston and Coles, 2002). Primary progressive MS generally affects the spinal cord, but can also affect the optic nerve, cerebrum, or cerebellum, though less frequently (Compston and Coles, 2002). Lesions in the cerebrum can lead to loss of sensation on one side of the body, movement impairments, depression, and cognitive impairments (Compston and Coles, 2002). In early stages, cognitive impairment results in attention, reasoning, and executive function deficits (Compston and Coles, 2002). Late stage cognitive impairment results in dementia (Compston and Coles, 2002). In rare cases, lesions in the cerebrum can also cause in epilepsy, or even focal cortical deficits (Compston and Coles, 2002). In contrast, lesions to the cerebellum and cerebellar pathways lead to postural, action, and/or speech tremors, clumsiness, and poor balance (Compston and Coles, 2002). Lesions to the optic nerve lead to painful visual impairments that include scotoma, reduced visual acuity, loss of colour, temporary blindness, and loss of vision in one side (Compston and Coles, 2002). The brainstem can also be affected by lesions, which results in diplopia and oscillopsia, dizziness, impaired speech and swallowing functions, and a sudden intensification of symptoms (Compston and Coles, 2002). Spinal cord

lesions cause upper motor neuron signs such as muscle weakness and stiffness, as well as painful spasms (Compston and Coles, 2002). Spinal cord lesions also result in bladder dysfunction, constipation, and erectile dysfunction (Compston and Coles, 2002). Finally, lesions in other areas can result in pain, fatigue, temperature sensitivity, and intolerance of exercise (Compston and Coles, 2002).

On average, the life expectancy for individuals with MS is a minimum of 25 years; however, differences in symptoms lead to variations in prognosis (Compston and Coles, 2002). Roughly 25 percent of all individuals with MS never experience ADL limitations, while severe disability occurs over a very short period of time in about 15 percent of individuals (Compston and Coles, 2002). Those who primarily have sensory symptoms tend to have a good prognosis, while prognosis is generally poor when symptoms involve disturbed coordination and balance (Compston and Coles, 2002). Due to the relapsing-remitting pattern of MS seen for the majority of individuals, treatment primarily consists of reducing the proximity between relapses (Compston and Coles, 2002). Treatments are also provided to prevent permanent disability, manage symptoms of fixed lesions, and treat symptoms caused by established progression (Compston and Coles, 2002). Individuals with MS generally remain in their homes and are cared for by spouses (Aronson et al., 1996). Only a fifth to a quarter of individuals with MS eventually admitted to nursing home (Stolp-Smith et al., 1998). Death for individuals with MS is typically unrelated to scleroses, but can be the result of dyspnea in some individuals (Gosselink et al., 1999).

Table 1: Relevant Clinical Variables, by Condition

Condition	Clinical Symptom	Resulting Effect
ADRD	**Cognitive Decline**	Memory impairment
		Acalculia
		Goal setting/planning
	Behavioral Symptoms	Aggression/Hostility
		Delusions/Hallucinations
		Agitation
		Depression/Anxiety
		Social Interaction
	Movement (Rigidity, Apaxia)	Gait Disturbance
		Fine motor movement
		Urinary Incontinence
		Falls
		Walking
		Stair use
	Comorbidity	Stroke
		Parkinsonism
		ALS
		MS
	Other	Pain
		Respiratory Difficulty
ALS	**Muscle Weakness and Rigidity**	ADL Difficulties
		Unsteady Gait/Falls
		Stairs
	Dysphagia/Dysarthria/Dyspnoea	Swallowing
		Malnutrition
		Weight Loss
		Shortness of breath
		Respirator use
		Other Respiratory Treatment
		Tracheostomy
	Indirect Symptoms	Depression/Anxiety
		Sleep Problems
		Constipation
		Drooling
		Pain
		Fatigue
	Comorbidities	Frontotemporal Lobe Dementia
MS	**Unilateral Optic Neuritis, diplopia**	Vision
	Gait Ataxia, limb weakness, clumsiness, chorea, rigidity, tremor, poor coordination	Falls
		Walking
		Stairs

		ADL Function
	Neurogenic Bladder/Bowel	Bladder Incontinence
		Bowel Incontinence
		Constipation
	Sexual Dysfunction	Erectile Dysfunction
	Cognitive Impairment	Dementia (rare)
		Emotional Lability
	Dysarthria/ Dysphagia	Speech
		Swallowing
	Vertigo	Dizziness
	Pain	Pain
	Fatigue	Fatigue
	Depression	Depression
	Other	Dementia (rare)
		Respiratory Problems

2.2 Home Care

Home care (HC) was first established in Canada as a publicly funded service in Ontario in 1970, and by 1988, had been established in all Canadian provinces and territories (Canadian Healthcare Association, 2009). Despite the presence of HC programs across Canada, HC services are not federally mandated, and as a result no national definition or standards on HC exist. Nevertheless, HC is understood to provide health and social services to help individuals with health conditions or functional limitations remain in their homes (MacAdam, 2004). Unlike other types of healthcare, HC is focused on prevention and maintenance, and can sometimes be a substitute for post-acute care, and long-term care (MacAdam, 2004; Canadian Healthcare Association, 2009). Even without federal mandate, HC programs are amongst the fastest growing components of the Canadian healthcare system. HC expenditures increased substantially in the 1990s, and at least some shift in expenditures from other post-acute and non-acute care settings to HC programs have been seen (Canadian Healthcare Association, 2009; Coyte, 2000; MacAdam, 2004). This trend is the result of a number of factors including the cost-effectiveness of HC programs relative to hospital and nursing home care, the use of HC to respond to the aging Canadian population, as well as the desire for Canadians to remain in their homes (Coyte, 2000; Ontario Home Care Assocation, 2010). Thus, despite the lack of federal involvement, HC is expected to be an increasingly healthcare service for Canadians with health conditions or functional limitations.

Home Care in Ontario

In Ontario, HC falls under the jurisdiction of the Ontario Ministry of Health and Long-Term Care, is funded by Local Health Integration Networks (LHINs), and is administered by Community Care Access Centers (CCACs). The CCACs act as the single-point entry for all HC

services in Ontario so that all individuals wishing to receive HC services must contact their CCAC, as defined by their geographical location. In total, there are 14 CCACs, which have the same geographic boundaries as the LHINs. The 14 CCACs are as follows: Erie St. Clair (ESC), South West (SW), Waterloo Wellington (WW), Hamilton Niagara Haldiman Brant (HNHB), Central West (CW), Mississauga Halton (MH), Toronto Central (TC), Central, Central East (CE), South East (SE), Champlain, North Simcoe Muskoka (NSM), North East (NE), and North West (NW). A map of the CCACs can be found in Appendix A.

Service Provision

While CCACs administer HC services, they do not provide direct services. The provision of services is instead contracted to individual service provider agencies that provide professional services. These services include nursing services, physiotherapy, occupational therapy, speech-language therapy, social work services, and nutritionist/dietitian services, as well as non-professional personal support services. In general, nursing services in HC include the promotion of health and the treatment of conditions (CCAC, 2006). These tasks encompass post-acute services such as wound care, and IV therapy. Dietetics services help to treat and prevent nutrition-related disorders, while speech language pathology services help with oral motor and communication functions (CCAC, 2006). Occupational therapists address adaptive behaviors to develop, maintain, or rehabilitate function for self-care, leisure, and productivity (CCAC, 2006). Physiotherapists treat physical dysfunction, injury, or pain, and help to develop, maintain, rehabilitate, or augment physical function, as well as relieve pain (CCAC, 2006). Social work services help individuals and their families with skills that optimize functioning within the home. Specific treatments can include assisting in adjustment to altered health state, behavioral problems, as well as providing support to informal caregivers (CCAC, 2006). Finally, personal

support workers provide homemaking services to individuals with functional limitations, which can include assistance with ADLs and IADLs, and account for the greatest proportion of HC service provision.

Eligibility

Eligibility for HC services is determined by case managers at CCACs based on the following criteria: individuals must require one of the professional services provided by HC, excluding social workers and dietitians; must require these services in order to remain in their homes; are unable to access these services at community based settings; live where services can be provided safely and effectively; and services provided are expected to lead to reasonable levels of rehabilitation (CCAC, 2006). In addition, the professional service required by individuals must be available in the CCAC in order to qualify for HC services. Finally, individuals must be insured under the Ontario Health Insurance Plan.

Clinical Assessment and Care Planning

Clinical assessments of HC users are conducted for all long-stay individuals requiring HC services for greater than 60 days. These assessments are performed using the RAI-HC instrument, which was first introduced to Ontario HC in 2004. HC services are not age restricted, but assessments are only required for individuals ages 18 and over. In addition, the RAI-HC is not used for those receiving palliative, acute, or rehabilitation services. For HC service users requiring clinical assessments, CCAC case managers perform assessments every 6 months, or more frequently if deemed necessary by the case manager. The purpose of these assessments is to provide clinicians with a comprehensive view of the strengths and needs of HC service users (Morris et al., 1997). Specifically, these assessments help case managers develop plans of service for HC service users based on these clinical assessments (CCAC, 2006). These plans of service

determine the amount of each service to be provided to HC service users, as well as the coordination of these services (CCAC, 2006). RAI-HC reassessments are also used to evaluate and revise plans of service as the clinical needs and preferences of service users change (CCAC, 2006).

Service Users

Formal HC service use is uncommon in the general population in Ontario (Hall and Coyte, 2001). For individuals with functional disabilities, however, HC is much more common as restriction in ADLs and IADLs are significantly and independently predictive of HC (Hall and Coyte, 2001). Other characteristics predictive of HC use include having a diagnosed health problem, being disabled, having poor self-reported health status, and older age (Hall and Coyte, 2001). In other jurisdictions, cognitive status has also been identified as a predictor of HC use (Hall and Coyte, 2001). Overall, individuals using HC services are likely to have greater levels of disability and functional impairments.

Due to the functional impairments and disability associated with ADRD, ALS, and MS, individuals with these neurological conditions are more likely to be HC service users. For example, Vazirinejad et al. (2008) have found that the proportion of individuals with MS seeking HC services such as home help, occupational therapy, speech language pathology, physiotherapy, nursing, and social work services all increased with disease severity. In contrast, general practitioner and specialist services remained consistent throughout the course of the condition (Vazirinejad et al., 2008). Similar patterns of resource use exist for individuals with ADRD (Kraft et al., 2010), and it is likely that patterns of resource use for persons with ALS are similar given the clinical similarities between the three conditions. Overall, HC services are also likely to command a great deal more resource utilization than primary care for individuals with

neurological conditions. Kraft et al. (2010) found that in Switzerland, visits to general practitioners accounted for 0.4 percent of the annual cost of dementia, while community care accounted for 4.8 percent of total annual costs. Based on HC service utilization literature available for ADRD and MS, it is likely that HC service use is a crucial part of the care plan for individuals with ADRD, ALS, and MS.

2.3 interRAI

The interRAI family of assessment instruments was designed for use on various vulnerable populations (Hirdes et al., 2008). The first of these instruments was the Resident Assessment Instrument for nursing home, which was developed to address the need for a national resident assessment system in nursing homes identified by the Omnibus Budget Reconciliation Act (OBRA) of 1987 (Morris et al., 1990). This instrument was developed by a consortium of researchers and clinicians that has since become the interRAI network. The goals of the interRAI nursing home instrument were to replace previous nonuniform assessment instruments, to encourage the integration of resident assessment and care planning information, to improve quality of life through better care planning and provision, and finally, to set a standard methodology for resident assessment that could be updated and adapted to suit future needs (Morris et al., 1990). Unlike previous resident assessment tools, the interRAI nursing home instrument was designed to measure physical and psychological well being, functional and cognitive status, quality of life, and resource need using a case-mix system approach (Morris et al., 1990). The interRAI consortium has since developed a number of other assessment instruments, all of which are guided by the same goals and methodology of the interRAI nursing home instrument (Hirdes et al., 2008). These instruments include assessments for use in mental

health, acute care, post-acute care, palliative care, and home care settings. Today, the interRAI collaborative network and its instruments are in use in over 30 countries worldwide.

The interRAI instruments were introduced to Canada on July 1, 1996 in Ontario complex continuing care (CCC) facilities (Hirdes et al., 2003). The interRAI nursing home instrument was implemented in CCC to improve the role of CCC in the Ontario healthcare system. The absence of CCC user data meant that there was little evidence to base funding and resident admissions criteria. The need for data to inform funding decisions for CCC responsibilities eventually led the Ontario Joint Policy and Planning Committee to establish the use of the interRAI nursing home instrument for patient level data collection. Although other assessment tools had also been considered, the interRAI tool was chosen because of its rigorous scientific testing, its sensitivity to clinical complexity, its use in international settings, and the inclusion of a clinical component that would be beneficial to patient need (Hirdes et al., 2003). Today, interRAI tools are used across different care settings in Canada. Some of these settings are nursing home, inpatient and community mental health, palliative care, as well as HC.

Resident Assessment Instrument for Home Care

The Resident Assessment Instrument for Home Care (RAI-HC) was designed to provide comprehensive assessment for the needs of individuals in HC using the same methods and principles as the interRAI nursing home instrument (Morris et al., 1997; Morris et al., 2000). Unlike most other community care assessment instruments, the RAI-HC is a multidimensional system that included measures of functional, social, disease, and environmental status (Morris et al., 1997). Where possible, the RAI-HC adopted assessment items that were compatible with measures in the nursing home instrument (Morris et al., 2000). Overall, 47 percent of the 223 items found in the RAI-HC were derived directly from the nursing home assessment instrument,

and included items in the cognition, communication, vision, mood, behavior, ADL, and continence areas (Morris et al., 1997). Items found in the RAI-HC that are not found in the nursing home assessment instrument include informal support, IADL self-performance, alcohol abuse, and environmental conditions (Morris et al., 1997). Like the interRAI nursing home instrument, the RAI-HC tool designed for use by trained professionals performing assessments to observe individuals in the nursing home environment, review available health documents, in addition to directly questioning the nursing home residents and his or her informal caregiver(s). Finally, the RAI-HC provides a substantial improvement on previous community care assessment tools as both the validity and reliability of this instrument has been confirmed in various studies (Morris et al., 1997; Kwan et al., 2000; Landi et al., 2000).

2.4 Methods for Estimating the Costs of Neurological Conditions

Accurate estimates of costs for neurological conditions are required in order to better allocate scarce health care resources. In some cases, estimates help to justify the costs of expensive new pharmacological treatments. In HC, cost estimates are typically required for policy planning uses such as estimating future service and resource needs. Indeed, understanding clinical characteristics that affect costs for the neurological conditions is important given evidence suggesting that for at least some neurological conditions, the cost of care is costlier care for those without this these conditions (Kang et al., 2006). It is important to note, however, that while the costs for individuals with neurological conditions can be estimated, it is not possible to estimate the costs caused by neurological conditions (Jönssen et al., 2006).

Cost of Illness

The most common method for estimating the cost of neurological conditions relies on the cost of illness (COI) methodology. COI research was the first economic evaluation technique

used in the health field that standardized the methodology for estimating the cost of illness and injury (Tarricone, 2006). Although studies estimating the economic impact of disease on society had been conducted for many years using average costs for all illnesses, Rice (1967) argued that estimating the costs of specific illnesses was favorable since it provided more accurate reflections of costs. Accurate estimates of cost could then be used to estimate the total economic burden of specific illnesses to society, as well as to help further programs of research, to control or justify program expenditures, and to compare the costs of different illnesses (Rice, 1967; Cooper and Rice, 1976; Byford et al., 2000). Since Rice's seminal paper, COI studies have shifted from measuring global disease costs, to measuring the combined social and financial costs of specific illnesses.

Estimation Procedure

COI is estimated by identifying components where costs are incurred, and then attributing a monetary value to those components (Tarricone, 2006). The components are measured by the individual services and resources consumed through the course of care, such as meals, medications, lab procedures, or surgeries. Opportunity costs, which are the values placed on the lost opportunity to select the next best options, are used to measure the monetary value in COI estimates so that the full economic cost can be estimated as best as possible (Hodgson and Meiners, 1982). The application of these opportunity costs is placed on components of care that include the utilization of specific services, as well as losses in productivity. Specific components of associated costs are described in greater detail below.

Direct Costs:

Direct costs can include both healthcare and non-healthcare related costs, and are invariably included in cost estimates for the neurological conditions. Healthcare related direct

costs refer to the services or goods consumed in the process of prevention, detection, treatment, rehabilitation, research, training, and capital investment in medical facilities of a given condition (Rice, 1967). Expenditure data for these health related activities are generally available through government bodies such as the United States National Health Accounts, or CIHI in Canada. Data are available for the majority of healthcare related activities, particularly, physician, hospital and drug costs. However, components such as research, training, and capital investment are difficult to allocate to specific illnesses since these activities are not necessarily carried out at the same time that other expenditures for care are made (Hodgson and Meiners, 1982). Non-healthcare related activities considered in direct costs include transportation related to health provider visits, relocation due to illness, and informal care (Tarricone, 2006). Costs incurred by family providing care to the ill are also considered as non-medical direct costs (Hodgson and Meiners, 1982). Although expenditures related to these non-healthcare related activities form an important component of the total direct costs, these expenditure data are not often systematically collected. As a result, non-medical direct costs generally rely on self-reported data collected for COI research purposes.

Indirect Costs:

Direct costs alone do not measure the full economic costs resulting from illness and injury; therefore, total COI estimates must also include indirect costs. Unlike direct costs, indirect costs are not measured by expenditures. Instead, these costs are measured by losses in output or productivity caused by illness, as in a human capital approach. Losses of output to the economy because of illness, disability, or premature death can be estimated by applying mean earnings to work-loss data for a given illness or condition to the time taken away from employment for both the ill and their informal caregivers (Rice, 1967). The basic premise behind

indirect cost estimates has been challenged for a number of reasons, however. One of the primary arguments challenges the assumption that illness is the cause for losses in productivity, and that had death or illness not interfered, all individuals of the same age and sex would have had the same employment experiences (Rice, 1967). Similarly, measuring indirect costs through losses in productivity have been critiqued for systemically favoring diseases that traditionally afflict higher income groups (Hodgson and Meiners, 1982). This bias is problematic as losses caused by these illnesses necessarily have greater valuations based on indirect cost calculations rooted in earnings data. Nevertheless, indirect costs continue to be included in the majority of COI studies.

Intangible Costs:

Although COI studies attempt to estimate the total burden of disease, certain aspects of disease cannot be valued using this approach. Specifically, these aspects include psychosocial costs associated with pain and suffering (Cooper and Rice, 1976). Common examples of these intangible costs include family conflict caused by disease, changes in personality, reduced self-esteem, and physical pain (Hodgson and Meiners, 1982). Intangible costs that are not captured by indirect costs also include loss of ability to engage in leisure activities, economic dependence, unwanted employment changes, as well as loss of opportunities for promotions or higher education (Hodgson and Meiners, 1982). Finally, grief due to death for family and friends cannot be quantified and included in COI estimates.

Other Considerations

Although the basic COI estimation procedure has been described above, a number of other methodological considerations must be made. These considerations pertain to the type of data that are used to estimate incidence or prevalence, individual or group level data, and finally

prospective as opposed to retrospective data. The corresponding advantages and disadvantages of each of these data types are discussed below.

Prevalence and Incidence:

As in epidemiological studies, disease temporality must be considered. Prevalence based COI studies estimate the direct and indirect costs associated with all cases of the given illness that occur over a specified period of time (Byford et al., 2000). On the other hand, incidence based studies include the direct and indirect costs of cases of illness beginning in the year in which an illness first begins (Byford et al., 2000). In general, prevalence based approaches to COI produce larger estimates of cost. This observation is particularly true of conditions with long sequelae, although the estimates from both approaches will be similar for illnesses with short durations (Tarricone, 2006). Nevertheless, both incidence and prevalence-based estimates are of value. Prevalence based studies are preferred for planning cost containment policies since they are better able to comprehensively show the costliest components of care (Tarricone, 2006). Meanwhile, incidence based studies are preferred when considering preventive measures, short-term illnesses or analyzing the cost of disease from onset until death (Tarricone, 2006).

Top down and Bottom up:

The top down and bottom up consideration in COI estimates describes whether population, or individual level data will be used. A bottom up approach requires the quantity of health services consumed to first be estimated, followed by an estimation of the service costs for each of the different services consumed. The consumed services are then multiplied by the service costs, and the costs of all the different consumed services are then summed (Tarricone, 2006). Finally, the number of individuals within a given sample or population is multiplied by the summed value in order to produce a gross estimate of health service expenditures (Olesen et

al., 2012). In top down studies, total expenditures for a given illness are used so that service use and service cost data are not required. The advantage of the top down approach is that the estimates of expenditures cannot be larger than the known total expenditure (Tarricone, 2006). However, the bottom up approach is considered the 'gold standard' of COI estimation due to its ability to accurately attribute costs to specific components of illness, and is preferred where reliable data of service utilization and cost are available (Akobundu et al., 2006).

Prospective and Retrospective:

Like incidence and prevalence, the prospective and retrospective approaches in COI estimates are derived from epidemiological concepts. Retrospective studies, which were described by both Rice (1967), and Cooper and Rice (1976), are most common, and involve data that have already been collected from relevant events. On the other hand, follow up data are used for prospective COI studies, and involve events that have not yet occurred (Tarricone, 2006). Due to the high costs associated with follow up studies, prevalence based studies are typically preferred where relevant data are available.

Limitations

Although COI estimates have become extremely popular since Rice (1967) first attempted to standardize burden of disease studies, some methodological problems limit the applicability of these studies for policy and planning purposes. According to Currie and colleagues (2000), COI studies cannot be used for priority setting purposes. As COI studies only involve the costs of an associated activity and not the benefits, true opportunity costs cannot be determined, and thus the activities that would best maximize health cannot be determined (Currie et al., 2000). In addition, COI estimates cannot be used to predict future COI, as drivers of cost are not generally included in these studies.

Case-Mix Systems

Although the COI methodology is effective in identifying components of cost, it is inadequate at identifying predictors of costs based on clinical and personal characteristics. Case-mix systems, on the other hand, are able to identify predictors of cost since they were initially designed for prospective payment systems. Unlike COI, which essentially estimates opportunity cost, case-mix systems view costs in terms of service utilization. The notion of cost in case-mix systems is based on the concept that healthcare resource use is driven by personal and clinical characteristics (Fetter, 1991). As a result, case-mix systems were designed to classify individuals into resource homogenous groups based on their personal characteristics, followed by the attachment of a ratio level value to describe group resource intensity (Fetter, 1999).

While case-mix systems are not strictly used for cost estimation purposes, case-mix does involve an element of cost measurement through its use as a reimbursement tool in hospitals and nursing homes (Fetter, 1999). In addition, case-mix systems were designed for use as predictors of resource utilization. Unlike the COI approach, which requires a number of considerations such as prevalence and incidence, top-down and bottom-up, and direct and indirect measures, case-mix estimates of cost are simple in that they are invariably prevalence-based, bottom-up, direct estimates of cost. Case-mix based estimates of cost only require data on service utilization and costs, in addition to clinical information. Depending on care setting, clinical information can come from ICD codes representing an individual's primary diagnosis, or from assessment instruments such as the RAI-HC described previously. It is generally accepted that measurements of service utilization only include services related directly to the medical treatment of a condition in hospitals, or to symptom management in non-acute settings (Fetter et al., 1991). As a result, only these services are included as part of resource intensity and cost estimates using the case-

mix approach (Fetter et al., 1991). Resource utilization information is typically obtained from administrative records. Based on the defined services, unit costs representative of the wages for each of the different services are applied to the services provided in order to produce the total cost of treatment or care for an individual. A mean cost of care can then be calculated to describe the mean cost of care across the entire population. This mean is used as the reference value, and the resource use for all case-mix groups are then compared to this value. Case-mix groups are identified based on a set of clinical characteristics that have been identified to drive resource utilization. Mean costs are then produced for each of the different clinically homogenous groups. These mean costs are then used to determine the resource intensity weights for each of the resource homogenous groups, as compared to the mean cost of the total population. Based on the cost measurement component of case-mix systems, individual- and group-level estimates of costs can be derived. Ratio measures of resource intensity are also available using the case-mix approach. Some level of variability is expected between the individual- and group-level cost estimates using these ratio measures; however, variability should be limited as much as possible. Effective case-mix systems are expected to have low levels of variation between an individual's group-level cost, and their individual-level cost (Fetter, 1991).

Though simple in comparison to the COI approach, the grouping of a case-mix system is paramount to the effectiveness of a given system at estimating resource use, and must be considered. Overall, case-mix systems must meet statistical, clinical, and incentive criteria (Fetter, 1999; Fries et al., 1994). As briefly mentioned above, the clinical criterion requires that individuals within a group are clinically homogenous and have similar expected resource utilization (Fries et al., 1994; Fetter et al., 1980). However, groupings must also be based on a number of different clinical variables in order to ensure that all individuals are identified by the

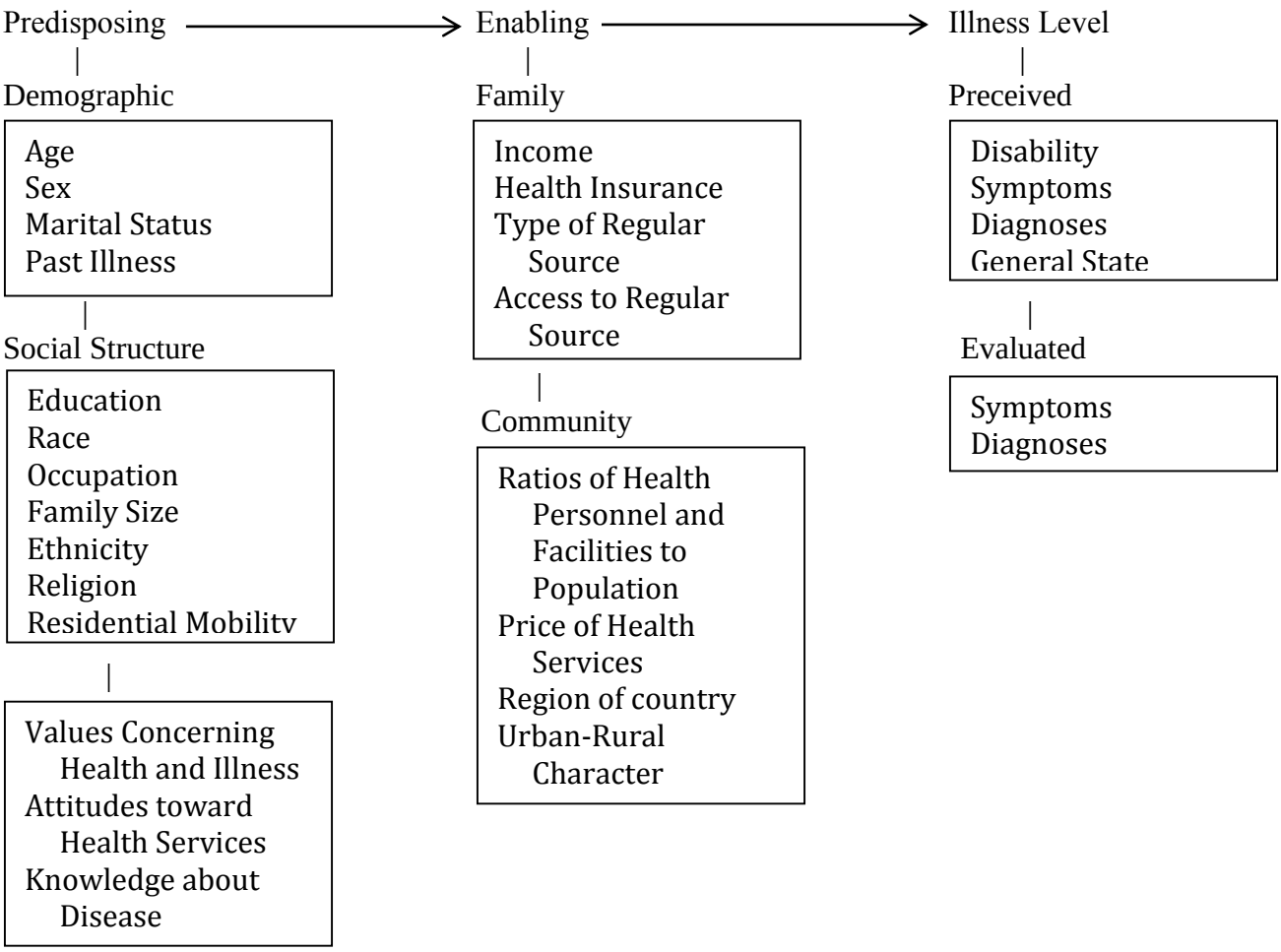
system, even if they are rare (Fries et al., 1994). For example, grouping based solely on illness is problematic since descriptive labels and treatments for the same illnesses can differ, and not all diseases are equally well understood (Fetter, 1991). The statistical criterion is also crucial in case-mix systems, and requires that variability within case-mix groups can be predicted (Fetter, 1991). Finally, as case-mix systems are primarily used for prospective payment systems, the incentive criterion is also important in ensuring that estimates of cost are reflective of actual needs-based costs, rather than desired costs (Fries et al., 1994; Botz, 1992).

Overall, the case-mix systems approach provides a reasonable alternative to the more traditional COI approach. Even though case-mix measurement can only be used to estimate costs in defined care settings, this approach is likely to produce more reliable estimates of cost within these settings because of incentive criteria, and use for funding purposes. As Fetter (1991) argues, clinical variables primarily lead to differences in costs. While individual clinical characteristics result in only small variations to the equipment and supply needs that make up the bulk of COI component variables, they result in substantial differences in service needs in care programs and facilities (Fetter, 1991). As a result, the case-mix approach is likely preferable when the purpose is to identify predictors of cost, rather than components of cost. Perhaps the only potential problem associated with the case-mix measurement system is its exclusion of services not directly related to care or treatment. In hospitals, non-acute care settings, and community care programs, this exclusion is likely to underestimate costs associated with administrative tasks required to ensure the provision of treatments and care. Assuming that these administrative tasks are in proportion with the resource intensity weights for the different resource homogenous groups, however, this exclusion of administrative services is not likely to greatly impact the resource utilization ratios generated using this system.

2.5 Conceptual Framework

Andersen and Newman proposed a framework for predicting the utilization of healthcare services. The framework includes both societal and individual determinants of service utilization. Societal determinants affect healthcare utilization through changes in treatment and technology, as well as the financing of healthcare (Andersen and Newman, 1973). Individual determinants, on the other hand, affect healthcare service utilization through individuals' predisposition to use services, the ability for individuals to access these services, and individuals' medical needs based on their conditions (Andersen and Newman, 1973). The predisposing determinants describe the likelihood for individuals use healthcare services irrespective of illness (Andersen and Newman, 1973). Enabling determinants are described as family and/or community resources that may affect resource utilization (Andersen and Newman, 1973). These include family attributes such as availability of caregiver, income, or insurance coverage (Andersen and Newman, 1973). Finally, according to Andersen and Newman (1973), illness is the most important determinant of healthcare service utilization, and is affected by the severity of the illness. The full list of predisposing, enabling, and illness components proposed by Andersen and Newman (1973) can be found in Figure 1. Across care settings, illness levels have the greatest relative importance for predicting resource utilization (Andersen and Newman, 1973). The relative importance of other characteristics depends on care setting. In HC, enabling factors have been found to have medium relative importance, while predisposing factors have low relative importance in predicting HC service utilization (Kempen et al., 1991).

Figure 1: Andersen and Newman, 1973 Characteristics Determining Resource Utilization in Healthcare



Source: Andersen and Newman, 1973

3.0 EXISTING COST LITERATURE FOR THE NEUROLOGICAL CONDITIONS

Existing research on the cost of neurological conditions such as ADRD, ALS, and MS have largely focused on identifying the annual costs associated with care for these conditions using COI methods. Indeed, the findings suggest substantial economic burden associated with each of the neurological conditions discussed in this project. According to the Canadian Alzhiemer Society (2010), for example, the total economic burden of ADRD was $15 billion in 2008. The lifetime cost per individual with MS, meanwhile, was estimated at $1.6 million (The Canadian Burden of Illness Study Group, 1998), and over $1 billion per year in Canada (MS Society of Canada, 2007). For MS, the most substantial component of cost has been attributed to lost productivity rather than costs incurred from health and social services (The Canadian Burden of Illness Study Group, 1998). For ADRD, the largest component of cost was due to informal care (Canadian Alzheimer Society, 2010). As very little cost research has been conducted for ALS, no estimates of economic burden could be found for Canadians with ALS. According to a Spanish estimate, the annual cost of ALS was €36 194 for 2004 ($58 065 CAD based on the November 30, 2004 exchange rate) (Lopez-Bastida et al., 2009).

While estimates of economic burden are helpful in identifying the components of costs, these traditional estimates of costs provide little insight into factors that drive healthcare resource use and funding needs for individuals with ADRD, ALS, and MS. In fact, very little research has specifically addressed the need for such information. Therefore, little is known about the mechanisms surrounding the high costs of care for ADRD, ALS, and MS. A review of existing literature was conducted through a MEDLINE search to identify previous studies that had attempted to identify predictors of costs for the three neurological conditions. In total, ten articles were identified that could be retrieved. These articles primarily looked at the relationships

between disease severity, and cost for individuals with ADRD, ALS, and MS. Of the ten articles that were identified, eight articles were dementia related. Only one study looked at MS, and one article looked at ALS. The full search strategy used to identify these articles can be found in Appendix B. Although non-scholarly literature was also searched for possible findings, no such reports could found.

3.1 Study Designs

Of the ten articles identified, six were cross-sectional (Reese et al., 2011; Leicht et al., 2011; Gustavsson et al., 2011b; McCrone et al., 2008; Grima et al., 2000; Kang et al., 2006; López-Bastida et al., 2009), three were prospective cohort studies (Andersen et al., 2003; Jönsson et al., 2006), and one had a case-control design (Rojas et al., 2011). The sample sizes for all studies were large by statistical standards, ranging from a sample size of 42 (Grima et al., 2000), to 3346 (Andersen et al., 2003). Study participants were recruited from disease registries (López-Bastida et al., 2009; McCrone et al., 2008), population registries (Andersen et al., 2003; Kang et al., 2006), and outpatient hospital, clinic and office visits for neurologists and general practitioners (Reese et al., 2011; Leicht et al., 2011; Grima et al., 2000; Jönsson et al., 2006; Gustavsson et al., 2011b).

Cost and clinical data were collected through interviews (Andersen et al., 2003; Gustavsson et al., 2011b; Kang et al., 2006; Reese et al., 2011; Rojas et al., 2011) by self-reported survey data (Jönsson et al., 2006; Leicht et al., 2011; McCrone et al., 2008; López-Bastida et al., 2009), and clinical assessment (Grima et al., 2000). Clinical data were measured exclusively through disease severity scales including the Mini Mental State Examination (Gustavsson et al., 2011b; Jönsson et al., 2006; Rojas et al., 2011), Disability Assessment for Dementia scale (Gustavsson et al., 2011; Reese et al., 2007), Expanded Disability Status Scale

(Grima et al., 2000), Neuropsychiatry Inventory (Gustavsson et al., 2011b; Reese et al., 2007; Jönsson et al., 2006) and ADL Functionality Scale (Kang et al., 2007; Reese et al., 2011; Andersen et al., 2003). The measurement of severity used was undefined in the López-Bastida et al. (2009) and Leicht et al. (2011) studies. In addition, some studies looked at quality of life using the Euro Quality of Life (EQ-5D) (López-Bastida et al., 2009; McCrone et al., 2008) and the Health Utility Index 2 (Grima et al., 2000), as well as time since onset. Only one study looked at depression using the Geriatric Depression Scale (Reese et al., 2007). Cost data were self-reported (López-Bastida et al., 2009; Andersen et al., 2003; Grima et al., 2000), identified using the Resource Utilization in Demential Lite questionnaire (Gustavsson et al., 2011b; Jönsson et al., 2006), administrative records (Kang et al., 2007; Reese et al., 2011; Leicht et al., 2011), or were estimated based on total population costs (Rojas et al., 2011; McCrone et al., 2008).

A variety of statistical analyses were also found across the ten studies. These included the use of t-tests, Fisher's exact tests, and chi-square tests to detect differences across groups (Reese et al., 2011; Leicht et al., 2011). Multivariate linear regression models were also common across the ten studies (Reese et al., 2011; Gustavsson et al., 2011b; McCrone et al., 2008; Andersen et al., 2003; Rojas et al., 2011). ANOVA methods were used by Jönsson et al., (2006) as well as Rojas et al., (2011). No details were provided on the statistical analyses for Grima et al. (2000), and López-Bastida et al. (2009).

Study Findings

The findings across the ten studies suggested that disease progression and severity were positively associated with cost. For individuals with ADRD, Jönsson et al. (2006), Andersen et al. (2003), Rojas et al. (2011), Leicht et al. (2011), and Gustavsson et al. (2011b) all found that

ADRD progression and corresponding severity, as well as level of cognitive impairment were all significantly associated with increasing cost. In addition, Gustavsson et al. (2011b) and Kang et al. (2006) reported that ADL function was the most important predictor of care costs in community dwelling individuals with ADRD. One study also found that costs and predictors of costs varied depending on the type of dementia (Rojas et al., 2011). Comparatively fewer studies looked at ALS or MS; however, MS type (primary or secondary progressive), years with MS, quality of life, and disability were all significantly associated with care cost for this condition (McCrone et al., 2008; Grima et al., 2000). Finally, Lopez-Bastida et al. (2009) found that disease severity was positively significantly associated with care costs for ALS. More information of study findings can be found in Appendix C.

3.2 Study Strengths and Limitations

The ten studies identified were generally of high quality. The sampling methodology used across the studies appeared to produce samples that were representative of the community-dwelling sample. The measurement of disease progression or severity, and cost were somewhat questionable for the studies using self-reported questionnaires (Jönsson et al., 2006; Leicht et al., 2011; McCrone et al., 2008; López-Bastida et al., 2009) due to possible misreporting biases. In addition, recall bias is also likely to result from studies of individuals with reduced cognitive performance. As no details were provided outlining the statistical analyses in the studies by Kang et al. (2006), Grima et al., (2000) and López-Bastida et al. (2009), the significance of their findings could not be assessed.

Although these studies do provide valuable insight into the clinical characteristics that are predictive of cost for the neurological conditions, it is clear that research in this area is still in its early stages. Primarily, research in this area has been constrained by the absence of high-quality

data. In many cases, the studies relied on self-reported questionnaire data for measurements of both disease progression and severity, and cost (Jönsson et al., 2006; Leicht et al., 2011; McCrone et al., 2008). In addition, the absence of high quality indicators of personal and clinical characteristics limited the numbers and types of variables that could be tested as potential predictors of cost. Finally, even though these studies were based almost exclusively on community-dwelling samples, only the studies by Grima et al. (2000), Gustavsson et al. (2011), and McCrone et al. (2008) included a range of community care services commonly used by individuals with ADRD, ALS, and MS. The majority of studies included only home care nursing and homemaking services, but excluded services by physiotherapists, occupational therapists, dietitians, social workers, and speech language pathologists (Andersen et al., 2003; Gustavsson et al., 2011; Jönsson et al., 2006; Kang et al., 2006; Leicht et al., 2011; Reese et al., 2011; Rojas et al., 2011).

4.0 STUDY RATIONALE

Individuals with ADRD, ALS, and MS form three distinct groups with neurological conditions within the HC population. Those with ADRD are the largest and oldest neurological groups within the HC setting. In contrast to individuals with ADRD, persons with MS form one of the youngest diagnostic groups within the HC population. They experience substantial functional loss over time but less cognitive loss. Finally, individuals with ALS have the greatest clinical complexity and disability levels within the HC population, and tend to fall between the ADRD and MS age groups. Although the populations of each of these conditions are quite different, formal and informal services are an important part of the care plan for individuals with each of these three conditions in HC (Yaffe et al., 2002; Krivicaks et al., 1997; Aronson et al., 1996). Individuals with these conditions overwhelmingly prefer community care to institutionalization where possible (Yaffe et al., 2002; Krivicaks et al., 1997; Aronson et al., 1996). In order to ensure individuals with these conditions are able to remain in their homes, it is necessary to understand their service needs and costs to ensure that appropriate levels of service provision are available. As a result, it is necessary to identify the factors affecting costs of care for individuals with these conditions.

As outlined in the background section, methodological and data limitations have affected the number and quality of studies examining the costs of these conditions. In particular, no research has looked specifically at the costs of ADRD, ALS, and/or MS in a HC setting. Existing studies were able to identify predictors of costs for these conditions; however, they were general measures of condition severity such as ADL functionality, cognitive performance, or scales related to disability. More specific clinical and personal characteristics that contribute to overall severity of these conditions were not examined in any of the previous studies identified. Finally,

only one of the ten studies identified dealt with Canadian samples (Grima et al., 2000). Therefore further research identifying the relationships between costs and the characteristics of persons with ADRD, ALS, and MS was warranted.

4.1 Purpose, Objectives, and Goals

Given the scarcity of cost research for the neurological conditions, the purpose of this proposed research study was to contribute to the current cost literature for the neurological conditions. More specifically, the objectives were to provide estimates of costs for individuals with ADRD, ALS, and MS in HC settings, and to determine whether specific predisposing, enabling, and illness factors could be used to predict HC care costs for individuals with ADRD, ALS, and MS. The specific goals of this project were to identify reliable weekly estimates of costs for individuals with ADRD, ALS, and MS in the Ontario HC program, as well as clinical and personal characteristics associated with the cost of care for individuals with these conditions.

4.2 Research Questions

The overarching questions being addressed in this project are as follows:

1. How do costs differ for individuals with ADRD, ALS, and MS?
2. What characteristics associated with ADRD, ALS, and MS affect HC care costs?
3. Is the existing RUGIII/HC categorization effective for predicting resource utilization for individuals with neurological conditions?
4. What factors identified by Andersen and Newman (1973) are most important in predicting the costs of care for individuals with ADRD, ALS, and MS after controlling for RUGIII-HC?

4.3 Relevance of Research

The need for an increased understanding of cost for the neurological conditions is undeniable. According to the WHO (2006, p.1), "a large body of evidence shows that policy-makers and health-care providers may be unprepared to cope with the predicted rise in the prevalence of neurological and other chronic disorders and the disability resulting from the extension of life expectancy and ageing of populations globally." In Canada, the research needed to cope with the expected rise in the prevalence of neurological conditions has been outlined by CIHI in their 2007 *Burden of Neurological Diseases, Disorders and Injuries in Canada* report. This report specifically called for research that can "examine how factors such as severity of illness, comorbidities and quality of care influence the patterns of healthcare utilization by patients with neurological conditions" (CIHI, 2007, p.16). The findings from this project will contribute to the research undertaken through the Innovations in Data, Evidence, and Applications for Persons with Neurological Conditions (ideas PNC) project. The ideas PNC project is aimed at identifying factors affecting individuals with neurological conditions such as changes to quality of life and resource utilization. This study will respond to the need for direct cost data beyond the current scope of hospital, physician, and drug expenditures. The addition of expenditure information for HC programs will improve the availability of information, and enhance understanding on the direct cost of healthcare for Canadians with neurological conditions.

5.0 METHODS

5.1 Basic Design

A secondary analysis of assessment data and costs from a 13-week study of home care was used in order to estimate, and determine the predictors of costs for ADRD, ALS and MS in the community. Assessment data were collected at a single point in time, while billed cost data were collected in the 13 weeks following initial assessment. Although secondary data analyses have many limitations, the availability of high quality data, the long duration of neurological conditions, and the low costs associated with this proposed study warrant the use of this approach. Indeed, one of the main advantages of this proposed project was the availability of comprehensive assessment and cost data from the Canadian Staff Time Resource Intensity Verification (CAN-STRIVE) project, which includes the entire population of individuals receiving long-stay HC in Ontario.

5.2 Ethics

This project was cleared for ethics by the Office of Research Ethics at the University of Waterloo on 10 December 2012 (Appendix D).

5.3 Study Sample and Setting

The sample for this project was drawn from the sub-study of CAN-STRIVE project dealing specifically with HC (Hirdes et al., 2010). This research project was based on a parallel US Staff Time and Resource Intensity Verification Project that was also intended to examine the RUG-III system. In both Canada and the United States, the aim was to validate the use of the RUG-III for the funding of institutionalized care, but only the Canadian study extended to research to HC. The CAN-STRIVE project was funded by Ontario Ministry of Health and Long

Term Care, and the Canadian Institute for Health Information (CIHI). Data for the CAN-STRIVE study were collected in Ontario, Canada for CCAC clients receiving long-stay services between April 2005 and June 2008. While long-stay individuals included those on service for 60 days or greater, individuals receiving palliative, acute, or rehabilitation home care services do not receive a RAI-HC assessment, and were therefore not included into this dataset. Since those requiring palliative services are likely to have more advanced illness, the population of HC users with ADRD, ALS, and MS in this study are likely to represent less severe cases. In addition, those requiring palliative care will probably require greater levels of service. In contrast, acute and rehabilitation clients may be more likely to have short-term needs for intense services but may have less advanced illness meaning less long-term need for services compared with long-stay HC users in this study. The data were provided by the Ontario Association of Community Care Access Centers (OACCAC), which collects RAI-HC assessment, admission and discharge records, and service billing/payment records from contracted agencies from the 14 individual CCACs that can be linked by encrypted health card number. The entire population of individuals using HC services was included into the HC portion of the CAN-STRIVE project, which included a total of 435,141 client assessments.

5.4 Measurements

Data for exposure, outcome, and other variables were also gathered in the CAN-STRIVE project. The exposure of interest in this research project was identified as having ADRD, ALS, or MS, while the outcome of interest were the total HC costs. Other variables required for this proposed study included personal and clinical characteristic items that were expected to affect costs. These items fall into Andersen and Newman's (1973) predisposing, enabling, and illness (need) determinants of healthcare utilization, and include demographic, geographical, and

clinical variables. The RUGIII/HC case-mix algorithm provides a validated classification system for understanding resource utilization in HC. Presence or absence of neurological condition, as well as predictors of cost were identified from the clinical component of the CAN-STRIVE data set. Cost data were available as part of the data collected on resource use, but instead came from linked administrative datasets.

Cost Measurement

In the HC sample, average weekly costs were calculated by aggregating service episodes up to 13 weeks after the date of a RAI-HC assessment, with services provided in at least three of those weeks. Where discharge or a follow-up RAI-HC occurred, the episode length used to calculate average weekly costs was determined by those dates. A single set of standard service costs that represented median wages from 2007/2008 for each of the various service categories were applied to billing/payment records from contracted agencies. Use of median wage rates were used since they permit the average weekly cost to represent the volume of services provided, rather than the actual expenditures. Depending on the type of service provided, costs were applied either hourly, or per visit. However, CCAC case manager time was not included as part of these service costs. The wage rates for the various services can be found in Appendix E. Informal care costs were also available for the HC sample. These costs were calculated by taking the reported hours of support for activities of daily living (ADL) and instrumental activities of daily living (IADL) over the 7 day period, and applying an hourly rate in order to valuate personal care. This estimation procedure was appropriate for the proposed research project since it has been used extensively in case-mix research (Björkgren et al., 2000; Poss et al., 2008). A wage rate of $14.09 was applied to the weekly personal care hours, which was half of the

standard hourly wage rate of a personal support worker. The total cost of HC was calculated by combining the informal care costs with the average weekly costs.

Exposure Measurement

Individuals with ADRD, ALS, and MS in the CAN-STRIVE HC sample were identified through the Disease Diagnoses section, section J, or the RAI-HC. ADRD (RAI-HC item J1g and J1h), and MS (J1k) were available from a pick list, while ALS diagnosis was written in using the International Classification of Disease (ICD) 10th Revision codes (J2a to J2d). These conditions were considered present if they were coded as either 1, indicating that the condition was present but did not require HC treatment or service, or 2, indicating that the condition was either being monitored or treated by a HC professional.

Independent (Predictor) Variables

Personal and clinical variables were identified as potential predictor variables based cost and clinical literature for ADRD, ALS, and MS. The findings from the literature reviewed can be found in the Neurological Conditions, and Review of Existing Literature sections. Any symptoms or clinical characteristics associated with ADRD, ALS, or MS, and any variables tested as a potential predictor of cost in the existing cost literature were included for analysis for all three conditions. All variables that were included in the analyses have been organized based on the Andersen and Newman classification of healthcare resource utilization.

Predisposing Variables:

Predisposing variables examined here included age and sex. Approximate age at time of assessment was estimated based on individuals' reported date of birth (BB2) and RAI-HC

assessment date (A1), and was kept as a continuous variable. Sex (BB1) was reported as a binary variable where zero indicated female, and one indicated male.

Enabling Variables:

Enabling variables are factors that permit or hinder individuals to access healthcare services. For this project, these included residence in one of the 14 CCACs, and cohabitation with a primary or secondary caregiver. Individuals' CCACs were available in the existing CAN-STRIVE HC dataset. The CCACs were coded as dummy variables, with the Hamilton Niagara Haldimand Brant CCAC as the reference category. Cohabitation with either a primary or secondary caregiver was identified using the 'lives with client' item in the RAI-HC (G1a and G1b). These items were collapsed into a single binary variable so that zero indicated that the individual did not live with either a primary or secondary caregiver, and one indicated cohabitation with either or both primary and secondary caregivers.

Need Variables:

In total, 39 different need variables were included for analysis. These consisted of variables reflecting ADRD, ALS, or MS symptoms and associated impairments (Ropper and Samuels, 2009; Compston and Coles, 2002; Kiernan et al., 2011; Krivickas et al., 1997; Ballard et al., 2011). These span across a number of categories including mobility, daily function, bladder and bowel, swallowing, breathing, speech, cognitive, behavioural, mental, and vision impairments, as well as pain, and dizziness. Neurological comorbidities that are common in the HC population were also considered. Health related quality of life (HRQOL) was also included as one of the variables of interest (McCrone et al., 2008; Grima et al., 2000). Finally, the RUGIII/HC case-mix ratios were also included due to its ability to explain resource intensity; however, this variable was also included to control for any clinical or personal characteristics not

associated with ADRD, ALS, or MS that may affect drive resource use. More detailed descriptions of the individual and scale measures can be found as follows.

Cognitive symptoms

Cognitive functions such as memory loss and communication can be affected by neurological disorder. For ADRD in particular, cognitive performance describes not only the progression of the condition, but also may predict the cost of care (Jönsson et al., 2006; Andersen et al., 2003). Although less common, cognitive impairment can also affect individuals with ALS and MS (Compston and Coles, 2002; Kiernan et al., 2011). As such, cognitive performance was included as a potential predictor variable, and was measured using the Cognitive Performance Scale (CPS). The CPS was originally created for use in nursing home settings and included seven measures of cognitive performance available in the RAI for nursing home settings. These items include comatose status, short-term memory, cognitive skills for daily decision-making, the ability to be understood by others and ADL self-performance in eating (Morris et al., 1994). The items included in the CPS were derived from the Mini-Mental State Exam (MMSE), which is a widely used test of cognitive status that has been found to be both valid and reliable, and the Test for Severe Impairment, which addresses sensitivity issues in the MMSE (Morris et al., 1994). The resulting test for cognitive performance is a hierarchical algorithm comprised of seven levels, from intact (scale score 0) to very severe impairment (scale score 6) that has been found to be both reliable and valid in both nursing home and HC settings (Morris et al., 1994; Landi et al., 2000). The CPS was kept in its original form with seven levels in this analysis.

Functional and Movement Symptoms

ADLs describe the ability to perform daily activities required for self-care. The first ADL classification was introduced by Katz (1963) to measure function as an indicator of care needs, as well as to evaluate the effectiveness of treatment. In this analysis, ADLs were used as indicators of self-care function affected by symptoms of ADRD, ALS and MS, such as gait ataxia, muscle weakness, spasticity and rigidity. In addition, difficulties performing ADLs have been identified in some studies to be associated with cost (Kang et al., 2006; Gustavsson et al., 2011). ADLs are typically evaluated using a summary measure that determines dependence for daily activities such as bathing, eating, dressing and locomotion. The RAI-HC includes ADL items on dressing (H2e and H2f in RAI-HC), personal hygiene (H2i), toilet use (H2h), locomotion (H2c and H2d), transfer (H2b), bed mobility (H2a), and eating (H2g). These items are then scaled from independent (0) to total dependence (4), with the additional option where the activity did not occur (8), which is also coded as total dependence (Morris et al., 1999). Although these scores can be used to produce summary scales, the hierarchical classification of ADLs was chosen for this project. This Self-Performance Hierarchy uses only four of the seven ADL items: eating, locomotion, personal hygiene and toilet use. These items were identified through factor analysis as the most consistent indicators of early, middle, and late loss of ADL self-performance (Morris et al., 1999). The Self-Performance Hierarchy has seven levels including independent, supervision, limited, extensive 1, extensive 2, dependent, and total dependence (Morris et al., 1999). The scoring criteria can be found in Appendix F. Although the Self Performance Hierarchy was originally created for the interRAI nursing home instrument, it it is both valid and reliable in HC settings as well (Landi et al., 2000).

Other movement and physical function characteristics are also present for ADRD, ALS, and MS, and can potentially affect the cost of care. These items include unsteady gait, falls, and stair climbing. The item for unsteady gait (K6a) was a binary variable and was considered present if coded as one. Stair climbing (H5) was measured as 'up and down stairs without help,' 'up and down stairs with help,' and 'not go up and down stairs,' and coded as zero, one, and two respectively. This variable was kept in its continuous form. Finally, falls were identified using the 'falls frequency' item (K5). This item was collapsed into a binary variable so that zero denoted that no falls had occurred, while a value of one denoted that at least one fall had occurred in the previous 90 days.

Like ADLs, IADLs also measure the self-maintenance abilities of individuals. While ADLs are generally physical functions that are more likely to be impaired in the elderly, IADLs are functions considered to be typical of normal activities prior to old age, such as shopping, using transportation, and managing finances (Lawton and Brody, 1969). Despite the relatively young onset for ALS and MS conditions, IADL limitations had not been identified in the review of clinical literature. Given some of the symptoms associated with ALS and MS, however, it was felt that at least some difficulty with IADLs could be expected in these individuals. IADL self-function impairments were specifically identified as symptoms of ADRD. As a result, IADL was considered a characteristic of interest across all three conditions. IADL function was measured using the interRAI IADL capacity scale, which is highly correlated with the original, validated Lawton and Brody (1969) scale in the HC setting. Items in the scale include self-performance in meal preparation (H1a), housework (H1b), use of the phone (H1e), transportation (H1g), shopping (H1f), managing finances (H1c), and taking medications (H1d) (Landi et al., 2000).

Breathing, Swallowing, and Expression (Speech)

Dysphagia was measured using the swallowing item (L3). The swallowing item was kept in its original form, which had the following levels: normal (0), requires diet modification to swallow solid foods (1), requires diet modification to swallow solid foods and liquids (2), combined oral and tube feeding (3), and no oral intake (4). In addition, since dysphagia can lead to malnutrition and associated weight loss, the severe malnutrition (L1b) and 'unintended weight loss of more than five percent in last 180 days' (L1a) items were also included as potential predictor variables. Verbal expression was measured using the 'making self understood' item (C2), and was kept as a continuous variable ranging from zero to four, as found in the RAI-HC. Zero indicated understood, while a value of four indicated rarely/never understood. Finally, dyspnea was also included and was measured using the shortness of breath (K3e), respirator use (P2b), other respiratory treatment (P2c), and tracheostomy (P2m) items. All of these items were kept in their original form as binary variables. Presence of any of these characteristics was identified where items were coded as one, and zero denoted absence of these characteristics.

Bladder and Bowel Symptoms

Bladder incontinence (I1a) and bowel incontinence (I3) were considered present if they were coded as greater than, or equal to three. Constipation was identified using the binary 'no bowel movement in three days' item (K3b), and was considered present if coded as one.

Visual Symptoms

Visual impairment was coded as a binary variable. It was considered present if the RAI-HC vision item (D1) was coded as moderately impaired (2), highly impaired (3), or severely impaired (4), or the visual limitation/difficulties item (D2) was coded in one. Although an item

was available measuring vision decline, it was not included as part of the visual symptoms item because vision decline over time was not a symptom of ADRD, ALS, or MS.

Pain symptoms

Pain was identified as a common symptom in individuals with MS and ALS. For the current project, pain was measured using the interRAI Pain Scale. The interRAI Pain Scale was developed using the Visual Analogue Scale (VAS) as the external standard because of its efficiency and sensitivity for various levels of pain (Fries et al., 2001). Pain frequency and intensity, as found in the interRAI tools, were predictive of VAS scored pain (Fries et al., 2001). Initial separation in the pain algorithm was split into the following groups: no pain, less than daily pain, and daily pain (Fries et al., 2001). The daily pain category was then further split by pain intensity, into daily mild to moderate pain, and horrible or excruciating daily pain (Fries et al., 2001). In total, the interRAI Pain Scale is comprised of four levels, and has high concordance with the VAS, which is considered the "gold standard" in the measurement of pain (Fries et al., 2001). The four levels are no pain (0), mild pain that is less than daily (1), moderate pain describing daily mild or moderate paint (2), and severe pain describing daily pain that is horrible or excruciating (3). Again, although the Pain Scale was originally devised for nursing home setting, the large proportion of similar items between the nursing home and HC instruments suggests that this scale should effectively identify various levels of pain in HC as well (Zyczkowska et al., 2007).

Behavioral, Mood, and Mental Symptoms

Mood disorders, primarily depression and anxiety, are common for individuals with neurological conditions. They can be part of the clinical characteristic of a given neurological condition, but can also be caused by diagnosis with the neurological condition. Depression and

anxiety associated with ADRD, ALS and MS were considered as potential clinical predictors of cost and were measured using the Depression Rating Scale (DRS). The DRS was validated against the Hamilton Depression Rating Scale, which is the current standard for psychiatric research, as well as the Cornell Scale for Depression in Dementia, which is relevant to the nursing home population (Burrows et al., 2000). According to Burrows et al., (2000), the DRS items include the following:

a. Resident made negative statements (E1a)
b. Persistent anger with self or others (E1b)
c. Expressions of what appear to be unrealistic fears (E1c)
d. Repetitive health complaints (E1d)
e. Repetitive anxious complaints, concerns (E1e)
f. Sad/pained/worried facial express (E1f)
g. Recurrent crying/tearfulness (E1g)

For each of these items, corresponding numerical scores were given to indicate that the behavior had not exhibited in last three days (0), had been exhibited in one to two of last three days (1), or exhibited on each of last three days (2). The DRS is a continuous variable with a range from zero to 14. The cutoff for mild to moderate depression is considered three on this scale (Burrows et al., 2000). The DRS has been used in a number of studies of HC clients (Dalby et al., 2008; Szczerbinska et al., 2011; Maxwell et al., 2008)

Other behavioral and mental symptoms not included in the RAI DRS are also common, particularly in individuals with ADRD. The following additional characteristics were therefore also included for analysis: aggression/hostility, delusions, hallucinations, and reduced social interaction. Aggression and/or hostility were categorized into a single binary variable. These symptoms were considered to be present if verbally abusive symptoms (E3b) or physically abusive symptoms (E3c) were coded as either one or two, or the openly expresses conflict with family of friends item (F1b) was coded as one. The delusions (K3f), hallucinations (K3g), and

social interaction (F1a) criteria were kept in their original binary form from the RAI-HC, and were considered present if they were coded as one.

Comorbidities

Since comorbidities are common for at least some of the neurological conditions, other neurological conditions in the HC dataset were included in the analyses. The neurological comorbidities tested include ADRD, ALS, cerebral palsy, epilepsy, Huntington's disease, MS, muscular dystrophy, Parkinson's disease, stroke, spinal cord injury, and traumatic brain injury. As with identifying the presence of ADRD, ALS, and MS, the presence of these other conditions could be identified from the Disease Diagnoses section of the RAI-HC. Traumatic brain injury (J1j) and Parkinson's disease (J1i) could be found in the pick list, while the remaining conditions were written in using ICD 10 codes. These comorbidities were included into the analyses as binary variables, with a value of one indicating presence of the comorbidity, and zero indicating absence of the comorbidity. As the aim of this project was to identify the effects of characteristics of neurological conditions on cost, non-neurological comorbidities were not tested in the analyses.

Health Related Quality of Life

Although HRQOL did not necessarily pertain to any specific clinical characteristic for ADRD, ALS or MS, it was included as a potential predictor of cost for each of the three conditions considered in this project. Primarily, HRQOL was included because it is an important outcome in health economics research, and therefore has the potential to affect the cost of care. HRQOL was measured in this project using the Health Status Index (HSI), which is a crosswalked version of the Health Utilities Index Mark Two (HUI2) (Wodchis et al., 2003; Wodchis et al., 2007). The HUI2 is a health status classification system that is based on seven

attributes and associated preference weights (Wodchis et al., 2003). The HSI is comprised of 33 items in HC, measuring sensation, mobility, emotion, cognition, self-care and pain (Wodchis et al., 2003). Unlike the HUI2, the HSI does not include fertility as a component of HRQOL (Wodchis et al., 2003). The scoring for each of the attributes in the HSI reflects the preferences of a representative community sample for each health state, and was derived from the HUI2 (Wodchis et al., 2003). A scoring function is then used to produce a total HSI score from zero to one, where one represents perfect health, and zero indicates death (Wodchis et al., 2003). The worst possible score is -0.03, and represents health states considered to be worse than death (Wodchis et al., 2003). The multiplicative preference scoring function, and the list of RAI-HC items included in the HSI scale can be found in Appendix G. Preliminary evidence suggests that the HSI has good validity, and can be used to substitute the HUI2 for group-level comparisons (Wodchis et al., 2003; Wodchis et al., 2007).

Case-mix

The RUGIII/HC was included in order to control for clinical characteristics not associated with the condition of interest. In addition, since case-mix systems are designed to group individuals into resource homogenous groups, it was also identified as a potential predictor of cost for ADRD, ALS, and MS. The RUGIII/HC system classifies clients into seven clinical levels: Special Rehabilitation, Extensive Services, Special Care, Clinically Complex, Impaired Cognition, Behaviour Problems, and Reduced Physical Functions (Björkgren et al., 2000). Inclusion in the Special Rehabilitation category was defined by use of over 120 minutes of physical, occupational, or speech therapy per week (Björkgren et al., 2000). Within each category, further subgroups were determined based on ADL items, as well as IADL items for the Special Rehabilitation, Clinically Complex, Impaired Cognition, Behaviour Problems, and

Reduced Physical Functions categories (Björkgren et al., 2000). In total, the RUGIII/HC system is comprised of 23 groups. The structure of the RUGIII/HC, as well as the inclusion criteria for each of the seven levels can also be seen in Appendix H. Ratio level values were then assigned to each of the 23 RUGIII/HC categories. These values were based on the case-mix index (CMI) values for combined formal and informal care time, derived from the HC sub-study of the CAN-STRIVE project (Hirdes et al., 2010). The RUGIII/HC categories and associated case-mix values can be found in Appendix I.

5.5 Statistical Analysis

Statistical analysis for this study was carried out using Statistical Analysis Software (SAS), Version 9.2 (SAS Institute, Cary, NC, USA). Descriptive and sample characteristics were calculated using frequency and means procedures. Bivariate regression analyses were performed for all independent variables across all three conditions. Multivariate linear regression analyses were carried out for each of the three conditions using the hypothesized characteristics. Since the analyses were performed on population-level data, and the population sizes were very large in all cases, it was expected that the multivariate regression models would be very robust. Nevertheless, since cost is typically heavily left skewed, a log-transformed model was also run. This log transformation was carried out in order to check the effects that any of the violation of assumptions for regression analysis had produced any misleading results.

Bivariate Analysis

Bivariate analyses were carried out between all hypothesized variables for each diagnosis group. These included frequency and means procedures used to produce descriptive statistics of the HC population, as well as mean costs. Descriptive statistics were also generated for a comparison group using the same bivariate analyses. The comparison group was composed of all

individuals within the HC sample without ADRD, ALS, and MS, as well as cerebral palsy, epilepsy, Huntington's disease, muscular dystrophy, Parkinson's disease, spinal cord injury, stroke, and traumatic brain injury. Simple linear regressions were also used to test the association between each of the hypothesized variables and cost in order to gain an appreciation of the independent associations between cost and all hypothesized variables for ADRD, ALS, and MS. However, nonsignificance in simple regression was not an excluding criteria for testing in multivariate regression analyses.

Multivariate Regression Analysis

For both the transformed and untransformed models, full models were initially run that included all 41 independent variables. Manual backward elimination was then performed in order to remove nonsignificant variables. A number of steps were taken to ensure that only truly nonsignificant variables were removed from the model. The cutoff for significance was intentionally set at a p-value of 0.10 during the initial removal of nonsignificant variables in order to keep as many variables within the model as possible. In addition, all variables that were removed during this initial stage were then individually re-added to the model and re-run to test for significance in the absence of the other removed variables. Variables that were significant at the 0.10 level were then reintroduced into the model simultaneously for further backward elimination. At this stage, the significance level was reduced to a maximum p-value of 0.05. All variables with p-values greater than 0.05 were then removed, and individually re-entered once again. All variables that were significant at the 0.05 level when individually reintroduced were again entered to the model simultaneously. These steps were repeated in cases where variables became nonsignificant when reintroduced to the model, or where other variables became nonsignificant after the reintroduction of the variables. Where more than one model was

identified, the explained variance value was used to identify the best model. In addition, possible alternative models were also checked using automatic forward selection, backward elimination, and stepwise procedures. As a final step, all removed variables were individually reintroduced into the model to ensure that as many variables remained in the models as possible.

Interaction terms between variables thought to be theoretically relevant were also included into the analyses after the final model had been identified. These interaction terms included age and ADL capacity, age and IADL capacity, age and cohabitation, sex and cohabitation, difficulty breathing and respirator, difficulty breathing and respiratory treatment other than respirator, and difficulty breathing and tracheostomy.

Model Diagnostics

Model diagnostics were performed by checking the residual plots for both the transformed and untransformed final models for each of the three conditions. In addition, collinearity was checked using the variance inflation factor (VIF). There is no standard cut point to suggest the presence of collinearity; however, VIF between four and ten have traditionally been suggested as appropriate cut points (O'Brien, 2007). Even based on the most conservative of cut points, there was no evidence to suggest that serious collinearity was present in any of the ADRD and ALS models. For the MS models, only the CMI and ADL variables had VIF greater than four, at 4.1 and 4.2, respectively. Given that the VIF for these variables remained on the low end of previously suggested cut point range, collinearity was not thought to be of concern even in the MS models. Based on even the most liberal VIF cut point of ten, however, all interaction terms had to be removed from all the models. VIF for the interaction terms as well as ADL, IADL, cohabitation, sex, breathing, respirator use, other respiratory treatment, and tracheostomy

variables ranged from 23 to 115, indicating very strong collinearity between interaction terms and these variables.

Missing Values

Models were checked for observations with missing values, and means procedures with the missing option were used to identify the variables with missing variables. These variables included ADL, speech, vision, and CMI for MS and ADRD. No missing values were found for any of the variables in ALS. Since the sample size for ADRD, and MS were both large and no variable had greater than four missing variables, any observations with missing values were excluded from multivariate regression analyses. SAS automatically excludes such observations from multivariate regression analyses. Therefore no additional steps had to be taken to remove these variables.

Sensitivity Analysis

As the CAN-STRIVE dataset includes repeat assessments for at least some individuals with ADRD, ALS, and MS, a one-way sensitivity analysis was conducted on all models to assess the impact of the repeat assessments in the dataset on the conclusions of the final models. For each of the conditions, the final untransformed and transformed models identified were re-run using a sample that excluded repeat assessments. Repeat assessments were removed so that only the most recent RAI-HC assessments were included into the analyses.

6.0 RESULTS

6.1 Sample Characteristics

Although 435,141 assessments were available, only 70,061 assessments were identified where ADRD, ALS, or MS were present. In total, ADRD was present in 59,310 assessments, representing 25,901 individuals; ALS was present in 991 assessments, representing 452 individuals; and MS was present in 9946 assessments, representing 3309 individuals. Within the total sample of individuals included in these analyses, the average age was 79. In comparison, the average ages for ADRD, ALS, and MS were 82, 63, and 58, respectively.

Table 2 shows the distribution of individuals across the predisposing variables. Distributions across the three neurological conditions and the comparison group show differences in the age and sex of individuals across these four groups. The comparison group was defined as individuals within the sample that did not have a diagnosis for ADRD, ALS, and/MS, as well as cerebral palsy, epilepsy, Huntington's disease, muscular dystrophy, Parkinson's disease, spinal cord injury, traumatic brain injury, and/or stroke. Individuals with MS and ALS were clustered in the 40 to 79 age groups. Meanwhile, individuals with ADRD and those without any of the 11 defined neurological conditions were primarily concentrated in the 60 to 99 age groups. Across conditions, a greater proportion of individuals were female; however, the sex distribution did vary across the groups.

Differences in the distribution of individuals across CCACs and the proportion of individuals living with caregivers were also evident across the four groups. Overall, the distribution of individuals across the CCACs was similar across the ADRD and MS groups, as compared to both the comparison group, and the entire HC population (not shown). The proportion of individuals with ADRD in WW was higher in comparison to the distribution of

individuals with other conditions in the WW CCAC. Individuals with ALS, however, were present in higher proportions as compared to the overall HC population in a number of CCACs. These CCACs included the MH, CW, Central, and Champlain CCACs. The rates of individuals cohabiting with caregivers were also different across the groups. In all cases, individuals with ADRD, ALS, and MS were more likely to live with a primary or secondary caregiver than individuals without neurological conditions. In particular, an overwhelming proportion of individuals with ALS lived with their caregivers. The distribution of these enabling factors can be found in Table 3.

The proportion of individuals experiencing problem symptoms or characteristics associated with their conditions were generally higher for ADRD, ALS, and MS as compared to individuals without any of the 11 identified neurological conditions listed above. Individuals with neurological conditions had greater difficulties with ADLs. Among the three neurological conditions considered in this project, the proportion of individuals with extensive ADL impairments (ADL score ≥ 3) was highest for those with ALS. A similar pattern was found for IADL impairments where again, individuals with ALS were concentrated in the highest impairment groups at the highest rates for IADL. In addition, greater proportions of individuals with ALS had unsteady gait, malnutrition, weight loss, respiratory treatment, breathing difficulty, and a larger proportion had experienced at least one fall in the past 180 days as compared to those with ADRD, MS, or without neurological conditions. Individuals with ADRD, ALS, and MS were unable to use stairs at higher rates than those without neurological conditions, although individuals with MS were most likely to be unable to use stairs. Individuals with MS also displayed the highest proportion of pain, dizziness, and constipation. Individuals with ADRD were likeliest to have bowel and bladder incontinence, as well as withdrawal from social

involvement, hallucinations, delusions, and other neurological comorbidities. The distributions of these need factors can be found in Table 4.

Table 2: Distribution of Predisposing Factors by Condition by Percent (number)

	ADRD	ALS	MS	Comparison Group
Age				
0-19	0.2 (92)	0.0 (0)	0.1 (9)	0.3 (679)
20-29	0.0 (22)	0.3 (3)	0.7 (70)	0.8 (2241)
30-39	0.1 (55)	2.7 (27)	5.8 (576)	1.4 (3908)
40-49	0.3 (147)	12.4 (123)	19.7 (1959)	3.8 (10,405)
50-59	1.1 (632)	23.7 (235)	31.0 (3084)	7.5 (20,581)
60-69	4.6 (2742)	26.8 (265)	24.6 (2448)	11.5 (31,438)
70-79	25.5 (15,140)	25.2 (249)	13.6 (1356)	23.9 (65,397)
80-89	53.1 (31,479)	8.7 (86)	4.3 (430)	38.0 (103,880)
90-99	14.9 (8806)	0.1 (1)	0.2 (16)	12.3 (33,545)
100-115	0.3 (193)	0.1 (1)	0.0 (0)	0.4 (1071)
Sex				
Male	36.1 (21,403)	46.2 (458)	24.5 (2433)	28.3 (77,273)
Female	63.9 (37,915)	53.8 (533)	75.5 (7515)	71.7 (195,929)

Table 3: Distribution of Enabling Factors by Condition by percent (number)

	ADRD	ALS	MS	Comparison Group
Cohabit				
Yes	66.1 (39,206)	83.4 (826)	70.6 (7018)	48.8 (133,298)
CCAC				
HNHB CCAC	17.3 (10,281)	14.5 (144)	17.6 (1754)	16.8 (45,867)
MH CCAC	5.0 (2968)	9.9 (98)	4.1 (406)	4.8 (13,030)
CW CCAC	2.6 (1555)	4.7 (47)	2.9 (285)	3.0 (8116)
TC CCAC	5.3 (3126)	4.2 (42)	4.1 (403)	6.2 (16,804)
CENT CCAC	10.6 (6270)	15.2 (151)	8.0 (792)	10.1 (27,471)
CE CCAC	11.3 (6676)	7.7 (76)	13.0 (1289)	11.9 (32,478)
SE CCAC	4.5 (2688)	3.1 (31)	3.7 (363)	3.7 (10,226)
CHAM CCAC	10.6 (6288)	12.6 (125)	9.2 (911)	7.4 (20,153)
NSM CCAC	3.5 (2073)	4.0 (40)	3.2 (315)	3.7 (10,202)
NE CCAC	5.3 (3157)	6.3 (62)	6.0 (598)	6.6 (17,987)
NW CCAC	2.8 (1679)	2.4 (24)	4.0 (396)	3.2 (8746)
ESC CCAC	5.6 (3339)	4.2 (42)	6.3 (628)	6.2 (17,027)
SW CCAC	8.2 (4870)	8.8 (87)	12.2 (1215)	11.6 (31,654)
WW CCAC	7.3 (4348)	2.2 (22)	6.0 (593)	4.9 (13,441)

Table 4: Distribution of Need Variables by Condition by percent (number)

	ADRD	ALS	MS	Comparison Group
ADL Hierarchy				
0	39.8 (23,577)	27.9 (276)	43.4 (4318)	77.5 (211,672)
1	19.2 (11,397)	6.8 (67)	6.5 (650)	7.5 (20,547)
2	18.3 (10,881)	16.5 (163)	10.6 (1053)	8.5 (23,364)
3	10.8 (6387)	7.7 (76)	17.2 (1715)	3.4 (9312)
4	5.7 (3383)	14.6 (145)	7.9 (788)	1.7 (4727)
5	3.6 (2134)	15.1 (150)	8.4 (840)	1.1 (2882)
6	2.6 (1558)	11.5 (114)	5.9 (583)	0.3 (797)
IADL Capacity				
0	1.1 (676)	1.1 (11)	1.1 (106)	5.8 (15,908)
1	2.5 (1459)	2.0 (20)	3.6 (361)	12.6 (34,321)
2	6.9 (4070)	5.2 (51)	11.5 (1145)	16.9 (46,222)
3	2.9 (1715)	1.0 (10)	0.4 (39)	0.9 (2512)
4	11.7 (6938)	14.6 (141)	22.7 (2259)	29.2 (79,750)
5	34.0 (20,157)	40.1 (397)	49.1 (4884)	29.9 (81,706)
6	41.0 (24,303)	36.4 (361)	11.6 (1154)	4.7 (12,783)
Unsteady Gait				
Yes	60.5 (35,860)	71.3 (707)	64.7 (6431)	56.0 (152,884)
Falls[1]				
Yes	31.0 (18,384)	40.8 (404)	33.7 (3355)	24.9 (67,999)
Stair Use				
Without Help	36.7 (21,740)	20.7 (205)	15.2 (1516)	41.0 (111,993)
With Help	22.2 (13,181)	18.8 (186)	11.1 (1108)	16.3 (44,431)
Does not use Stairs	41.1 (24,397)	60.6 (600)	73.6 (7324)	42.7 (116,778)
Difficulty Swallowing				
Normal	89.6 (53,165)	43.7 (433)	83.1 (8269)	94.7 (258,687)
Diet modifications for solid foods	7.1 (4226)	20.4 (202)	11.1 (1105)	3.7 (10,185)
Diet modifications for solid foods and liquids	2.7 (1605)	18.6 (184)	3.5 (351)	0.8 (2294)
Oral and tube feeding	0.2 (94)	7.2 (71)	0.9 (85)	0.4 (1171)
No oral intake	0.4 (228)	10.2 (101)	1.4 (138)	0.3 (862)
Malnutrition				
Yes	0.8 (465)	2.1 (21)	0.8 (78)	1.0 (2585)
Weight Loss[2]				
Yes	5.4 (3208)	15.3 (152)	3.3 (329)	6.9 (18,868)
Respirator Use				
Yes	0.2 (89)	6.5 (64)	0.4 (39)	0.5 (1323)
Other Respiratory				

[1] Yes denotes those with at least one fall in the previous 90 days.
[2] Yes denotes unintended weight loss of at least 5% of body weight in previous 120 days.

Treatments[3]				
Yes	2.9 (1689)	12.1 (12)	3.6 (355)	5.9 (16,076)
Tracheostomy				
Yes	0.0 (22)	2.3 (23)	0.2 (21)	0.3 (840)
Breathing Difficulty				
Yes	15.5 (9204)	29.0 (287)	10.1 (245)	28.5 (77,786)
Expression				
Understood	44.5 (26,396)	69.6 (69)	82.7 (8221)	91.1 (248,786)
Usually understood	25.5 (15,136)	11.6 (115)	11.4 (1137)	6.8 (18,638)
Often understood	15.4 (9147)	4.4 (44)	3.0 (302)	1.2 (3248)
Sometimes understood	9.9 (5884)	9.9 (98)	2.0 (198)	0.7 (1813)
Rarely/never understood	4.6 (2751)	4.4 (44)	0.9 (89)	0.3 (707)
CPS				
0	2.5 (1456)	61.5 (609)	60.0 (5966)	68.6 (187,531)
1	7.7 (4573)	24.9 (247)	19.1 (1902)	16.6 (45,281)
2	47.6 (28,222)	8.4 (83)	16.4 (1631)	12.5 (34,206)
3	21.8 (12,919)	2.9 (29)	2.3 (228)	1.3 (3545)
4	3.4 (2042)	0.5 (5)	0.1 (14)	0.2 (479)
5	13.9 (8292)	0.1 (1)	0.8 (79)	0.6 (1651)
6	3.1 (1814)	1.7 (17)	1.3 (128)	0.2 (508)
DRS				
0	58.7 (34,797)	48.9 (485)	64.3 (6397)	66.3 (181,173)
1	14.5 (8627)	17.4 (172)	11.6 (1156)	12.5 (34,144)
2	11.0 (6481)	13.0 (129)	9.5 (945)	8.7 (23,778)
3	5.1 (3015)	6.8 (67)	4.9 (488)	4.3 (11,649)
4	4.2 (2490)	4.8 (48)	3.4 (341)	3.2 (8808)
5	2.0 (1177)	2.0 (20)	1.9 (193)	1.5 (4069)
6	1.9 (1113)	3.4 (34)	1.9 (186)	1.6 (4257)
7	0.8 (487)	0.7 (7)	0.7 (73)	0.6 (1644)
8	0.8 (490)	2.0 (20)	0.7 (67)	0.6 (1554)
9	0.3 (198)	0.4 (4)	0.3 (29)	0.3 (671)
10	0.4 (246)	0.2 (2)	0.5 (45)	0.3 (673)
11	0.1 (74)	0.1 (1)	0.2 (17)	0.1 (234)
12	0.1 (76)	0.0 (0)	0.0 (7)	0.1 (331)
13	0.0 (20)	0.1 (1)	0.0 (3)	0.0 (78)
14	0.1 (27)	0.1 (1)	0.0 (1)	0.1 (137)
Pain Scale				
No pain	52.0 (30,814)	40.9 (405)	34.3 (3409)	25.7 (70,061)
Mild pain (not daily)	14.8 (8778)	10.4 (103)	11.4 (1132)	12.6 (34,326)
Mild/moderate pain (daily)	28.6 (16,987)	37.9 (376)	38.4 (3823)	45.3 (123,833)
Severe pain (daily)	4.6 (2738)	10.8 (107)	15.9 (1584)	16.5 (44,971)
Constipation				
Yes	0.81 (481)	1.1 (11)	2.5 (245)	1.0 (2700)

[3] Other respiratory treatments, excluding respirator use.

Bowel Incontinence				
Yes	19.6 (11,602)	6.1 (60)	17.8 (1766)	4.97
Bladder Incontinence				
Yes	44.4 (26,340)	14.9 (148)	39.3 (3907)	22.0 (60,154)
Vision[4]				
Yes	13.8 (8193)	4.8 (48)	13.0 (1292)	11.9 (32,444)
Dizziness				
Yes	11.7 (6948)	7.7 (77)	13.8 (1374)	16.6 (45,212)
Aggression				
Yes	18.8 (11,170)	11.1 (110)	13.2 (1309)	10.4 (28,322)
Social Withdrawal				
Yes	9.2 (5440)	3.4 (34)	2.8 (281)	2.7 (7478)
Hallucinations				
Yes	6.2 (3690)	0.2 (2)	0.4 (42)	0.7 (1897)
Delusions				
Yes	4.2 (2463)	0.2 (2)	0.2 (20)	0.4 (1146)
Comorbidities				
ADRD	N/A	1.9 (19)	1.7 (167)	0.0 (0)
Amyotrophic Lateral Sclerosis	0.0 (19)	N/A	0.1 (10)	0.0 (0)
Cerebral Palsy	0.0 (15)	0.0 (0)	0.2 (19)	0.0 (0)
Epilepsy	1.3 (756)	0.5 (5)	1.6 (156)	0.0 (0)
Huntington's Disease	0.1 (47)	0.0 (0)	0.0 (3)	0.0 (0)
Multiple Sclerosis	0.3 (167)	1.0 (10)	N/A	0.0 (0)
Muscular Dystrophy	0.0 (8)	0.0 (0)	0.1 (5)	0.0 (0)
Parkinson's Disease	6.8 (4048)	1.1 (11)	0.7 (70)	0.0 (0)
Spinal Cord Injury	0.0 (1)	0.1 (1)	0.1 (8)	0.0 (0)
Stroke	23.7 (14,068)	6.3 (62)	4.9 (484)	0.0 (0)
Traumatic Brain Injury	1.4 (829)	0.7 (7)	1.2 (117)	0.0 (0)
Health Status Index				
-0.03-0.13	3.9 (2317)	2.7 (27)	2.5 (247)	1.7 (4567)
0.14-0.30	18.7 (11,108)	15.4 (153)	13.6 (1354)	11.6 (31,764)
0.31-0.47	24.6 (14,614)	29.5 (292)	26.0 (2575)	19.2 (52,307)
0.48-0.64	26.8 (15,902)	28.1 (278)	35.1 (3496)	33.5 (91,394)
0.65-0.81	19.8 (11,747)	17.8 (176)	19.9 (1977)	23.1 (63,020)
0.82-1	6.1 (3630)	6.6 (65)	3.0 (299)	11.0 (30,150)

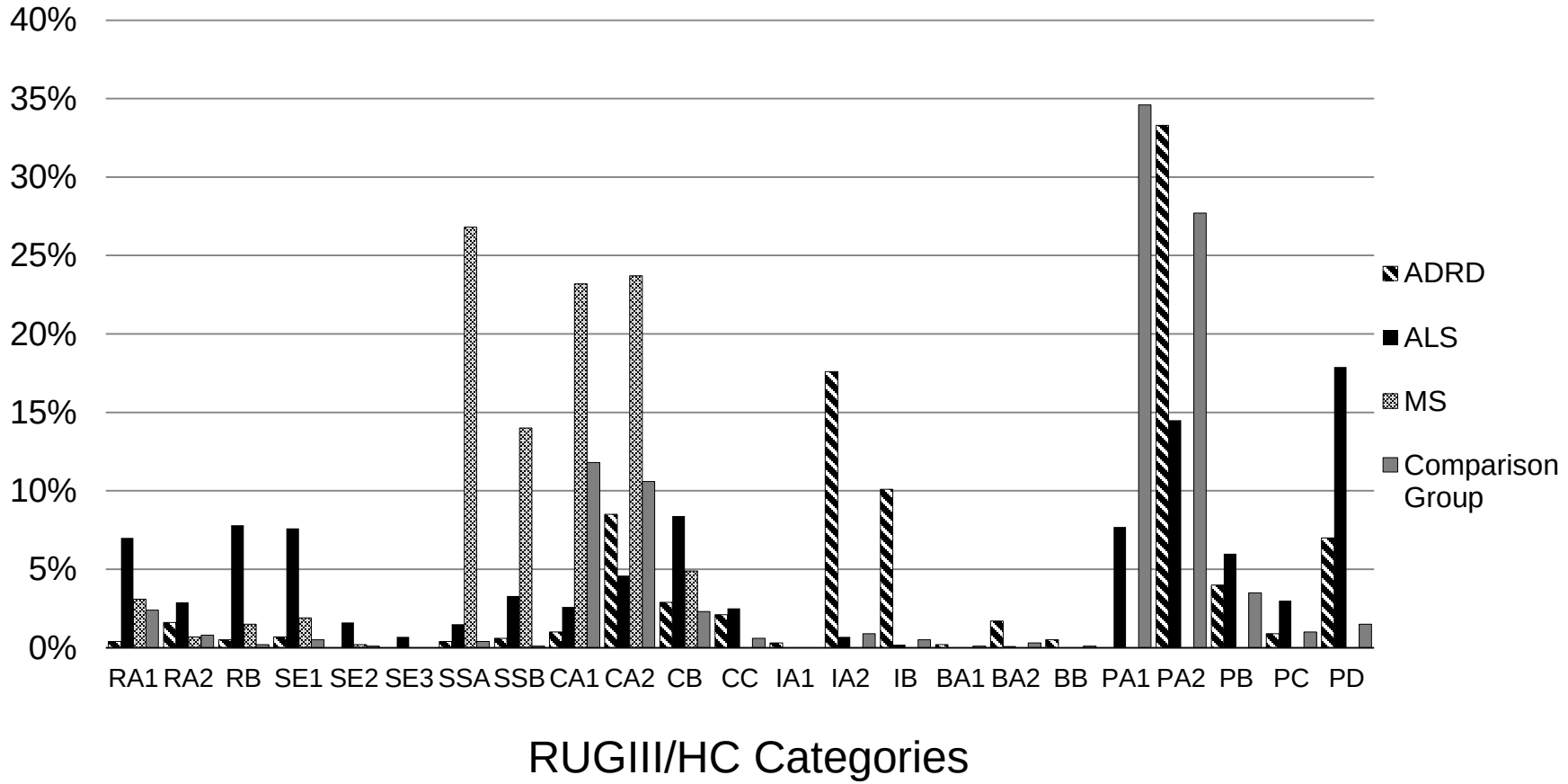
[4] Yes denotes any visual impairment, limitations, or difficulties.

Table 5 displays the distribution of individuals across the 23 RUGIII/HC CMI groups by condition. Individuals with ADRD were primarily clustered in the PA2, IA2, IB, CA2, PD, and PA1 categories, accounting for roughly 82 percent of the ADRD population. Not surprisingly, those with MS were also highly clustered in some case-mix groups due to the inclusion of the MS diagnosis criteria in the RUGIII/HC algorithm. These categories included CA1, CA2, SSA, and SSB. The clustering of the ALS population was less evident. Nevertheless, these individuals were present in higher proportions in the PPD, PA2, PA1, CB, SE1, and RA1 groups. The graphical distribution these populations across the RUGIII/HC can be found in Figure 2.

Table 5: Distribution of Population across RUGIII/HC CMI Groups by percent (number)

Formal and Informal RUGIII/HC Case-Mix Groups	ADRD	ALS	MS	Comparison Group
0.485 (PA1)	6.0 (3562)	7.7 (76)	0.0	34.6 (94,517)
0.593 (BA1)	0.2 (102)	0.0 (0)	0.0	0.1 (393)
0.609 (CA1)	1.0 (600)	2.6 (20)	23.2 (2310)	11.8 (32,173)
0.839 (IA1)	0.3 (152)	0.0 (0)	0.0	0.0 (115)
0.933 (PA2)	33.3 (19,742)	14.5 (144)	0.0	27.7 (75,768)
0.967 (RA1)	0.4 (223)	7.0 (69)	3.1 (305)	2.4 (6451)
1.126 (CA2)	8.5 (5050)	4.6 (46)	23.7 (2357)	10.6 (29,082)
1.281 (BA2)	1.7 (1004)	0.1 (1)	0.0	0.3 (848)
1.379 (PB)	4.0 (2344)	6.0 (59)	0.0	3.5 (9455)
1.609 (RA2)	1.6 (937)	2.9 (29)	0.7 (69)	0.8 (2287)
1.637 (IA2)	17.6 (10,422)	0.7 (7)	0.0	0.9 (2363)
1.660 (CB)	2.9 (1709)	8.4 (83)	4.9 (485)	2.3 (6384)
1.718 (BB)	0.5 (275)	0.0 (0)	0.0	0.1 (184)
1.755 (PC)	0.9 (513)	3.0 (30)	0.0	1.0 (2812)
1.878 (SSA)	0.4 (205)	1.5 (15)	26.8 (2661)	0.4 (1147)
2.121 (IB)	10.1 (5993)	0.2 (2)	0.0	0.5 (1273)
2.417 (PD)	7.0 (4129)	17.9 (177)	0.0	1.5 (3949)
2.498 (SE1)	0.7 (421)	7.6 (75)	1.9 (187)	0.5 (1298)
2.586 (CC)	2.1 (1257)	2.5 (25)	0	0.6 (1583)
2.743 (RB)	0.5 (313)	7.8 (77)	1.5 (153)	0.2 (625)
2.791 (SSB)	0.6 (345)	3.3 (33)	14.0 (1397)	0.1 (332)
4.240 (SE2)	0.0 (10)	1.6 (16)	0.2 (23)	0.1 (125)
5.151 (SE3)	0.0 (0)	0.7 (7)	0.0 (1)	0.0 (36)

Figure 2: Distribution of Population across RUGIII/HC CMI Groups



Across the three conditions, the average CMI was 1.42, and the average total weekly cost was $594.81. These means were close in value to the ADRD population, where the CMI, and weekly cost were 1.40, and $593.32, respectively. In contrast, both MS and ALS displayed substantially different sample characteristics. For the MS population, CMI and total weekly costs 1.52 and $574.92. ALS, meanwhile, had a case-mix ratio value of 1.77 and a mean total weekly cost of $898.41. In contrast, individuals in the comparison group had a mean CMI of 0.87, and an average weekly cost of $337.16. Across the CMI groups too, it appeared that individuals with ADRD, ALS, and MS generally had higher mean costs than the comparison groups. Table 6 shows the mean costs for each condition broken down by CMI.

Table 6: Mean costs (in dollars) by Formal and Informal RUGIII/HC Case-Mix Groups

Formal and Informal RUGIII/HC Case-Mix Groups	ADRD	ALS	MS	Comparison Group
0.485 (PA1)	243.47	292.25	N/A	198.27
0.593 (BA1)	332.83	N/A	N/A	218.29
0.609 (CA1)	271.17	355.67	247.64	253.10
0.839 (IA1)	405.16	N/A	N/A	282.66
0.933 (PA2)	412.52	451.96	N/A	352.49
0.967 (RA1)	440.88	701.97	467.31	386.98
1.126 (CA2)	539.92	529.77	426.52	436.96
1.281 (BA2)	562.22	130.54	N/A	430.11
1.379 (PB)	568.01	752.54	N/A	519.19
1.609 (RA2)	656.53	834.49	722.29	575.61
1.637 (IA2)	647.59	814.73	N/A	569.69
1.660 (CB)	833.27	592.80	592.08	610.98
1.718 (BB)	698.60	834.14	N/A	604.70
1.755 (PC)	665.72	1102.55	N/A	669.93
1.878 (SSA)	869.15	1218.86	719.17	817.19
2.121 (IB)	814.39	1138.55	N/A	792.45
2.417 (PD)	1017.16	1454.55	N/A	866.73
2.498 (SE1)	1070.48	1543.51	1076.54	880.74
2.586 (CC)	1098.48	1336.76	N/A	989.51
2.743 (RB)	1082.30	N/A	989.57	1053.43
2.791 (SSB)	1253.40	1431.36	982.51	1117.96
4.240 (SE2)	899.33	2309.47	1856.09	1356.81
5.151 (SE3)	N/A	2737.15	686.63	2430.60

6.2 Bivariate Regression Models

Bivariate linear regression models identified a number of variables that were significantly ($p \leq 0.05$) and independently associated with cost. For the ADRD group, independent associations were seen for all variables, with the exception of pain, difficulty breathing, dizziness, MD, SCI, and TBI. Huntington's disease was borderline significant at 0.056. Similar results were found in the log-adjusted model; however, tracheostomy and epilepsy became nonsignificant in this model.

For individuals with ALS, variables that were independently associated with cost included CMI, HSI, age, cohabitation with caregiver, ADL, IADL, falls, help with stairs, does not use stairs, trouble swallowing, respirator use, respirator treatment other than respirator, tracheostomy, depression, pain, cognitive performance, constipation, trouble with expression, vision, aggression, bladder and bowel constipation, and ADRD. In the log-transformed model, sex became significant, while malnutrition and vision became nonsignificant.

For MS, variables significantly associated with cost included the following: CMI, HSI, sex, cohabitation with caregiver, ADL, IADL, unsteady gait, falls, help with stair use and does not use stairs, difficulty swallowing, weight loss, malnutrition, respiratory treatment other than respirator, tracheostomy, pain, cognitive performance, constipation, communication, vision, dizziness, bladder and bowel incontinence, social involvement, hallucinations, delusions, ALS, ADRD, CP, SCI, TBI, and CCACs. Variables that were significant were similar in the log-transformed model, but also included age, respirator use, as well as Huntington's disease, and excluded social involvement, delusions, and ALS.

6.3 Multivariate Regression Models

The final untransformed multivariate regression model for ADRD can be found in Table 7. ALS diagnosis and cohabitation with caregiver contributed the greatest increases in cost. Respirator use, hallucinations, falls, and CMI also led to substantial increases in costs for individuals with ADRD. Only age, the does not use stairs, and the MH and SW CCACs were negatively associated with care costs. Results from the sensitivity analyses were displayed alongside the results of the final models for all conditions in order to demonstrate the effect of the repeat assessments within the CAN-STRIVE dataset in this project. A few items became nonsignificant in the sensitivity analysis used to assess the effects of the repeat assessments in the dataset on the predictors of costs that were identified.

Table 7: Untransformed multivariate regression model for ADRD

			Sensitivity Analysis	
	Model R^2=35.18% df=37 F-value=869.43 Model P-value=<.0001 N=59 310		Model R^2=30.34% df=37 F-Value=307.55 Model P-Value=<.0001 N=25 900	
	Parameter Estimate (s.e)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	13.80 (17.45)	0.43	39.51 (26.33)	0.13
Predisposing				
Age	-1.24 (0.19)	<.0001	-1.40 (0.29)	<.0001
Enabling				
MH CCAC	-21.25 (8.19)	0.01	-2.34 (12.31)	0.85
CW CCAC	42.35 (10.68)	<.0001	51.84 (15.16)	0.0006
TC CCAC	60.26 (8.04)	<.0001	44.26 (11.94)	0.0002
CENT CCAC	-2.43 (6.35)	0.70	-7.29 (10.22)	0.48
CE CCAC	17.18 (6.16)	0.01	26.11 (9.83)	0.01
SE CCAC	31.63 (8.47)	0.00	44.89 (12.81)	0.0005
CHAM CCAC	13.41 (6.28)	0.03	14.87 (9.61)	0.12
NSM CCAC	109.70 (9.42)	<.0001	78.67 (13.82)	<.0001
NE CCAC	113.17 (7.96)	<.0001	107.20 (12.76)	<.0001
NW CCAC	33.66 (10.31)	0.00	11.98 (17.71)	0.50
ESC CCAC	84.98 (7.8)	<.0001	71.61 (12.12)	<.0001
SW CCAC	-26.74 (6.82)	<.0001	-23.76 (10.77)	0.03
WW CCAC	2.47 (7.08)	0.73	-2.24 (11.24)	0.84
Cohabit	291.12 (3.75)	<.0001	277.47 (5.75)	<.0001
Need				
Formal and Informal RUGIII/HC Case-Mix	81.33 (5.27)	<.0001	85.91 (8.21)	<.0001
ADL Hierarchy	53.07 (1.74)	<.0001	47.32 (2.72)	<.0001
IADL Capacity	20.18 (1.46)	<.0001	20.49 (2.12)	<.0001
Unsteady Gait	8.10 (3.70)	0.03	15.63 (5.71)	0.01
Falls	8.22 (3.64)	0.02	8.16 (5.52)	0.14
Requires Help with Stairs	42.96 (4.67)	<.0001	40.44 (7.22)	<.0001
Does not use Stairs	-28.08 (4.24)	<.0001	-31.68 (6.52)	<.0001
Difficulty Swallowing	51.66 (3.7)	<.0001	44.84 (6.17)	<.0001
Weight Loss	38.55 (7.16)	<.0001	26.54 (9.79)	0.01
Respirator Use	99.44 (41.84)	0.02	-8.66 (70.79)	0.90
DRS	9.51 (0.91)	<.0001	8.28 (1.37)	<.0001
Pain Scale	9.82 (1.72)	<.0001	6.65 (2.64)	0.01
CPS	44.59 (1.8)	<.0001	42.57 (2.83)	<.0001
Breathing Difficulty	22.15 (4.53)	<.0001	21.74 (7.08)	0.002
Aggression	16.06 (4.83)	0.0005	18.03 (7.78)	0.02

Bowel Incontinence	15.23 (4.37)	0.00	16.22 (6.62)	0.01
Hallucinations	15.17 (4.77)	0.00	12.07 (7.7)	0.12
Delusions	71.74 (7.08)	<.0001	66.76 (10.74)	<.0001
Vision	19.52 (8.49)	0.02	13.05 (12.64)	0.30
Stroke Comorbidity	17.85 (3.85)	<.0001	19.07 (6.16)	0.002
Parkinson's Disease Comorbidity	36.06 (6.56)	<.0001	30.37 (10.86)	0.01
ALS Comorbidity	602.97 (90.48)	<.0001	223.21 (115.55)	0.05

Two untransformed models were identified for ALS. These multivariate regression models can be found in Table 8 and Table 9. Both models had similar levels of explained variance, as well as significant covariates. The only difference between the two models was the significance of IADL capacity in the first ALS model, and the significance of unsteady gait in the second model. Both unsteady gait and IADL capacity became borderline nonsignificant when both variables were kept in the same model with p-value=0.054 and 0.056, respectively.

In both models, many covariates were associated with large increases in cost. Tracheostomy use was the single greatest contributor to cost, although ADRD, bowel incontinence, the NSM and NW CCACs, and cohabitation with caregiver were also associated with substantial increases in cost. In fact, with the exception of age, none of the covariates were negatively and significantly associated with cost. In the sensitivity analysis, respirator use, bowel incontinence, age, unsteady gait, difficulty swallowing, tracheostomy, cognitive performance, trouble breathing, and ADRD comorbidity all became nonsignificant.

Table 8: Untransformed Multivariate Regression Model for ALS

			Sensitivity Analysis	
	Model R^2=55.21% df=27 F-value=42.62 Model P-value=<.0001 N=991		Model R^2=48.95% df=27 F-Value=16.10 Model P-Value=<.0001 N=482	
	Parameter Estimate (s.e.)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	-18.23 (120.56)	0.88	-18.81 (151.24)	0.90
Predisposing				
Age	-3.17 (1.30)	0.01	-2.96 (1.70)	0.08
Enabling				
MH CCAC	66.82 (63.50)	0.30	6.91 (81.88)	0.93
CW CCAC	103.27 (81.25)	0.20	61.20 (105.55)	0.56
TC CCAC	12.49 (84.54)	0.88	8.51 (100.57)	0.93
CENT CCAC	138.63 (56.95)	0.02	50.89 (80.53)	0.53
CE CCAC	-32.69 (68.67)	0.63	-99.80 (86.90)	0.25
SE CCAC	-54.01 (97.76)	0.61	-90.19 (122.87)	0.46
CHAM CCAC	175.82 (59.30)	0.003	119.06 (79.09)	0.13
NSM CCAC	265.55 (85.88)	0.002	108.63 (131.28)	0.41
NE CCAC	347.60 (73.37)	<.0001	266.39 (107.26)	0.01
NW CCAC	93.32 (106.21)	0.38	-2.29 (157.68)	0.99
ESC CCAC	67.68 (85.54)	0.43	50.51 (115.72)	0.66
SW CCAC	-55.78 (66.66)	0.40	-34.74 (91.93)	0.71
WW CCAC	-14.67 (109.31)	0.89	-17.01 (139.40)	0.90
Cohabit	186.95 (44.77)	<.0001	146.56 (60.15)	0.02
Need				
Formal and Informal RUGIII/HC Case-Mix	96.24 (34.34)	0.01	186.18 (47.79)	0.0001
ADL Hierarchy	99.49 (13.26)	<.0001	54.85 (18.04)	0.003
IADL Capacity	32.36 (15.75)	0.04	44.81 (18.36)	0.02
Needs Help with Stairs	121.93 (51.65)	0.02	156.42 (61.83)	0.01
Does not use Stairs	167.45 (45.89)	0.0003	176.96 (56.31)	0.002
Difficulty Swallowing	33.02 (13.11)	0.01	27.25 (19.20)	0.16
Respirator Use	169.21 (77.61)	0.03	201.44 (115.56)	0.08
Tracheostomy Use	644.08 (132.18)	<.0001	277.09 (175.61)	0.12
CPS	74.37 (17.26)	<.0001	11.22 (25.11)	0.66
Breathing Difficulty	75.55 (35.63)	0.03	68.40 (48.14)	0.16
Bowel Incontinence	327.91 (70.82)	<.0001	156.64 (102.21)	0.13
ADRD Comorbidity	308.85 (121.36)	0.02	210.89 (144.62)	0.15

Table 9: Alternate Untransformed Multivariate Regression Model for ALS

			Sensitivity Analysis	
	Model R^2=55.20% df=27 F-value=43.94 Model P-value=<.0001 N=991		Model R^2=48.31% df=27 F-Value=15.72 Model P-Value=<.0001 N=482	
	Parameter Estimate (s.e.)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	22.41 (114.31)	0.20	77.62 (147.16)	0.60
Predisposing				
Age	-3.00 (1.30)	0.02	-2.83 (1.71)	0.10
Enabling				
MH CCAC	66.54 (63.50)	0.29	6.86 (82.37)	0.93
CW CCAC	110.97 (81.24)	0.17	56.66 (106.21)	0.59
TC CCAC	21.53 (84.44)	0.80	8.53 (101.35)	0.93
CENT CCAC	134.46 (57.05)	0.02	51.50 (81.38)	0.53
CE CCAC	-20.83 (68.50)	0.76	-92.96 (87.39)	0.29
SE CCAC	-38.98 (97.60)	0.69	-76.50 (123.44)	0.54
CHAM CCAC	185.26 (59.21)	0.002	129.49 (79.45)	0.10
NSM CCAC	270.78 (85.82)	0.002	103.62 (132.46)	0.43
NE CCAC	342.15 (73.45)	<.0001	265.28 (108.29)	0.01
NW CCAC	105.13 (105.83)	0.32	3.37 (159.13)	0.98
ESC CCAC	86.46 (85.58)	0.31	54.61 (116.62)	0.64
SW CCAC	-56.74 (66.70)	0.39	-33.54 (92.97)	0.72
WW CCAC	-2.68 (109.24)	0.98	-8.03 (140.16)	0.95
Cohabit	194.24 (44.62)	<.0001	157.50 (60.31)	0.01
Need				
Formal and Informal RUGIII/HC Case-Mix	102.32 (34.33)	0.003	195.59 (47.98)	<.0001
ADL Hierarchy	109.32 (12.73)	<.0001	66.92 (17.59)	0.0002
Unsteady Gait	70.51 (35.27)	0.05	37.73 (47.75)	0.43
Needs Help with Stairs	123.46 (51.56)	0.02	177.75 (62.64)	0.01
Does not use Stairs	175.92 (45.07)	0.0001	202.73 (55.98)	0.0003
Difficulty Swallowing	35.31 (13.12)	0.01	32.65 (19.25)	0.08
Respirator Use	159.40 (77.53)	0.04	188.36 (116.18)	0.11
Tracheostomy Use	656.43 (132.36)	<.0001	270.81 (177.39)	0.13
CPS	76.81 (17.17)	<.0001	12.87 (25.28)	0.61
Breathing Difficulty	80.57 (35.45)	0.02	84.43 (47.92)	0.08
Bowel Incontinence	324.92 (70.83)	<.0001	154.98 (102.82)	0.13
ADRD Comorbidity	321.73 (121.43)	0.01	235.96 (145.27)	0.11

The untransformed multivariate regression model for MS found 19 variables significantly associated with cost. No predisposing characteristics were significantly associated with cost. These variables and their corresponding parameter estimates and p-values can be found in Table 10. Tracheostomy use was associated with the greatest increase in cost. HSI, depression, pain, dizziness, and trouble communicating were the only clinical characteristics negatively associated with cost. Negative associations were also found for all eight CCACs that had significant levels of association with cost. Items that became nonsignificant in the sensitivity analysis included pain, unsteady gait, needs help with stairs, weight loss, tracheostomy, and verbal expression.

Table 10: Untransformed Multivariate Regression Model for MS

			Sensitivity Analysis	
	Model R^2=46.76% df=32 F-value=272.07 Model P-value=<.0001 N=9946		Model R^2=44.40% df=32 F-Value=81.72 Model P-Value=<.0001 N=3307	
	Parameter Estimate (s.e.)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	105.87 (30.35)	0.001	133.42 (53.11)	0.01
Enabling				
MH CCAC	-46.22 (18.46)	0.01	-47.58 (29.7)	0.11
CW CCAC	-2.34 (21.36)	0.91	18.78 (33.06)	0.57
TC CCAC	-43.21 (18.57)	0.02	-40.09 (29.22)	0.17
CENT CCAC	-18.85 (14.39)	0.19	-6.21 (25.37)	0.81
CE CCAC	-13.67 (12.24)	0.26	-8.85 (22.81)	0.70
SE CCAC	58.48 (19.24)	0.002	80.48 (33.74)	0.02
CHAM CCAC	-5.27 (13.65)	0.70	-35.46 (22.78)	0.12
NSM CCAC	-17.98 (20.45)	0.38	-29.75 (32.62)	0.36
NE CCAC	30.31 (15.79)	0.05	19.12 (28.39)	0.50
NW CCAC	-61.99 (18.57)	0.001	-69.75 (36.39)	0.06
ESC CCAC	40.33 (15.54)	0.01	71.42 (27.11)	0.01
SW CCAC	-78.32 (12.5)	<.0001	-75.03 (23.25)	0.001
WW CCAC	-10.58 (15.88)	0.51	-25.52 (27.01)	0.34
Cohabit	247.97 (7.83)	<.0001	231.64 (13.72)	<.0001
Need				
Formal and Informal RUGIII/HC Case-Mix	119.21 (9.08)	<.0001	98 (16.41)	<.0001
HSI	-254.39 (29.13)	<.0001	-233.87 (50.16)	<.0001
ADL Hierarchy	47.7 (3.45)	<.0001	56.01 (6.15)	<.0001
IADL Capacity	25.32 (3.34)	<.0001	24.09 (5.42)	<.0001
Unsteady Gait	21.18 (8.03)	0.01	12.71 (14.28)	0.37
Needs Help with Stairs	50.4 (13.55)	0.0002	25.5 (21.33)	0.23
Does not use Stairs	55.2 (10.65)	<.0001	43.72 (17.35)	0.01
Difficulty Swallow	56.03 (5.76)	<.0001	55.22 (10.85)	<.0001
Weight Loss	97.56 (18.92)	<.0001	40.79 (28.52)	0.15
Respiratory Treatment other than Respirator	61.64 (18.58)	0.001	90.4 (36.29)	0.01
Tracheostomy Use	695.41 (75.87)	<.0001	171.17 (143.77)	0.23
DRS	-5.36 (1.99)	0.01	-9.82 (3.3)	0.003
Pain Scale	-21.22 (4.12)	<.0001	-11.5 (7.12)	0.11
CPS	17.7 (4.46)	<.0001	19.65 (7.99)	0.01
Difficulty with Expression	-20.09 (7.21)	0.01	-6.63 (13.28)	0.62
Dizziness	-27.87 (10.21)	0.01	-33.84 (16.96)	0.05

Bowel Incontinence	40.01 (9.67)	<.0001	57.37 (17.75)	0.001
Delusions	166.73 (76.7)	0.03	269.14 (94.12)	0.004

The log-transformed model for ADRD produced similar results to the untransformed model. However, fewer significant covariates were found in this model. Respirator use, vision problems, bowel incontinence, and delusions became nonsignificant. ALS was also nonsignificant in this model, but epilepsy became significantly associated with cost for those with ADRD. With the absence of ALS in this model, cohabitation with caregiver became the single largest contributor to cost. The presence of hallucinations, weight loss, as well as reductions in ADL functionality also contributed greatly to cost. In the sensitivity analysis of this model, only aggression and epilepsy became nonsignificant. The results of this model and its sensitivity analysis can be found in Table 11.

Table 11: Log-transformed multivariate regression model for ADRD

			Sensitivity Analysis	
	Model R^2=44.48% df=33 F-value=1461.77 Model P-value=<.0001 N=59 310		Model R^2=41.16% df=33 F-Value=548.20 Model P-Value=<.0001 N=25 900	
	Parameter Estimate (s.e.)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	2.078 (0.012)	<.0001	2.119 (0.018)	<.0001
Predisposing				
Age	-0.001 (0)	0.0001	-0.001 (0)	<.0001
Enabling				
MH CCAC	0.003 (0.006)	0.59	0.018 (0.009)	0.03
CW CCAC	0.022 (0.007)	0.0026	0.036 (0.010)	0.0006
TC CCAC	0.044 (0.006)	<.0001	0.034 (0.008)	<.0001
CENT CCAC	0.004 (0.004)	0.33	0.003 (0.007)	0.72
CE CCAC	0.021 (0.004)	<.0001	0.029 (0.007)	<.0001
SE CCAC	0.008 (0.006)	0.17	0.023 (0.009)	0.01
CHAM CCAC	0.004 (0.004)	0.34	0.004 (0.007)	0.60
NSM CCAC	0.068 (0.007)	<.0001	0.061 (0.010)	<.0001
NE CCAC	0.065 (0.006)	<.0001	0.068 (0.009)	<.0001
NW CCAC	0.043 (0.007)	<.0001	0.025 (0.012)	0.04
ESC CCAC	0.043 (0.005)	<.0001	0.038 (0.008)	<.0001
SW CCAC	-0.021 (0.005)	<.0001	-0.021 (0.007)	0.0043
WW CCAC	0.004 (0.005)	0.36	-0.002 (0.008)	0.84
Cohabit	0.319 (0.003)	<.0001	0.309 (0.004)	<.0001
Need				
Formal and Informal RUGIII/HC Case-Mix	0.077 (0.004)	<.0001	0.080 (0.006)	<.0001
ADL Hierarchy	0.032 (0.001)	<.0001	0.028 (0.002)	<.0001
IADL Capacity	0.024 (0.001)	<.0001	0.023 (0.001)	<.0001
Unsteady Gait	0.018 (0.003)	<.0001	0.023 (0.004)	<.0001
Falls	0.011 (0.003)	<.0001	0.010 (0.004)	0.01
Needs Help with Stairs	0.030 (0.003)	<.0001	0.027 (0.005)	<.0001
Does not use Stairs	-0.055 (0.003)	<.0001	-0.054 (0.005)	<.0001
Difficulty Swallowing	0.021 (0.003)	<.0001	0.018 (0.004)	<.0001
Weight Loss	0.036 (0.005)	<.0001	0.036 (0.007)	<.0001
DRS	0.007 (0.001)	<.0001	0.007 (0.001)	<.0001
Pain Scale	0.012 (0.001)	<.0001	0.009 (0.002)	<.0001
CPS	0.025 (0.001)	<.0001	0.022 (0.002)	<.0001
Breathing Difficulty	0.016 (0.003)	<.0001	0.015 (0.005)	0.0016
Aggression	0.011 (0.003)	0.0003	0.006 (0.005)	0.18
Hallucinations	0.043 (0.005)	<.0001	0.037 (0.007)	<.0001

Stroke Comorbidity	0.020 (0.003)	<.0001	0.020 (0.004)	<.0001
Parkinson's Disease Comorbidity	0.026 (0.005)	<.0001	0.026 (0.008)	0.0006
Epilepsy Comorbidity	-0.026 (0.010)	0.01	-0.029 (0.017)	0.09

Two potential final models were identified when cost was log-transformed for ALS. Both models had very similar levels of explained variance. The results from the first of these models can be found in Table 12. As compared with the untransformed model, a number of covariates became nonsignificant in this model including ADRD, difficulty breathing, cognitive performance, trouble with stairs, difficulty swallowing, respirator use, and age. Central CCAC also became nonsignificant in this model. Hallucinations and HSI became significant in the log-transformed model. Hallucinations were associated with a substantial decrease in cost, as was HSI; however, it is important to note that a one-unit increase in HSI represents a change from death to perfect health. Cohabitation with a caregiver was associated with the greatest increase in cost. All CCACs significant in the model were positively associated with cost. A number of variables became nonsignificant in the sensitivity analysis. These variables included respirator use and bowel incontinence. Hallucinations also became borderline significant.

The alternative log-transformed model was relatively similar. Bowel incontinence and hallucinations became non-significant in the alternative model, and instead, cognitive performance and difficulty with stairs, and needs help with stairs became significant. A sensitivity analysis was also performed for this model. Respirator use and cognitive performance became nonsignificant in this model. The results of this alternate mode can be found in Table 13.

Table 12: Log-transformed Multivariate Regression Model for ALS

			Sensitivity Analysis	
	Model R^2=59.06% df=22 F-value=63.48 Model P-value=<.0001 N=991		Model R^2=52.39% df=22 F-Value=22.95 Model P-Value=<.0001 N=482	
	Parameter Estimate (s.e.)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	2.190 (0.052)	<0.0001	2.178 (0.075)	<0.0001
Enabling				
MH CCAC	0.014 (0.030)	0.64	-0.035 (0.044)	0.42
CW CCAC	0.061 (0.038)	0.11	0.009 (0.057)	0.88
TC CCAC	0.001 (0.040)	0.97	-0.024 (0.054)	0.66
CENT CCAC	0.043 (0.027)	0.11	0.001 (0.043)	0.98
CE CCAC	-0.028 (0.032)	0.38	-0.042 (0.047)	0.37
SE CCAC	-0.025 (0.046)	0.58	-0.061 (0.065)	0.35
CHAM CCAC	0.085 (0.028)	0.00	0.045 (0.043)	0.30
NSM CCAC	0.121 (0.040)	0.00	0.028 (0.070)	0.69
NE CCAC	0.150 (0.035)	<.0001	0.120 (0.058)	0.04
NW CCAC	-0.046 (0.050)	0.35	-0.105 (0.085)	0.22
ESC CCAC	0.050 (0.040)	0.22	0.039 (0.062)	0.53
SW CCAC	-0.017 (0.031)	0.58	-0.023 (0.049)	0.65
WW CCAC	0.060 (0.052)	0.25	0.036 (0.075)	0.63
Cohabit	0.160 (0.021)	<0.0001	0.160 (0.032)	<0.0001
Need				
Formal and Informal RUGIII/HC Case-Mix	0.089 (0.015)	<.0001	0.120 (0.024)	<0.0001
ADL Hierarchy	0.045 (0.006)	<.0001	0.031 (0.009)	0.001
IADL Capacity	0.047 (0.007)	<.0001	0.048 (0.010)	<.0001
Unsteady Gait	0.049 (0.017)	0.01	0.056 (0.025)	0.03
Respirator Use	0.087 (0.032)	0.003	0.063 (0.055)	0.25
HSI	-0.161 (0.43)	0.01	-0.127 (0.063)	0.04
Bowel Incontinence	0.081 (0.032)	0.04	0.000 (0.053)	1.00
Hallucinations	-0.330 (0.160)	0.04	-0.323 (0.170)	0.06

Table 13: Alternate Log-transformed Multivariate Regression Model for ALS

			Sensitivity Analysis	
	Model R^2=59.13% df=23 F-value=60.82 Model P-value=<.0001 N=991		Model R^2=52.85% df=23 F-Value=22.32 Model P-Value=<.0001 N=482	
	Parameter Estimate (s.e.)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	2.131 (0.054)	<.0001	2.111 (0.08)	<.0001
Enabling				
MH CCAC	0.022 (0.030)	0.46	-0.029 (0.044)	0.51
CW CCAC	0.072 (0.038)	0.06	0.016 (0.056)	0.78
TC CCAC	0.006 (0.040)	0.88	-0.019 (0.054)	0.72
CENT CCAC	0.048 (0.027)	0.07	0.003 (0.043)	0.94
CE CCAC	-0.021 (0.032)	0.52	-0.035 (0.046)	0.45
SE CCAC	-0.023 (0.046)	0.61	-0.059 (0.065)	0.36
CHAM CCAC	0.090 (0.028)	0.00	0.052 (0.042)	0.22
NSM CCAC	0.129 (0.040)	0.00	0.045 (0.070)	0.52
NE CCAC	0.161 (0.035)	<.0001	0.134 (0.057)	0.02
NW CCAC	-0.030 (0.050)	0.54	-0.074 (0.085)	0.39
ESC CCAC	0.056 (0.040)	0.17	0.048 (0.062)	0.44
SW CCAC	-0.013 (0.031)	0.69	-0.011 (0.049)	0.82
WW CCAC	0.054 (0.052)	0.29	0.038 (0.075)	0.61
Cohabit	0.163 (0.021)	<.0001	0.166 (0.032)	<.0001
Need				
Formal and Informal RUGIII/HC Case-Mix	0.087 (0.015)	<.0001	0.119 (0.024)	<.0001
ADL Hierarchy	0.045 (0.006)	<.0001	0.028 (0.009)	0.003
IADL Capacity	0.042 (0.007)	<.0001	0.043 (0.010)	<.0001
Unsteady Gait	0.047 (0.017)	0.01	0.045 (0.026)	0.08
Respirator	0.085 (0.033)	0.01	0.061 (0.055)	0.27
CPS	0.020 (0.008)	0.01	0.006 (0.013)	0.62
Needs Help with Stairs	0.057 (0.025)	0.02	0.090 (0.034)	0.01
Does not use Stairs	0.053 (0.022)	0.02	0.074 (0.031)	0.02
HSI	-0.103 (0.047)	0.03	-0.068 (0.069)	0.32

The log-transformed model for MS can be found in Table 14. Very similar covariates were found in this model as compared to the untransformed model, with the exception of other respiratory treatments, tracheostomy use, and delusions, which became nonsignificant, and bowel incontinence and breathing difficulties became significant in the log-transformed model. Cohabitation with caregiver was associated with the greatest increase in cost. Increasing CMI and decreasing HSI were also associated with substantial increases in cost. Many of the CCACs became nonsignificant in the log-transformed model, including MH, CW, TC, Central, CE, and Champlain CCACs. The SW, and NE CCACs became significant in this model. In the sensitivity analysis for this model, a number of variables became nonsignificant, including unsteady gait, falls, pain, and trouble breathing.

Table 14: Log-transformed Multivariate Regression Model for MS

			Sensitivity Analysis	
	Model R^2=52.92% df=30 F-value=371.45 Model P-value=<.0001 N=9946		Model R^2=49.47% df=30 F-Value=106.91 Model P-Value=<.0001 N=3307	
	Parameter Estimate (s.e)	**P-Value**	**Parameter Estimate (s.e.)**	**P-Value**
Intercept	2.079 (0.023)	<.0001	2.109 (0.041)	<.0001
Enabling				
MH CCAC	-0.021 (0.014)	0.13	-0.01 (0.023)	0.67
CW CCAC	0.015 (0.016)	0.34	0.045 (0.026)	0.08
TC CCAC	-0.022 (0.014)	0.12	-0.022 (0.023)	0.34
CENT CCAC	-0.007 (0.011)	0.54	-0.001 (0.02)	0.95
CE CCAC	-0.004 (0.009)	0.69	0.011 (0.018)	0.53
SE CCAC	0.057 (0.015)	<.0001	0.059 (0.026)	0.02
CHAM CCAC	-0.003 (0.01)	0.77	-0.016 (0.018)	0.36
NSM CCAC	-0.011 (0.015)	0.47	-0.021 (0.025)	0.41
NE CCAC	0.039 (0.012)	0.001	0.024 (0.022)	0.28
NW CCAC	-0.066 (0.014)	<.0001	-0.086 (0.028)	0.002
ESC CCAC	0.003 (0.012)	0.77	0.039 (0.021)	0.06
SW CCAC	-0.049 (0.009)	<.0001	-0.039 (0.018)	0.03
WW CCAC	-0.015 (0.012)	0.21	-0.003 (0.021)	0.89
Cohabit	0.286 (0.006)	<.0001	0.277 (0.011)	<.0001
Need				
Formal and Informal RUGIII/HC Case-Mix	0.107 (0.007)	<.0001	0.088 (0.013)	<.0001
HSI	-0.218 (0.022)	<.0001	-0.214 (0.038)	<.0001
ADL Hierarchy	0.029 (0.003)	<.0001	0.034 (0.005)	<.0001
IADL Capacity	0.039 (0.003)	<.0001	0.04 (0.004)	<.0001
Unsteady Gait	0.017 (0.006)	0.01	0.015 (0.011)	0.17
Falls	0.016 (0.006)	0.005	0.016 (0.01)	0.12
Needs Help with Stairs	0.099 (0.01)	<.0001	0.066 (0.017)	<.0001
Does not use Stairs	0.078 (0.008)	<.0001	0.051 (0.013)	0.0001
Difficulty Swallowing	0.013 (0.004)	0.002	0.02 (0.008)	0.01
Weight Loss	0.055 (0.014)	0.0001	0.043 (0.022)	0.05
DRS	-0.003 (0.002)	0.03	-0.005 (0.003)	0.04
Pain Scale	-0.009 (0.003)	0.003	-0.006 (0.005)	0.27
Breathing Difficulty	-0.023 (0.009)	0.01	-0.028 (0.015)	0.07
Difficulty with Expression	-0.019 (0.004)	<.0001	-0.023 (0.008)	0.01
Bladder Incontinence	0.022 (0.005)	<.0001	0.021 (0.01)	0.03
Bowel Incontinence	0.027 (0.007)	0.0003	0.037 (0.014)	0.01

A summary table for all models showing the effects of the independent variables on cost can be found in Table 15.

Table 15: Summary Table of the Effects of Independent Variables

	Untransformed				Log-Transformed			
	ADRD	ALS 1	ALS 2	MS	ADRD	ALS 1	ALS 2	MS
Predisposing								
Age	---	--	-	NS	---	NS	NS	NS
Sex	NS	NS	NS	NS	NS	NS	NS	NS
Enabling								
MH CCAC	--	NS	NS	-	NS	NS	NS	NS
CW CCAC	+++	NS	NS	NS	++	NS	NS	NS
TC CCAC	+++	NS	NS	-	+++	NS	NS	NS
CENT CCAC	NS	+	+	NS	NS	NS	NS	NS
CE CCAC	++	NS	NS	NS	+++	NS	NS	NS
SE CCAC	++	NS	NS	++	NS	NS	NS	+++
CHAM CCAC	+	++	++	NS	NS	++	++	NS
NSM CCAC	+++	++	++	NS	+++	++	++	NS
NE CCAC	+++	+++	+++	NS	+++	+++	+++	++
NW CCAC	++	NS	NS	--	+++	NS	NS	---
ESC CCAC	+++	NS	NS	++	+++	NS	NS	NS
SW CCAC	---	NS	NS	---	---	NS	NS	---
WW CCAC	NS	NS	NS	NS	NS	NS	NS	NS
Cohabit	+++	+++	+++	+++	+++	+++	+++	+++
Need								
Formal and Informal RUGIII/HC Case-Mix	+++	++	++	+++	+++	+++	+++	+++
HSI	NS	NS	NS	---	NS	--	-	---
ADL Hierarchy	+++	+++	+++	+++	+++	+++	+++	+++
IADL Capacity	+++	+		+++	+++	+++	+++	+++
Unsteady Gait	+	NS	+	++	+++	++	++	++
Falls	+	NS		NS	+++	NS		++
Help with Stairs	+++	+	+	++	+++	NS	+	+++
Does not use Stairs	---	++	+++	+++	---	NS	+	+++
Diff. Swallowing	+++	+	+	+++	+++	NS	NS	++
Malnutrition	NS	NS	NS	NS		NS	NS	
Weight Loss	+++	NS	NS	+++	+++	NS	NS	+++
DRS	+++	NS	NS	--	+++	NS	NS	-
Pain Scale	+++	NS	NS	---	+++	NS	NS	--
CPS	+++	+++	+++	+++	+++	NS	++	NS
Respirator Use	+	+	+	NS	NS	++	++	NS

Respiratory Treatment Other than Respirator Use	NS	NS	NS	++	NS	NS	NS	NS
Tracheostomy Use	NS	+++	+	+++	NS	NS	NS	NS
Breathing Diff.	+++	+	+	NS	+++	NS	NS	--
Bladder Incontinence	NS	NS	NS	NS	NS	NS	NS	+++
Bowel Incontinence	++	+++	+++	+++	NS	+	NS	++
Social Withdrawal	NS	NS	NS	NS	NS	NS	NS	NS
Diff. with Expression	NS	NS	NS	--	NS	NS	NS	---
Aggression	++	NS	NS	NS	++	NS	NS	NS
Hallucination	+++	NS	NS	NS	+++	-	NS	NS
Delusions	+	NS	NS	+	NS	NS	NS	NS
Dizziness	NS	NS	NS	--	NS	NS	NS	NS
Vision	++	NS	NS	NS	NS	NS	NS	NS
ADRD	N/A	+	+	NS	N/A	NS	NS	NS
ALS	+++	N/A	N/A	NS	NS	N/A	N/A	NS
CP	NS	NS	NS	NS	NS	NS	NS	NS
MD	NS	NS	NS	NS	NS	NS	NS	NS
SCI	NS	NS	NS	NS	NS	NS	NS	NS
TBI	NS	NS	NS	NS	NS	NS	NS	NS
MS	NS	NS	NS	N/A	NS	NS	NS	N/A
Huntington's Disease	NS	NS	NS	NS	NS	NS	NS	NS
Stroke	+++	NS	NS	NS	+++	NS	NS	NS
Parkinson's Disease	+++	NS	NS	NS	+++	NS	NS	NS
Epilepsy	NS	NS	NS	NS	--	NS	NS	NS

+++ positive association with p-value ≤.0001
++ positive association with p-value ≤.01
+ positive association with p-value ≤.05
--- negative association with p-value ≤.0001
-- negative association with p-value ≤.01
- negative association with p-value ≤.05
NS not significant at 0.05 level
N/A not applicable to the model

Table 16 shows the explained variances for each of the multivariate regression models. In addition, the explained variance for the RUGIII/HC case-mix, predisposing variables, enabling variables, and all other need variables can be found for each of the three neurological conditions included in this project. Overall, ALS had the highest explained variances, regardless of variables, or whether the dependent variable had been log-transformed. The models looking only at the effect of the predisposing variables on cost had the lowest explained variance scores, while the final models identified in this project had the highest of explained variances. The models with CMI had high explained variance, independent of other variables; however, the enabling and other need variables tested within the models also contributed to the explained variance in the final model. In fact, these other variables, independent of CMI and CCAC variables, had explained variances that were very close to those found in the final models. Finally, the log-transformed models consistently showed higher levels of explained variance.

Table 17 shows the incremental effect of the addition of the other variables on the explained variance. Even with the presence of the RUGIII/HC case-mix variable, the other need variables and the enabling variables resulted in substantial increases in the levels of explained variances across all conditions and models.

Table 16: Explained Variances (R^2) by Model and Condition

	ADRD	ALS 1	ALS 2	MS
Untransformed Models				
Final Model	35.2%	55.2%	55.2%	46.8%
Predisposing Variables	0.0%	0.0%	0.0%	0.0%
Enabling Variables	17.9%	14.1%	14.1%	16.2%
Need Variables				
Formal and Informal RUGIII/HC Case-Mix	20.8%	39.2%	39.2%	33.4%
All other Need Variables	27.1%	50.9%	50.9%	39.0%
Log-Transformed Models				
Final Model	44.9%	59.1%	59.1%	52.9%
Predisposing Variables	0.0%	0.0%	0.0%	0.0%
Enabling Variables	30.5%	20.0%	20.0%	25.9%
Need Variables				
Formal and Informal RUGIII/HC Case-Mix	21.0%	43.1%	43.1%	33.7%
All other Need Variables	29.2%	51.8%	51.7%	38.6%

Table 17: Incremental Effect of Independent Variables on Explained Variances by Model and Condition

	ADRD	ALS 1	ALS 2	MS
Untransformed Models				
Formal and Informal RUGIII/HC Case-Mix	20.8%	39.2%	39.2%	33.4%
All other Need Variables	+6.8%	+12.1%	+12.1%	+6.8%
Predisposing Variables	+0.3%	+0.4%	+0.3%	0.0%
Enabling Variables	+7.2%	+3.3%	+3.3%	+6.1%
Log-Transformed Models				
Formal and Informal RUGIII/HC Case-Mix	20.7%	43.2%	43.1%	33.7%
All other Need Variables	+9.2%	+11.1%	+11.0%	+7.7%
Predisposing Variables	+0.4%	0.0%	0.0%	0.0%
Enabling Variables	+14.4%	+4.9%	+5.1%	+11.8%

7.0 DISCUSSION

Overall, the results from this project demonstrate substantial costs associated with ADRD, ALS, and MS. While previous studies were only able to assess the relationship between cost and the three conditions using broad measures of severity, this project was able to identify a number of specific clinical characteristics that affect severity and clinical complexity for individuals with ADRD, ALS, and MS. In addition, this project is the first Canadian study to identify the HC service costs for individuals with ADRD, ALS, and MS.

7.1 Descriptive Characteristics

Distribution of Populations Across Characteristics

The distributions of HC service users with each of the three conditions across the predisposing, enabling, and need factors support previous findings that persons with ADRD, ALS, and MS are more clinically complex than individuals without these conditions across the clinical items included in this study. The distributions across age groups for each of the conditions appeared to be reflective of the typical ages of onset and duration for each of the conditions, as are the sex distributions. It was interesting to see that a slightly greater proportion of HC users with ALS were female, since males typically have a higher incidence of disease (Kiernan et al., 2011); however, the difference in proportions was not large. The average age for the ALS population found here was also slightly older than expected, since the average age of onset for ALS is 56, and progression occurs rapidly (Kinsley and Siddique, 2012). It is likely that both these observations can be explained by the higher propensity of women and older individuals to use HC services.

It was also interesting to see that while the distribution of individuals across the CCACs were similar for ADRD, MS, and the non-neurological comparison group, the population of HC users with ALS seemed to be distributed differently across the 14 CCACs. The larger proportion of individuals in the more urbanized MH, CW, Central, and Champlain CCACs as compared to the general CCAC population may reflect the greater availability of resources for individuals with ALS in these regions of Ontario. These resources include specialized ALS clinics, neurologists, and ongoing ALS clinical trials (ALS Society of Ontario, n.d). Although specialized clinics are also available for those with ADRD and MS, these clinics can be found in a greater number of regions across Ontario (Alzheimer Society of Ontario, n.d.; MS Society of Canada, n.d).

Finally, it was unsurprising to see the high proportion of individuals with ADRD, ALS, and MS residing with their caregiver(s). In particular, the clinical complexity and high levels of functional limitations observed in these individuals would necessitate the availability of a cohabiting caregiver in order for these individuals to remain in the community (Aronson et al., 1996). Despite the clinical complexity associated with these conditions, however, some individuals were able to remain in their homes without either a primary or secondary caregiver. One explanation is that these individuals were still at the early stages of their conditions and did not yet require the help of a live-in caregiver. The absence of a cohabiting primary or secondary caregiver does not preclude the regular presence of an informal caregiver, but the volume of support available may be diminished.

The proportions of individuals with ADRD, ALS, and MS affected by the characteristics tested in this study had not previously been reported. The findings here did seem to correspond with the clinical progression and characteristics for each of the three conditions described in the

literature previously (Gustavsson et al., 2011; McCrone et al., 2008; Grima et al., 2000; Kang et al., 2006; López-Bastida et al., 2009; Reese et al., 2011; Leicht et al., 2011; Jönsson et al., 2006; Rojas et al., 2011). At least across the clinical characteristics considered in this project, these conditions are likely more complex than non-neurological conditions. The higher concentrations of individuals with ADRD, ALS, and MS in higher ADL and IADL levels suggest substantial disability for these individuals. This finding corresponds with existing literature on the neurological conditions, which has identified neurological conditions as one of the leading causes of disability in Canada and worldwide (CIHI, 2007; WHO, 2006).

For those with ADRD, the concentration of individuals in the two to three ranges on the CPS is indicative of early-stage ADRD. Although the proportion of individuals with aggression, delusions, hallucinations, bladder incontinence, and bowel incontinence were invariably the highest for those with ADRD across the three neurological conditions and the comparison group, the presence of these problem characteristics was low, relative to the proportion of individuals with these characteristics across all individuals with ADRD (Bassiony et al., 2000; Skelly and Flint, 1995; Lyketsos et al., 2002). Since these characteristics are generally associated with moderate to severe stages of dementia (Tyas and Gutmanis, 2008; Gauthier, 2002), the lower rates of aggression, delusions, hallucinations and incontinence in this HC population are likely due to the higher proportion of individuals with early-stage dementia in the HC population. This finding resembles the care setting patterns for those with ADRD found in prior studies (Yaffe et al., 2002; Tyas and Gutmanis, 2008).

For those with ALS, a greater proportion of individuals reported having problem characteristics than those with ADRD or MS. Further, those with ALS also reported greater severity for many of these characteristics. Unlike those with ADRD, the ALS population in this

study displayed similar rates of problem characteristics including cognitive performance to the overall ALS population (Ringholz et al., 2005). While this observation is reflective of the clinical complexity and severity associated with ALS, it is also likely reflective of the pattern of care corresponding to the progression for each of the three conditions. Individuals with ADRD and MS may be more likely to be admitted to nursing home (Stolp-Smith et al., 1998; Aronson et al., 1996; Krivickas et al., 1997; Tyas and Gutmanis, 2008), while comparatively few individuals with ALS end up in nursing home settings (Kehyayan et al., submitted). Beyond the scope of these diagnostic groups, the clinical variation across the ADRD, ALS, and MS HC populations demonstrate that even though persons with these conditions are often defined by their diagnoses, clinical characteristics are much more descriptive of individuals with these conditions.

Mean Costs and Resource Use

No prior estimates of HC costs for individuals with ADRD, ALS, and MS in HC could be found. Therefore, comparisons to prior estimates could not be made. In studies looking beyond the scope of HC care costs, annual costs appeared to be greatest for MS, followed by ADRD, then ALS (López-Bastida et al., 2009; Andersen et al., 2011; McCrone et al., 2008). The discrepancy between the findings of this project and prior findings can likely be explained by the inclusion of indirect costs in prior studies. In particular, MS is known to have substantial effects on productivity loss due to its young age of onset (Grima et al., 2008). Further, very few studies have focused on costs related to ALS. As a result, it is difficult to gauge the reliability of the cost estimates provided by López-Bastida et al. (2009). It is also important to note that comparisons of cost across the three neurological conditions cannot accurately be made due to the differing methodologies used in prior studies. Interestingly, although the mean costs of care and mean CMI values were similar in both the initial analyses and sensitivity analyses for ADRD and MS,

the mean cost of care and mean CMI value were substantially lower for ALS in the sensitivity analysis. This finding suggests that repeat assessments were likely to have led to an oversampling of individuals with higher resource use. Overall, the mean cost trends identified in this project are likely to be reflective of HC care costs, but these trends are unlikely to be representative of total cost of illness patterns. It is clear from the mean dollar and CMI values, however, that HC costs and service utilization are higher for individuals with ADRD, ALS, and MS than for those without any neurological conditions. Further, it is evident that the levels of resource utilization for individuals with each of the three conditions are different.

7.2 Regression Models

The multivariate regression models in this project identified a number of variables associated with cost of care in HC beyond the characteristics identified in previous studies. Predictors of costs identified in both models were assumed to be valid even though the normality assumptions had been violated in the untransformed models. The multivariate models were expected to be quite robust so that these violations were not expected to seriously affect the validity of the findings in those models. In addition, the higher number of covariates and the lower explained variances in the untransformed models as compared to the log-transformed models suggests that the normality violations would merely cause a weakening of effects. Therefore, unless unsupported by the literature review, the findings in both the untransformed and transformed models were thought to be valid. Detailed discussions of findings for each of the different conditions follow.

Predisposing Factors

Alzheimer's Disease and Related Dementias

Age was the only predisposing characteristic that was significantly associated with cost. Interestingly, age was predictive of lower HC costs. The significance and direction of this association was surprising since age of onset is not known to affect the severity of ADRD, which typically drives costs (Huff et al., 1987). One possible explanation for this finding may be the greater proportion of individuals with frontotemporal lobe dementia in younger populations (Jeffries and Agrawal, 2009). Unlike Alzheimer's disease, which is the most common form of dementia in elderly populations, early symptoms of frontotemporal lobe dementia are associated with greater behavioral changes and speech impairments (Jeffries and Agrawal, 2009). Although speech impairments are not known to increase care costs (Fries et al., 2001), behavioral difficulties can be associated with increased costs. It is possible that the progression of frontotemporal lobe dementia may result in greater costs during the early stages of the condition. Given that the present results show decreased resource use with age after controlling for need and enabling factors, an alternative explanation may be ageism within the HC setting, whereby older individuals are systematically provided with fewer resources. Although ageism in HC is also possible, the ability to conclusively determine ageism is beyond the scope of this project. A final possibility may be that more caregiver support is more accessible for younger persons with ADRD in HC, and so younger individuals with higher levels of severity are able to remain within their homes for longer periods of time. Regardless of the explanation, it is clear that more attention should be paid to ensuring that the needs of older individuals with ADRD in HC are met.

Amyotrophic Lateral Sclerosis

Only age was significantly associated with lower costs for individuals with ALS in HC, and was predictive of lower care costs. Age had previously been identified as significant predictor of costs in previous studies. Although age of onset is not known to affect the severity or progression of ALS, it is possible that younger individuals with ALS may be associated with greater costs due to the increased likelihood of frontal temporal dementia at earlier ages (Kiernan et al., 2011). It may also be possible that the significance of age is reflective of ageism in the distribution of HC services. Like ADRD, however, the ability to ascertain ageism is beyond the scope of this project. Since age was only significant in the untransformed model, it is possible that the significance of this variable is the result of violations to the normality assumption in multivariate regression models.

Multiple Sclerosis

Neither predisposing factor was significantly associated with cost for individuals with MS. Although previous findings have suggested that age and sex are likely to affect productivity losses associated with MS, the cost measurement in this study looks exclusively at care costs. As a result, the nonsignificance of both variables was expected since age and sex are not known to affect the severity or progression of the condition.

Enabling Factors

Both enabling variables tested in this project were associated with costs across all conditions and models. Like previous studies, cohabitation with a caregiver resulted in substantial increases in care costs (Moore et al., 2001). Cohabitation with a caregiver was likely predictive of increased care costs because of the availability of a caregiver for greater durations of time. In cases where cohabitation with the caregiver was the result of ADRD, ALS, or MS

diagnosis, cohabitation may also indicate greater disease severity, which is associated with increased care costs (Jönsson et al., 2006; Andersen et al., 2003; Rojas et al., 2011; Leicht et al., 2011; Gustavsson et al., 2011; López-Bastida et al., 2009; McCrone et al., 2008; Reese et al., 2011).

The significance of some CCACs in predicting care costs for ADRD, ALS, and MS was also expected. Compared with the HNHB CCAC, the NE CCAC, in particular, was associated with the greatest increase in cost, followed by the NSM CCAC for both ADRD and ALS. Interestingly, both these CCACs encompass northern and primarily rural regions of Ontario, where access to alternative medical services may be difficult. As a result, it is possible that the ADRD and ALS care costs in these regions are higher because of the absence or limited accessibility of other care services in these regions. In the NE CCAC region, in particular, limited nursing homes and the clustering of these homes in the more urban regions of this CCAC may necessitate HC services for those living outside the proximity of these homes (NSM LHIN, 2013; NE LHIN, 2013). In contrast, CCACs were predictive of lower care costs for those with MS. The greatest reduction in care costs relative to the HNHB CCAC was seen in the NW CCAC. Since all CCACs significantly associated with cost were negatively associated, it is possible that the HNHB CCAC had the highest regional costs for MS care. Finally, the variations in costs across at least some of the CCACs are very likely the result of regional variations in care costs and practice patterns that are common across health conditions and jurisdictions.

Illness Factors

Although illness factors were consistently the most important in predicting care costs across ADRD, ALS, and MS, the results of the multivariate models demonstrate that clinical characteristics have a far greater effect on HC costs. In particular, ADL function, IADL function, cognitive performance, unsteady gait, and the RUGIII/HC CMI were most consistent in their ability to strongly predict costs.

Cognitive Performance

Cognitive performance was predictive of increased costs across the conditions. Level of cognitive impairment had been identified as a predictor of costs for ADRD in previous studies (Gustavsson et al., 2011; Jönsson et al., 2006; Rojas et al., 2011). Cognitive impairment in persons with ALS and MS most likely also indicate dementia (Compston and Coles, 2002; Mitchell and Borasio, 2007). The increase in costs for each unit increase in CPS appeared to vary across the conditions; however, CPS was predictive of the highest increase in costs for ALS, and lowest for MS. The differences in costs associated with CPS appeared to reflect the different types of dementia that are common in ADRD and ALS. Since individuals with ALS that develop dementia tend to develop frontotemporal lobe dementia, it was unsurprising that cognitive performance was associated with the greater increases in care costs than those with ADRD. According to Rojas et al. (2011), frontotemporal lobe dementia is costlier than Alzheimer's disease, which is the primary form of dementia afflicting those with an ADRD diagnosis (Geldmacher and Whitehouse, 1996). No specific type of dementia had been associated with dementia found in individuals with MS, but the low rates of dementia for these individuals may explain the small increases in costs associated with CPS.

Functional- and Movement-Related Characteristics

ADL functionality was positively associated with costs across all conditions and models. The association between ADLs and cost had been identified in studies on ADRD (Kang et al., 2006; Reese et al., 2011; Andersen et al., 2001), but had not been established for MS or ALS. Across conditions, however, impaired ADL functions are likely to increase care costs because of the extensive assistance required by individuals to complete these activities. In particular, the daily aspect of these activities, including eating, locomotion, personal hygiene and/or toilet use explains the significance of ADL functionality in predicting HC care costs across conditions. The costs associated with these activities are most likely driven by assistance provided to individuals with these conditions by PSWs, and family members providing informal care.

While a single-unit increase in ADL score resulted in similar increases in care costs for ADRD and MS, the increase in care costs associated with ALS were about two times as high as the increases for ADRD and MS. This finding is likely representative of the much higher proportion of individuals with ALS in the highest ADL levels, as compared to those with ADRD and MS, since the cost of care is likely to increase exponentially across the ADL levels. This finding demonstrates that even a minimal increase in ADL limitation for those with ALS is associated with much greater care costs in HC. Nevertheless, the presence of any ADL impairment for those with any of the three neurological conditions considered in this project represents substantial increases in weekly care costs.

IADL functionality was also positively associated with HC costs across all three conditions and models. The increases in care costs were similar across the three conditions and were consistently smaller than the increases in care costs associated with ADL impairments. As with ADL functionality, IADL impairments require extensive daily assistance. IADL limitations

are also therefore likely to increase care costs because of the large amount of personal support and informal caregiving required to perform these activities for individuals with ADRD, ALS, and MS. Although a single-unit increase in IADL level appeared to result in small increases in costs for all three conditions, the high concentration of these individuals in the highest IADL levels suggests that assistance with IADL tasks contributed enormously to the high costs of these conditions.

Unsteady gait was significantly and positively associated with HC costs for all three conditions. Falls were also significantly and positively associated with costs in some of the ADRD and MS models. Since muscle weakness and rigidity are part of the clinical course for all three conditions, the association between these characteristics and cost was unsurprising. Again, it was interesting to see that both unsteady gait and falls resulted in relatively similar increases in cares costs for ADRD and MS, while unsteady gait resulted in increased care costs that were more than three times higher for ALS. Unlike ADRD and MS, muscle weakness and rigidity affect a large proportion of individuals with ALS during early stages (Kiernan et al., 2011). The rapid progression of ALS also probably means that regardless of onset, limb muscle weakness and rigidity affects most individuals with ALS quite severely over a short period of time (Kiernan et al., 2011). In addition, while individuals with ADRD and MS tend to enter nursing homes when their conditions become complex, individuals with ALS are more likely to remain in their homes. As a result, the higher costs associated with unsteady gait in individuals with ALS are likely a reflection of the severity of limb muscle rigidity and weakness in these individuals, as well as the likelihood for these individuals to remain in HC throughout the progression of the condition. Management of unsteady gait would therefore require much more

extensive assistance by a combination of HC professionals, PSWs, and family members for individuals with ALS in HC.

Finally, needing help with stairs was also associated with increased care costs across all conditions. The costs associated with the need for assistance with stairs was highest for those with ALS, and lowest for those with ADRD. Since stair use is an indication of muscle weakness and rigidity, and these signs are common and progress most rapidly in those with ALS (Kiernan et al., 2011), this finding is likely reflective of the rapid and severe functional decline found in those with ALS. As assistance with stairs is also likely to require care provided by PSWs, informal caregivers, PTs, and OTs, it is unsurprising to find that assistance of stairs is associated with increased care costs across all three conditions. Interestingly, those with MS and ALS that did not use stairs were also associated with increased care costs while individuals with ADRD that did not use stairs were associated with decreased care costs. Unlike individuals with ALS and MS, those with ADRD are likely to develop motor rigidity and coordination problems in much later stages. As a result, it is possible that the decreased costs associated with those with ADRD not using stairs reflect transition of many of these individuals to long-term care facilities, while those with ALS and MS continue to require substantial HC resources.

Breathing, Swallowing, and Expression (Speech)

Shortness of breath was predictive of costs in at least one model across the three conditions. For ADRD and ALS, breathing difficulties were associated with increased care costs, while it was associated with reduced care costs for those MS. Care costs associated with respiratory difficulties were much higher for individuals with ALS, while associated costs for those with ADRD were comparatively low. This finding may be explained by the fact that respiratory difficulties are generally more severe for individuals with ALS. While respiratory

challenges tend to occur during sleep for individuals with ADRD (Pond et al., 1990), it is a continuous symptom for individuals with ALS (Mitchell and Borasio, 2007). In addition, death is caused by pneumonia with related shortness of breath in the majority of individuals with ALS. As a result, substantial anxiety and pain is caused by this characteristic in those with ALS (Kiernan et al., 2011). It is likely that distress over respiratory difficulties results in greater levels of service use for these individuals since they are likelier to seek treatment for this symptom.

In contrast, the lower costs associated with respiratory difficulties for ADRD are likely to reflect the differences in the progression for this condition. Since dyspnea is associated with late progression for ADRD, it is likely that these individuals are transitioned into institutionalized settings before respiratory difficulties become associated with substantial increases in cost. For MS, trouble breathing rarely becomes severe as the disease progresses (Gosselink et al., 1999); as a result, HC services are unlikely to be sought by these individuals for this problem. Further, individuals with MS may choose to do less, and therefore require less PSW or informal caregiver assistance, while they are experiencing respiratory challenges. Nevertheless, the small decrease in costs associated with respiratory difficulties may require further investigation to ensure that adequate care is being provided for those with MS experiencing shortness of breath.

At least one form of respiratory treatment was predictive of increased costs for all three conditions. Non-respirator treatments were associated with the lowest increases in care costs, while tracheostomy was associated with the largest increase in care costs. Respirator use was also predictive of substantial increases in costs for those with ADRD and ALS, but was two times higher for those with ALS. The overall significance of the association between respiratory therapies and HC costs has been identified in previous studies (Sevick et al., 1996). The high costs of these therapies are typically driven by the need for increased monitoring by nurses,

PSWs, as well as informal caregivers within the HC setting (Sevick et al., 1996). In addition, Rowland and Schneider (2001) have found that mechanical ventilators cause significant burden to the families of those with ALS. It is possible that the larger increases in costs associated with respiratory treatments for those with ALS are also reflective of the pattern of institutionalization for individuals with ADRD and MS requiring respiratory therapy (Buchanan et al., 2002). Since individuals with ALS are much likelier to remain in their homes even when permanent respirator use is required, it is unsurprising that respirators are associated with the greatest increase in cares costs for individuals with ALS.

Dysphagia was predictive of increased costs for ADRD, ALS, and MS. Associated weight loss was also significantly associated with increased costs for those with ADRD and MS, although malnutrition was not significant in any of the conditions. Since treatment for dysphagia is addressed by modifications to diet and feeding, increased costs may be caused by the need for dietitians to assist in planning dietary modifications, as well as assistance from PSWs and family caregivers to assist and supervise in feeding. Speech therapy is also used as part of dysphagia therapy (Compston and Coles, 2002), and it is likely that the majority of HC costs are associated with use of speech language pathology services. However, specific service costs were not identified in this project, so the contribution of speech language pathology, dietitian and PSW services to HC costs for individuals with these conditions could not be determined.

Finally, difficulty with expression was predictive of reduced care costs for individuals with MS. As Fries et al. (2001) discussed, the negative association between resource use and verbal expression difficulties may be caused by the under-treatment of symptoms without physical cues. The negative association between pain and cost observed in those with MS

seemed to correspond with the assertion by Fries et al. (2001) that difficulties with communication can result in reduced pain management.

However, it was interesting that verbal expression was not significantly predictive of costs for individuals with ADRD, or ALS. It is possible that ADRD and ALS are associated with fewer symptoms with nonphysical cues and so difficulties with expression were not predictive of costs for these conditions. Detection of nonphysical cues may also be better for individuals with ADRD and ALS. Regardless of the mechanism causing this finding, more attention should be paid to ensure the needs of individuals with MS and difficulties with communication are adequately addressed.

Bladder and Bowel Incontinence

Bowel incontinence was associated with care costs for all three conditions. Although bowel incontinence was predictive of higher costs for all conditions, the costs associated with bowel incontinence were very substantially higher in those with ALS. No previous studies had identified bowel incontinence as a significant predictor of cost for these conditions, but the problem is known to be associated with substantial care costs (Miner, 2004). Though many of the costs for bowel incontinence are related to treatments and therapies that are beyond the cost measurements included in this project, the day-to-day management bowel incontinence is known to result in substantial costs as well (Miner, 2004). The much higher cost of bowel incontinence for those with ALS is likely to reflect the increased likelihood for individuals with ADRD and MS to enter nursing homes since bowel incontinence is known to be one of the primary predictors of nursing home admission from home care across most conditions (Miner, 2004; Tsuji et al., 1995; O'Donnell et al., 1992). For individuals with ADRD and MS with bowel incontinence, it is most likely that they are admitted to nursing home before the symptom

becomes associated with large care costs. Interestingly, incontinence is not a clinical characteristic associated with ALS since involuntary bladder and bowel muscles are not generally affected (Williams and Windebank, 1991). One possible explanation may be that bowel incontinence found in these individuals is the result of concomitant dementia. The concomitant dementia would also explain why bowel incontinence contributes so much more cost in these individuals with ALS, although CPS and ADRD comorbidity are already part of the ALS multivariate regression models.

Bladder incontinence was a significant predictor of increased care costs for MS only. Since bladder incontinence in those with MS is generally caused by a combination of failure to empty the bladder, and failure to store urine, treatment generally requires drug therapy and self-catheterization where necessary (Compston and Coles, 2002). Increased costs surrounding bladder incontinence for these individuals are likely to be caused by increased costs associated with the management of medications, and assistance with catheter use by nurses, PSWs, and family members. Bladder incontinence is also a symptom of ADRD, but was not significantly associated with cost in this project. Since urinary incontinence can sometimes lead to early nursing home admissions (Thom et al., 1997), it may again be possible that individuals with ADRD developing urinary incontinence were transitioned to nursing homes before the problem became demanded substantial increases in costs. Indeed, individuals with ADRD are more likely to transition to nursing home than those with MS.

Visual Symptoms

Vision was only predictive of cost for individuals with ADRD, but did not contribute greatly to the cost of care for these individuals. The significance of vision in predicting ADRD cost was likely caused by cognitive disturbances that are the result of visual misperceptions and

poor vision in these individuals (Geldmacher and Whitehouse, 1996). Since these vision problems are already likely to exacerbate cognitive impairment, which was already present in all models, it was unsurprising that these symptoms led only to a small increase in care costs.

Visual symptoms also affect individuals with MS; however, the vision variable was nonsignificant across MS models. Vision impairments caused by MS typically involve temporary blindness, unilateral optic neuritis, diplopia, or flashes of light (Noseworthy et al., 2000). Since the measurement of vision in the RAI-HC only measures vision status in the previous three days, these temporary visual impairments may be undetected. In addition, since these visual symptoms are generally temporary, it is unlikely that they result in long-term increases in care costs. Finally, HC professionals are unlikely to be able to address the types of vision problems experienced by individuals with MS, so changes in care use would not be significant.

Pain

Pain was significantly associated with cost for ADRD and MS. A one-unit increase on the RAI pain scale was responsible for small increases in costs. Pain was not expected to contribute substantial care costs to the treatment of individuals with ADRD since pain treatment generally consists of drug therapy (Scherder et al., 2009). The small increase in costs could reflect an increased need for help with housework, since pain may limit the performance of some of these tasks. It is possible that the increases in costs associated with pain were small since the ADL and IADL scales already captured the majority of these needs. Finally, increased costs associated with pain for these individuals were likely limited since higher levels of cognitive impairment are known to reduce reports of pain and pain severity, and therefore limit treatment for the pain.

In contrast, pain was predictive of reduced costs in individuals with MS. This finding was alarming since pain in individuals with MS can be treated, but typically requires some combination of physical therapy and pharmacological treatment (Noseworthy et al., 2000). As discussed earlier, it is possible that pain may be unreported because of difficulties with verbal expression, and so treatment and assistance is not provided. Pain may also cause withdrawal resulting in reduced demands for both formal and informal care. Regardless of the mechanism, the pattern of decreased costs and pain was consistent in the bivariate analysis, across the MS models, and in the sensitivity analysis for the untransformed model, suggesting that this finding was unlikely to be the result of a statistical error. This finding is alarming, however, and may reflect an under-treatment of pain symptoms for individuals with MS in HC.

It was surprising to see that pain was not significantly associated with cost for individuals with ALS. Pain in ALS can generally be addressed by a combination of drug and physical therapy for musculoskeletal pain and pain from joint stiffness (Mitchell and Borasio, 2007). Pain caused by skin pressure can also be addressed through care provided by nurses (Mitchell and Borasio, 2007). One explanation is that pain may not be significant because it is incorporated in the HSI index, which was significant in the log-transformed models for those with ALS. Addition of the pain scale could have over-explained the significance of the association between pain and care costs. Nevertheless, pain for individuals with ALS in HC may not be adequately addressed, and further investigation is required in future studies.

Behavioral, Mood, and Mental Symptoms

Depression was significantly predictive of small changes in HC costs for ADRD and MS. ADRD was positively associated with costs, while MS was negatively associated with costs. Depression is part of the clinical course for ADRD and MS (Geldmacher and Whitehouse, 1996;

Sadovnick et al., 1996). Since depression can present through social withdrawal, reductions in care costs for those with MS may not be surprising. Combined with reduced capacity to verbally express needs and fatigue, individuals with depression may make fewer demands on HC professionals, PSWs, and informal caregivers. Since verbal expression was not predictive of care costs for those with ADRD, and fatigue is not generally associated with ADRD, it is possible that these individuals were more likely to continue expressing some care needs. Although depression is common in individuals with ALS following initial diagnosis, full-fledged depression rarely develops (Mitchell and Borasio, 2007). As a result, it was unsurprising that depression was not predictive of care costs in those with ALS.

Delusions were significantly associated with increased care costs for individuals with ADRD and MS. The increase in care costs caused by delusions was almost ten times higher for those with MS than those with ADRD. Delusions can cause individuals to act dangerously or violently, so costs for delusions in those with ADRD and MS are likely driven by supervision provided by family caregivers. Delusions for individuals with ADRD tend to result in interpersonal conflicts related to accusations of theft and marital infidelity, as well as perceived persecution by caregivers (Geldmacher and Whitehouse, 1996; Hwang et al., 1999). Costs for those with ADRD may also be lower since aggression is also significantly predictive of costs in the model for these individuals. Therefore, some of the costs caused by delusions may also be predicted by costs associated with aggressive behaviours. The delusions present in individuals with MS have not been typified; however, psychosis for those with MS is generally associated with late-stage disease, an increased number of lesions, and dementia (Feinstein et al., 1992). As a result, the substantial increase in cost associated with delusions for those with MS is likely driven by the disease severity.

Psychosis through hallucinations was also predictive of care costs for ADRD and ALS. Hallucinations were associated with substantial increases in costs for those with ADRD, while they were associated with substantial decreases in care costs for those with ALS. As with delusions, increased care costs for those with ADRD may be incurred through supervisory needs, as well as coping and comforting of individuals with hallucinations. It was interesting to see that hallucinations contributed far greater costs than delusions for these individuals.

In contrast, hallucinations are not generally part of the clinical profile of individuals with ALS, but can be caused by associated frontotemporal lobe dementia (Kiernan et al., 2011). Since hallucinations for individuals with ALS may begin experiencing these symptoms during the terminal phase, it is possible that the reduced care costs may reflect the reduction of service use during this time (Kiernan et al., 2011). Instead, ALS is generally treated through respiratory therapies, and drug therapies for restlessness and anxiety during the terminal phase (Mitchell and Borasio, 2007). Further, although the terminal phase is associated with multidisciplinary care, the costs of these services and therapies may be beyond the measurements of cost in this project. As a result, it may appear that hallucinations are predictive of significantly lower care costs for individuals with ALS when it in fact reflects disease progression and its corresponding care.

Comorbidities

Neurological comorbidities were only predictive of costs for individuals with ADRD and ALS. For individuals with ALS, ADRD was significantly associated with very substantial increases in care costs. The large increase in care costs was unsurprising since ADRD diagnosis for these individuals causes increasing clinical complexity.

For individuals with ADRD, ALS, stroke, and Parkinson's disease were all associated with increased care costs. This finding corresponded with neurological conditions that typically

have dementia comorbidities. The direction of the association was also expected since clinical complexity is known to increase care costs. ALS comorbidity increased care costs considerably, and corresponded with the high costs associated with ALS identified in this project. Interestingly, epilepsy diagnosis was associated with lower care costs.

Health Status Index

Increases in the HSI were associated with very substantial reductions in HC costs for individuals with ALS and MS. This finding corresponded with previous findings that improved HRQOL was predictive of lower care costs for ALS and MS (López-Bastida et al., 2008; Grima et al., 2000).

RUGIII/HC Case Mix Index

The CMI variable was significantly predictive of increased care costs for all conditions, and had the highest level of explained variance of any individual item tested in this project. It was unsurprising that the CMI variable was significantly predictive of costs for individuals with ADRD since this population was older, and similar to the population in which the RUGIII/HC was initially derived. Although the RUGIII/HC had demonstrated reasonable reliability and validity, this CMI had not been designed specifically for use in populations with neurological conditions. Therefore, it was interesting to see that the RUGIII/HC CMI was also significant in predicting care costs for individuals with ALS and MS. In addition, individuals with ALS and MS were much younger than the average ages of those with ADRD, and in the general HC population. It is probable that the RUGIII/HC CMI was significantly predictive of costs for ALS and MS because of the suitability of many RUGIII/HC algorithm items for individuals with these two conditions. In particular, the inclusion of a number of characteristics that are common to the neurological conditions within the RUGIII/HC likely contributed to the effectiveness of this CMI

on these groups. These characteristics include tracheostomy and respirator use, respiratory therapy, cognitive performance, ADL function, and IADL function. In addition, a number of specific items within the CPS, ADL, and IADL scales were relevant to ADRD, ALS, and MS, and also likely contributed to the effectiveness of the RUGIII/HC for individuals with these conditions. These items included the ability to be understood by others, cognitive skills for daily decision-making, the ability to manage personal finances, and locomotion.

Across all conditions, CMI consistently had the greatest level of explained variance in the bivariate analyses. CMI had less predictive power compared to the explained variances found for the remaining variables within the models, excluding CCACs. This finding suggests that some additions to the RUGIII/HC algorithm may increase the predictive power of this case-mix system, at least for the neurological population. Clinical characteristics identified in the models that were not already included in the RUGIII/HC algorithm included the following: stair use, unsteady gait, falls, difficulty with speech, difficulty with breathing, difficulty with speech, bladder incontinence, bowel incontinence, pain, depression, delusions, ALS, Parkinson's disease, epilepsy and stroke diagnosis, and visual problems. A number of these items were predictive of cost across all three conditions. Addition of even some of these items to the RUGIII/HC algorithm is likely to improve the predictive abilities of the case-mix system for individuals with neurological conditions, although it is expected that it would also improve the performance of this CMI in the general HC population.

7.3 Andersen and Newman Framework

The findings from this project both support and expand on the findings from previous studies identifying predictors of costs for ADRD, ALS, and MS. Grounded in Andersen and Newman's framework for healthcare service utilization, this project found that the enabling and

illness factors were consistently predictive of cost in a HC setting. In particular, a couple of clinical characteristics were significant across all conditions and models, including sensitivity analyses, suggesting that these characteristics are highly predictive of resource utilization for individuals with ADRD, ALS, and MS in the HC setting. These characteristics included CMI, and ADL functionality. Other characteristics were also consistently found across conditions, though not necessarily across all models. These characteristics included cognitive impairment, IADL capacity, unsteady gait, assistance with stairs and no stair use, difficulty swallowing, respiratory challenges, and bowel incontinence. As a result, it is likely that beyond neurological diagnoses, these specific characteristics have high relative importance for predicting resource utilization for individuals with these conditions in HC. Similarly, cohabitation was significantly predictive of higher costs across models for all three conditions; therefore, cohabitation with a caregiver is also very likely to have high relative importance for predicting HC resource use for these individuals. At least one CCAC was also consistently significant across all conditions and models; however, the directions of the associations and the significance of specific CCACs varied across the models. As a result, geographic location is likely to have medium relative importance, contingent on the specific CCAC.

Interestingly, although Andersen and Newman identified predisposing characteristics to have medium relative importance in predicting healthcare utilization, this assertion was not supported by the findings in this project for the HC setting. Only age was significant within the ADRD and ALS models, and became nonsignificant in the log-transformed models for ALS. In addition, the direction of the association for age did not support Andersen and Newman's (1973) argument that older age can lead to increased healthcare seeking behavior. As a result, predisposing variables were of low relative importance in predicting healthcare utilization in this

project. Overall, the high relative importance of the illness factors found in this project, along with the medium relative importance of the enabling factors, and low relative importance of the demographic predisposing factors for individuals with ADRD, ALS, and MS in HC were consistent with the conclusions of a previous study on the Andersen and Newman framework in a HC setting (Kempen and Suurmeijer, 1991).

7.4 Limitations

Although the results from this project expand on previous findings, a few limitations should be noted. The relationship between a number of symptoms related to ADRD, ALS, and MS and cost could not be studied because these symptoms were not available in the RAI-HC tool. These symptoms included fatigue, emotional lability, sexual dysfunction, agitation, and drooling. However, the absence of these symptoms from the analyses in this project is not expected to be of great concern since these symptoms are unlikely to drive HC service costs substantially.

Another limitation in this project had to do with the diagnosis measurement. In particular, the analyses in this project did not differentiate between the various subtypes of ADRD, ALS, and MS. Since the progression and severity for each of these conditions differ based on subtype, differentiation between these subtypes may have identified other predictors of costs. Similarly, no measure of time since onset of disease was available. Although time since onset is associated with severity for ADRD, ALS, and MS, it nevertheless would have been interesting to see the relationship between time since onset, and HC care costs.

Finally, though this analysis provides accurate estimates of costs based on HC service utilization, the measurement of cost in this project was somewhat limited. Though the purpose of this project was to identify predictors of HC service costs, the predictors identified in this project

may not be reflective of the predictors of combined direct and indirect costs. Further, even though this project identified the majority of direct costs including formal and informal care, this project did not differentiate between the care types, or the formal service types. Within the formal costs as well, the measure of cost in this project excluded drug costs, which are likely to be particularly substantial for ALS and MS. Case manager time was not included in these analyses. Since case managers play substantial roles in the formal care provision for individuals in HC, the inclusion of costs associated with case manager time would have provided more reliable estimates of HC care costs for each of the conditions. While it is unlikely that the absence of case manager time would have altered the predictor variables in the model, inclusion of case manager time would have also provided a more accurate portrait of predictors of HC care costs for those with ADRD, ALS, and/or MS. Assuming that case manager time is proportionate to HC service utilization, the absence of case manager costs is unlikely to be of concern.

7.5 Strengths

The primary strength of this project was the data from the CAN-STRIVE HC study. As detailed earlier, these data include RAI-HC assessments for the entire Ontario HC population between 2005 and 2008. Clinical data are expected to be accurate since trained healthcare professionals perform RAI-HC assessments. In addition, incentives were put in place to ensure that data collected in the RAI-HC reflected the actual needs of individuals in HC. Accurate cost data were also available from this dataset. Resource utilization data were obtained from administrative records, and mean wage rates from across the province for each these services were applied.

Similarly, the use of the case-mix approach to estimating cost was also an important strength in this project. The review of cost literature suggested that this is the first project to use

a case-mix approach to estimating and predicting care costs for individuals with ADRD, ALS, or MS. The use of the case-mix approach allowed service use and cost to be understood in the context of resource need. In addition, the RUGIII/HC categorization provided a ratio-level estimate of resource use, so that the costs associated with ADRD, ALS, and MS can be understood in comparison to other neurological conditions, as well as to the general HC population. The use of the same methodology across the cost estimate and prediction procedures also ensure that the findings from this project can be compared across conditions.

Another strength of the project was the identification of a number of specific clinical characteristics and their relationships to cost. Although prior studies had explored the effect of severity on cost, these previous findings had only been able to measure severity through ADL impairment ratings, Expanded Disability Status Scale, or cognitive impairment scales. The results from this project allow severity to be measured much more comprehensively through the availability of specific clinical characteristics. At this time, it does not appear that any other study has been able to identify such detailed clinical predictors of cost for ADRD, ALS, or MS.

A final strength of this project is the generalizability of these findings. Since the findings from this project were based on the entire population of HC service users with ADRD, ALS, and MS between April 2005 and June 2008, the predictors of costs identified for these conditions are very likely to be generalizable towards other HC jurisdictions in Canada. In addition, some of the clinical predictors of costs identified this project are likely to be generalizable across at least some of the neurological conditions. The specific clinical characteristics that were predictive of cost across ADRD, ALS, and MS, including stair use, difficulty swallowing, respiratory challenges, and incontinence, are common across a number of other neurological conditions, and are likely to be significant predictors of costs for these conditions as well. These conditions

include cerebral palsy, with symptoms such as limb function impairment and bladder incontinence (Koman et al., 2004); Huntington's disease and muscular dystrophy, which are associated with movement impairment and swallowing symptoms (Walker, 2007; Emery, 2002); and finally Parkinson's disease, where urinary incontinence and difficulty swallowing are also common symptoms (Lees et al., 2009). However, since each of these conditions are associated with a number of other symptoms occurring at different stages in the disease progression, further research on these conditions is still required to identify predictors of costs for other neurological conditions. Further, the amount of cost that is contributed by these characteristics cannot be estimated based on the findings from this project.

7.6 Implications

The findings from this project correspond with the aims of the Innovations in Data, Evidence and Applications for Persons with Neurological Conditions (ideasPNC). The purpose of ideasPNC was the first major study linking interRAI data to the neurological conditions in order to better understand the needs of persons with neurological conditions across the continuum of care. In this particular project, a number of predictors of HC service needs associated with ADRD, ALS, and MS in a HC setting were identified. Some of these predictors included ADL functionality and cognitive performance, which had been identified in other studies, and are predictive of HC use in the general population as well. Other characteristics, such as difficulty with stairs, swallowing, and breathing, had not previously been identified as predictors of costs for these conditions.

The identification of these clinical and personal characteristics are expected to contribute to care planning for individuals with these conditions. At the policy level, these findings can help ensure that adequate resources are available to provide HC services for these individuals.

Ensuring the availability of HC services for individuals with ADRD, ALS, and MS involves the ability to predict care needs, as well as resources required to address these needs. The findings from this project provide the necessary data to ensure adequate funding and service provision for these individuals to remain in the community for as long as possible. In particular, the main finding from this project is that clinical complexity found in individuals with ADRD, ALS, and MS invariably result in increases in costs. As individuals with these conditions increasingly choose to remain in their homes for longer periods of time, the HC costs can be expected to increase, reflecting the increasingly complex characteristics of service users. Increased funding for the Ontario HC program will be required should the proportion of HC service users with neurological conditions increase, as has been predicted.

Another implication of this project is the applicability of the RUGIII/HC algorithm for the neurological populations. Overall, the RUGIII/HC demonstrated reasonable effectiveness in its ability to predict resource utilization levels for individuals with ADRD, ALS, and MS. Mean costs for individuals with these conditions were higher than the mean costs for individuals without neurological conditions within the same RUGIII/HC group. This finding does indicate that variability between the predicted and observed levels of resource utilization is higher for individuals with these neurological conditions. As a result, future versions of the RUGIII/HC should consider the inclusion of additional characteristics predictive of cost for these individuals. The need for revisions or adaptations of the RUGIII/HC for the neurological populations will be especially important given the expected rise in prevalence for neurological conditions. Beyond the neurological populations, the focus on this population is also likely to improve the RUGIII/HC for the general population. Many individuals with and without these conditions prefer to remain in their homes for as long as possible and so an increase in HC service users can

be expected. Improved ability to predict the levels of resource utilization will therefore be crucial to the success of HC service provision in Ontario.

Finally, the findings from this project provide further support that illness factors in the Andersen and Newman model are the most important predictors of healthcare service use in HC settings. These findings also suggests that the factors driving healthcare service use is at least somewhat different in HC than in other care settings. In addition, it is important to note that the relative importance of predisposing and enabling factors were similar between HC service users with neurological conditions, and the general HC population. This finding implies that although revisions to the framework proposed by Andersen and Newman (1973) are required in the HC setting, no specific revisions are required to describe individuals with neurological conditions.

7.7 Future Directions

The findings from this project are expected to improve the understanding of mechanisms driving care costs for ADRD, ALS, and MS. Future studies of cost for these conditions should consider addressing the weaknesses identified in this project. Indeed, while the findings from this project provide a broad understanding of the clinical mechanisms driving costs for the neurological conditions, more specific analysis would help to identify predictors of cost for specific HC services. Future HC cost research would also benefit from the addition of case manager time and drug expenditures into the cost measurement. The inclusion of these costs would provide a more complete estimate of direct costs associated with these conditions. Finally, since some predictors of costs were nonsignificant when repeat assessments were removed, future research should also determine whether clinical predictors of costs are different in populations with repeat assessments removed.

Beyond the three neurological conditions discussed in this project, the relationship between cost and the neurological conditions is still not well understood across other neurological conditions and other non-acute care settings. Future research estimating and identifying predictors of costs should continue to apply high-quality cost and clinical data to other neurological conditions. In particular, this project has demonstrated the excellent research capabilities for data collected using the interRAI instruments. Future cost studies for other neurological conditions care settings should therefore consider the use of data collected through interRAI instruments. These other care settings may include nursing home, palliative care, as well as mental health facilities. Other conditions with adequate sample sizes across these care settings include cerebral palsy, epilepsy, Huntington's disease, muscular dystrophy, Parkinson's disease, stroke, spinal cord injury, and traumatic brain injury.

7.8 Conclusions

Overall, individuals with ADRD, ALS, and MS displayed greater clinical complexity and higher care costs. The mean costs of care for ADRD, ALS, and MS were higher than for individuals without neurological conditions in HC. This finding was demonstrated by the mean costs of care, as well as through the mean CMI values found for each of the three conditions. In addition, these conditions displayed greater proportions of severity or presence of problem characteristics.

Characteristics predictive of costs across the three neurological conditions were fairly consistent. These characteristics included CMI, cohabitation with caregiver, ADL, IADL, unsteady gait, difficulty with stair use, respiratory challenges, difficulty swallowing, impaired cognitive performance, and incontinence. In almost all cases, these characteristics were predictive of higher costs. Many of these characteristics had previously been associated with the

three neurological conditions, but had not been identified as predictors of costs. Nevertheless, these results were consistent with prior findings that disease severity was predictive of increased care costs.

Taken together, these findings suggest that individuals in HC with ADRD, ALS, and MS have greater clinical complexity than individuals without neurological conditions. It is these clinical complexities that result in higher care costs for individuals with these conditions.

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APPENDICES

Appendix A: CCAC/LHIN Map

(Source: Community Care Access Centres, n.d.)

1. Erie St. Clair
2. South West
3. Waterloo Wellington
4. Hamilton Niagara Haldimand Brant
5. Central West
6. Mississauga Halton
7. Toronto Central
8. Central
9. Central East
10. South East
11. Champlain
12. North Simcoe Muskoka
13. North East
14. North West

Appendix B: MEDLINE PubMed Search Strategy

The purpose of this literature review was to examine existing cost literature for the neurological conditions that specifically studied predictors of cost. As little research has been conducted looking at predictors of cost for the neurological conditions exist, a very general search statement was used in order to capture as many articles as possible.

Search Statement:

("multiple sclerosis"[MeSH Terms] OR "dementia"[MeSH Terms] OR "alzheimer disease"[MeSH Terms] OR "amyotrophic lateral sclerosis"[MeSH Terms]) AND ("economics"[Subheading] OR "economics"[All Fields] OR "cost"[All Fields] OR "costs and cost analysis"[MeSH Terms])

Exclusion and Inclusion Criteria:

Results from the search statement were further filtered to exclude non-English articles, non-human studies, and articles without an available abstract and full text. Articles on the predictors of cost had to be manually selected. During manual selection, only original research articles that explicitly stated determining predictors or drivers of cost were included.

Results:

A total of 3590 articles were found using the search statement above. After applying exclusion criteria, 2192 articles remained. Through manual selection, only ten articles were found that fit the inclusion criteria.

Appendix C: Summary of Existing Cost Literature

Author Condition Region	Purpose	Design	Sample	Analysis	Findings
Reese et al., 2011b **Alzheimer's Disease (AD)** **Germany**	Report service use and costs for patients with AD, and to explore influence of sociodemographic and illness-related determinants in ambulatory setting.	Cross Sectional	N=395 outpatient memory clinic, office based neurologists, general practitioner clinics, mental disorder clinic	t-tests, chi-square tests, and Mann-Whitney U-tests used to test differences in demographic characteristics. Multivariate analyses used to create model of independent predictors of higher costs including age, gender, and ADL.	Model explained only 7-43% of variability in costs. Direct medical care costs and LTC costs related differently to patient's clinical characteristics.
Leicht et al., 2011 **Dementia** **Germany**	Estimate net costs of dementia by degree of severity. Include detailed assessment of costs of care.	Cross Sectional	N=348 Sample from German Study on Ageing and Dementia in Primary Care Subjects	Differences in proportions tested using Chi-square test or Fisher's exact test. Group differences tested with t-tests.	Across disease stages, nursing care accounted for 75% of total costs. Half of these nursing costs resulted from informal care.
Gustavsson et al., 2011 **Alzheimer's Disease (AD)** **Spain, Sweden, UK, US**	Identify measures of disease severity that are most important predictors of societal costs of care.	Cross Sectional	N=2444 1222 and their caregivers in community or residential settings.	Descriptive means and standard deviations. Multivariate model used to identify important drivers of costs of care.	ADL ability most important predictor of cost of care for community dwelling patients. Predictors of costs were the same across countries.
Rojas et al., 2011 **Dementia** **Argentina**	Compare costs between various subtypes of dementia.	Case-Control Direct	N=79 Patients diagnosed with dementia presenting to hospital between 2002 and 2008	Means and standard deviation for descriptive data. Differences among dementia subtypes estimated using ANOVA.	Different types of dementia have different costs.
McCrone et al., 2008 **Multiple Sclerosis (MS)**	Investigate links between service use, costs, QOL, and disability.	Cross Sectional	N=1942 members of the MS Society of Great Britain and Northern Ireland	Spearman's correlations given between costs and other continuous variables in addition to Pearson's correlations. Regression models using least squares method used to identify	Age, being divorced/separated, MS type, and years with MS were all significantly associated with higher service costs ($p=<0.05$).

United Kingdon				patient characteristics that were significantly associated with cost.	
Andersen et al., 2003 **Dementia** **Denmark**	Use longitudinal data to examine changes in healthcare costs with disease progression.	Prospective cohort study	N=3346 Community sample randomly drawn from Danish Population Register living in Odense	Multivariate linear regression used to analyze impact of disease progression on healthcare costs.	Change in costs over time increased with disease progression, particularly in the severe stage of dementia. Functional abilities also important factor for exampling changes in costs.
Grima et al., 2000 **Multiple Sclerosis (MS)** **Canada**	Quantify cost of MS and examine the influence of disability on patient utility and healthcare costs.	Cross Sectional	N=42 Patients recruited during regularly scheduled visits to MS clinics either at the Montreal Neurological Institute, or the London Health Sciences Centre.	Patient costs were calculated for each category. No methods for statistical analysis were provided.	Strong positive relationship observed between EDSS and remission healthcare costs.
Jönsson et al., 2006 **Alzheimer's Disease (AD)** **Sweden, Denmark, Norway, Finland**	Estimate the costs of formal and informal care and identify determinants of care costs.	Prospective cohort study	N=272 Recruited from memory clinics.	Non-parametric rank analogue of ANOVA used to test for differences in costs of care between patients in different MMSE states.	Dementia costs related to dementia severity, as well as presence of behavioral disturbances.
Kang et al., 2006 **Dementia** **Korea**	Analyze healthcare expenditures and cost per dementia patient.	Cross Sectional	N=609 Randomly selected patients from nationwide claim database of the Korean National Health Insurance Corporation	Descriptive means for direct and indirect costs calculated. Testing comparisons were performed on the three levels of severity.	Limitations on ADL significantly affects service use ($p=<0.05$).

Appendix D: University of Waterloo Office of Research Ethics, Ethics Clearance

UNIVERSITY OF WATERLOO

OFFICE OF RESEARCH ETHICS

Notification of Ethics Clearance of Application to Conduct Research with Human Participants

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Faculty Supervisor: John Hirdes **Department:** Health Studies & Gerontology
Student Investigator: Clare Cheng **Department:** Health Studies & Gerontology

ORE File #: 18599

Project Title: Identifying Predictors of Costs for the Neurological Conditions in an Ontario Home Care Population

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This certificate provides confirmation that the additional information/revised materials requested for the above project have been reviewed and are considered acceptable in accordance with the University of Waterloo's Guidelines for Research with Human Participants and the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans. Thus, the project now has received ethics clearance.

***Note 1**: This ethics clearance from the Office of Research Ethics (ORE) is valid for one year from the date shown on the certificate and is renewable annually, for four consecutive years. Renewal is through completion and ethics clearance of the Annual Progress Report for Continuing Research (ORE Form 105). A new ORE Form 101 application must be submitted for a project continuing beyond five years.*

***Note 2**: This project must be conducted according to the application description and revised materials for which ethics clearance has been granted. All subsequent modifications to the project also must receive prior ethics clearance (i.e., Request for Ethics Clearance of a Modification, ORE Form 104) through the Office of Research Ethics and must not begin until notification has been received by the investigators.*

***Note 3**: Researchers must submit a Progress Report on Continuing Human Research Projects (ORE Form 105) annually for all ongoing research projects or on the completion of the project. The Office of Research Ethics sends the ORE Form 105 for a project to the Principal Investigator or Faculty Supervisor for completion. If ethics clearance of an ongoing project is not renewed and consequently expires, the Office of Research Ethics may be obliged to notify Research Finance for their action in accordance with university and funding agency regulations.*

***Note 4**: Any unanticipated event involving a participant that adversely affected the participant(s) must be reported immediately (i.e., within 1 business day of becoming aware of the event) to the ORE using ORE Form 106.*

Maureen Nummelin, PhD
Director, Office of Research Ethics

OR
Susanne Santi, MMath
Senior Manager, Research Ethics

OR
Julie Joza, MPH
Manager, Research Ethics

12/10/2012
Date

Appendix E: Standardized Service Wage Rates in Ontario Home Care (2007-8)

Service	**Cost (per visit or hourly)**
Nursing Visit	$55.56
Nursing Hourly	$47.31
Personal Care Hourly	$28.18
Physical Therapy Visit	$100.02
Occupational Therapy Visit	$111.60
Speech Language Pathology Visit	$101.31
Social Work Visit	$140.85
Nutrition/Dietetic Visit	$112.22
Respite Hourly	$31.36

(Source: Hirdes et al., 2010**)**

Appendix F: Scoring Rules for the RAI ADL Self-Performance Hierarchy

Scoring rules—Note the four items used to score this scale are the same as the four items used to score the MDS ADL–Short Form Scale: personal hygiene, toileting, locomotion, eating	Category Score Value	Category Label	Percent of Residents in the Cross-State MDS Sample in Category
All 4 ADLs	0	Independent	8.6
MDS ADL–Short Form range > 0 AND All four ADLs < 2	1	Supervision	7.4
All four ADLs < 3 AND One or more of the four ADLs = 2	2	Limited	13.2
Both eating and locomotion < 3 AND Either or both of personal hygiene and toilet > 2	3	Extensive 1	24.9
Either eating or locomotion = 3 AND Neither of these 2 ADLs = 4	4	Extensive 2	12.1
One or both of eating and locomotion = 4	5	Dependent	17.7
All four ADLs = 4	6	Total dependence	16.1

(Source: Morris et al., 1999)

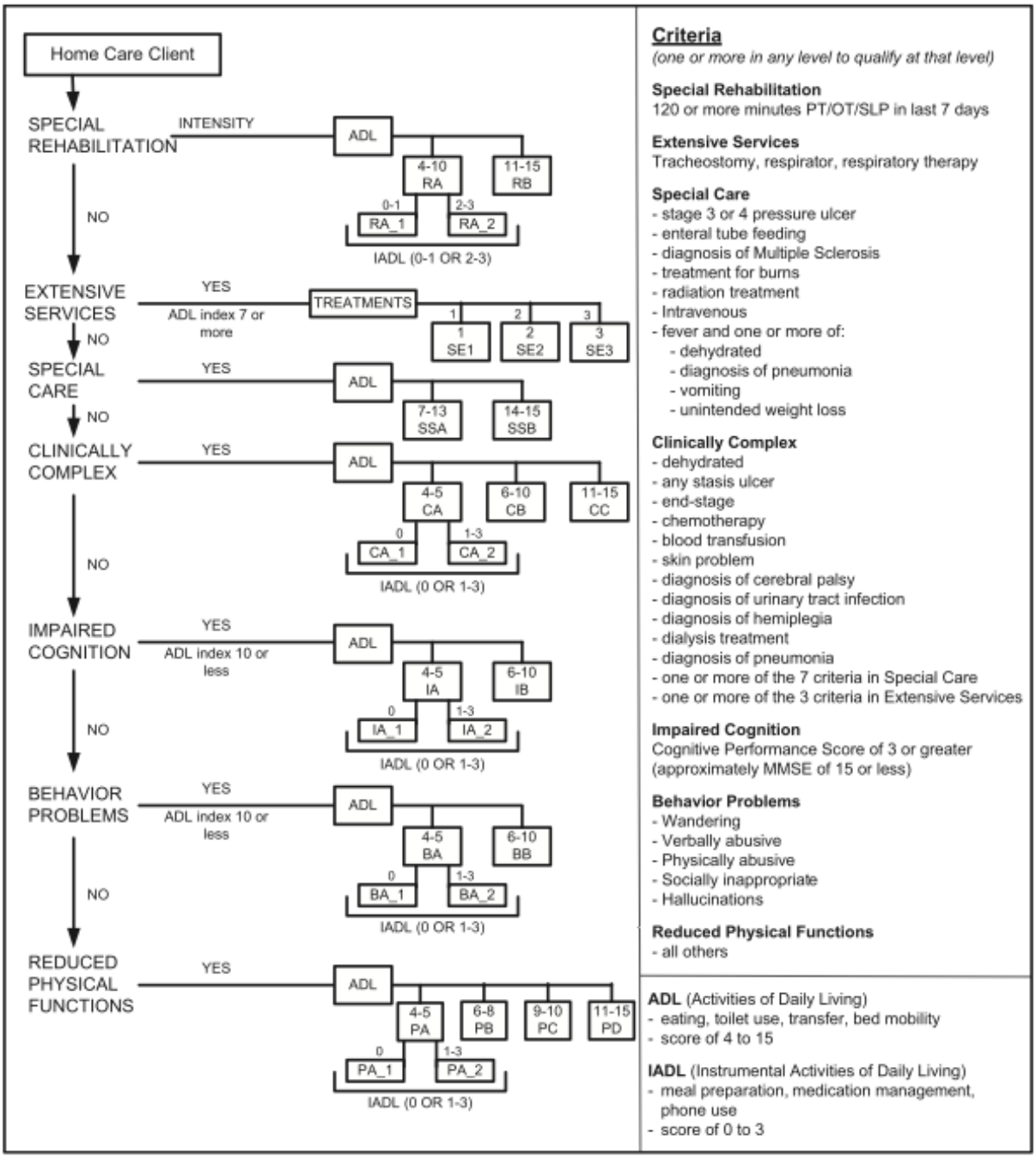
Appendix G: HSI Multiplicative Function and Mapping

$$\mathrm{HSI}_i = \left(\alpha * \prod_{k=1}^{6} \mathrm{w}_k(\mathrm{h}_{i,k}) \right) - \beta$$

Attribute	MDS variable	HUI2 level, description, and preference weight[a]	
Sensation (vision, hearing, speech)	No vision, hearing, or communication difficulty	1. Able to see, hear, and speak normally for age	1.00
	Minimal impairment (with or without aids)	2. Requires equipment to see or hear or speak	0.95
	Highly impaired (with or without aids)	3. Sees, hears, or speaks with limitations, even with equipment	0.86
	Severely impaired (with or without aids)	4. Blind, deaf, or mute	0.61
Mobility	Independent in locomotion	1. Able to walk, bend, lift, jump and run normally for age	1.00
	Supervision but no physical help from others & no devices	2. Walks, bends, lifts, or jumps with some limitations; no help required	0.97
	No physical help & self-supporting devices (cane, walker, self-wheel)	3. Requires mechanical equipment (such as canes, crutches, braces or wheelchair) to walk or get around independently	0.84
	Physical help from others & use of devices	4. Requires the help of another person to walk or get around and requires equipment	0.73
	Total dependence on others	5. Unable to control or use arms and legs	0.58
Emotion	No negative mood indication in last 30 days (5 indicators)	1. Generally happy and free from worry	1.00
	Up to two indications exhibited 1–5 days per week	2. Occasionally fretful, angry, irritable, anxious, depressed, or suffering "night terrors"	0.93
	Any one daily or at least three exhibited 1 to 5 days per week	3. Often fretful, angry, irritable, anxious, or depressed	0.81
	Two or three indicators exhibited daily	4. Almost always fretful, angry, irritable, anxious, or depressed	0.70
	Four or five indicators exhibited daily	5. Extremely fretful, angry, irritable, anxious, or depressed usually requiring hospital or psychiatric care	0.53
Cognition	No problem with memory or decision-making	1. Learns and remembers normally for age	1.00
	Memory problem or mild impairment in decisionmaking	2. Learns and remembers more slowly than peers	0.95
	Moderate impairment in decision-making	3. Learns and remembers very slowly	0.88
	Severe impairment in decision-making	4. Unable to learn and remember	0.65
Self-care	Independence in all these activities	1. Eats, bathes, dresses, and uses toilet normally for age	1.00
	Supervision by others but no assistance for any of these activities	2. Eats, bathes, dresses, or uses the toilet independently with difficulty	0.97
	Limited assistance in any of these activities	3. Requires equipment to eat, bathe, dress, or use the toilet independently	0.91
	Extensive assistance or total dependence in any of these activities	4. Requires help of another person to eat, bathe, dress, or use the toilet	0.80
Pain	No pain	1. Free from pain and discomfort	1.00
	Pain less than daily, not requiring prescribed medications	2. Occasional pain. Discomfort relieved by nonprescription drugs or self control without activity disruption	0.97
	Pain daily, not intense, disrupts activities, and relieved by medication	3. Frequent pain. Discomfort relieved by oral medicines with occasional disruption of normal activities	0.85
	Pain daily, intense, disrupts activities, and relieved by medication	4. Frequent pain; frequent disruption of normal activities. Discomfort requires prescription narcotics for relief	0.64
	Pain daily, intense, disrupts activities, and not relieved by medication	5. Severe pain; pain not relieved by drugs and constantly disrupts normal activities	0.38

(Source: Wodchis et al., 2000)

Appendix H: RUGIII/HC Grouping Criteria



(Source: Poss et al., 2008)

Appendix I: RUGIII/HC Case-Mix Index Values

RUGIII/HC Groups	CMI Values
RB	2.743
RA2	1.609
RA1	0.967
SE3	5.151
SE2	4.240
SE1	2.498
SSB	2.791
SSA	1.878
CC	2.586
CB	1.660
CA2	1.126
CA1	0.609
IB	2.121
IA2	1.637
IA1	0.839
BB	1.718
BA2	1.281
BA1	0.593
PD	2.417
PC	1.755
PB	1.379
PA2	0.933
PA1	0.485

(Source: Hirdes et al., 2010)