Healthcare utilisation and costs of home mechanical ventilation

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ABSTRACT

Background Individuals using home mechanical ventilation (HMV) frequently choose to live at home for quality of life, despite financial burden. Previous studies of healthcare utilisation and costs do not consider public and private expenditures, including caregiver time.

Objectives To determine public and private healthcare utilisation and costs for HMV users living at home in two Canadian provinces, and examine factors associated with higher costs.

Methods Longitudinal, prospective observational cost analysis study (April 2012 to August 2015) collecting data on public and private (out-of-pocket, third-party insurance, caregiving) costs every 2 weeks for 6 months using the Ambulatory and Home Care Record. Functional Independence Measure (FIM) was used at baseline and study completion. Regression models examined variables associated with total monthly costs selected a priori using Andersen and Newman's framework for healthcare utilisation, relevant literature, and clinical expertise. Data are reported in 2015 Canadian dollars (\$C1=US\$0.78=£0.51=€0.71).

Results We enrolled 134 HMV users; 95 with family caregivers. Overall median (IQR) monthly healthcare cost was \$5275 (\$2291–\$10 181) with \$2410 (58%) publicly funded; \$1609 (39%) family caregiving; and \$141 (3%) out-of-pocket (<1% third-party insurance). Median healthcare costs were \$8733 (\$5868–\$15 274) for those invasively ventilated and \$3925 (\$1212–\$7390) for non-invasive ventilation. Variables associated with highest monthly costs were amyotrophic lateral sclerosis (1.88, 95% CI 1.09 to 3.26, $P<0.03$) and lower FIM quintiles (higher dependency) (up to 6.98, 95% CI 3.88 to 12.55, $P<0.0001$) adjusting for age, sex, tracheostomy and ventilation duration.

Conclusions For HMV users, most healthcare costs were publicly supported or associated with family caregiving. Highest costs were incurred by the most dependent users. Understanding healthcare costs for HMV users will inform policy decisions to optimise resource allocation, helping individuals live at home while minimising caregiver burden.

INTRODUCTION

Individuals using home mechanical ventilation (HMV) who choose to live at home are a small yet rapidly growing population with prevalence rates ranging from 6.6¹ to 20.0² per 100 000 adults (12.9/100 000 in Canada).³ These individuals have unique and variable healthcare needs requiring a

Key messages

What is the key question?

- What are the public and private healthcare utilisation and costs for home mechanical ventilation (HMV) and what factors are associated with higher costs?

What is the bottom line?

- For HMV users, most healthcare costs were publicly supported or associated with family caregiving with the highest costs incurred by the most dependent users.

Why read on?

- This prospective study rigorously detailed healthcare costs and confirms the substantial public healthcare cost savings of facilitating mechanical ventilation in the home even when family caregiver time is properly costed, yet indicates that the burden to family caregivers is substantial and needs to be considered in future policy decisions related to HMV.

coordinated strategy for delivery of appropriate and cost-effective care.⁴ Many choose to live at home to promote quality of life, despite the potential physical, psychological and financial burden for themselves and their families.^{5 6}

Current data describing healthcare utilisation and costs for HMV are limited, with few studies considering aspects of *both* public and private expenditures.^{7 8} Most studies were published before 2000^{7 9 10} and are therefore less valid today. Furthermore, most studies do not provide a monetary value for time devoted to caregiving by family caregivers, or for missed time from employment, or household work for either family caregivers or HMV users.^{11 12} Additional limitations of existing studies include retrospective designs leading to increased risk of recall bias.

Few studies examine factors associated with increased healthcare costs for HMV. In the 1990s, Seveck and Bradham analysed US labour costs of home care services purchased by 277 HMV users and found these increased with older age, greater dependence with activities of daily living and greater technical complexity (medical and equipment) of home care. Labour costs were inversely associated with the number of family caregiving

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hours per day.⁷ Although, this was the largest study of healthcare costs for HMV users to date, it did not consider direct costs associated with emergency department and physician visits, hospitalisations, and ventilator rental and maintenance.⁷

In Canada, few studies describe healthcare utilisation and costs of HMV users, and no study considers both public and private costs.^{4 12} Data on HMV user and caregiving factors influencing healthcare utilisation rates and costs are unavailable. These data are essential for informing policy when allocating limited healthcare resources to facilitate optimal support and quality of life for these individuals while minimising caregiver burden. Therefore, our objectives were to determine public and private healthcare utilisation rates and costs for adults using HMV in two Canadian provinces, and to examine factors associated with higher costs.

METHODS

Design, setting and participants

We conducted a longitudinal, prospective, observational cost analysis study in two Canadian provinces, Ontario and British Columbia (BC), from April 2012 to July 2014 (Ontario) and June 2014 to August 2015 (BC). In Ontario, the most populous province (approximately 13 million), public and private healthcare services for HMV users vary across 14 different health networks.³ Ontario has a ventilator equipment pool (VEP), which provides publicly funded mechanical ventilators, related equipment and supplies. In BC (estimated population 5 million), a province-wide and publicly funded HMV programme supports adult HMV users with ventilators and equipment and also respiratory care services and education in the home. Each province is responsible for the administration and delivery of services under federal government regulation.¹³

In Ontario, we recruited participants using convenience sampling from the VEP and West Park Healthcare Centre's Long-Term Ventilation Centre of Excellence. In BC, we recruited from the BC Association for Individualized Technology and Supports for People with Disabilities, Provincial Respiratory Outreach Program (PROP). Three participants transitioning to adult care (18 years old) were recruited from the Children's Hospital of Eastern Ontario (CHEO). At the time of the study, the sampling frame was approximately 1100 (700 from VEP/West Park/CHEO and 400 from PROP). Potential study participants were identified by the organisation's staff and mailed or handed a recruitment letter. Participants who were interested in obtaining more information or taking part in the study were contacted by study investigators.

Inclusion criteria for HMV users were: ≥ 18 years old; invasive or non-invasive bilevel (pressure or volume) mechanical ventilation for ≥ 6 hours a day; and living in a private dwelling (including attendant care apartments) for at least 6 months after transition. We selected this time frame as we were interested in relatively stable home care costs as opposed to those incurred during transition. Both HMV user/caregiver dyads and individuals without a family caregiver were included. HMV users were interviewed directly unless they were unable to communicate. Inclusion criteria for caregivers were: ≥ 18 years old; and family member or friend most responsible for providing and/or coordinating care without financial compensation. Exclusion criteria were: using only CPAP and unable to read/speak English/French. Written and informed consent was obtained from all study participants.

Measures

We collected demographic data including age, sex, living arrangement, marital status, highest education level, employment status, diagnosis requiring ventilation, date initiated, ventilation interface (eg, tracheostomy, mask), ventilation hours per day, number of ventilators and household income. We documented chronic conditions using the Canadian Community Health Survey's Chronic Conditions module¹⁴ and measured functional status using the Functional Independence Measure (FIM)¹⁵ at study enrolment and at 6 months. The FIM measures disability based on burden of caring, with 13 motor and 5 cognition domains. It has been validated and standardised in individuals with spinal cord injury, traumatic brain injury and multiple sclerosis.¹⁵ A higher FIM score (scale 18–126) represents more independent function. Caregiver demographic information included age, sex, relationship to HMV user, marital status, highest education level and employment status.

To minimise recall bias, we collected cost and utilisation data every 2 weeks for 6 months using the Ambulatory and Home Care Record (AHCR)¹⁶ by telephone interview. The AHCR assesses both public and private healthcare expenditures and has been reliably used in various populations¹⁷ including palliative care¹⁸ and people living with chronic conditions.¹⁹ Data collected comprised all costs covered by the public sector including ambulatory/outpatient, inpatient and home-based health services. In Canada, most (approximately 70%) medically necessary health services are publicly insured, exceptions include some medications, vision care, dental care and allied health services. We collected data on health services covered by the private sector including third-party insurance and out-of-pocket spending, caregiving costs (time) incurred by family caregivers and costs to employers due to caregiver or HMV user absenteeism.

Calculation of costs

We used established methods for generating costs.^{16 20} We aggregated all costs over the 6 months and reported median (IQR) monthly costs (rate per 30 days) in 2015 Canadian dollars (\$C). We grouped costs into three categories: publicly funded health services; privately funded health services; and time costs incurred by family caregivers. Categories considered costs associated with personnel, equipment, supplies and medications (see online supplementary material).

We obtained physician and laboratory costs from the Ontario Health Insurance Schedule of Benefits, Schedule of Benefits for Laboratory Services (Ontario) and the Medical Services Plan (BC). We obtained medication costs from the government-sponsored Ontario Drug Benefit (ODB) programme, PharmaCare (BC) and RxTx (formerly eCPS and eTherapeutics). Payment rates from public home care agencies were used to calculate costs associated with home care personnel. Disposable supply costs were derived from the Ontario Assistive Devices Program product manuals, BC Ministry of Health's payment schedules, a publicly funded homecare agency, PROP's suggested yearly inventory and a previous study.¹⁸

Unpaid caregiver time costs were calculated using the human capital approach.^{21 22} We applied 2011 Canadian census age/sex-based earnings estimates, adjusted for 15% nominal earnings growth to 2015, multiplied by 1.20 for employer paid benefits, and further multiplied by 52/46 to account for vacation days and holidays. Time lost from household work/leisure was calculated using the 2011 Canadian census estimated earnings of a homemaker and adjusted for earnings growth, fringe benefits, vacation days and holidays. For each participant, the total time

cost was the product between the monetary value of one unit of time and the total time of caregiving.¹⁶

Sample size

Based on recommendations for fitting multivariable models, a minimum of 50+8m (where m is the number of predictors) study participants is required to avoid overfitting.²³ Therefore, a minimum sample of 122 participants was required to assess the association of nine independent variables with 30-day total costs (dependent variable).

Data analysis

We used descriptive statistics to describe healthcare utilisation for the overall study cohort, according to duration of ventilation duration per day, ventilatory interface (invasive or non-invasive), disease and province. Because the different cost categories did not sum to the total statistically derived median cost, the proportion of each category was calculated using the sum of median public+caregiver lost time+private out-of-pocket+third-party insurance as the denominator. A priori variables considered associated with increased healthcare costs were selected using Andersen and Newman's²⁴ framework for healthcare utilisation, relevant literature and clinical expertise. We generated a multivariate linear regression model of monthly total costs with age, sex, functional status (quartiles of FIM total score, the lowest of the two baseline and study completion scores), ventilation status (invasive vs non-invasive); maximal daily ventilation duration (hours/day), diagnosis (amyotrophic lateral sclerosis (ALS), COPD, muscular dystrophies, other) and province (Ontario, BC) as independent variables. Exponentiated estimates and their 95% CIs were determined. As participants provided data at multiple time points, we controlled for within-person correlation using a first-order autoregressive covariance structure.

RESULTS

Demographics

Between April 2012 to July 2014 (Ontario) and June 2014 to August 2015 (BC), 134 HMV users (86 Ontario, 53 BC) were enrolled. Ninety-five (71%) (68 Ontario, 27 BC) were identified as family caregivers, 69 living with them. Mean (SD) age was 56.5 (17.2) years, 76 (57%) were male; 45 (34%) were invasively ventilated via tracheostomy. Participants required HMV for a median (IQR) of 8.0 (2.5–15.2) years and now required 10.0 (8.0–24.0) hours on average per day (table 1). The median (IQR) total, cognitive and motor FIM scores at baseline were 88.1 (51.5–115.3), 33.5 (32.0–34.0) and 56.5 (18.0–82.0), respectively. Scores were lowest across all domains for individuals with ALS (n=17) of 49.3 (44.5–72.9), 33.0 (31.0–34.0) and 18.5 (13.0–38.9). Mean (SD) age of primary caregivers was 60 (11) years; 72 (76%) were female (table 1).

Monthly healthcare costs

The overall median (IQR) monthly healthcare cost per individual was \$5275 (\$2291–\$10 181). Over half (58%, \$2410, \$955–\$361) of the overall monthly healthcare costs were publicly funded; 39% (\$1609, \$0–\$5527) were family caregiver time costs; 3% (\$141, \$67–\$438) out-of-pocket costs; and less than 1% (\$0, \$0–\$21) third-party insurance (table 2).

Median (IQR) cost for non-invasively ventilated individuals was \$3925 (\$1212–\$7390); \$8733 (\$5868–\$15 274) for those invasively ventilated. The per cent breakdown of publicly funded, caregiver time costs, out-of-pocket costs and third-party insurance was similar to the overall cohort (table 2). With respect to

daily duration of ventilation, the highest median (IQR) monthly cost was incurred by individuals ventilated more than 20 hours/day (n=41) at \$8551 (\$6131–\$17 090) (table 2). When examining costs associated with diagnosis, ALS incurred the highest costs at \$12 876 (\$8495–\$17 565), followed by Duchenne muscular dystrophy (DMD) at \$8419 (\$5868–\$10 297) and other muscular dystrophies at \$8197 (\$3942–\$13 970) (table 3). We identified large provincial differences in the proportion of costs assigned to different cost categories with HMV users in BC using more publicly funded healthcare at 98% (\$4000 (\$1546–\$5983)) vs 39% (\$1500 (\$629–\$4516)) in Ontario. Caregiving time costs were 0% (\$0 (\$0–\$4950)) in BC vs 56% (\$2141 (\$62–\$6262)) in Ontario (online supplementary tables S1 and S2). For most of the HMV user dyads, there was an inverse cost relationship of public healthcare utilisation and caregiver lost time, for example, for individuals with DMD, 98% of costs were publicly incurred and 0% were associated with caregiving; for those with kyphoscoliosis, 27% of costs were publicly incurred and 64% were related to caregiving (figure 1).

In our multivariable model, variables associated with the highest monthly costs included ALS (estimate 1.88, 95% CI 1.09 to 3.26, P=0.03) representing an 88% increase in median monthly cost compared with those with COPD, and the three lower FIM score quintiles (lowest quintile estimate 6.98, 95% CI 3.88 to 12.55, P<0.001) representing a 300%–700% increase in median costs compared with the highest functioning group (highest quartile). This model adjusted for sex, age, invasive/non-invasive ventilation and duration of daily ventilation, and explained 55% of the variation in total monthly cost (table 4).

DISCUSSION

To our knowledge, this study is the first to provide a comprehensive description of public and private healthcare costs of HMV including costs associated with caregiving. Median monthly healthcare cost (2015 \$C) per individual was \$5275 with 58% derived from public funding and 39% associated with family caregiving time costs. Multivariable modelling demonstrated that highest monthly healthcare costs were associated with ALS and the lower levels of functional independence. Although we found higher costs for invasive ventilation compared with non-invasive ventilation and increasing hours of daily mechanical ventilation, these were not associated with higher costs in this model.

The monthly median cost of \$5275 for our cohort is equivalent to \$63 300 each year; considering the most costly group, ALS, the median annual cost is estimated at \$154 512. Previous Canadian reports estimate the per diem costs of individuals requiring ventilation and cared for in an intensive care unit would range from \$2328 to \$4435 (\$849 720–\$1 618 775 per year) with costs of \$921–\$1587 (\$336 165–\$579 255) (2015 \$C) in a chronic respiratory, weaning, step-down or rehabilitation unit.²⁵ We found an inverse relationship in costs associated with public healthcare utilisation and costs incurred through family caregiving, that is, in Ontario HMV users accessed more publicly funded healthcare services (39%) and less family caregiving (56%). Conversely in BC, which has a province-wide and publicly funded HMV programme that includes access to personnel as well as equipment, public healthcare costs comprised 98% whereas caregiver costs were 0% of total median costs. Although this is a Canadian example, it illustrates investing more in healthcare policy for provision of healthcare services that enable more HMV users to live and stay at home while providing appropriate levels of support to family caregivers may be a wise public healthcare investment that may result in substantial cost savings, when

Table 1 Demographic characteristics of HMV users and caregivers

HMV user (n=134)	n (%), unless specified	Caregiver (n=95)	n (%), unless specified
Age (mean (SD)), years	56.5 (17.2)	Age (mean (SD)), years	60.0 (11.1)
Male	73 (56.7)	Female	72 (75.8)
Diagnosis		Marital status	
COPD or other chronic lung disease	21 (15.7)	Married	75 (78.9)
Amyotrophic lateral sclerosis	17 (12.7)	Never married	4 (4.2)
DMD	9 (6.7)	Divorced/separated	5 (5.3)
Muscular dystrophies other than DMD	14 (10.4)	Widowed	3 (3.2)
Postpolio syndrome	11 (8.2)	Missing	8 (8.4)
Spinal cord injury	14 (10.4)	Education level (n=11 missing)	
Central hypoventilation syndrome	5 (3.7)	High school or less	24 (25.3)
Sleep apnoea	3 (2.2)	Any vocation/college	33 (34.7)
Diaphragmatic paralysis	2 (1.5)	Any university	23 (24.2)
Kyphoscoliosis/scoliosis	4 (3.0)	Postgraduate	4 (4.2)
Other*, unknown or missing	34 (25.3)	Employment status (n=9 missing)	
Ventilation interface		Retired	38 (40.0)
Tracheostomy	45 (33.6)	Full-time	19 (20.0)
Mask (face, nasal)	68 (50.8)	Not employed	15 (15.8)
Mouthpiece, diaphragmatic pacer	4 (2.9)	Part-time/casual	7 (7.4)
Multiple interfaces	17 (12.7)	Student, disability, on leave, self-employed	7 (7.4)
Duration of HMV, years (median (IQR))	8.0 (2.5–15.2)	Relationship to HMV user	
Daily duration of HMV, hours/day (median (IQR))	10.0 (8.0–24.0)	Wife	38 (40.0)
Living arrangement		Parent	23 (24.2)
With spouse	60 (44.8)	Husband	13 (13.7)
Alone	43 (32.1)	Daughter/daughter-in-law, son	9 (9.5)
With parents	21 (15.7)	Extended family, friend/neighbour	12 (12.6)
With friends, children or others	10 (7.4)		
Marital status			
Married	61 (45.5)		
Never married	49 (36.6)		
Divorced/separated	8 (6.0)		
Widow	16 (11.9)		
Education level			
High school or less	44 (32.8)		
Any vocational/college	38 (28.4)		
Any university	39 (29.1)		
Postgraduate	13 (9.7)		
Number of chronic conditions (median (IQR))	4.0 (2.0–6.0)		
Household income			
≤\$25 000	28 (20.9)		
\$25 001–\$45 000	29 (21.6)		
\$45 001–\$65 000	18 (13.4)		
≥\$65 001	29 (21.6)		
Don't know/refused	30 (22.3)		

*Other diagnoses include: *neuromuscular*: multisystem atrophy, giant axonal neuropathy, Charcot-Marie-Tooth disease, polymyositis autoimmune disease; *non-neuromuscular*: ankylosing spondylitis, glucosamide amino acid deficiency, spina bifida+scoliosis, pulmonary alveolar hypoventilation, haemangioblastoma, multifocal motor neuropathy, brain tumour, brainstem stroke, severe restrictive lung disease, Larsen syndrome, Arnold-Chiari, morbid obesity.

DMD, Duchenne muscular dystrophy; HMV, home mechanical ventilation.

compared with institutionalisation. Such an investment should include infrastructure such a centralised HMV programme providing equipment that includes access to respiratory

specialists in the home and appropriate levels of support for publicly funded caregivers as well as caregiver respite, education, training programmes and day-to-day assistance.²⁶²⁷ The

Table 2 Monthly healthcare costs for the entire cohort by daily duration of ventilation and by ventilator interface

Item	Entire cohort (n=134)	Non-invasive (n=89)	Invasive (n=45)	<10 hours (n=54)	10–19 hours (n=39)	≥20 hours (n=41)
	Median (IQR), % of total summed cost*					
Total public	2410 (955–5361) 58%	1380 (572–3440) 53%	5246 (2730–7333) 54%	1222 (423–3970) 76%	1542 (762–3228) 39%	5800 (4000–8021) 62%
Home-based visits	622 (27–3723)	101 (15–1002)	3780 (2036–5106)	62 (12–692)	162 (25–2109)	3825 (2346–5634)
Ambulatory-based clinic visits	144 (73–249)	153 (85–252)	106 (69–189)	183 (86–284)	153 (99–200)	91 (13–173)
Hospitalisation ED hospice care	0 (0–1002)	0 (0–288)	37 (0–1092)	0 (0–648)	0 (0–288)	0 (0–1098)
Medications	175 (65–392)	173 (36–344)	184 (123–487)	159 (46–303)	158 (40–392)	240 (147–507)
Supplies	173 (38–410)	68 (9–408)	244 (157–410)	52 (1–249)	203 (50–473)	280 (165–574)
Caregiver lost time	1609 (0–5527) 39%	1108 (0–3559) 42%	4262 (0–6586) 44%	236 (0–2547) 15%	2257 (126–6880) 58%	3380 (0–6262) 36%
Total private out-of-pocket	141 (67–438) 3%	136 (73–438) 5%	142 (67–403) 1%	151 (73–365) 9%	125 (63–403) 3%	148 (78–502) 2%
HCP visits (in or out of home)	17 (0–92)	30 (0–79)	0 (0–98)	19 (0–58)	17 (0–97)	8 (0–159)
Travel	30 (13–81)	45 (18–85)	26 (10–55)	44 (15–97)	45 (18–82)	27 (10–45)
Medications	4 (0–18)	3 (0–15)	13 (0–28)	9 (1–18)	1 (0–16)	9 (0–25)
Supplies	30 (4–149)	24 (3–134)	51 (6–178)	20 (2–104)	27 (8–131)	69 (5–264)
Total third-party insurance	0 (0–21) 0%	0 (0–29) 0%	0 (0–7) 0%	0 (0–33) 0%	0 (0–37) 0%	0 (0–6) 0%
Home-based visits	0	0	0	0	0 (0–5)	0
Medications	0	0	0	0	0	0
Supplies	0	0	0	0	0	0
Total healthcare costs	5275 (2291–10181)	3925 (1212–7390)	8733 (5868–15274)	2997 (950–5304)	4980 (2291–10181)	8551 (6131–17090)

Monthly healthcare costs for HMV users in the entire cohort, those ventilated <10, 10–19 and ≥20 hours/day, and those ventilated non-invasively or invasively in 2015 \$C.

*Because different cost categories did not sum to the total statistically derived median cost, the proportion of each category was calculated using the sum of median public+caregiver lost time+private out-of-pocket+third-party insurance as the denominator.

ED, emergency department; HCP, healthcare provider; HMV, home mechanical ventilation.

median monthly healthcare costs we report differ substantially from those in other studies; however, comparison is problematic due to differences in study methods as well as differences in funding mechanisms and patterns of healthcare provision. In the US cross-sectional survey, we highlight in our introduction⁷ the total *labour* median monthly cost of HMV home care (private and public) was \$8201 (1995 US\$) (\$16 234 (2015 \$C)). This is greater than costs incurred by the most dependent patients in our study and likely represents differences in healthcare funding and provision between the USA and Canada. Another reason for disparate findings may be differences in our HMV cohort. Thirty-four per cent of our cohort required tracheostomy ventilation with a median monthly healthcare cost of \$8733 compared with \$3925 for those on non-invasive ventilation. Previous studies have included a higher proportion of individuals on tracheostomy ventilation.^{7,12} Klein and Forshew similarly reported much lower costs for non-invasive ventilation at \$435 (1996 US\$) per month (\$825 (2015 \$C)) compared with \$16 625 (1996 US\$) for tracheostomy ventilation (\$32 260 (2015 \$C)) for individuals with ALS, though this difference is markedly greater than found in our cohort.²⁸ Another reason for the disparity in costs compared with those reported in previous studies is that our cohort is well established using ventilation at home, on average 10 years. Therefore, our data are representative of stable costs of HMV as they exclude those incurred during the highly expensive transition to home period. These costs that include items such as home renovation, new equipment and caregiver training have been shown to be much higher, especially if ventilation is commenced in the hospital.^{12,29,30} As an example, a previous

Canadian report¹² describing only healthcare personnel costs during transition to home, found that the mean monthly cost was \$11 093 (2015 \$C).

We found that the highest monthly healthcare costs were associated with ALS and low levels of functional ability. Previous studies support this finding.⁷ In a review of 12 studies describing costs associated with ALS (some were HMV users), Gladman and Zinman identified drivers of cost included more severe disease, lower levels of functional ability defined as increased support with activities of daily living and mechanical ventilation.³¹ Similarly, studies with HMV users with ALS³² and mixed diagnoses¹⁰ indicate that increased healthcare costs were incurred by individuals with more complex health needs and higher functional dependency defined as the need for skilled healthcare personnel, respiratory equipment and supplies.

We found that 39% of monthly healthcare cost was incurred through family caregiver time. As noted earlier, few studies have considered family caregiving when calculating the cost of HMV; the 13 studies we found had highly variable costs due to different calculation methods. Seveck and Bradham⁷ reported that median cost of home care rose by 41% (\$2006–\$3409 (1995 US\$); \$4564 (2015 \$C)) when family caregiving time was considered, using opportunity costing methods (market-based earning capacity of the caregiver based on education and skill).⁷ Overall caregiving costs were higher compared with our cohort but were similar for those receiving tracheostomy ventilation or ventilated >20 hours/day. Variability in caregiving time cost reporting may also be due to cohort characteristics. Two studies, one with an international DMD cohort, the other a Spanish ALS

Table 3 Monthly healthcare costs for HMV users by diagnosis

Diagnosis	Total public	Caregiver lost time	Total private out-of-pocket	Total third-party insurance	Total healthcare costs
Amyotrophic lateral sclerosis (n=17)	5042 (2040–7903) 44%	5890 (3805–6997) 51%	656 (178–1987) 6%	0 (0–99) 0%	12 876 (8495–17 565)
DMD (n=9)	5394 (4432–8021) 99%	0 (0–5907) 0%	46 (20–187) 1%	0 (0–10) 0%	8419 (5868–10 297)
Other muscular dystrophies (n=14)	4040 (2494–8334) 73%	1425 (0–4950) 26%	103 (25–208) 2%	1 (0–13) 0%	8197 (3942–13 970)
Spinal cord injury (n=14)	4869 (4000–6396) 93%	0 (0–8096) 0%	372 (63–719) 7%	0 (0–7) 0%	6093 (5190–13 473)
Chronic lung disease (n=21)	1380 (965–2664) 39%	2003 (1108–2984) 57%	116 (79–219) 3%	0 (0–33) 0%	3554 (2291–4980)
Postpolio syndrome (n=11)	651 (157–1587) 88%	0 (0–95) 0%	86 (41–196) 12%	0 (0–19) 0%	735 (265–2283)
Central hypoventilation syndrome (n=5)	3174 (174–3970) 96%	0 (0–327) 0%	133 (108–241) 4%	0 0%	4538 (307–9469)
Kyphoscoliosis/scoliosis (n=4)	627 (502–832) 27%	1503 (149–3065) 64%	196 (84–316) 8%	34 (10–51) 1%	2571 (1117–3892)
Sleep apnoea (n=3)	572 (103–1083) 91%	0 (0–60) 0%	60 (18–91) 9%	0 (0–38) 0%	591 (224–1212)
Diaphragmatic paralysis (n=2)	376 (227–525) 28%	815 (9–1621) 60%	167 (120–213) 12%	8 (0–16) 1%	1366 (356–2375)
Other* (n=34)	1465 (423–4469) 90%	2137 (62–5527) 9%	139 (84–365) 1%	0 (0–23) 0%	5968 (1436–8802)

Median and IQR (upper numbers) and per cent of total (lower numbers) of monthly healthcare costs for HMV users by diagnosis in 2015 \$C.

*Other diagnoses: see table 1. Because different cost categories did not sum to the total statistically derived median cost, the proportion of each category was calculated using the sum of median public+caregiver lost time+private out-of-pocket+third-party insurance as the denominator.

DMD, Duchenne muscular dystrophy; HMV, home mechanical ventilation.

cohort (not all of whom received mechanical ventilation in the home), report costs attributable to family caregiver time represented 18%–31% annual costs for DMD³³ (mean \$13 370–\$18 530 (2012 US\$); \$13 977–\$19 371 (2015 \$C)) and 53% of costs for ALS³⁴ (mean €19 125 (2004); \$14 381 (2015 \$C)). In our cohort, caregiving time costs for DMD were much lower (0% of total median cost, median (IQR) \$0 (0–5907)), though we only recruited nine individuals with this diagnosis (moderate function with a mean total FIM score 55 out of 126). Caregiving time cost for our HMV cohort with ALS was 51% of monthly costs, which is comparable to those of the Spanish cohort.

Our study provides detailed information on the public and private healthcare costs of HMV which can inform policymaking decisions on funding allocation. For instance, those individuals with higher functional dependence and/or a diagnosis of ALS may warrant more publicly funded support. Additionally, costs associated with family caregiver time were equivalent to 39% of all healthcare costs and therefore highlight the reliance of the current public healthcare funding model on provision of care by family caregivers. However, this reliance is likely to result in physical, psychological and financial burden on caregivers. Liu and colleagues⁶ found greater physiological and social burden for family caregivers of HMV users living at home in Taiwan compared with caregivers of those living in a chronic respiratory care ward. Current financial supports for family caregivers including tax credits,³⁵ employment insurance benefits,^{35 36} home modification and vehicle modification programmes,^{37 38} and paid leaves for family caregiving³⁹ in Canada are limited.

However, many HMV users live years at home and require daily care, requiring the presence of family caregivers for much more extended time periods.

Strengths and limitations

Irrespective of differences in study design and country of origin, our study offers a comprehensive estimate of HMV home healthcare costs considering both publicly and privately funded health services, and time costs incurred by family caregivers as well as prospectively measuring costs every 2 weeks using a well-established and validated tool over 6 months to minimise recall bias. However, our study has limitations. First, despite collecting healthcare utilisation data every 2 weeks using a validated tool, our data may be subject to recall bias. Second, participants may have provided data that fit with perceived social norms, for example, overestimating or underestimating the time provided for family caring (social desirability bias).⁴⁰ Third, our costing estimates may not be completely accurate as participants did not always know the costs of items, especially those paid out-of-pocket or by third-party insurance, despite our best efforts to locate all costs. Additionally, third-party insurance costs may have been underestimated because the majority of participants may not have possessed it (we did not collect these data). Fourth, study sample characteristics may differ from the broader Canadian HMV user population³ as invasive HMV users are over-represented, and under-representing HMV users with ALS, central hypoventilation syndrome, muscular dystrophies and chest wall

Figure 1 Graphical representation showing an inverse relationship in the proportion of public and caregiver lost time for most HMV user cohorts. *Because different cost categories did not sum to the total statistically derived median cost, the proportion of each category was calculated using the sum of median public+caregiver lost time+private out-of-pocket+third-party insurance as the denominator. ALS, amyotrophic lateral sclerosis; CHS, central hypoventilation syndrome; DMD, Duchenne muscular dystrophy; HMV, home mechanical ventilation; NIV, non-invasive ventilation; PPS, postpolio syndrome; resp, respiratory; SCI, spinal cord injury.

deformities.³ Finally, our data are not generalisable to jurisdictions without publicly funded programmes for HMV users where there will be greater reliance on private health insurance and/or the family caregiver, with the possibility that these individuals may be unable to return home thus incurring greater public healthcare costs.

CONCLUSION

This study, which rigorously detailed publicly and privately funded monthly healthcare costs for HMV including time costs incurred

by family caregivers, demonstrates substantial cost savings to the public healthcare system when these individuals live at home when compared with estimates of costs that occurred in acute or long-term care. Although public funding accounted for the highest proportion of costs, over one-third were due to caregiving time provided by family members. Healthcare costs were highest for those with the lowest functional independence and those with ALS. Our findings confirm the substantial public healthcare cost savings of facilitating mechanical ventilation in the home even when family caregiver time is properly costed, yet indicate that the burden to family caregivers is substantial and needs to be considered in future policy decisions related to HMV.

Table 4 Variables associated with total monthly healthcare costs

Variable	Exponentiated estimate	95% CI	P value
Intercept			<0.0001
ALS diagnosis (ref=all other diseases)	1.88	1.09 to 3.26	0.03
FIM (ref=76th–100th percentiles)			
0th–25th percentiles	6.98	3.88 to 12.55	<0.0001
26th–50th percentiles	6.83	4.05 to 11.51	<0.0001
51st–75th percentiles	2.93	1.91 to 4.50	<0.0001
Male (ref=female)	1.12	0.82 to 1.52	0.47
Age	1.00	0.99 to 1.01	0.81
Tracheostomy (ref=non-invasive interfaces)+	1.41	0.89 to 2.25	0.15
Ventilation duration	1.00	0.97 to 1.03	0.95

We present exponentiated estimates as we log transformed 30-day healthcare costs. Adjusted $R^2=55\%$.

ALS, amyotrophic lateral sclerosis; FIM, Functional Independence Measure.

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