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Journal of the Chinese Medical Association 76 (2013) 390–394

www.jcma-online.com

Original Article

The utilization of hospice care among patients with motor neuron diseases: The experience in Taiwan from 2005 to 2010

Shih-Chao Kang^{a,b}, Shinn-Jang Hwang^{b,c,*}, Pin-Yuan Wu^c, Ching-Piao Tsai^d^a Division of Family Medicine, National Yang-Ming University Hospital, Yilan, Taiwan, ROC^b Department of Family Medicine, Faculty of Medicine, National Yang-Ming University School of Medicine, Taipei, Taiwan, ROC^c Department of Family Medicine, Taipei Veterans General Hospital, Taipei, Taiwan, ROC^d Department of Neurology, Taipei Veterans General Hospital, Taipei, Taiwan, ROC

Received August 19, 2012; accepted December 10, 2012

Abstract

Background: The nature and extent of how patients with motor neuron diseases (MNDs) utilize hospice care in Taiwan remains unclear. This study aims to investigate the use of hospice care in Taiwan by patients with MND, and those factors that affect the extent, the cost, and the quality of their hospice treatment and care.

Methods: We analyzed data from hospice care inpatient claims of MNDs, using the National Health Insurance Research Database of Taiwan during 2005–2010.

Results: Thirty patients and 58 related discharges were enrolled into our study, which consisted of 13 males and 17 females, with a mean age 58.3 years. Of that total, 27 of them (90%) had amyotrophic lateral sclerosis, and four (13.3%) had comorbid cancers; 17 died during hospice care. Acute low respiratory conditions (31.0%) accounted for the most common acute comorbidity. Noninvasive ventilation care was performed in only 13 (22.4%) of the discharges. In contrast to nasogastric intubation (40 discharges, 69.0%), no gastrostomy/jejunostomy was noted. These procedures bore no relationship to results observed in the discharges. Family physicians provided most inpatient hospice services (74.1%). Respiratory problems were the major causes of death (70.6% of decedents). The mean inpatient costs of hospice care were noticeably reduced from previously established nationwide mean costs.

Conclusion: Hospice care can save costs for patients with terminal MNDs, and family physicians play a valuable role in caring for these patients. However, respiratory and feeding problems are prevalent, yet there are proven benefits when noninvasive ventilation care and gastrostomy/jejunostomy are promoted.

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Keywords: cause of death; hospice; motor neuron diseases; respiratory failure; Taiwan

1. Introduction

Motor neuron diseases (MNDs) are a group of neurodegenerative illnesses involving motor nerves with preserved sensory functions. Because of their irreversible progression and lack of available curative treatment, MNDs have been a topic of considerable discussion in the literature.¹ Studies in many

academic fields have focused on a range of domains such as therapeutic developments,² psychological health issues,³ quality of life and care,⁴ the safety of narcotic prescriptions,^{5,6} and ethical issues surrounding respiratory or feeding care.^{7,8}

In Taiwan, National Health Insurance (NHI) has been established since 1995. The NHI program designates MNDs as catastrophic illnesses and patients are enrolled for hospice care payments. Under the standard criteria and diagnosis by neurologists, those who are identified as having a catastrophic illness such as an MND would be exempt from the copayments incurred due to their corresponding disease in both outpatient and inpatient services.⁹ Furthermore, a complete registry of

* Corresponding author. Dr. Shinn-Jang Hwang, Department of Family Medicine, Taipei Veterans General Hospital, 201, Section 2, Shih-Pai Road, Taipei 112, Taiwan, ROC.

E-mail address: sjhwang@vghtpe.gov.tw (S.-J. Hwang).

MNDs has been established. Because the enumeration of patient distribution among individual hospitals has been limited, critical studies of MNDs have also been relatively limited in Taiwan as well. In the late 1990s, Wu¹⁰ presented a descriptive study of care for patients with MNDs using single-hospital data. After 2000, Lai and Tseng¹¹ reported the first epidemiologic report of MNDs in Taiwan, demonstrating the increasing incidence with age and the increasing inpatient costs yearly. The estimated prevalence and annual incidence of MNDs in Taiwan are 1.63–2.79/10⁵ population and 1.05/10⁵ population/year respectively, which are lower than those comparable numbers as reported in the West.¹¹ However, the characteristics of hospice care utilization among patients with MND in Taiwan remain unclear. The aim of this study was to examine how patients with MND use and are integrated into the hospice care system in Taiwan, looking at quality of services provided, costs, patient decision-making, and other issues.

2. Methods

2.1. Design and source of study

This study was conducted using retrospective claim-based analyses, and collected hospice claims data of discharges among patients with MNDs from the National Health Insurance Research Database (NHIRD) from 2005 to 2010. Maintained by the National Health Research Institute of Taiwan, the NHIRD provides anonymous and encrypted NHI claims data for local researchers.¹² Under NHIRD authorization, we screened in-patients' claims data over 6 years (DD2005.DAT, DD2006.DAT, DD2007.DAT, DD2008.DAT, DD2009.DAT, and DD2010.DAT) and collected hospice care discharges of patients with MND by the International Classification of Diseases, Ninth Revision (ICD-9) code of 335.2x. Details of patients' therapeutic procedures were extracted by linking the corresponding ordering files (DO2005.DAT, DO2006.DAT, DO2007.DAT, DO2008.DAT, DO2009.DAT, and DO2010.DAT).

2.2. Parameters

Acute and chronic comorbidities were screened by the corresponding ICD-9 codes such as diabetes mellitus (250), stroke (430-438), cancers (140-239), gastroenterological illnesses (530-537), hypertensive disease (401-405), acute respiratory failure (518.81), acute low respiratory conditions (ALRC, 480-487), and urological infections (590, 595, 597, 599, and 601). The causes of death were extracted according to the corresponding lethal ICD-9 codes of discharges and cause-effect relationships. For instance, we defined "pneumonia" as the cause of death if the decedent had coexisting diagnosis of "pneumonia", "acute respiratory failure", and "sepsis". The mean cost of each hospice admission was calculated in New Taiwan Dollars (NTD).

2.3. Statistical analysis

Data were expressed as mean \pm standard deviation and percentage (%). SPSS software (SPSS version 17.0, SPSS Inc.,

Chicago, IL, USA) was used for statistical analyses, and descriptive statistics were performed in this study. Chi-square and Fisher exact tests were used for comparison of results of discharges and procedures; $p < 0.05$ was taken as the criterion for statistical significance. This study was conducted with the approval of the National Health Research Institute, Republic of China (Taiwan) (No. 100257).

3. Results

During the study period, a total of 30 patients and 58 related discharges were enrolled, including 13 males and 17 females with a mean age 58.3 years (range 32–86 years). Twenty-seven of the patients (90%) had amyotrophic lateral sclerosis; the other three had progressive bulbar palsy, pseudobulbar palsy, and other MND. Four patients (13.3%) suffered from comorbid cancers, including three with ovarian cancer and one with gastric cancer (Table 1). Seventeen patients died during hospice care.

Among the 58 discharges of patients with MND, acute low respiratory conditions (ALRC, 31.0%) and acute respiratory failure (20.7%) were the most common acute comorbidities. Noninvasive ventilation care and tracheostomy were performed only on 13 (22.4%) and 2 (3.4%) of those admitted, respectively, whereas no cardiopulmonary resuscitation or endotracheal intubation was performed. Nasogastric intubation was required for 40 of those admitted (69.0%), but no percutaneous endoscopic gastrostomy/jejunostomy was performed for feeding. Correlations between the aforementioned procedures and expiry or nonexpiry of discharges did not achieve statistical significance (all $p > 0.05$). Most inpatient hospice care for patients with MND was provided by family physicians (74.1%), and only 5.2% by neurologists (Table 2).

Among the 17 deceased patients with MND, pneumonia was the most common cause of death (41.2%). Respiratory problems, including pneumonia, aspiration pneumonitis/suffocation, and acute respiratory failure without involvement of pneumonia, constituted the major causes of death (70.6% of deceased patients in total; Table 3).

The mean costs for hospice care inpatients with MNDs were 47,180 \pm 42,009 NTD. These tabulations were much

Table 1
The demographic characteristics of patients with motor neuron diseases who received hospice care in 2005–2010 ($n = 30$).

Parameters	
Mean age (y)	58.3 \pm 14.2
Sex (male/female)	13/17
Classification (%)	
Amyotrophic lateral sclerosis	27 (90.0)
Progressive bulbar palsy	1 (3.3)
Pseudobulbar palsy	1 (3.3)
Other motor neuron disease	1 (3.3)
Chronic comorbidities (%)	
Diabetes mellitus	2 (6.7)
Stroke	1 (3.3)
Cancers	4 (13.3)
Gastroenterologic illness	1 (3.3)
Hypertensive diseases	1 (3.3)

Table 2
The demographic characteristics of admission records for patients with motor neuron diseases who received hospice care in 2005–2010 ($n = 58$).

Parameters	
Mean hospital stay (days)	12.8 ± 10.8
Results of admissions (discharged/transferred/died)	37/4/17
Noninvasive ventilation care (%) ^a	13 (22.4)
Tracheostomy (%)	2 (3.4%)
Nasogastric intubation (%)	40 (69.0)
Percutaneous endoscopic gastrostomy or jejunostomy (%)	0 (0)
Acute comorbidities (%)	
Acute low respiratory conditions	18 (31.0)
Acute respiratory failure	12 (20.7)
Urologic infections	6 (10.3)
Sections of admissions (%)	
Family medicine	43 (74.1)
Neurology	3 (5.2)
Internal medicine	7 (12.1)
Oncology/hematology	1 (1.7)
Radiation oncology	4 (6.9)

^a No cardiopulmonary resuscitation or endotracheal intubation was performed.

lower than the national mean costs that had been reported (47,180 NTD vs. 121,392 to 180,897 NTD).¹¹

4. Discussion

Amyotrophic lateral sclerosis is the most common type of MND and is present in up to 60% of Taiwanese patients with MND.¹¹ In our current study, the distribution of amyotrophic lateral sclerosis reflected a centralizing trend (90%) among those under hospice care. For patients with terminal MND, this phenomenon implied problems likely to be faced in hospices such as swallowing difficulty, limb and skin care, and respiratory complications. In contrast to the sex distribution shown in a previous study,¹¹ more female than male patients with MND tend to be admitted to hospices. Consequently, it could be worth investigating the existence and extent of sex differences in attitude toward hospice care among patients with MND in Taiwan.

Interestingly, the proportion of chronic comorbidities was low among our patients under hospice care except for comorbid cancers (up to 13.3%). We suppose there might be a

Table 3
The causes of death among deceased hospice-care patients with motor neuron diseases in 2005–2010 ($n = 17$).

Cause of death (%)	
Bacteremia/sepsis, origin undetermined	1 (5.9)
Pneumonia	7 (41.2)
Aspiration pneumonitis/suffocation	1 (5.9)
Acute respiratory failure, without indication of pneumonia	4 (23.5)
MNDs ^a	2 (11.8)
Other or unclear causes ^b	2 (11.8)

MND = motor neuron disease.

^a The ICD-9 code of these two deceased patients' records was registered only as MND.

^b The comorbidities of these two deceased patients were registered as gastric cancer and liver cirrhosis.

registration bias as a result of screening via ICD-9 codes. In order to satisfy claims protocol requirements, the diagnoses of hospice care inpatients may focus on the major diagnosis indicated for hospice care together with relevant complications. Linking and comparing the corresponding outpatient databases or medical records could be a possible solution.

For respiratory care, our results show a low utilization of noninvasive ventilation (22.4%). In developed countries, it has been shown that noninvasive ventilation care is advantageous for maintaining quality of life among patients with MND, even in hospices or under home care.^{13,14} Furthermore, noninvasive ventilation has been recommended for patients with respiratory failure.¹⁵ According to the results of our investigation, this procedure did not affect the prognosis of hospice care; i.e., it maintained patients' care quality rather than prolonged lives. Because of its noninvasive characteristics, we suggest that noninvasive ventilation care should be promoted before tracheostomy among Taiwanese patients with MND.

The prevalence of invasive feeding procedures varied greatly by type in our study. For instance, 69.0% of hospice admissions underwent nasogastric intubation, but no patient underwent gastrostomy or jejunostomy for feeding. We suppose that this difference can be attributed to traditional concepts of end-of-life body image, which are common among the elderly in Taiwan. Patients are more willing to endure uncomfortable nasogastric insertions and related complications rather than undergoing gastrostomy in order to maintain a complete body image at the ends of their lives. Similar considerations of end-of-life body image may also explain the low incidence of tracheostomy in our study, although tracheostomy care has been demonstrated to increase the long-term survival of patients with MND.^{16,17} Experience in developed countries shows that early involvement of palliative gastrostomy enhances the quality of care for patients with MND.⁸ Gastrostomy has been shown to increase the survival rate of patient with MND.¹⁸ Dysphagia and related malnutrition correlate with a poor prognosis among such patients.¹⁹ We suggest that palliative gastrostomy/jejunostomy should be promoted for patients with MND in Taiwan.

Among acute comorbidities, ALRC accounted for a third of the admission records for hospice care patients with MND. Furthermore, one-fifth of those admissions suffered acute respiratory failure. This shows that respiratory complications and infections remain the major challenges for these patients in hospices. On the basis of informed consent and symptomatic palliation, hospice care could solve some of the ethical dilemmas and controversies that arise in acute ward care for patients with MND.⁷

In Taiwan, family physicians and radiation oncologists provide most of the hospice care among patients with cancers.²⁰ The characteristics of physician manpower in hospices can be traced back to the inception of hospice care in Taiwan. Essentially, hospice care was introduced by radiation oncologists in Taiwan in the 1980s, with later legislated and enrolled NHI payment since 2000. By the end of 2010, there were 43 hospital-affiliated hospices and 647 ward beds for such services.²¹ Our results revealed that family physicians provided

most of the hospice care for patients with MND. In contrast, neurologists only provided 5.2% of such care. The decision-making process among terminal noncancer patients regarding their choice of specialties for hospice care in Taiwan remains unclear. However, derived from our own experience of caring for patients with MND, we suppose that family physicians could play a similar hospice care role for noncancer patients, and relevant future studies to explore this are indicated.

The causes of death among patients with terminal MND have been described in developed countries.^{22,23} In developing countries, dysphagia-related malnutrition due to the underlying scarcity of medical resources also contributes noticeably to death.²⁴ As in the aforementioned studies, respiratory problems including pneumonia, aspiration pneumonitis/suffocation, and acute respiratory failure without involvement of pneumonia are the most common causes of death in our hospice care patients. This again demonstrates the importance of adequate respiratory care for symptomatic palliation in patients with terminal MND. In addition, there are still sudden deaths and unclear causes of death among these patients. There is a wide variation in prevalence among different endemic studies (0.7% in France, 3.6% in People's Republic of China, and 12.6% in Italy). Our results indicated 11.8% among the 17 deceased patients. This phenomenon reflects underlying patient–physician controversies in acute ward care, and increasing difficulties in preparing for death in hospice care. Physicians and healthcare workers should keep this in mind and explain the terminal phase of patient care carefully to patients with MND and their families.

It is known that patients with MND incur high medical costs. In the United States, a systemic sampling study demonstrated higher inpatient costs for MNDs than for non-MND illnesses.²⁵ Another nationwide cohort study revealed an increasing trend of inpatient costs over 15 years.²⁶ A similar increasing trend of inpatient costs was found among patients with MND in Taiwan.¹¹ Compared with mean costs in Taiwan nationwide as shown in previous reports, our current results show much lower mean inpatient costs under hospice care. Furthermore, if we focused on 2005 data (DD2005.DAT) and conducted our calculations using previous epidemiologic results,¹¹ the mean inpatient costs were much lower in patients with MND under hospice care than those under acute ward care (73,734.5 vs. 121,914.0 NTD). Most patients with MND underwent acute ward care instead of hospice care (365 vs. 4 admission records in 2005). The phenomenon reflected the nonaggressive role of hospice care for patients with MND at that time, which might be viewed as preying preparation or removal of ventilation care. Under the fixed payment of NHI, hospice care has previously been shown to be cost saving in local terminal cancer patients.²⁷ Notwithstanding the aforementioned results and epidemiologic reports, the utilization of hospice care remains extremely low among patients with MND in Taiwan. Because of the symptomatic palliation it provides and considerations of cost-efficiency, there is room to promote hospice care for patients with terminal MND.

There are several inevitable limitations to this study. Descriptive statistics were performed instead of comparisons

among variables because of the limited number of patients. Registration bias could not be excluded by screening ICD-9 codes, especially for chronic comorbidities, which may be viewed as less important for claims than acute comorbidities. Psychological issues could not be approached by claim-based analyses. Additionally, epidemiologic data after 2005 are still unavailable in Taiwan. Issues concerning prescriptions were not investigated because of technical difficulties, such as measuring dosages and durations. It must be emphasized that this study is a preliminary investigation illustrating the characteristics of patients with MND under hospice care in Taiwan. Further in-depth investigations are needed.

In conclusion, this study presents an initial approach to the utilization of hospice care among patients with MND. In Taiwan, respiratory and feeding problems accounted for a large measure of the terminal care provided to these patients, and there is a basis for promoting noninvasive ventilation care and gastrostomy/jejunostomy as well. Additionally, family physicians played and continued to have important roles in hospice care for patients with MND.

Acknowledgments

The authors wish to thank the National Health Research Institute for permission and authorization to utilize the dataset. The first author especially thanks Ms. Tsai-Mei Chen for sharing her experiences. This study does not represent the official viewpoints of the National Health Research Institute, Republic of China (Taiwan).

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