Guidelines for the economic analysis of a telematic platform for Parkinson's disease monitoring

Jorge Cancela, Maria T. Arredondo and Olivia Hurtado

Abstract—PERFORM (A sophisticated multi-parametric system FOR the continuous effective assessment and monitoring of motor status in Parkinson's disease and other neurodegenerative diseases) is an European project which aim is to provide doctors with a telematic tool for the remote monitoring of Parkinson's disease (PD) patients at their homes. The technical performance of PERFORM has been addressed in previous publications. The aim of this work is to introduce the best well-known methodologies and indicators used in the health economics field, normally linked to the assessment of drugs or devices, and provide a set of guidelines for the use of these methodologies in the assessment of telehealth systems focused on PD patients. PD will also be introduced in economic terms, i.e. burden and cost of PD. This study will be done with special emphasis in European countries.

Index Terms—Parkinson's disease, health economics, telehealth, telemedicine

I. INTRODUCTION

HRONIC diseases are prolonged conditions that normally do not improve with time and are rarely cured completely. Parkinson's disease (PD) is one of the most common neurodegenerative disorders. It occurs in about 1% of the population over the age of 60 and its prevalence increases with age. Advancements in treatment for chronic diseases have resulted in reduced length of hospital stay, and in some cases, the avoidance of hospital visits. Telemedicine brings healthcare delivery to the home environment by connecting the patient with medical professionals. It is not intended to replace health professional care, but rather to enhance the level of care [1]. Nevertheless, there is a widespread perception that most telemedicine applications are not as widely used as might be expected. The limited availability of information on large-scale performance and economic impact of telemedicine might account for some of this perception [2].

The major motor disturbances in PD are bradykinesia (i.e. slowed movement), hypokinesia (small amplitude movements), resting tremor, rigidity and postural instability. These major motor features of PD are associated with, and are largely a result of, the loss of dopaminergic innervation of the basal ganglia. These motor changes in PD often restrict functional independence and are a major cause of morbidity and mortality among these patients [3–6]. PD is typically characterized by severe, unpredictable and abrupt changes in the patient motor performance whereby OFF periods, characterized by the temporary loss of drugs effectiveness, alternate, sometimes within minutes, with ON periods, during which the medication effectively attenuate motion symptoms.

II. THE PERFORM APPROACH

PERFORM platform was composed by a set of wearable sensors for the continuous recording of the motion signals and a set of software algorithms for the signal processing. The hardware was formed by a set of four tri-axial accelerometers positioned at each patient limb used to record signals from legs and arms; a belt sensor, composed by an accelerometer and a gyroscope, used to record body movement accelerations and angular rate; and a data logger used to receive and store all recorded signals in a SD card. All sensors transmit data using Zigbee protocol to the logger device, with 62.5 Hz sampling rate before a synchronization phase. The software sub-system is called Local Base Unit (LBU) and is responsible for the identification and quantification of the patient symptoms and the recording of other useful information for the evaluation of the patient status. For each symptom, a dedicated algorithm processes the relevant signals, detects the symptom episode and quantifies it into a severity scale from 0 to 4, according to the UPDRS scale for PD's patients. About the technical performance of the system it shows an accuracy of up to 93.73% of accuracy for the classification of levodopa induced diskynesias (LID) severity [7], an 86% for the classification of bradykinesia severity [8] and 87% for tremor severity [9]. Also, a specific module was developed for the assessment of gait [10].

III. PD ECONOMY AND HEALTH ECONOMICS

A. Introduction

Health economics is a branch of economics concerned with issues related to efficiency, effectiveness, value and behavior in the production and consumption of health and health care. Economic evaluation of telemedicine applications is required to provide decision makers in health care with appropriate information on costs and benefits of this information and communications technology [2]. Health

Manuscript received June 26, 2013.

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economics theories and methodologies are widely used in the healthcare for the evaluation of new interventions (i.e. new drugs, devices or procedures) in order to evaluate them and provide policy makers and program administrators with enough information to make decisions, nevertheless, economic evaluations of telemedicine remain rare [11]. Usually a telemedicine program evolves through a known number of stages [2]: a) Pilot or introductory phase, focus on demonstrate the feasibility of the system (limited numbers of patients); b) a routine telemedicine service and c) deploy of the telemedicine application across an entire health system or expanded network. Each of these stages in the life of a telemedicine program requires a specific economic assessment, in order to analyze its feasibility. The requirements change as the application matures, including changes in the focus of evaluation and the sources of information. There are four principal analytic techniques for economic evaluation [12]:

- Cost minimization analysis (CMA), this methodology is simply a comparison of costs of comparator therapies. CMA is only appropriate if the effects associated with all therapies are identical [13].
- 2) Cost effectiveness analysis (CEA). In most of the cases the clinical outcomes associated with treatments will differ and CMA would not be feasible. In a CEA, the cost effectiveness of a therapy is assessed in terms of the incremental cost per outcome gained [14].
- 3) Cost utility analysis (CUA) is a form of CEA which incorporates all effects of treatment (i.e. morbidity and mortality). Outcome is expressed as QALY, a standard unit for this kind of measures that will be detailed later. By adopting a common outcome measure, CUA allows the assessment of the cost effectiveness of therapy relative to treatments for other diseases [15], [16].
- 4) Cost-benefit analysis (CBA). It is the most comprehensive type of economic evaluation and allows the study of interventions with multiple outcomes [11]. However, there are measurements difficulties within CBA which has limited its adoption. Few telemedicine evaluations have conducted BCAs because this type of evaluation is data intensive and technically sophisticated [11]. A true CBA requires that all outcomes are expressed in monetary values [17].

PD therapies tend to impact the severity of the disease but not life expectancy. CUA is particularly pertinent for the evaluation of therapies where there is a considerable impact on the quality of life of patients through changes in the severity of disease. [12]. An analysis of the economic impact of home telehealth must include costs from both the patient and population levels [1].

B. Quality-Adjusted Life Year (QALY) vs. Daily Adjusted Live Years (DALY)

QALY is a measure including both the quality and the quantity of life lived. It is used in assessing the value for money of a medical intervention [18]. The QALY is based on the number of years of life that would be added by the intervention. Each year in perfect health is assigned the value of 1.0 down to a value of 0.0 for death. If the extra years would not be lived in full health, e.g. if the patient have to use a wheelchair, then the extra life-years are given a value between 0 and 1 to account for this. This utility score is obtained from a structured set of questions to patients in whom they rank different states of health.

DALY instead can be thought of as one year of healthy life lost, and the overall disease burden can be thought of as a measure of the gap between current health status and the ideal health status, where the individual lives to old age free from disease and disability [19]. DALY is the result of adding years of life lost (YLL) and years lived with disability (YLD). It is a composite measure of premature mortality and disability, equivalent to years of healthy life lost due to a given condition. YLL is direct related with the number of deaths (with PD as primary cause) and the standard life expectancy at each year of age. On the other hand, YLD is function of the population susceptible to PD at each age (a value between 0 - 1), the PD incidence at each age, the average duration of PD by age of onset (measured in years) and the disability weight (range 0-1) [20].

C. Discounting and age-weighting modulation

Any economic evaluation where costs and benefits occur over a number of years should consider discounting. Discounting adjusts for costs (and benefits) occurring at different points in time. There is a preference for benefits today rather than in the future. Additionally a euro today would be a higher value than one in the future. Therefore a time discounting rate should be applied [21]. Economic evaluation is concerned with the summation of costs and benefits over time, but, individuals and society tend to defer costs to the future than incur them now and individuals and society tend to prefer benefits now rather than in the future. A similar approach is used for age analysis. An ageweighting modulation factor (K) is usually applied. The ageweighting modulation factor quantifies the perception that a year of healthy life has greater social value during early adulthood (K = 1) than during earlier or later life (K = 0when healthy life values equally).

D. Burden of Parkinson's disease

Disease burden is the impact of a health problem on an area measured by financial cost, mortality, morbidity, or other indicators. It is often quantified in terms of DALYs, which quantify the number of years lost due to disease [19]. The burden of illness associated with PD is related not only to the disease itself, but also to the progressive disability that patients experience as their disease advances. Studies indicate that quality of life is affected not only by the motor symptoms of PD, including bradykinesia, tremor rigidity and degradation of gait, but also by depression and cognitive state [22–24]. Non-motor complications also increase over time and dementia, depression and other neuropsychiatric disorders are commonly reported comorbidities [25–27].

This decline in quality of life affects not only patients and caregivers, but also has been associated with increased economic of illness [28].

Based on the World Health Report 2001 on mental health [29], PD DALY contributed 0.1% of the Global Burden of Disease (GBD) in the world, 0.6% in the European A subregion (European countries with very low children and adult mortality according to WHO classification), and 0.5% of the GBD in Spain [20]. In accordance with such data for the European A subregion, PD represented approximately 1/200 of the GBD. In Spain, based on the extended work done by Cubo et al. [20], (discounting rate 3%, K=1), PD generated 67,582 DALY, comprising 6,351 (9.4%) YLL and 61,231 (90.6%) YLD. Most PD DALY (57.5%) occurred in the population 60 to 74 years of age. In order to calculate the YLD value PD was modeled as a progressive condition, with affected people passing through the three stages described in a previous study [30]. On the basis of that it was assumed that two thirds of total PD duration was spent in the mild stage, with a disability weight of 0.48 (0.31 - 0.64), and the rest in the intermediate and high end stages, with disability weight levels of 0.79 (0.72-0.85) and 0.92 (0.89-0.95), respectively.

E. Cost of Parkinson disease

Cost of illness, the cost of different diseases and where those costs occur, are complementary to burden of disease studies, and they are indispensable for policy makers [31]. A cost-of-illness analysis can be conducted from several different perspectives e.g. an individual hospital, insurance company or government [31]. The perspective chosen determines which costs are included in the analysis.

- 1) Direct healthcare costs, are costs for goods and services used in the prevention, diagnosis, treatment and rehabilitation of the illness, disease or disorder in question, e.g. costs for medical visits, hospitalization and pharmaceuticals. Direct non-medical costs include all other resource use related to a disease, for example transportation, social services, etc. [31].
- 2) Indirect costs are defined as the value of the output that is lost because people with a certain illness, disease or disorder are impaired and too ill to work, either shortterm or long-term [32]. Some examples are costs of loss of production due to short-term absence from work, early retirement or reduced productivity at work due to illness. There are also intangible costs, which include pain, psychosocial suffering, and changes in social functioning and activities of daily living. Intangible costs are in general not included in currently available cost of illness studies due to difficulties in quantifying these costs [31].

Direct healthcare cost due to brain disorders in Europe amounted to $\notin 135$ billion, corresponding to 35% of the total cost. The cost for hospital care is the dominating healthcare cost, reaching $\notin 78$ billion in 2004 (20% of the total cost and 57% of the healthcare cost). The cost of outpatient care amounted to a total of \notin 45 billion, making up 12% of the total cost of disorders of the brain. Drug cost totaled \notin 13 billion (3% of the total cost). The largest non-medical resource component was cost of social services due to brain disorders, amounting to \notin 52 billion (13% of total cost) also closely related with the informal care. It refers to the unpaid care provided by family members, friends or voluntary workers to disabled and impaired individuals in the community[33].

IV. DISCUSSION

Most studies agree that telemedicine reduces the use of health services such as physician office visits, emergency department visits, number of hospitalizations, hospital readmissions, home visits, length of hospital stay, use of ambulance services, number of referrals, duration of consultations, number of laboratory tests, and avoided transfers/evacuations [11]. Progression of the disease leads to higher costs, and there are substantial gains to be made from slowing down this progression [33]. To estimate the economic benefits of telemedicine programs, clinical and social outcomes must be translated into monetary values using reliable conversion factors. Monetary conversion factors should be as precise as possible in terms of the location and context of the program and, when available, more than one factor should be used for each outcome to allow for sensitivity analyses [11]. Dávalos et al. [11] propose a wide range of outcomes that can be monitoring to analyze the outcome of the telemedicine system and how to convert them into monetary units. Also it was already mentioned the model proposed by Stouthard et al. [30] and used by Cubo et al. [20] to estimate the burden of PD. Assessing how PERFORM or other telehealth platform impact on the disability levels is an excellent starting point to obtain the impact of the platform in the global burden of the PD. Also, some works have implemented models to evaluate the economic impact. Coyle et al. [12] propose an economic analysis of entacapone using a Markov model. Three health states were adopted to reflect disease progression depending on the time spending on Off state per day. The transition probability relation to the progression of disease from moderate to severe disease was estimated based in data obtained from a cross sectional observational study [27].

PERFORM system is focus on increasing the performance of the medication through a better and more objective monitoring of the patients' symptoms, reducing costs by utilizing communications technology, which eliminates the need for transportation, reduces the waiting and/or consultation time can be reduced via telemedicine consultations, transferring health knowledge transfer from the practitioner to the patient, ability to self-care, and medication adherence. Coyle's model together with Davalos's guidelines for monetary transformation will be the initial approach to estimate the impact on the costs related to PD. While Stouthard will be adapted in order to estimate the impact of PERFORM in the global burden of PD.

V.CONCLUSION

Telemedicine programs can reduce healthcare utilization through early detection of a worsening condition, timely treatment, and the avoided need for further tests. Nevertheless further proofs are required in order to provide policy makers and healthcare providers of the necessity of the deployment of telemedicine systems. A close collaboration with the health economics field is required in order to translate the already known methodologies for the economic evaluation of drugs and medical devices to the telehealth systems i.e. translate the system outcomes into monetary values both for the individual/patient, hospital/healthcare provider or societal/policy makers perspectives.

ACKNOWLEDGMENT

Research by Jorge Cancela has been partly supported by a PICATA predoctoral fellowship of the Moncloa Campus of International Excellence (UCM-UPM).

Part of this work was done while the first author was visiting the Program in Health Services and Systems Research, Duke-NUS Graduate Medical School Singapore supported by the Moncloa Campus of International Excellence (UCM-UPM).

This work is partially funded by the European Commission under the PERFORM project (7th Framework Programme, Grant Agreement 215952). The authors would like to thank the PERFORM consortium.

REFERENCES

- J. Polisena, D. Coyle, K. Coyle, and S. McGill, "Home telehealth for chronic disease management: a systematic review and an analysis of economic evaluations.," International Journal of Technology Assessment in Health Care, vol. 25, no. 3, pp. 339–349, 2009.
- [2] D. Hailey and P. Jennett, "The need for economic evaluation of telemedicine to evolve: the experience in Alberta, Canada.," Telemedicine journal and ehealth the official journal of the American Telemedicine Association, vol. 10, no. 1, pp. 71–76, 2004.
- [3] M. E. Morris, R. Iansek, T. A. Matyas, and J. J. Summers, "The pathogenesis of gait hypokinesia in Parkinson's disease.," Brain: A journal of neurology, vol. 117 (Pt 5, no. 5, pp. 1169–1181, 1994.
- [4] B. R. Bloem, J. M. Hausdorff, J. E. Visser, and N. Giladi, "Falls and freezing of gait in Parkinson's disease: a review of two interconnected, episodic phenomena.," Movement disorders official journal of the Movement Disorder Society, vol. 19, no. 8, pp. 871–884, 2004.
- [5] Y. Balash, C. Peretz, G. Leibovich, T. Herman, J. M. Hausdorff, and N. Giladi, "Falls in outpatients with Parkinson's disease: frequency, impact and identifying factors.," Journal of Neurology, vol. 251, no. 1, pp. 1310– 1315, 2004.
- [6] R. M. Pickering, Y. A. M. Grimbergen, U. Rigney, A. Ashburn, G. Mazibrada, B. Wood, P. Gray, G. Kerr, and B. R. Bloem, "A meta-analysis of six prospective studies of falling in Parkinson's disease.," Movement disorders official journal of the Movement Disorder Society, vol. 22, no. 13, pp. 1892–1900, 2007.
- [7] M. G. Tsipouras, A. T. Tzallas, G. Rigas, P. Bougia, D. I. Fotiadis, and S. Konitsiotis, "Automated Levodopa-induced dyskinesia assessment," Engineering in Medicine and Biology Society EMBC 2010 Annual International Conference of the IEEE, vol. 2010, pp. 2411–2414, 2010.
- [8] M. Pastorino, J. Cancela, M. T. Arredondo, M. Pansera, L. Pastor-Sanz, F. Villagra, M. A. Pastor, and J. A. Martin, Assessment of bradykinesia in Parkinson's disease patients through a multi-parametric system. IEEE, 2011, pp. 1810–1813.
- [9] G. Rigas, A. T. Tzallas, M. G. Tsipouras, P. Bougia, E. E. Tripoliti, D. Baga, D. I. Fotiadis, S. G. Tsouli, and S. Konitsiotis, "Assessment of Tremor Activity in the Parkinson's Disease Using a Set of Wearable Sensors," Information Technology in Biomedicine, IEEE Transactions on, vol. 16, no. 3, pp. 478–487, 2012.
- [10] J. Cancela, M. Pastorino, M. T. Arredondo, M. Pansera, L. Pastor-Sanz, F. Villagra, M. A. Pastor, and A. P. Gonzalez, Gait assessment in Parkinson's

disease patients through a network of wearable accelerometers in unsupervised environments. IEEE, 2011, pp. 2233–2236.

- [11] M. E. Dávalos, M. T. French, A. E. Burdick, and S. C. Simmons, "Economic evaluation of telemedicine: review of the literature and research guidelines for benefit--cost analysis," Telemedicine and e-Health, vol. 15, no. 10, pp. 933–948, 2009.
- [12] D. Coyle, M. Barbeau, M. Guttman, and J.-F. Baladi, "The economic evaluation of pharmacotherapies for Parkinson's disease.," Parkinsonism related disorders, vol. 9, no. 5, pp. 301–307, 2003.
- [13] R. Robinson, "Costs and cost-minimisation analysis.," BMJ British Medical Journal, vol. 307, no. 6906, pp. 726–728, 1993.
- [14] R. Robinson, "Cost-effectiveness analysis.," The Journal of family practice, vol. 44, no. 6, pp. 793–795, 1993.
- [15] A. Nicholls, "Cost-utility analysis.," BMJ British Medical Journal, vol. 307, no. 6908, pp. 859–862, 1993.
- [16] G. W. Torrance and D. Feeny, "Utilities and quality-adjusted life years.," International Journal of Technology Assessment in Health Care, vol. 5, no. 4, pp. 559–575, 1989.
- [17] R. Robinson, "Cost-benefit analysis.," BMJ British Medical Journal, vol. 307, no. 6909, pp. 924–926, 1993.
- [18] J. S. Pliskin, D. S. Shepard, and M. C. Weinstein, "Utility Functions for Life Years and Health Status," Operations Research, vol. 28, no. 1, pp. 206–224, 1980.
- [19] A. Pruess-Ustun and C. Corvalan, Preventing disease through healthy environments: Towards an estimate of the environmental burden of disease. SciELO Brasil, 2006.
- [20] E. Cubo, E. Alvarez, C. Morant, J. De Pedro Cuesta, P. Martínez Martín, R. Génova, and J. M. Freire, "Burden of disease related to Parkinson's disease in Spain in the year 2000.," Movement disorders official journal of the Movement Disorder Society, vol. 20, no. 11, pp. 1481–1487, 2005.
- [21] M. F. Drummond, B. J. O'Brien, G. W. Torrance, and G. L. Stoddart, Methods for the economic evaluation of health care programmes, vol. 3rd, no. 1. Oxford University Press, 1987, p. 379.
- [22] E. A. Chrischilles, L. M. Rubenstein, M. D. Voelker, R. B. Wallace, and R. L. Rodnitzky, "The health burdens of Parkinson's disease.," Movement disorders official journal of the Movement Disorder Society, vol. 13, no. 3, pp. 406–413, 1998.
- [23] A. Schrag, M. Jahanshahi, and N. Quinn, "What contributes to quality of life in patients with Parkinson's disease?," Journal of Neurology, Neurosurgery & Psychiatry, vol. 69, no. 3, pp. 289–290, 2000.
- [24] A. M. Kuopio, R. J. Marttila, H. Helenius, M. Toivonen, and U. K. Rinne, "Quality of life in Parkinson's disease," Nippon Rinsho, vol. 58, no. 2, pp. 17–21, 2000.
- [25] L. J. Findley, "The economic impact of Parkinson's disease.," Parkinsonism related disorders, vol. 13 Suppl, no. S1, pp. S8–S12, 2007.
- [26] D. M. Huse, K. Schulman, L. Orsini, J. Castelli-Haley, S. Kennedy, and G. Lenhart, "Burden of illness in Parkinson's disease.," Movement disorders official journal of the Movement Disorder Society, vol. 20, no. 11, pp. 1449–1454, 2005.
- [27] M. Guttman, P. M. Slaughter, M.-E. Theriault, D. P. DeBoer, and C. D. Naylor, "Burden of parkinsonism: a population-based study.," Movement disorders official journal of the Movement Disorder Society, vol. 18, no. 3, pp. 313–319, 2003.
- [28] T. Keränen, S. Kaakkola, K. Sotaniemi, V. Laulumaa, T. Haapaniemi, T. Jolma, H. Kola, A. Ylikoski, O. Satomaa, J. Kovanen, E. Taimela, H. Haapaniemi, H. Turunen, and A. Takala, "Economic burden and quality of life impairment increase with severity of PD.," Parkinsonism related disorders, vol. 9, no. 3, pp. 163–168, 2003.
- [29] B. Saraceno, "The WHO World Health Report 2001 on mental health.," Epidemiologia e Psichiatria Sociale, vol. 11, no. 2, pp. 83–87, 2002.
- [30] M. E. A. Stouthard, M. L. Essink-Bot, G. J. Bonsel, J. J. M. Barendregt, P. G. N. Kramers, H. P. A. de Water, L. J. Gunning-Schepers, and P. J. Van Der Maas, Disability weights for diseases in the Netherlands. Inst. Sociale Geneeskunde, 1997.
- [31] P. Andlin-Sobocki, B. Jönsson, H.-U. Wittchen, and J. Olesen, "Cost of disorders of the brain in Europe,," European journal of neurology the official journal of the European Federation of Neurological Societies, vol. 12 Suppl 1, no. 2. Blackwell, pp. 39–44, 2005.
- [32] B. R. Luce and A. Elixhauser, "Estimating costs in the economic evaluation of medical technologies.," International Journal of Technology Assessment in Health Care, vol. 6, no. 1, pp. 57–75, 1990.
- [33] P. Lindgren, S. Von Campenhausen, E. Spottke, U. Siebert, and R. Dodel, "Cost of Parkinson's disease in Europe.," European journal of neurology the official journal of the European Federation of Neurological Societies, vol. 12 Suppl 1, no. 4, pp. 357–365, 1991.
- [34] M. C. Sokol, K. A. McGuigan, R. R. Verbrugge, and R. S. Epstein, "Impact of medication adherence on hospitalization risk and healthcare cost.," Medical Care, vol. 43, no. 6, pp. 521–530, 2005.