Author: A'Campo, Laura Eva Ingeborg
Title: A patient and caregiver education program : in Parkinson's disease, Huntington's disease, and other chronic diseases
Issue Date: 2012-06-05

1

General introduction

The worldwide transition from acute disease towards chronic disease has created the challenge and need to develop new treatment approaches next to the technical biomedical treatment forms.[1-3] Patients have to adjust their activities of daily life, work and social participation to their decreased functional abilities. They also have to learn to cope emotionally with physical problems and disability. Chronic disease treatment strategies aim to improve adaptation and maximizing patients' quality of life (Qol). Quality of life refers to the patients' own perception and self-evaluation regarding the effects of an illness and its consequences on his or her life. In accordance with the biopsychosocial model,[4] Qol comprises physical, psychological and social wellbeing. Chronic disease has a negative impact on psychosocial wellbeing. Also, long-term psychosocial stress can have detrimental effects on the physical health.[5] Patients with a chronic disease are more at risk for psychiatric problems, mostly depression and anxiety disorders.[6] Furthermore, psychiatric disorders are a risk factor for non-compliance in medical treatment.[7] Most patients with chronic disease live in the community. Therefore, long-term responsibility of day-to-day management with the disease and its psychosocial consequences has become a new challenge for patients together with their informal caregivers.

Self-Management Interventions

To help patients and caregivers manage, Self-Management Interventions (SMIs) have been developed for many chronic diseases.[8] Their aim is to increase patients' and caregivers' involvement and control in treatment and its effects on their lives [8]. SMIs has been defined as: practicing the skills necessary to carry on an active and emotionally satisfying life in the face of a chronic illness, including making informed choices, adopting new perspectives and skills, practicing new health behaviours, and maintaining or regaining emotional stability.[9]

SMI is a common term in health education literature,[10] but other terms are used also, e.g. 'education programs', 'psychosocial interventions', or 'cognitive-behavioral programs'.[8,11-13] Examples of SMIs are The Arthritis Self-Management Program,[14] The Diabetes X-PERT Programme,[15] and the Chronic Disease Self-Management Program (CDSMP).[16] All SMIs have in common being relatively brief interventions (about 4-10 weeks) and often being provided in a group format. A group format has economic benefits and participants learn from each other as being fellow-sufferers (modelling). However, SMIs differ in specific contents of the programs [8]. Also, two applications of SMIs exist: 1) a disease-specific or 2) a generic approach. The former is the most often used SMI and has been described in the literature for several chronic diseases, mostly in arthritis, diabetes and asthma.[8] Some relatively rare diseases like Huntington's disease have never been assessed on the feasibility of SMI use. A generic SMI is based on the premise that a generic set of skills is helpful in coping with every chronic disease [8]. Studies showed that it is feasible and effective to place patients with different chronic diseases in the same program.[17]

The SMI development started with the recognition of the importance of information provision. In traditional patient education, disease-specific information was provided in the didactic format and with technical skills.[3,8] Because information proved to be important, but often not sufficient for behavior change and for improvement of psychosocial wellbeing, new strategies were developed with help of psychological theories.[1,8] A transition occurred from viewing the patient as a dependent and passive care recipient to an active autonomous patient with own responsibility and possibilities for self-management of the disease. This transition leads to a more collaborative care between patient and health care professionals. Therefore, patients have to learn problem-solving and helpful coping skills. Helpful coping can minimize emotional problems.[8,18] Important influences came from psychological theories, like the Transactional model from Lazarus and Folkman. This theory comprises that the way a person copes is not dependent on the situation per se but on how the situation is appraised.[8,19] Techniques from the cognitive-behavioral therapy (CBT) gave important treatment possibilities for SMIs. The use of CBT techniques in SMIs has proven to be especially effective.[8] They have benefits when treating mental health problems in patients with chronic disease and in caregivers.[8,20-26]

The Patient Education Program for Parkinson's disease

The Patient Education Program for Parkinson's disease (PEPP) was developed as a disease-specific SMI between 2003 and 2005 by a European consortium called EduPark. The consortium comprised researchers, professionals and lay organizations from seven countries (Estonia, Finland, Germany, Italy, The Netherlands, Spain and The United Kingdom). Also, patients with Parkinson's disease (PD) and caregivers contributed to the development. The EduPark consortium defined the program as: 'A systematic and professional approach to support patients and caregivers by teaching them knowledge and skills in order to improve their quality of life, complementing the medical treatment'. The aim of the program is to improve the coping skills of patients and caregivers to empower them in dealing with psychosocial stressors caused by PD. The key element of the program comprises techniques from the cognitive behavioral therapy, like cognitive restructuring, systematic relaxation training, situational behavioral analysis and training in social skills. The eight weekly sessions of ninety minutes each have a specific theme. An important benefit of this program for research is that the content is fully standardized and published in manuals in six languages, including English and Dutch.[27-32]

Participants of the program

The PEPP consists of two parallel programs for patients and caregivers, who both participate in groups of four to seven participants. An important characteristic of the PEPP is that in contrast with most SMIs, the caregiver receives an equal form of treatment. Caregivers are included in the PEPP program for several reasons. The (healthy) partner becomes the informal caregiver providing long-term daily care. Informal caregivers enable patients to live in the community and delay institutionalization.[33] Also, for the patient, social support within a spousal relationship is preventive for depression and the quality of a relationship has a positive influence on coping with PD.[34] However, caregiving has adverse effects on the health of the caregivers: they often experience long term strain including emotional, social and financial burden.[35-37] They have significantly higher rates of depressive (6.3 % vs. 4.2%) and anxiety (17.5% vs. 10.9%) disorders than non-caregivers and use health services for mental health problems nearly twice as frequent.[38]

The program was originally developed for Parkinson’s disease (PD), and thereafter adapted for another neurodegenerative disease, namely Huntington’s disease (HD), and eventually changed into a generic program for chronic disease.

Parkinson’s disease

PD is one of the most common neurodegenerative diseases; it has an incidence over the age of 65 of about 160 per 100.000.[39] Mean age of onset is between 55 and 75 years. The cause of PD is still unknown, but decreased dopamine levels are found due to gradual degeneration of dopamine-producing cells in the substantia nigra. The core features of the disease are a resting tremor, rigidity, bradykinesia/akinesia and postural disturbances. Other PD symptoms include speech disturbances, sleep problems, sexual dysfunction, autonomic dysfunction, cognitive deficits and neuropsychiatric problems. Cognitive impairments in PD are common, even in early stages of the disease.[40,41] About 40% will develop dementia.[42] Neuropsychiatric symptoms (as diagnosed with the DSM-IV) have been reported in 87% of PD patients (versus 31% in controls), including panic disorders (30%), major depression (21%), and dysthymia (19%).[43] All symptoms and signs have much impact on daily life activities[44] and often cause psychosocial stress in patients.[45,46] Psychosocial aspects comprise the psychological experience (thoughts and feelings) of living with PD and the impact on social functioning. Psychological experience may comprise feelings as anxiety, low mood, worthlessness, low-self-esteem, and loss of control and autonomy.[47] Social functioning may directly be hampered by the physical limitations and communication problems. Social anxiety is also common because of feelings of shame for visible signs as the tremor. Above this, psychosocial stress negatively influences the severity of signs (e.g. an increase of tremor). About half of the PD patients reported not being able to cope adequately with PD and one third stated that psychological support would be beneficial to them.[45] The use of new coping strategies may have a mediating function on QOL.[48] Psychosocial wellbeing has been found to be an important predictor of self-reported quality of life in PD patients.[49] Increased psychosocial problems and depressive symptoms are associated with more decline of Qol over time and are therefore often recommended to be included in PD treatment.[49-51]

Huntington's disease

Huntington's disease (HD) is a rare autosomal dominant inherited neurodegenerative disorder with a mean age of onset between the age of 30 and 50 years. Diagnosis is confirmed with DNA analysis. The mutation is an expanded trinucleotide (CAG) repeat in the HD (*HTT*) gene.[52] Its prevalence is 5-10 per 100,000 in the Caucasian population.[53,54] The disease is characterized by progressive motor, psychiatric and cognitive symptoms, causing functional decline.[53,54] Among the psychiatric symptoms, depression, anxiety, apathy and irritability have a prevalence rate varying across studies from 33% to 69%.[55] Many psychosocial stressors described in PD are also relevant in HD. However, some extra challenges due to the inherited nature of HD exist. The psychosocial stressors due to HD often begin in childhood, growing up in a family with HD with one of the parents being ill.[56] Often the care for the affected parent relies on the family, including the children. Furthermore a child with an affected parent has a 50% chance of carrying the HD gene themselves which often causes anxiety. With the discovery of the HD gene,[52] premanifest testing became available. Individuals now have the choice whether or not to be tested. A test result disclosing the HD gene mutation may cause anxiety for disease onset.[54,57] It also may influence important future planning issues, like reproductive decisions. In manifest HD, patients have reported to experience more burden by psychosocial consequences than physical aspects of HD.[58] This indicates the importance of attention for these psychosocial aspects of HD in treatment.

Patient Education Program for Parkinson's disease research thus far

The first studies assessed the Patient Education Program for Parkinson's disease (PEPP) in a formative evaluation. Macht et al. evaluated the participation of the patients in the seven countries and found significantly reduced psychosocial burden en need for help after the program.[59] Simons et al.[60] evaluated the data from the English patients and caregivers as part of the same uncontrolled study. They did not found any improvements on Qol, psychosocial problems and depression. The data of the complete caregiver group have not been assessed yet. In Finland, a non-randomized but controlled study has been performed with PD patients only.[13] Benefits were found for the PEPP participants regarding general Qol and social support. No randomized controlled trial had been performed yet to assess the effectiveness in PD patients and caregivers. Furthermore, the effectiveness of this particular program has not been assessed in other chronic diseases yet.

Aims and outline of this thesis

The general aim of this thesis was to assess the effectiveness of a standardized patient and caregiver education program, originally developed for Parkinson's disease. Then, the aim was to explore the feasibility of two adapted versions of the program: a disease-specific approach for another neurodegenerative disease, namely Huntington's disease, and a generic approach for chronic diseases with co-morbid psychiatric problems.

To assess if the psychosocial needs of PD patients are sufficiently met, a cross-sectional study was performed with data from medical records of PD patients who were attending an outpatient multidisciplinary assessment center. Patients' psychosocial burden and need for help were explored relative to their current use of psychosocial treatment and their actual request for help (chapter 2).

The Patient Education Program for Parkinson's disease (PEPP) was developed in seven European countries and was thereafter evaluated in a formative evaluation by means of a single group pre-post study design. We aimed to describe the results of the PD caregivers who participated in this study, next to the data of the PD patients which were published earlier (chapter 3).

Thereafter, a randomized controlled trial was performed with PD patients and caregivers. Participants allocated to the PEPP were compared with participants in a control group receiving usual care (chapter 4). More insight in characteristics of participants who benefit most from the program may help to improve referring advices and to achieve more (cost-) effectiveness. Therefore, we searched for treatment effect modifiers by means of secondary analyses of data from the randomized controlled trial (chapter 5).

Since the program is freely available in a manual and health care professionals are trained to provide the program in their own health care setting, we assessed the effectiveness of the program in the uncontrolled 'real world' clinical practice (chapter 6). Also, the effectiveness of the program in clinical practice at six-month follow-up was assessed.

We adapted the program for use in Huntington's disease (HD), another neurodegenerative disease. The program was used in manifest and premanifest HD stages and assessed on its feasibility (chapter 7). The study was followed by a six-month follow-up (chapter 8).

Finally, the program was extended for use in other chronic diseases: we changed the program from a disease-specific to a generic approach. We conducted a pilot study to assess the feasibility of the program when used in a heterogeneous group of patients with a chronic disease and co-morbid psychiatric problems (chapter 9).

The main conclusions of this thesis are discussed (chapter 10).

References

1. Deccache A, Aujoulat I. A European perspective: common developments, differences and challenges in patient education. Patient Educ Couns. 2001; 44:7-14
2. Fava GA, Sonino N. The biopsychosocial model thirty years later. Psychother Psychosom. 2008; 77:1-2
3. Bodenheimer T, Lorig,K, Holman, H et al. Patient self-management of chronic disease in primary care. JAMA. 2002; 288:2469-2475
4. Engel GL. The need for a new medical model: a challenge for biomedicine. Science. 1977; 196:129-136
5. Katon W, Ciechanowski P. Impact of major depression on chronic medical illness. J Psychosom Res. 2002; 53: 859-863
6. Polsky D, Doshi JA, Marcus S et al. Long-term risk for depressive symptoms after a medical diagnosis. Arch Intern Med. 2005; 165:1260-1266
7. DiMatteo MR, Lepper HS, Croghan TW. Depression is a risk factor for noncompliance with medical treatment: meta-analysis of the effects of anxiety and depression on patient adherence. Arch Intern Med. 2000; 160: 2101-2107
8. Newman S, Steed L, Mulligan K. Self-management interventions for chronic illness. Lancet. 2004; 364:1523-1537
9. Gruber R, Elman J, Huijbregts MPJ. Self-management Programs for People With Parkinson's Disease. Topics in Geriatric Rehabilitation. 2008; 24:141-150
10. Groessl EJ, Cronan TA. A cost analysis of self-management programs for people with chronic illness. Am J Community Psychol. 2000; 28:455-480
11. Cooper H, Booth K, Fear S et al. Chronic disease patient education: lessons from meta-analyses. Patient Educ Couns. 2001; 44:107-117
12. Schneider S. Moyer A, Knapp-Oliver S et al. Pre-intervention distress moderates the efficacy of psychosocial treatment for cancer patients: a meta-analysis. J Behav Med. 2010; 33:1-14
13. Tihonen S, Lankinen A, Viemero V. An evaluation of a Cognitive-Behavioral Patient Education Program for persons with Parkinson's Disease in Finland. Nordic Psychology. 2008; 60:316-331
14. Lorig KR. Arthritis self-management: a patient education program. Rehabil Nurs. 1982; 7: 16-20

15. Deakin TA, Cade JE, Williams R et al. Structured patient education: the diabetes X-PERT Programme makes a difference. Diabet Med. 2006; 23:944-954

16. Lorig KR, Sobel DS, Ritter PL et al. Effect of a self-management program on patients with chronic disease. Eff Clin Pract. 2001; 4:256-262

17. Lorig KR, Sobel DS, Stewart AL et al. Evidence suggesting that a chronic disease self-management program can improve health status while reducing hospitalization: a randomized trial. Med Care. 1999; 37:5-14

18. Bisschop MI, Kriegsman DM, Beekman AT et al. Chronic diseases and depression: the modifying role of psychosocial resources. Soc Sci Med. 2004; 59: 721-733

19. Folkman S, Lazarus RS. Coping as a mediator of emotion. J Pers Soc Psychol. 1988; 54:466-475

20. Secker DL, Brown RG. Cognitive behavioural therapy (CBT) for carers of patients with Parkinson's disease: a preliminary randomised controlled trial. J Neurol Neurosurg Psychiatry. 2005; 76:491-497

21. Cole K, Vaughan FL. The feasibility of using cognitive behaviour therapy for depression associated with Parkinson's disease: a literature review. Parkinsonism Relat Disord. 2005; 11:269-276

22. Mohlman J, Reel DH, Chazin D et al. A Novel Approach to Treating Anxiety and Enhancing Executive Skills in an Older Adult with Parkinson's Disease. Clin Case Stud. 2010; 9:74-90

23. Macht M, Smith Pasqualini MC, Taba P. Cognitive-behavioral Strategies for Parkinson's Disease: A Report of Three Cases. J Clin Psychol Med Settings. 2007; 14:165-176

24. Dobkin RD, Allen LA, Menza M. Cognitive-behavioral therapy for depression in Parkinson's disease: a pilot study. Mov Disord. 2007; 22:946-952

25. Farabaugh A, Locascio JJ, Yap L et al. Cognitive-behavioral therapy for patients with Parkinson's disease and comorbid major depressive disorder. Psychosomatics. 2010; 51:124-129

26. Butler AC, Chapman JE, Forman, EM et al. The empirical status of cognitive-behavioral therapy: a review of meta-analyses. Clin Psychol Rev. 2006; 26:17-31

27. Spliethoff-Kamminga NGA. Patiënt Educatie Programma Parkinson. Harcourt Publishers, Amsterdam, 2006

28. Smith Pasqualini MC, Simons G. Patient education for people with Parkinson's disease and their carers: a manual. John Wiley & Sons, Chichester, 2006
29. Ellgring H, Gerlich C, Macht M et al. Psychosoziales Training bei neurologischen Erkrangkungen-Schwerpunkt Parkinson. W. Kohlhammer, Stuttgart, 2006
30. Lankinen A, Viemero V, Lehtonen A et al. Psykososiaalinen valmennusohjelma Parkinsonin tautia sairastaville ja heidän Iäheisilleen. Suomen Parkinson-liitto ry, Turku, 2005
31. Ricci Bitti PE, Candini L, Carlini M et al. Intervento psicoeducativo nella malattia di Parkinson. Erickson, Gardolo, 2006
32. Taba P, Krikmann U, Kanarik E et al. Koolitusprogramm Parkinsoni tovega inimestele ja nende tugiisikutele. Tartu Parkinsoni Haiguise Selts, Tartu, 2005
33. Martinez-Martin P. Benito-Leon J, Alonso F et al. Quality of life of caregivers in Parkinson's disease. Qual Life Res. 2005; 14:463-472
34. Seiler S, Perleth B, Ulm G et al. Partnership and depression in Parkinson's disease. Behavioural Neurology. 1992; 5:75-81
35. Roland KP, Jenkins ME, Johnson AM. An exploration of the burden experienced by spousal caregivers of individuals with Parkinson's disease. Mov Disord. 2010; 25;189-193
36. Aarsland D, Larsen JP, Karlsen K et al. Mental symptoms in Parkinson's disease are important contributors to caregiver distress. Int J Geriatr Psychiatry. 1999; 14:866-874
37. Thommessen B, Aarsland D, Braekhus A et al. The psychosocial burden on spouses of the elderly with stroke, dementia and Parkinson's disease. Int J Geriatr Psychiatry. 2002; 17:78-84
38. Cochrane JJ, Goering PN, Rogers JM. The mental health of informal caregivers in Ontario: an epidemiological survey. Am J Public Health. 1997; 87:2002-2007
39. De Rijk MC, Tzourio C, Breteleret MM et al. Prevalence of parkinsonism and Parkinson's disease in Europe: the EUROPARKINSON Collaborative Study. European Community Concerted Action on the Epidemiology of Parkinson's disease. J Neurol Neurosurg Psychiatry. 1997; 62:10-15
40. Brown RG, Marsden CD. Cognitive function in Parkinson's disease: from description to theory. Trends Neurosci. 1990; 13; 21-29

41. Foltynie T, Brayne CE, Robbins TW et al. The cognitive ability of an incident cohort of Parkinson's patients in the UK. The CamPaIGN study. Brain. 2004; 127: 550-560

42. Cummings JL. Intellectual impairment in Parkinson's disease: clinical, pathologic, and biochemical correlates. J Geriatr Psychiatry Neurol. 1988; 1:24-36

43. Nuti A, Ceravolo R, Piccinniet A et al. Psychiatric comorbidity in a population of Parkinson's disease patients. Eur J Neurol. 2004; 11:315-320

44. Jankovic J. Parkinson's disease: clinical features and diagnosis. J Neurol Neurosurg. Psychiatry. 2008; 79:368-376

45. Macht M, Schwarz R, Ellgring H. Patterns of psychological problems in Parkinson's disease. Acta Neurol Scand. 2005; 111:95-101

46. Ellgring H. Seiler S, Perlethet B al. Psychosocial aspects of Parkinson's disease. Neurology. 1993; 43:S41-S44

47. Groves MS, Forrest DV. Parkinson's disease as a model for psychosocial issues in chronic neurodegenerative disease. Adv Neurol. 2005; 96:65-83

48. Frazier LD. Coping with disease-related stressors in Parkinson's disease. Gerontologist. 2000; 40:53-63

49. Visser M. Van Rooden SM, Verbaan D et al. A comprehensive model of health-related quality of life in Parkinson's disease. J Neurol. 2008; 255:1580-1587

50. Visser M, Verbaan D, Van Rooden SM et al. A Longitudinal Evaluation of Health-Related Quality of Life of Patients with Parkinson's Disease. Value Health. 2008; 12:392-396

51. Forsaa EB, Larsen JP, Wentzel-Larsen T et al. Predictors and course of health-related quality of life in Parkinson's disease. Mov Disord. 2008; 23:1420-1427

52. The Huntington's Disease Collaborative Research Group. A novel gene containing a trinucleotide repeat that is expanded and unstable on Huntington's disease chromosomes. Cell. 1993; 72:971-983

53. Bates G, Harper PS, Jones L. Huntington's disease. University Press, Oxford, 2002

54. Roos, RA. Huntington's disease: a clinical review. Orphanet J Rare Dis. 2010; 5:40

55. Van Duijn E, Kingma EM, Van der Mast RC. Psychopathology in verified Huntington's disease gene carriers. J Neuropsychiatry Clin Neurosci. 2007; 19: 441-448

56. Van der Meer L, Timman R, Trijsburg W et al. Attachment in families with Huntington's disease. A paradigm in clinical genetics. Patient Educ Couns. 2006; 63:246-254

57. Timman R, Roos R, Maat-Kievit A et al. Adverse effects of predictive testing for Huntington disease underestimated: long-term effects 7-10 years after the test. Health Psychol. 2004; 23:189-197

58. Helder DI, Kaptein AA, Van Kempen GM et al. Impact of Huntington's disease on quality of life. Mov Disord. 2001; 16:325-330

59. Macht M. Gerlich C, Ellgring H et al. Patient education in Parkinson's disease: Formative evaluation of a standardized programme in seven European countries. Patient Educ Couns. 2007; 65:245-252

60. Simons G, Thompson SB, Smith Pasqualini MC. An innovative education programme for people with Parkinson's disease and their carers. Parkinsonism Relat Disord. 2006; 12:478-485