

# Neuropalliative Care in Patients with Progressive Neurological Disease in Czech Republic: Study Protocol

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**Abstract**—Introduction: Currently, there has been an increasing concern about the provision of palliative care in non-oncological patients in both professional literature and clinical practice. However, there is not much scientific information on how to provide neurological and palliative care together. The main objective of the project is to create and to verify a concept of neuro-palliative and rehabilitative care for patients with selected neurological diseases in an advanced stage of the disease and also to evaluate biopsychosocial and spiritual needs of these patients and their caregivers related to the quality of life using created standardized tools. Methodology: Triangulation of research methods (qualitative and quantitative) will be used. A concept of care and assessment tools will be developed by analyzing interviews and focus groups. Qualitative data will be analyzed using grounded theory. The concept of care will be tested in the context of the intervention study. Using quantitative analysis, we will assess the effect of an intervention provided on the saturation of needs, quality of life, and quality of care. A research sample will be made up of the patients with selected neurological diseases (Parkinson's syndrome, motor neuron disease, multiple sclerosis, Huntington's disease), together with patients' family members. Based on the results, educational materials and a certified course for health care professionals will be created. Findings: Based on qualitative data analysis, we will propose the concept of integrated care model combining neurological, rehabilitative and specialist palliative care for patients with selected neurological diseases in different settings of care and services. Patients' needs related to quality of life will be described by newly created and validated measuring tools before the start of intervention (application of neuro-palliative and palliative approach) and then in the time interval. Conclusion: Based on the results, educational materials and a certified course for doctors and health care professionals will be created.

**Keywords**—Multidisciplinary approach, neuropalliative care, research, quality of life.

## I. INTRODUCTION

**S**ELECTED progressive neurological diseases (e.g. multiple sclerosis, neurodegenerative diseases, and motor neuron disease) result in a patient's severe disability and in many cases also in their death. It includes a heterogeneous group of patients with different dynamics of progressive deterioration. Advanced stages of the disease may last for several months or even years. Chronic, ever-growing movement handicap brings a patient and their family a lot of health as well as psychosocial problems, and requires multidisciplinary care and cooperation. Patients with

progressive neurological disease belong to the group of patients for whom palliative care may be indicated [1].

### A. Multiple Sclerosis

According to WHO, multiple sclerosis is one of the most common neurological disorders and causes of disability in young adults (20-30 years). The median of presumed prevalence of MS (worldwide) is 30 per 100,000 individuals, with the highest incidence in Europe (80 per 100,000). The Czech Republic is one of the countries with the highest prevalence, affecting 130 per 100,000 inhabitants [2]. The patients reach the stage of secondary chronic progression with already developed neurological disability after 10-15 years. Approximately 3% of patients show a relapsing-progressive course, with a very rapid disability and progression [3].

### B. Neurodegenerative Brain Disease

Neurodegenerative brain disease also belongs to the neurological diseases with a progressive course without the possibility of an effective therapeutic management. The pathological action with a differently rapid progression results in a characteristic syndrome which usually combines cognitive impairment with movement and behavioral disorders. Cognitive impairment may also occur in an early stage of the disease [4]. Typical examples of neurodegenerative diseases are different forms of dementia or Parkinson's disease with an incidence in Europe and the USA from 10 to 20 cases per year per 100,000 individuals. The average age of onset of the disease is about 60 years. Since the late sixties of the 20<sup>th</sup> century mortality rate dropped significantly and the median survival time has been extended to 14 years [3]. Advanced stages of neurodegenerative diseases are characterized by complex symptoms and care needs [4].

### C. Motor Neuron Disease

Motor neuron disease involves several forms of the disease; the most common is amyotrophic lateral sclerosis (ALS), with an incidence of 2 per 100,000 individuals [5]. A common feature for the motor neuron disease is a relatively fast progression of weakness, which, in a few years, or even months, results in a patient's full dependency on other people's care, possibly in the application of nutritional probes and artificial pulmonary ventilation. This is a typical disease requiring palliative care [5]. According to some epidemiological studies, the mean duration of ALS is 2-4.3 years. Around 25% of patients will survive 5 years and 8-16% will live for 10 years. The rate of progression is very variable

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[3], in the final stage shortness of breath occurs [6]. Mental function remains intact in most of the patients.

#### *D. The Progression of Diseases*

The progression of these neurological diseases may be characterized in four phases: 1<sup>st</sup> phase: a diagnostic phase, 2<sup>nd</sup> phase: a maintenance phase focused on the prevention of complications, 3<sup>rd</sup> phase: complex phase focused on the optimal management of medication and 4<sup>th</sup> phase: a palliative phase focused on relief of symptoms and distress [7]. Significant factors that improve the patients' quality of life include timely medical care, supportive rehabilitative care and individualization of care that promote quality of life [2]. The quality of life of these patients and their families should be maintained also in an advanced stage of the disease. Patients who have truthful information about their disease and its progression at an early stage of the disease can plan for future care, discuss appropriate support in an advanced and terminal stage of the disease, including endotracheal intubation and the introduction of artificial pulmonary ventilation with regard to their own and their loved ones' preferences and values. Planning for future care is very important at a time when the patient is still able to communicate their wishes and preferences. The family and the patient may find useful to discuss with health professionals the issues such as a patient's admission to hospital, resuscitation issues in case of the deterioration of a patient's health condition, the introduction of artificial nutrition and hydration, or their preferred place of death [4].

#### *E. Palliative Care in Neurology*

The aim of palliative care is to provide a physical, psychosocial, emotional and spiritual care for patients with a progressive and serious disease and for their family members [8]. The main objective of specialist palliative care is to alleviate symptoms, prevent complications, minimize distress, preserve human dignity and offer counseling. Some research shows that patients with progressive neurological diseases have unmet physical and psychosocial needs [8]. Unmet needs have been identified also in their family members [9]. Family caregivers perceive low social support, especially in malignant forms of MS and in the advanced stages of the disease [10], even though training materials for family caregivers have been created. A lot of patients with progressive long-term neurological conditions and their caregivers could benefit from palliative care services. There is not enough scientific evidence for combining these two concepts of care [8].

Some studies prove the possibility of combining rehabilitative care with neurological and palliative care to optimize the management of care for patients in the advanced stage of chronic neurological disease [11], [12]. Sutton [13] also states that the basis of good care is combining care of a specialist neurologist, rehabilitation and palliative care services for the whole duration of the disease until the patient's end of life. In this approach, neurological care focuses on the diagnosis, examination and treatment, while palliative care supports end-of-life decisions, care planning

and the management of symptoms causing distress. Rehabilitative care includes ongoing treatment and disability management, the coordination of multidisciplinary team interventions, assistance and provision of necessary equipment [12]. The emphasis is put on support for both the patient and their family [11]. Specialist palliative care may be available at any stage after the diagnosis has been made, not just at the end of life. The patient may be referred to specialist palliative care team with unmanageable symptoms, distress, management of complex needs, difficulties in care coordination, respite care, end of life decisions and planning.

In the terminal stages of progressive neurological diseases two main problems usually have to be solved – to provide sufficient and comfortable nutrition and to prevent a patient from choking. Weakness, immobility, self-insufficiency, anxiety, depression, dysarthria, pain, spasms and spasticity have to be also addressed [5].

The concept of neuropalliative and rehabilitative care has not yet been described in the Czech literature and professional practice. More attention is paid to palliative care issues for patients with oncological diseases, where it is easier to distinguish a curative and palliative treatment phase. Patients with progressive neurological conditions belong to a group of patients who are not dying but are facing a life-threatening disease and need high-quality symptom management, appropriate communication promoting and facilitating decisions about further treatment and care and an assistance with coordination of the provision of care in different care settings before reaching the terminal stage of the disease [14]. Medical specialists are not often sufficiently educated in palliative medicine and they do not pay enough attention to pain issues and other difficult symptoms [15]. A specific method of identifying patients with non-oncological diseases who would be eligible for some form of palliative care is not available at the moment.

## II. METHODOLOGY

The main priority of the project is to ensure the highest possible quality of life for individuals suffering from diseases of the nervous system through follow-up continuous neuro-rehabilitative, psychosocial, educational, and modern community social care linked with a palliative approach in an advanced stage of the disease.

#### *A. Aims of Project*

The main objective of the project is further elaborated in partial aims:

1. To find out the current state of the provision of health and social care to patients with selected neurological diseases and of the support given to their carers from the beginning of diagnosis until death and to identify unmet care needs.
2. To create the concept of an integrated care model combining neurological, rehabilitative and specialist palliative care for patients with selected neurological diseases in different health and social care settings.
3. To process a literary review of current instruments evaluating needs, quality of care and quality of life of

patients with selected neurological diseases in an advanced stage of the disease and their caregivers.

4. To adapt existing tools or to create our own tools for identifying needs related to patients' and family members' quality of life in order to provide individualized care focused on promoting the quality of life of patients and their caregivers. Further, to develop an assessment tool for identifying patients with selected neurological diseases who would be eligible for early initiation of specialist palliative care to improve their quality of life. To evaluate a patient's health condition in an advanced stage of the disease according to the ICF - International Classification, functional abilities and health.
5. To evaluate psychometric properties of created questionnaires and to standardize them for use in the Czech Republic.
6. To establish the level of saturation of bio-psychosocial and spiritual needs of patients and their caregivers related to the quality of life in different care settings before and after the intervention (neuro-palliative and rehabilitative approach). To compare the quality of care from the perspective of patients or family members in the intervention and control groups. To determine other potential factors that could affect the saturation of needs.
7. To create educational materials for healthcare professionals, patients and family members. To create a certified training course for medical and paramedical professions focused on the issue of neuro-palliative and rehabilitative care.

### *B. Design*

The project objectives will be met by triangulation of research methods, i.e. by interconnecting of methods of qualitative and quantitative research and by provision of literature search. They will be implemented in four main stages.

Qualitative research will be used for identification of unmet needs of patients with selected neurological diseases and their family members. Data collection method will be in-depth dialogues and focus groups. Focus groups and in-depth dialogues belong to main methods of qualitative research providing qualitative information acquired through open questions. The principle of focus groups method is a controlled discussion managed by a trained moderator whose task is to conduct the discussion on selected topic among approximately 7 – 8 people. The aim is to explore the attitudes, views and opinions of focus group. This method is suitable for investigating new and still unmapped areas, for evaluation planning and needs analysis. Questions for a target group are asked in a way that it is possible to identify important topics based on the answers of the group or to detect their opinions and attitude towards certain phenomenon [16], in this case towards provided health and social care and unmet needs of the patients and also their caregivers.

### *C. Sample*

The sample will be made up of the patients with selected

neurological diseases such as multiple sclerosis, selected neurodegenerative diseases (e.g. Parkinson's syndrome - multiple system atrophy, progressive supranuclear palsy, corticobasal degeneration, dementia with Lewy bodies) and selected motor neuron disease (e.g. amyotrophic lateral sclerosis, progressive bulbar palsy, progressive muscular dystrophy). Criteria for inclusion of patients in the sample will be: selected neurological disease in an advanced stage of the disease, age > 18 years, informed consent. Family members of the patients will be also included in the sample. Criteria for inclusion will be: 18 years or older, signed informed consent, a person giving a patient the main support.

Patients and family members will be selected from various care settings - hospitals (University Hospital Ostrava – Neurological clinic - inpatient and outpatient departments); follow-up care (follow-up care department of Bílovecká hospital, a.s, LDN Radvanice, LDN Klokočov); private neurological clinics; Hospic sv. Lukáše; Mobile hospice Ondrášek; social service facilities (Radvanice and Hrabyně, respite services, homes for the elderly). These will be patients who will be in the period of implementation of data collection (2017 - 2019) hospitalized at the Neurological clinic of the University Hospital Ostrava or registered at a surgery of this clinic. They will sign an informed consent form for inclusion in the study during the first contact at the University Hospital Ostrava. We assume that min. 100 patients and 100 family members will participate.

### *D. Data Collection*

Questionnaires and scales for evaluation of needs and patient's quality of life at the end stage of disease and their family members will be searched in electronic databases: MEDLINE, Scopus, Proquest Public Health, Web of Knowledge, CINAHL and EMBASE (a period 1985-2017) according to defined key words in English. Evaluation will include systematic reviews and empiric studies stating development of evaluation tools for measuring needs or patient's quality of life at the end stage of disease and their family members, providing at least two psycho-metric attributes from following: validity (sensitivity, specificity), discrimination validity, validity of evaluated criteria, reliability, sensitivity, factorial analysis and consistency.

The creation of assessment tool for evaluation of needs and quality of life will be based on literature search and qualitative analysis. Focus group method will also be used in order to obtain the material to formulate the items. Proposal of particular items of the questionnaire and scale of answers for a certain area will be created based on focus group discussion. A method of expert consensus (Delphi method) will then be used, when the questionnaire will be sent to selected specialists from the practice in order to obtain their opinion on questions created. Their comments will be worked into the final version of the questionnaires. Number of tools created in individual fields of needs (bio-psycho-social and spiritual), for patients with cognitive and non-cognitive disorder, in different care settings will be specified in focus groups. Patients in advanced stage of neurological disease will be evaluated also

according to ICF – international classification of functional ability, disability and health, based on bio-psycho-social model of disease conception. ICF is detailed classification including over 1 400 codes divided into three domains (body functions, activity and participation, environmental factors). It provides the most extensive language for the description of disability and other related factors [20].

#### *E. Data Analysis*

Qualitative data analysis will be executed by grounded theory method developed by Glaser and Strauss [17]. This approach was modified by Straus and Corbin [18]. Analysis will lead to identification of main category of the researched material being a centre of hierarchical category network. Identification and refinement of categories will be done in several intersecting phases during repeated processing of analysed material (theoretical coding). The aim of the method is a theory proposal (theory of needs and consequent creation of neuro-palliative and rehabilitative approach), which will include formulation of categories and their mutual relationship. Revealing data for interpretation, conceptualisation and new integration will be done by open, axial and selective coding [19].

Quantitative data will be described using basic descriptive statistics (arithmetic mean, median, standard deviation, and frequency tables). Confirmation factor analysis (maximum likelihood method), exploration factor analysis (method of the main components), inner rate consistency (Cronbach coefficient alpha), test retest reliability, construct validity will be used for the evaluation of psychometric attributes of questionnaires created. Parametric and non-parametric test for paired data will be used for the evaluation of intervention effect. Methods of multidimensional analysis which take in the account time effect and other factors influencing the intervention result will further be used. Program Stata, v. 10. and SPSS program, v. 21. will be used for data analysis.

### III. RESULTS

#### *A. The Description of Individual Phases*

In the first phase, we will assess the current situation and identify unmet needs and on their basis a concept of integrated care model will be created. In this part available statistical data on the provision of care to patients with selected neurological diseases will be analyzed. We will use the methods of qualitative research, such as in-depth interviews and focus groups. Selected healthcare professionals (doctors, nurses, and physiotherapists), social workers, selected patients with neurological diseases and their caregivers will participate in the qualitative research. In focus groups, ideas of interested parties on an integrated care model will be discussed, including the inclusion of specialist palliative care and planning end of life care, and identifying unmet needs of patients and their families in the current model of provision of health and social care. Qualitative data will be analyzed using a method of grounded theory.

In the second phase, we will develop and validate

assessment tools for patients in individual stages of the disease and for their caregivers based on a systematic approach (analysis of theoretical starting points, measurement tools and focus groups). In the first stage, we will conceptualize and operationalize the components of individual needs reflecting the specifics of the target population. The second stage will deal with, on the basis of qualitative methodology (Delphi method, controlled focus groups), the proposal of the domains and individual items of a pilot version of the questionnaires followed by the verification of content validity. Emphasis will be put on creating assessment tools for evaluation of the specific needs of patients with progressive neurological disease (dyspnoea, pain, nutrition, fatigue, immobility, self-insufficiency, anxiety, depression, dysarthria, and assessment of ethical aspects of care, psychosocial and spiritual needs).

A pilot version of the tools created will be tested on a group of patients with selected neurological diseases in an advanced stage of the disease in the care setting and of their caregivers. Patients will be divided into two groups. The first group will be made up of the patients who will be able, with regard to their health status, to participate in completing the questionnaire. In the second group of patients, the evaluation will be carried out by another person (a healthcare professional or a family member).

For evaluating the psychometric properties of the questionnaire, we will use the selected methods recommended for measuring the psychometric properties of the questionnaires used in health care, namely content validity, construct validity and reliability [21].

In the third phase, within an intervention study, we will evaluate the difference in a saturation of needs and quality of care in patients who have received care in the intervention group as part of an integrated concept of neuro-palliative and rehabilitative care compared with a control group of patients with standard care. The concept will be also evaluated by qualitative approach (a grounded theory method). The concept of neuro-palliative and rehabilitative care will be implemented within an intervention study in selected inpatient and outpatient health care facilities. Patients in an advanced stage of the disease will be also evaluated according to the International Classification of Functioning, Disability and Health.

In the fourth phase, a certified educational course will be created with the aim of training doctors and paramedical staff about the possibilities of neuro-palliative and rehabilitative care for patients with advanced progressive neurological disease. Educational materials for doctors, patients and family members will be also created.

### IV. CONCLUSION

We expect to write three publications in the journals with an IF, six publications in the journals in the Scopus database and writing a monograph.

Other project outputs will be a proposal of a neuropalliative and rehabilitative care concept for patients with selected progressive neurological diseases; educational materials for doctors, health care workers and family members; a certified

course for medical and paramedical professions with focus on the use of a neuropalliative and rehabilitative approach in clinical practice.

#### ACKNOWLEDGMENT

Supported by project "A neuropalliative rehabilitation approach to preserve the quality of life in patients with an advanced stage of selected neurological diseases" (AZV MZ ČR 17-29447A).

**Radka Bužgová** has long been interested in needs analysis, measuring instrument standardization, assessment of quality of life and anxiety/depression related to treatment and illness, particularly in adult patients and in palliative care. She is the main author of two the Czech Ministry of Health grants IGA MZ ČR NT 13417-4/2012 (palliative care issues), AZV MZ ČR 17-29447A (neuropalliative care) and a co-author of the Czech Technology Agency project TB02MPSV015 (the issues of health and social care in the elderly). Her articles have been published in journals with impact factors. She is a member of a multidisciplinary team in a mobile hospice Ondrášek in Ostrava.

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