

Silver Linings: The Experience of Hope in Dyads with Motor Neuron Disease. An Explorative Study

Sandra De Morée^{1,2*}, Ellen M Smets², Leonie NC Visser², Reinout O Van Vliet³, Esther T Kruitwagen-van Reenen⁴, Hepke F Grupstra¹, Sotice Pieters⁵, Marianne De Visser⁶, Hanneke C De Haes², Frans Nollet¹ and Anita Beelen⁴

1. Department of Rehabilitation, Amsterdam University Medical Centers, AMC (Academic Medical Center), The Netherlands

2. Department of Medical Psychology, Amsterdam University Medical Centers, AMC (Academic Medical Center), The Netherlands

3. Roessingh Centre for Rehabilitation, Enschede, The Netherlands

4. Department of Neuroscience, University Medical Centre Utrecht, Utrecht, The Netherlands

5. Sophia Rehabilitation Centre, The Hague, The Netherlands

6. Department of Neurology, Amsterdam University Medical Centers, AMC (Academic Medical Center), The Netherlands

Introduction

Amyotrophic lateral sclerosis (ALS) and progressive muscular atrophy (AMP), collectively known as motor neuron disease (MND), are rapidly evolving fatal neurodegenerative diseases. There is no proven curative treatment. MND causes progressive muscle weakness and stiffness, limited mobility, slurred speech, difficulty swallowing and possibly respiratory problems. Most patients with MND die from respiratory failure. The median survival time from the onset of symptoms is three years. Being diagnosed with MND naturally leads to an existential shock in patients and their caregivers, forcing them to reassess their lives. Psychological reactions to the diagnosis can range from heartache and anger to the desire for hasty death. To date, the results on the prevalence of psychological problems are inconsistent, but it would seem safe to conclude that only a minority of patients suffer from clinical depression and anxiety, and that most people with MND while having problems end of life find a way to cope with the diagnosis and demonstrate psychological resilience. In this context, the feeling of hope can play a role, as shown by patients with other terminal illnesses. Herth defined hope as "an inner power which facilitates the transcendence of the current situation and the movement towards a new consciousness and an enrichment of being". In fact, hope has been identified as a central need for terminally ill patients and their caregivers. To date, research in MND on the theme of hope is mostly limited to the perspective of patients. Some studies on hope in MND have been conducted, confirming the relevance of the concept and the need for healthcare professionals to take the time to explore approaches that promote hope in their patients. Deepening our understanding of hope in dyads with MND could help enrich communication between patients, caregivers and professionals, and help improve supportive care as in other terminal illnesses.

The purpose of this qualitative study is to explore if and how hope is felt in the dyads of MND patients and their caregivers at different stages of the disease.

Methods

Participants

MND patients attending four large specialized MND multidisciplinary care teams in the Netherlands were approached to participate in this study. Patients inclusion criteria were:-

- Having a diagnosis of ALS or PMA according to El Escorial criteria [19];
- Being able to communicate in Dutch. An associated diagnosis of Fronto-Temporal Dementia (FTD) was an exclusion criterion. Patients were sampled purposively to capture diversity in: gender, age, diagnosis, first symptoms, marital situation, religion and cultural background. Additionally, patients in different illness phases were selected;
- 'early MND', from four weeks after diagnosis up till one year in MND care;
- 'later MND', defined as more than one year after onset of MND care;
- 'end of life', as determined by the physician of the MND care team and generally considered to be the last months of life. Patients invited an informal caregiver of their choice to participate in the interviews. Inclusion criteria for the caregivers were
 - 18 years or older;
 - Able to communicate in Dutch, and
 - Invited by the recruited patient.
- The appointed caregivers all appeared to be Family Caregivers (FCs). Recruitment of patients continued until data saturation was reached i.e. no new themes emerged from the interviews.
- Twenty-eight eligible patients were invited to participate. Of these, 2 considered themselves too emotionally unstable to participate, 2 experienced severe physical deterioration, and 2 FCs were not willing to participate, leaving 22 participating dyads in the study. The Medical Ethical Review Committees of the 4 participating centers approved the study. Written informed consent was obtained from all participants.