The impact of oropharyngeal dysphagia and dysphonia on health-related quality of life in Parkinson's disease

Summary

The research presented in this thesis discusses the impact of oropharyngeal dysphagia (OD) and dysphonia on the quality of life of patients with idiopathic Parkinson's disease (IPD). On the one hand, we evaluate the importance of patient-reported outcome measures (PROMs) regarding OD and dysphonia in patients with IPD. On the other hand, we are evaluating treatment options for OD and dysphonia, in particular surface electrical stimulation (SES) of the neck to improve dysphonia in patients with IPD.

IPD is characterized by several distinct symptoms: rest tremor, rigidity and postural disturbances. 1% of the total world population over the age of 60 suffers from IPD. More than 80% of patients with IPD also develop OD and/or dysphonia. **Chapter 1** elaborates on the problems that patients with IPD and their loved ones may experience and the challenges that healthcare professionals face with regard to the diagnosis and treatment of OD and dysphonia in IPD.

PROMs in patients with IPD and OD and dysphonia
In addition to Flexible Endoscopic Evaluation of Swallowing (FEES) and videofluoroscopic swallowing study (VFSS), PROMs are important outcome measures in diagnosing OD and dysphonia from a holistic point of view. PROMs are any status report of a patient's health coming directly from the patient. PROMs can be used to evaluate the individual's perception of their health. There are many different validated PROMs that are usually specific to a particular patient population with a particular symptom or disease. In **Chapter 2**, we validated the MD Anderson Dysphagia inventory (MDADI) in Dutch for patients with OD of a neurogenic etiology. The MDADI was already validated for the head and neck oncology population. Due to the validation for patients with OD of a neurogenic etiology, it is possible to use the same PROM to the vast majority of the patient population that comes to the ENT outpatient clinic with OD.

Chapter 3 describes the exploratory, prospective clinical study regarding whether changes in OD and dysphonia-specific quality of life are associated with progression of IPD. Both the outcomes of the OD (MDADI and DSS; dysphagia severity scale) and the dysphonia-specific PROMs (VHI; Voice Handicap Index and VAS Voice) worsen as IPD progresses. Remarkably, the decline in PROM outcomes is not proportional to the decline in IPD. Although OD and dysphonia may result in a decreased outcome on PROMs in the earliest stage of IPD (Hoehn and Yahr scale 1), in the patient's perception, these symptoms start worsening at a mor advanced stage of IPD (Hoehn and Yahr stage 3). In addition, this study shows that the outcomes of the OD and dysphonia-specific PROMs are related to each other. Thus, with a worsening of OD-specific PROM outcomes, there is most likely a similar deterioration in dysphonia-specific PROM outcomes.

In **chapter 4** we've compared the outcomes of OD-specific PROMs (MDADI and DSS) with the results of the FEES and VFS by using a neural network analysis. Neural network analysis is a modern statistical approach to processing complex data. The neural network must be trained to predict the outcome of the FEES and VFS based on the outcome of the PROM of a patient. By comparing the predicted outcome with the actual outcome of the FEES and VFS, the neural network learns from the error and predicts better next time. By repeating this experiment often, with different patients, a reliable neural network can be created, since the margin of error is getting smaller. This study included 90 patients with IPD and OD. After training the neural network, we conclude that there is only limited agreement between the outcomes of the PROMs and FEES and VFS. To obtain a better insight in this limited agreement, a two-step cluster analysis was performed. A two-step cluster analysis categorizes patients into one or more clusters, based on all available data. This creates clusters with patients that have many similarities in the given data. For example, we found two clusters of patients who showed no signs of laryngeal penetration or aspiration on FEES or VFS, where one of the two clusters had a relatively high outcome on PROMs and the other cluster had a relatively low outcome. In addition, there was a cluster that did show signs of laryngeal penetration or aspiration on FEES or VFS with a similar low outcome compared to the cluster with no abnormalities on FEES or VFS. Based on the outcome of the PROMs used, it is therefore impossible to predict what the outcome will be based on FEES or VFS. It is therefore important to include both parameters in the analysis of OD in patients with IPD, because the absence of complaints does not mean that there are no abnormalities.

An update on treatment of dysphagia and dysphonia in Parkinson's disease
A systematic literature review on the different treatment options for OD in patients with IPD is described in **Chapter 5**. Twelve studies were included regarding pharmacological and rehabilitation treatments. Of these, only 4 studies were randomized and the methodological quality of a number of studies was limited. No high-quality evidence that pharmacological of rehabilitation treatments improve OD in patient with IPD was found. However, a clinical trend was found that conventional swallowing therapy using rehabilitative and compensatory techniques has a positive effect on OD in patients with IPD. The role of pharmacological treatments such as dopamine replacement still remains unclear. A person-focused conventional swallowing therapy seems to be the best treatment strategy for the time being. Person-focused conventional swallowing therapy should be based among others on IROMs during FEES or VFSS, patients' physical and cognitive capabilities, additional IPD-related needs and comorbidity, and OD-specific PROMs

Chapter 6 describes the effects of SES of the suprahyoid region of the neck as a new adjunctive treatment in voice therapy for patients with IPD and dysphonia. Patients were quasi-randomized into 3 groups. The first group received standard voice therapy only, the second group received standard voice therapy and SES at the motor level, and

the third group received standard voice therapy and SES at the sensory level. Patients were referred to one of the eighty-five participating speech and language pathologists (SLPs) affiliated with ParkinsonNet. The person-focused standard voice therapy included Lee Silverman Voice Treatment (LSVT), airway and breathing exercises and oral motor exercises. We found that after 15 sessions of standard voice therapy of 30 minutes per day for three weeks, a statistically significant improvement on the outcomes of dysphonia-specific PROMs (VHI) and stroboscopy was found. SES as an adjunctive treatment did not result in an additional improvement of dysphonia-specific outcomes.