Measurement and clinical evaluation of oropharyngeal dysphagia; a multidimensional approach

Heijnen, B.J.

1

General Introduction

This first chapter explains the main topic of the thesis, oropharyngeal dysphagia, and the aspects related to its assessment. The chapter presents the research questions addressed in the thesis and briefly introduces the contents of each chapter.

Deglutition

Swallowing, or rather deglutition, is a highly organized sequence of movements and sensory processes that involves fine coordination regulated by both cortical and brainstem processes.[1, 2] Figure 1 shows the various anatomical structures that are involved in swallowing: the lips(1), teeth(2), oral cavity(3), tongue(4), palate(5), pharynx(6), epiglottis(7), larynx(8), esophagus(9), and vocal folds(10).[2] Muscle innervation and sensory feedback are provided by the cranial nerves V, VII, IX, X, XI, and XII.[2, 3]

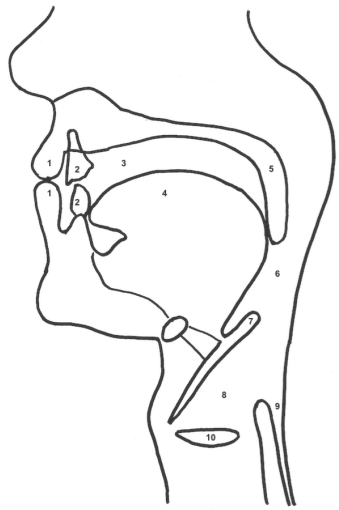

Figure 1. Anatomical structures involved in swallowing.

Swallowing occurs in four phases, starting with the oral preparatory phase (Figure 2A). This phase consists of voluntary actions required before a swallowing reflex is initiated, e.g. mastication and bolus placement. The second is the oral phase (Figure 2B), comprising the oral transport of the bolus to the pharynx, which is the last voluntary act before the swallowing reflex is initiated. The third is the pharyngeal phase (Figure 2C). In this phase, the bolus is transported through the pharynx, the larynx is elevated, the vocal folds are closed, and the tongue and the pharyngeal muscles transport the bolus towards the esophagus. In the fourth and last phase, the esophageal phase

(Figure 2D), the bolus enters the esophagus and is transported towards the stomach by peristaltic movements.[2, 4]

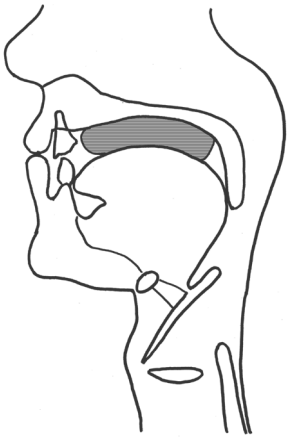

Figure 2A. Oral preparatory phase.

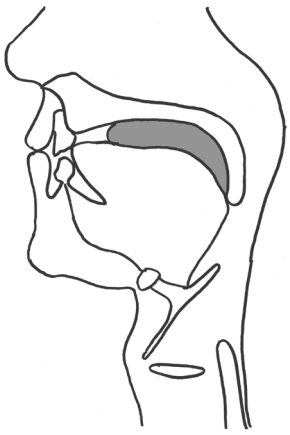

Figure 2B. Oral phase.

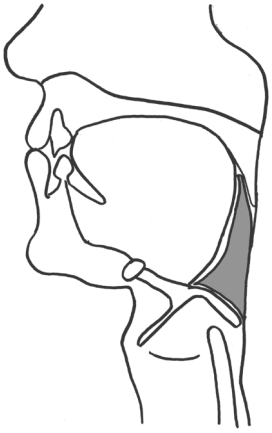

Figure 2C. Pharyngeal phase.

Figure 2D. Esophageal phase.

Swallowing disorders (dysphagia)

The concept of oropharyngeal dysphagia (OD) captures the arduous disruption of the process of transporting solids or liquids from the mouth to the esophagus.[5] This concept includes penetration into the larynx or aspiration of the bolus below the vocal fold level into the trachea[6]. OD can also take the form of oropharyngeal residue or pooling after swallowing.[7] It is associated with malnutrition, dehydration, aspiration

pneumonia, and sudden death. [8-11] Dysphagia is also known to have severe negative impacts on the quality of life. [12, 13].

Prevalence and patient populations

OD is often secondary to iatrogenic, metabolic, myopathic, neurogenic, or structural conditions. The most common causes are neurological, due to stroke or neurodegenerative diseases; the second most common ones are iatrogenic, resulting from surgery or radiation. [7] The prevalence of OD, as reported in the literature, varies from 2 to 16% in a normal population, and older age is associated with higher prevalence.[14] The prevalence of OD varies by medical condition: a prevalence of 8-80% was found after stroke, of 11-81% in Parkinson's disease, and of 27-30% after head trauma.[15]

Screening for dysphagia

The aim of screening for dysphagia is to identify at-risk patients. According to the World Health Organization a screening tool is a simple test to identify a subject with a disease or complaint but without the symptoms. [16] The screening methods should be easy to administer and quick, avoiding burden and invasiveness for the patients. [17] An important feature of a screening tool is that it strikes the right balance between sensitivity and specificity. Sensitivity refers to the proportion of positive-tested subjects who actually have the disease. Specificity is the opposite, referring to the proportion of negative-tested subjects in whom the disease is absent. A sensitivity of 70% and a specificity of 60% is considered a minimal requirement for an OD screening tool. [18]

In everyday medical practice a clinician often starts to investigate the presence of dysphagia by asking the patient whether there are swallowing complaints.[19] In addition, patients can be subjected to a (bedside) dysphagia screening, of which there are several types. Clinical features of aspiration can be noted with or without using a standardized form to determine whether a patient is at risk for dysphagia. Also a trial swallow using water or a substance in different viscosities can be combined with testing oxygen desaturation to detect patients at risk for OD.[17, 18, 20] Examples of evidence-based screening methods are the 3-oz. water swallow test[21], a V-VST (volume-viscosity swallowing test)[22, 23], a TOR-BSST (Toronto Bedside Swallowing Screening Test)[23], and a cough test[24]. A positive result indicates that further assessment for dysphagia is needed. To illustrate, consider the procedure of the 3-oz. water swallow test. A patient is instructed to drink a glass of water (90 cc) all at once. Meanwhile, the patient is observed for signs of aspiration; those may take the form of coughing, choking, clearing the throat, watery eyes, or shortness of breath.

When the subject undergoing screening test scores positive on one of the signs, further assessment for dysphagia is needed.

Assessment for dysphagia

After failing the initial screening, the patient should undergo further dysphagia assessment.[17] The two gold standards are fiberoptic endoscopic evaluation of swallowing (FEES) and videofluoroscopy (VFS). The latter is a radiographic examination of the swallowing act, which allows the investigator to visualize the complete trajectory of the swallowing act from mouth to stomach. VFS may show abnormal structures, pathological muscle activity, penetration, aspiration, and passage problems (Figure 3A). A disadvantage of VFS is the X-ray load for the patient.[2] The other gold standard, FEES, is the observation of swallowing in real time with a flexible nasendoscope introduced via the nose into the pharynx. By showing the swallowing act in motion, FEES gives information about the occurrence of penetration, aspiration, or passage problems (Figure 3B). In contrast to VFS, however, it only shows the swallowing act in the pharyngeal phase, not after passing through the upper esophageal sphincter and below. Another disadvantage derives from the short interval during swallowing in which there is a white-out due to muscle contractions.[25]

Patient-perceived aspects of dysphagia

Functional Health Status (FHS) is the ability to perform tasks in multiple domains [26] and it serves as an indicator of the influence of a given disease on particular functional

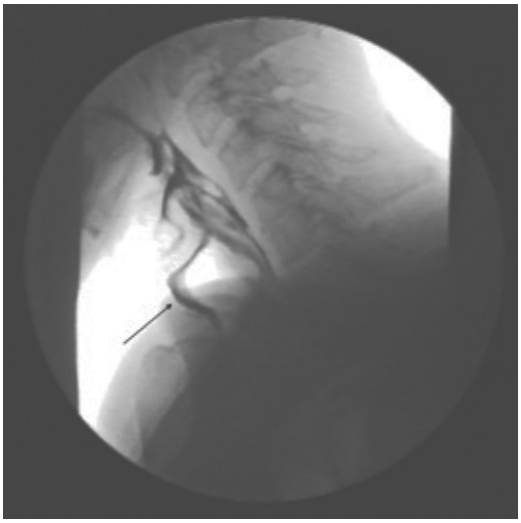

Figure 3A. VFS shows aspiration of thin liquids.

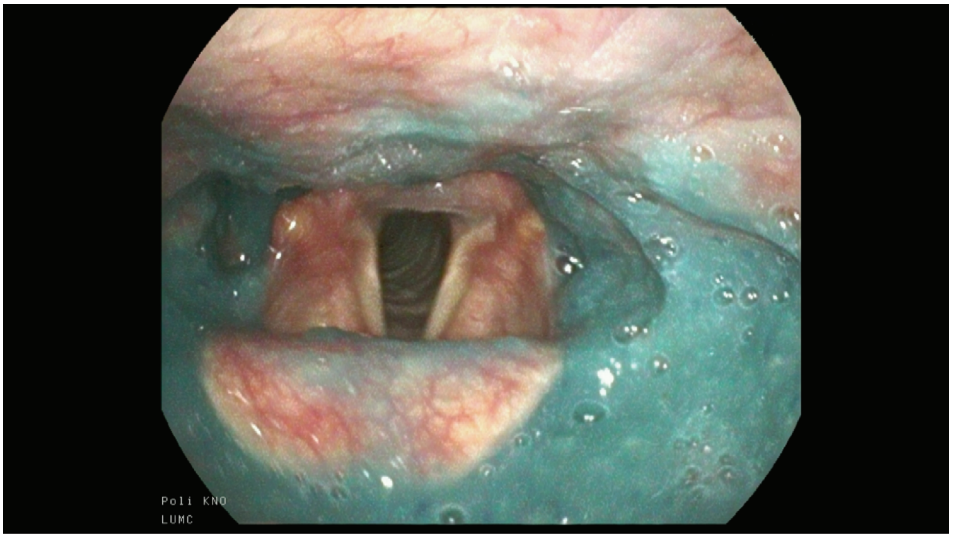

Figure 3B. FEES shows residue after swallowing with penetration of blue-dyed thick liquids. The residue creates the risk of aspiration.

aspects of a person. In dysphagia, FHS assessment quantifies the severity of the symptoms. This measure is complemented by Health-Related Quality of Life (HRQoL), which refers to the unique perception individuals may have of their health, taking into account social, functional, and psychological issues.[26] HRQoL can be divided into generic HRQoL and disease-specific HRQoL. The latter evaluates HRQoL in patients with a specific diagnosis and takes the characteristics and conditions of a disease into account. It has to be sensitive to measure the effects of a condition or intervention. [27] Generic HRQoL is able to measure a wide variety of characteristics, including clinical and sociodemographic features.

The severity that the consequences of dysphagia may have on different aspects of a patient's life, as measured in terms of either FHS or HRQoL, is usually ascertained from patient self-report questionnaires. Before a questionnaire can be used in everyday clinical practice or for research purposes, however, its measurement properties (its reliability, validity, and responsiveness) should be evaluated and be judged sufficient. [28, 29] These aspects are, among other characteristics, described in psychometrics.

Psychometrics

Psychometrics refers to the construction and validation of measurement instruments. [30] The main domains for checking whether a questionnaire is a reliable and valid form of measurement, as determined by the COSMIN taxonomy of measurement

properties and definitions for health-related patient-reported outcomes (HR-PRO), are reliability, validity, and responsiveness.[28] Reliability is the degree to which a measure is free from measurement error. Validity is the degree to which an HR-PRO instrument measures the construct it purports to measure. Responsiveness is the ability of an HR-PRO instrument to detect change over time in the construct to be measured.[28] Only measures or questionnaires with psychometric properties that are deemed sufficient should be used to evaluate a patient's status or treatment outcome.

Treatment of dysphagia

When the assessments show significant abnormalities, treatment is indicated. There are six categories of treatment modalities for OD.[31] The first is bolus modification and management, which includes adjusting the viscosity, volume, temperature, and/or acidity of the bolus. The second comprises behavioral techniques such as oral motor exercises. The third is sensory and neurophysiologic stimulation, for example taste or temperature adaptation or electrical stimulation. The fourth is postural adjustment, e.g. chin-tuck or head-turn maneuvers. The fifth consists of swallow maneuvers such as effortful swallow or Mendelsohn maneuver, the latter referring to prolonging the larynx in a high position to prevent aspiration. And the last is adjunctive biofeedback, which gives a patient visual information on the swallowing act.[2, 31-33]

AIM AND SCOPE

The aim of this thesis is to improve the measurement and evaluation of OD, focusing on the use of patient self-report measures.

The first study, presented in chapter 2, investigates the effects of radiochemotherapy on dysphagia, voice, speech, and trismus in patients with head and neck cancer (HNC). HNC patients suffer from various functional, physical, and emotional impairments due to both the primary illness and the secondary consequences of the tumor treatment [34]. Head and neck oncological treatments can have a severe impact on the anatomical structures, organ function, and quality of life [35].

A systematic review was conducted to study the effects of radiotherapy or chemotherapy on functions of the upper aerodigestive tract, including swallowing, in HNC patients. Data on patient characteristics, interventions, outcome measures, and treatment effects were summarized. Furthermore, the available evidence on interventions by speech pathologists was examined.

In chapter three, therapy effects are compared to determine the effects of traditional therapy and neuromuscular electrical stimulation (NMES) on patient self-reported

HRQoL in patients with Parkinson's disease and OD. NMES may be a therapeutic adjunct to known interventions in the treatment of OD [36-38]. The rationale underpinning NMES is that stimulation of muscle fibers by stimulating the nerve and the motor end plate would result in a re-education of the functional muscle-contraction patterns[38, 39].

In a randomized controlled trial the effects of adjunctive NMES in dysphagic Parkinson's disease patients were compared to traditional speech language therapy for dysphagia treatment with HRQoL as the primary outcome measure. It was hypothesized that NMES would not only lead to significant improvement of the swallowing function but would also contribute to an increased HRQoL in these patients.

Chapter 4 makes a comparison between daily clinical practice and the use of screening tools. It determines the diagnostic performance of daily clinical practice versus patient self-report tools in patients with OD. In daily clinical practice, a single question such as 'What about swallowing?' is frequently used without any additional standardized testing. The purpose of this study was to compare the diagnostic performance of a single question on swallowing with the FHS questionnaire EAT-10[40] as reference test. It was hypothesized that a single question, 'What about swallowing?', would show poor diagnostic performance when compared to the EAT-10. It was expected that the single question would have insufficient sensitivity and specificity to identify patients at risk of dysphagia.

In chapter 5 the reliability and validity of common self-report measures in OD are determined. Quality of life is considered to be an important patient-reported outcome measure in objectifying the current health status or therapy effects in patients with OD. In this study, the validity and reliability of the Dutch version of the Deglutition Handicap Index (DHI) and the MD Anderson Dysphagia Inventory (MDADI) were determined in oncological patients with OD.

Chapter 6 explores the predictive value of self-report measures for aspiration. The presence of aspiration is the most critical clinical sign of OD. Screening for and detecting OD may be done in several ways, but the gold standard is FEES or VFS. Unfortunately, the gold standard is often not available when dysphagia has to be assessed. The purpose of this study was to build a model to forecast aspiration in patients with OD using common patient self-evaluation questionnaires and oral intake status. Logistic regression was used to build a model to predict aspiration. Performance of the model was evaluated by constructing receiver operating characteristic (ROC) curves and computing the area under the curve (AUC).

Chapter 7 provides a general summary and draws some conclusions. On that basis, a number of recommendations for future research are made.

REFERENCES

1. Miller, A., *Deglutition.* phys. rev, 1982(62): p. 129-184.
2. Logemann, J., *Evaluation and Treatment of Swallowing Disorders.* 1984.
3. Shaker R., B.P., Postma G., Easterling C.,, *Principles of Deglutition: A Multidisciplinary Text for Swallowing and its Disorders*. 2012: Springer Science & Business Media.
4. Ekberg, O., *Dysphagia, Diagnosis and Treatment*. 2017: Springer.
5. Verin, E., et al., *Oropharyngeal dysphagia: when swallowing disorders meet respiratory diseases.* Eur Respir J, 2017. **49**(4).
6. Rosenbek, J.C., et al., *A penetration-aspiration scale.* Dysphagia, 1996. **11**(2): p. 93-8.
7. Cook, I.J. and P.J. Kahrilas, *AGA technical review on management of oropharyngeal dysphagia.* Gastroenterology, 1999. **116**(2): p. 455-78.
8. Kirshner, H.S., *Causes of neurogenic dysphagia.* Dysphagia, 1989. **3**(4): p. 184-188.
9. Mari, F., et al., *Predictive value of clinical indices in detecting aspiration in patients with neurological disorders.* Journal of Neurology, Neurosurgery & Psychiatry, 1997. **63**(4): p. 456-460.
10. Martin, L., et al., *Effect of electrical stimulation training on the contractile characteristics of the triceps surae muscle.* European Journal of Applied Physiology and Occupational Physiology, 1993. **67**(5): p. 457-461.
11. McHorney, C.A., et al., *The SWAL-QOL and SWAL-CARE outcomes tool for oropharyngeal dysphagia in adults: III. Documentation of reliability and validity.* Dysphagia, 2002. **17**(2): p. 97-114.
12. Gustafsson, B. and L. Tibbling, *Dysphagia, an unrecognized handicap.* Dysphagia, 1991. **6**(4): p. 193-199.
13. Plowman-Prine, E.K., et al., *The relationship between quality of life and swallowing in Parkinson's disease.* Movement Disorders, 2009. **24**(9): p. 1352-1358.
14. Kertscher, B., et al., *Prevalence of oropharyngeal dysphagia in the Netherlands: a telephone survey.* Dysphagia, 2015. **30**(2): p. 114-20.
15. Takizawa, C., et al., *A Systematic Review of the Prevalence of Oropharyngeal Dysphagia in Stroke, Parkinson's Disease, Alzheimer's Disease, Head Injury, and Pneumonia.* Dysphagia, 2016. **31**(3): p. 434-441.
16. World Health Organization. *Early detection of cancer*. 2017; Available from: http://www.who.int/cancer/detection/en/.
17. Speyer, R., *Oropharyngeal dysphagia: screening and assessment.* Otolaryngol Clin North Am, 2013. **46**(6): p. 989-1008.
18. Kertscher, B., et al., *Bedside screening to detect oropharyngeal dysphagia in patients with neurological disorders: an updated systematic review.* Dysphagia, 2014. **29**(2): p. 204-12.
19. Heijnen, B.J., et al., *'What about swallowing?' Diagnostic performance of daily clinical practice compared with the Eating Assessment Tool-10.* Dysphagia, 2015. **32**: p. 214-22.
20. Bours, G.J., et al., *Bedside screening tests vs. videofluoroscopy or fibreoptic endoscopic evaluation of swallowing to detect dysphagia in patients with neurological disorders: systematic review.* J Adv Nurs, 2009. **65**(3): p. 477-93.
21. Suiter, D.M., S.B. Leder, and D.E. Karas, *The 3-ounce (90-cc) water swallow challenge: a screening test for children with suspected oropharyngeal dysphagia.* Otolaryngol Head Neck Surg, 2009. **140**(2): p. 187-90.

22. Clave, P., et al., *Accuracy of the volume-viscosity swallow test for clinical screening of oropharyngeal dysphagia and aspiration.* Clin Nutr., 2008. **27**: p. 806-15.

23. Martino R, S.F., Teasell R, Bayley M, Nicholson G, Streiner DL, Diamant NE., *The Toronto Bedside Swallowing Screening Test (TOR-BSST): development and validation of a dysphagia screening tool for patients with stroke.* Stroke, 2009. **40**: p. 555-61.

24. Wakasugi Y, T.H., Hattori F, Motohashi Y, Nakane A, Goto S, Ouchi Y, Mikushi S, Takeuchi S, Uematsu H. , *Screening test for silent aspiration at the bedside.* Dysphagia, 2008. **23**: p. 364-70.

25. Baijens, L.W., et al., *FEES protocol derived estimates of sensitivity: aspiration in dysphagic patients.* Dysphagia, 2014. **29**(5): p. 583-90.

26. Ferrans, C.E., et al., *Conceptual model of health-related quality of life.* J Nurs Scholarsh, 2005. **37**(4): p. 336-42.

27. Briançon, S., et al., *Disease-Specific Versus Generic Measurement of Health-Related Quality of Life in Cross-Sectional and Longitudinal Studies: an Inpatient Investigation of the SF-36 and Four Disease-Specific Instruments*, in *Statistical Methods for Quality of Life Studies: Design, Measurements and Analysis*, M. Mesbah, B.F. Cole, and M.-L.T. Lee, Editors. 2002, Springer US: Boston, MA. p. 87-99.

28. Mokkink, L.B., et al., *The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: an international Delphi study.* Qual Life Res, 2010. **19**(4): p. 539-49.

29. Speyer, R., et al., *Psychometric properties of questionnaires on functional health status in oropharyngeal dysphagia: a systematic literature review.* Biomed Res Int, 2014: p. 458678.

30. Ginty, A.T., *Psychometric Properties*, in *Encyclopedia of Behavioral Medicine*, M.D. Gellman and J.R. Turner, Editors. 2013, Springer New York: New York, NY. p. 1563-1564.

31. Speyer, R., *Behavioral Treatment of Oropharyngeal Dysphagia.* Med Radiol Diagn Imaging, 2017.

32. Newman, R., et al., *Effect of Bolus Viscosity on the Safety and Efficacy of Swallowing and the Kinematics of the Swallow Response in Patients with Oropharyngeal Dysphagia: White Paper by the European Society for Swallowing Disorders (ESSD).* Dysphagia, 2016. **31**(2): p. 232-49.

33. Speyer, R., et al., *Effects of therapy in oropharyngeal dysphagia by speech and language therapists: a systematic review.* Dysphagia, 2010. **25**(1): p. 40-65.

34. Kreeft, A.M., et al., *Speech and swallowing after surgical treatment of advanced oral and oropharyngeal carcinoma: a systematic review of the literature.* Eur Arch Otorhinolaryngol, 2009. **266**(11): p. 1687-1698.

35. Rogers, S.N., S.A. Ahad, and A.P. Murphy, *A structured review and theme analysis of papers published on 'quality of life' in head and neck cancer: 2000–2005.* Oral Oncology, 2007. **43**(9): p. 843-868.

36. Bogaardt, H., et al., *Use of Neuromuscular Electrostimulation in the Treatment of Dysphagia in Patients with Multiple Sclerosis.* Annals of Otology, Rhinology & Laryngology, 2009. **118**(4): p. 241-246.

37. Bulow, M., et al., *Neuromuscular Electrical Stimulation (NMES) in Stroke Patients with Oral and Pharyngeal Dysfunction.* Dysphagia, 2008. **23**(3): p. 302-309.

38. Freed ML, F.L., Chatburn RL, Christian M. , *Electrical stimulation for swallowing disorders caused by stroke.* Respir. Care, 2001. **46**: p. 466-474.

39. Freed ML, W.Y., *VitalStim Certification Program. Training manual for patient assessment and treatment using VitalStim electrical stimulation.* 2003, Hixson TN: Chattanooga Group.

40. Belafsky, P.C., et al., *Validity and reliability of the Eating Assessment Tool (EAT-10).* Ann Otol Rhinol Laryngol, 2008. **117**(12): p. 919-24.