FACULTY OF SOCIAL SCIENCES
UNIVERSITY OF COPENHAGEN

PhD thesis

Trine Schow Hansen

Research in rehabilitation treatment for patients with severe Traumatic Brain Injury: Mapping complex therapeutic procedures and developing measures of adherence and treatment outcome

REHABILITATION IS LIKE HORSE TRAINING:

IT IS ESSENTIAL TO PROVIDE THE RIGHT FEEDBACK AT THE RIGHT TIME IN AN INSPIRING ENVIRONMENT; HAVING A MEANINGFUL GOAL AHEAD!

Academic advisor: Jesper Mogensen
Submitted 18.12.2009

TABLE OF CONTENTS

PREFACE

The work described in this thesis was carried out between June 2006 and December 2009 at the Department of Occupational Therapy and the Traumatic Brain Injury Unit, Hvidovre University Hospital, and at the Department of Psychology, University of Copenhagen, Denmark. The work is part of the requirements for the degree of PhD.

This work consists of the five following papers:

I) Hansen, T.S., Engberg, A.W., Larsen K. Functional Oral Intake and time to reach unrestricted dieting for patients with severe Traumatic Brian Injury. Archives of Physical Medicine and rehabilitation: 2008; 89, 8, 1556-62
II) Hansen, T.S., Larsen, K., Engberg A.W. The association of functional oral intake and pneumonia in patients with severe TBI. Archives of Physical Medicine and Rehabilitation: 2008; 89, 11, 2114-20
III) Hansen, T.S., Jakobsen, D. A decision algorithm defining the rehabilitation approach: Facial Oral Tract Therapy (FOTT) (Accepted for publication 14.december 2009 in the journal: Disability and Rehabilitation)
IV) Hansen, T.S., Jakobsen, D., Nowack, D'A, Whyte, J. Development of an adherence measure for a complex neurorehabilitation approach (submitted to the American Journal of Physical Medicine and Rehabilitation)
V) Hansen, T.S., Jakobsen, D., Westergaard, L., Speyer, R. FOTT versus FEES. A clinical evaluation of swallowing, feasible for patients with severe TBI and low level of functioning (preliminary manuscript)

Reading guide:

This thesis is addressed to both clinicians and researchers. Thus chapters 2 and 3 may be of interest to clinicians who are not very familiar with the area of rehabilitation research and chapters 4 and 5 may be more relevant for the researcher who might not know the world of the clinician. Chapter 1, 6 and 7 should be of equal interest for both groups.

This work has been a journey in rehabilitation research and also in personal development. Most times I have enjoyed the ride, sometimes I felt lonely and at others frustrated. There is no doubt in my mind that this journey has given me a great professional and personal experience that I'm very happy to have explored. However, this thesis would never have been possible without a lot of external support from people to whom I would like to show my deepest gratitude:

ACKNOWLEDGEMENTS

First I would like to thank all of the patients for participating in the studies and the relatives for giving consent that the data could be used in this PhD thesis; without them this study would never have been possible, and I wish you all the best of luck.

This work would never have been initiated without the support and recognition of my advisor Prof. Jesper Mogensen, to whom I am grateful for believing in this PhD project.

My deepest gratitude goes to the head of the Department of Occupational Therapy, Johanne Drachmann, for supporting me in starting this project and for her professional and caring support in all the ups and downs I have been through.

I would also like to thank the head of the Traumatic Brain Injury Unit, Annette Nordenbo for supporting me in starting this project and offering her guidance along the way. I'm grateful to Aase Engberg, who helped me design and write the two first studies in this thesis, and Lars Westergaard for engaging himself in the world of swallowing! I am also very indebted to John Whyte for giving me the opportunity to do a study at MossRehab, USA, for lots of professional advices and not least for "taking good care of me" in a foreign country. I'm also grateful to the staff at MossRehab for helping me with my study and making me feel welcome.

Huge thanks go to the staff at the Traumatic Brain Injury Unit, especially the occupational therapists and their leader, Marianne Falkengaard, for participating in this project and for supporting and criticizing my endless ideas. The work in this thesis is our work! Special thanks go to senior FOTT instructor Daniela Jakobsen, for letting me put her dear FOTT in boxes and for contributing significantly to the work in this thesis. My sincere appreciation goes to Kay Coombes for supporting and helping me in my work with her treatment (FOTT), and to the FOTT instructors in the FormaTT group. I would also like to thank Rainer Seidl and Rickie Müller-Busch for sharing their world of FEES.

Moreover I would like to dedicate special thanks and big hugs to the people in the research department to the TBIU, Ingrid Poulsen, Anne Norup, Karin Kristensen and Jakob Lorentzen, for

creating a great environment at the job and for supporting me, in your individual ways, both personally and professionally.

Last but not least I would also dedicate the biggest gratitude to friends and family, who have put up with me through this challenging phase of work and life, for supporting me through all times and for understanding my periodic absences. This goes especially to my mother Birthe, my sister Mette and dear friend Kirsten. Very special thanks goes also to Kristoffer Hansen, who has believed in me more than everybody else and at all hours been ready to discuss all aspects of my work and give valuable advice, even though rehabilitation research is a field that, at least in the beginning, he knew absolutely nothing about.

Trine Schow Hansen

ABBREVIATIONS AND DEFINITIONS:

Abbreviations

CIMT: Constrained Induced Movement Therapy

FEES: Fibreoptic Endoscopic Evaluation of Swallowing

FIM: Functional Independence Measure

FOIS: Functional Oral Intake Scale

FOTT: Facial Oral Tract Therapy

ICF: International Classification of Functions, Disability and Health

OT: Occupational Therapist

RCT: Randomised Controlled Trial

SSED: Single-Subject Experimental Designs

TBI: Traumatic Brain Injury

TBIU: Traumatic Brain Injury Unit

VFSS: Videofluoroscopic Swallowing Study

WHO: World Health Organisation

Definitions

Active ingredients: The essential components of the treatment that are hypothesized to cause change

Efficacy: the impact of an intervention as determined through a clinical trial

E*ffectiveness*: the impact of an intervention in real world situations

External validity: the extent to which the study can be generalized into broader or other rehabilitation settings

Internal validity: the extent to which conclusions about causes of relations are likely to be true, in view of the treatment, measures and research design used.

RESUME

På Afdeling for Højt Specialiseret Neurorehabilitering/Traumatisk Hjerneskade, Hvidovre Hospital indlægges patienter med svær traumatisk hjerneskade (TBI) i et tidligt rehabiliterings forløb. Behandlingen varetages af et tværfagligt team bestående af ergo- og fysioterapeuter, plejepersonale, læger, neuropsykologer, audiologopæder, socialrådgivere samt pædagoger.

Forskning i rehabilitering af patienter med TBI er stadig i en tidlig opstart og meget forskning mangler før det er muligt at sammensætte et program til alle patienter som beror udelukkende på evidens baseret behandling. Derfor er det heller ikke alt den terapeutiske behandling som anvendes på afdelingen som er evidens baseret. Der er særlige vilkår der gør det en udfordring at undersøge effekten af den eksisterende behandling. Dette er bl.a. at behandlingen, patienterne og mål for behandlingen er sparsomt eller dårligt defineret, hvilket gør det meget vanskeligt hvis ikke umuligt at undersøge behandlingens effekt. Samtidig er patienterne meget forskellige i skadens fysiologi og omfang, hvilket gør det vanskeligt at sammenligne patienterne og der er svære etiske problematikker i forhold til at lave kontrollerede undersøgelse.

En af de ”ikke evidens baserede” behandlinger som anvendes, primært af ergoterapeuterne, på afdelingen er Facio Oral Trakt Terapi (FOTT). Denne behandling retter sig mod patienter med vanskeligheder i at synke og spise, mundhygiejne, og verbal og nonverbal kommunikation. FOTT er udbredt i det meste af Europa med meget sparsom viden om dets effekt.

Det primære formål med dette projekt var at åbne en af de ”sorte bokse” i TBI rehabilitering og komme skridt nærmere evidens baseret behandling.

5 studier danner basis for denne afhandling:
Studie I og II undersøger omfanget og sværhedsgraden af patienternes vanskeligheder med at spise og drikke, samt komplikationer med lungebetændelse i en retrospektiv undersøgelse af 173 patienter. Vi fandt at patienter med svær TBI hyppigt har vanskeligheder med at spise og drikke, samt at over halvdelen kommer sig fuldstændigt indenfor en periode på 3 måneder. Vi fandt desuden at 43% af patienterne indlægges med lungebetændelse og 12% udvikler lungebetændelse i

løbet af indlæggelsen. Næsten alle de som udvikler lungebetændelse får ingen oral ernæring. Derudover fandt vi en sammenhæng mellem lavt kognitivt niveau og lavt bevidsthedsniveau ved indlæggelsen og chancen for at komme sig til fuld oral ernæring samt risikoen for at udvikle lungebetændelse.

Studie III og IV definerer behandlingen Facio Oral Trakt Terapi i en behandlings manual samt udvikler og tester en metode til at måle om terapeuterne følger manualen. Behandlingsmanualen udviklet i studie III definerer de essentielle komponenter af FOTT i 4 flowchart diagrammer, ét for hvert område af FOTT, med en tilhørende manual. Manualen guider terapeuterne i gennem beslutningsprocessen omkring valg af terapeutisk strategi og metode i en beslutnings algoritme. Den giver struktur til behandlingen og viser de forskellige valgmuligheder af terapeutisk adfærd indenfor de forskellige områder af FOTT. Det er en manual som samtidig efterlader terapeuterne med en fleksibilitet der gør det muligt at tilpasse behandlingen individuelt. Studie IV beskriver en metode til at måle om terapeuterne følger manualen. Resultaterne viser at målemetoden kan måle om en terapeut anvender de ”aktive komponenter” i FOTT samt at FOTT adskiller sig væsentligt fra den metode der anvendes til at behandle synkevanskeligheder på et Rehabiliterings center i USA.

Studie V udvikler og tester en klinisk metode til at evaluerer synkefunktionen hos patienter med svær TBI i relation til FOTT i et pilotstudie på 20 patienter. Interrater reliabiliteten af denne kliniske undersøgelse var god både for terapeuter med lidt og megen erfaring indenfor FOTT. Sammenlignet med en guld standard til bestemmelse af synkefunktionen, Fiberoptisk Endoskopisk Evaluering af Synkning (FEES), overestimerer den kliniske undersøgelse antallet af patienter som fejlsynker men bedømmer risiko for fejlsynkning, i form af rester i svælget efter synkning og materiale i øvre luftveje tilfredsstillende.

Med dette ph.d. studie er vi kommet skridt nærmere at undersøge effekten af en kompleks behandlingstilgang anvendt (bl.a.) til patienter med svær TBI. Flere studier er nødvendige før FOTT er evidensbaseret, men manualen der definerer FOTT, målemetoden som kan dokumentere om behandlingen anvendes som designet, samt den kliniske undersøgelse bidrager alle til at et fremtidigt effekt studie kan blive muligt.

SUMMARY

At the Traumatic Brain Injury Unit (TBIU), Hvidovre Hospital patients with severe traumatic brain injury (TBI) are admitted for early sub-acute rehabilitation. Treatment begins at the first day of admission by interdisciplinary teams comprising occupational therapist, physiotherapist, neuropsychologist, nurse, social worker, speech and language therapist and educator.

Research in rehabilitation of patients with TBI is still in an early phase, and much more research is needed before it is possible to put together a treatment programme based exclusively on evidence. Thus few of the rehabilitation approaches used at the department are evidence-based. However, special circumstances make it challenging to investigate the efficacy of existing treatments. There is limited or no clear definition of: the treatment used, the patients and of the treatment outcome. This makes it difficult if not impossible to investigate treatment efficacy and effectiveness. In addition, patients with TBI are very different in terms of the brain damage and functional impairments making it difficult to compare them in clinical trials. Also ethical issues make it difficult to do controlled trials.

One of the “non-evidence based” treatments, primarily used by the occupational therapists at the TBIU, is Facial Oral Tract Therapy (FOTT). This treatment is used for patients with difficulty in swallowing and eating, oral hygiene, and verbal and nonverbal communication. FOTT is widespread in most of Europe, with limited knowledge of its efficacy.

The primary objective of this project was to open one of the "black boxes" in rehabilitation and move a step closer to having evidence-based treatment.

Five studies form the basis for this thesis:
Studies I and II examine the extent and severity of patients’ difficulties with eating and drinking, as well as complications with pneumonia in a retrospective study of 173 patients. We found that patients with severe TBI frequently have difficulties with eating and drinking, and nearly two thirds (64%) recover completely within a period of 3 months. We also discovered that 43% of patients admitted to the TBIU have pneumonia at admission and 12% develop pneumonia during their stay.

Almost all who develop pneumonia have no oral nutrition. In addition, we found an association between low cognitive level, low level of consciousness on admission and the chance to reach unrestricted oral intake (study I), and the risk of developing pneumonia (study II).

Studies III and IV define the treatment Facial Oral Tract Therapy in a decision algorithm (treatment manual) and the development and testing of a method to measure how therapists follow the manual (measure of adherence). The algorithm defines the essential components of FOTT in four flow chart diagrams, one for each area of FOTT, with an accompanying manual. It guides the therapists through the decision-making process of therapeutic method within the different areas of FOTT. It gives structure to the treatment and shows the different options of therapeutic behaviour while leaving the therapist with the flexibility to adjust treatment for each individual patient. Study IV describes a method for measuring adherence. The results show that this measure can assess whether a therapist uses the "active components" in FOTT and that FOTT differs significantly different from the method used to treat swallowing problems at a rehabilitation centre in the USA.

Study V describes development and testing of a clinical tool to evaluate swallowing function in patients with severe TBI in a pilot study of FOTT on 20 patients. Interrater reliability for this evaluation tool was good for therapist both highly experienced- and less experienced in FOTT. Compared with FEES the clinical examination overestimated the number of patients with aspiration. However, retention and penetration were estimated with a satisfying level of sensitivity and specificity.

In this PhD study we have come steps closer to understanding the effect of FOTT, a complex treatment approach used (among others) for patients with severe TBI. More studies are needed before FOTT is truly evidence-based, however the treatment manual, adherence measure as well as the outcome measure al contribute to a future efficacy or effectiveness study.

INTRODUCTION

Progress in basic and clinical neuroscience highlight the potential for rehabilitation of function, even for patients with severe brain injury [1-4]. This knowledge has evolved through experimental studies in animals and humans demonstrating that the adult brain maintains the ability to reorganize itself throughout life even after severe injury. This phenomenon is known as a type of neuroplasticity [5].

Although neuroscience research has made major progress in understanding how the brain functions, it is still not possible simply to replace lost neurons and neural connections even this may be the most effective way to restore impaired functions. The natural way for the brain to restore lost functions is via learning. The brain continuously remodels its neural circuitry in order to encode new experiences and enable behavioural change to take place[4, 6]. These responses are valuable when planning rehabilitation programmes for those suffering from brain injury. However, translation into useful treatments of the new knowledge that has evolved in neuroscience is slow. In order to succeed this process needs close cooperation between clinicians and researchers. Thus bridging this gap between research and clinical practice is a highly important subject for the immediate future.

One explanation for this gap may be that the understanding of what drives neuroplasticity is not sufficiently advanced nor proven in human studies to construct a total rehabilitation programme. Still, one might think that the fully developed knowledge that is available should be integrated into existing treatments. The classic model of a translational pipeline where evidence based knowledge is integrated into clinical treatments involves four steps: 1. Basic science in the laboratory– 2. Proof of principle studies – 3. Clinical trials- 4. Health service research and delivery. However this process is less linear in rehabilitation science. Among many things it requires clear definitions of the treatments that already exist in order to combine them with laboratory research. Turning this around, by exploring existing treatment in laboratory research, one also needs a clear definition of the active ingredients of the treatment and a clear measure of treatment outcome. The new knowledge developed in the laboratory is typically clearly defined and tested in small studies, including animal models[7-8]. However, treatments used in the clinic are very different from and rarely as simple as “laboratory treatments”. Existing clinical treatments are experience-based,

developed over years or decades by interacting with patients and observing their responses. How these treatments have been developed and what hypothesis that has been formed along the way is rarely documented. Neither is the theoretical foundation or the definition of what is believed to be the active ingredients. However, this is not the same as suggesting that existing treatments are not effective. Thus until other treatments can be proven to be superior and actually possible to implement in the clinics, the existing treatment should be appreciated and acknowledged. Hypothetically, existing experience- based treatments might be superior to new laboratory treatments. It requires considerable effort to answer whether or not this is true. Detailed descriptions of the established "standard treatments" are an important step for 1.knowing how to combine these treatments with new evidence based knowledge and 2.being able to investigate their efficacy and effectiveness. Thus, clear definitions of the content of these treatments and attached therapeutic behaviour will pave the way toward evidence-based treatment programmes[9]!

Another issue adding to the gap between laboratory research and clinical practice may be that of communication and collaboration. Clinicians may not always understand the results of science and how to implement and combine it with their own treatment, and science does not always understands the patient's needs and the language of the people working with them. The world of the brain injured patient and the factors influencing rehabilitation offers a less rigorous framework for research than is available in the laboratory. Clinical work is influenced by many factors that are hard to control. Few treatments have made it all the way through the 4 steps in the translation pipeline to adoption by clinics. Constrained Induced Movement Therapy (CIMT) [10-11] is one example of a treatment that has accomplished that. CIMT was first tried in small animal studies and has now been taken all the way to a Phase III study in the USA that showed the effectiveness of the treatment. However, widespread implementation of CIMT in clinical settings is limited[12]. One reason might be that CIMT is directed towards a single impairment and may be difficult to combine with the many factors influencing rehabilitation of neurological patients. These patients' disabilities and functional problems are often very complex. Several different treatments are used and combined differently according to the patients needs. If neither the patients' needs nor goals and how to reach them are clearly defined, then implementing a new method such as CIMT may appear somewhat problematic. The extent to which new and existing treatments already overlap and agree in principle is not known. This is due to lack of definitions in the clinics and maybe also lack of openness to each other's work.

So the essence of productive translational science, and the key to bridging the gap between clinicians and research, may be improving communication between the partners involved and by mutual recognition of the different "cultures". Scientists should take more interested in understanding the whole patient and respect the clinical reality faced in the world of rehabilitation. On the other hand clinicians should be more open to what the scientific field can offer. In order to communicate the "land of clinical work" the established treatments should make an effort to defining clearly the content and theories of the active ingredients and test them in clinical trials. Otherwise it is difficult, if not impossible, to combine them in rehabilitation programmes and develop them in line with latest scientific understanding.

This PhD thesis balances somewhere within the translational pipeline [12]; by exploring the clinical field in order to translate it towards science. The research that underlies this thesis was carried out in subacute rehabilitation for patient with severe traumatic brain injury (TBI), dealing with a complex experience-based treatment approach, already well established in clinics around the world[13-14]. Objectives of this study were chosen based on the needs and wishes of the occupational therapists working at the TBIU, combined with the demands of evidence-based treatments. The choices made in this clinical research have also been decided jointly with clinicians in order to include and pay attention to the reality involved when working with these patients. This has resulted in a study combining epidemiological, descriptive, observational and experimental based research. It is my hope that it will interest both researchers and clinicians and help to bridge the gap between the clinician and scientist.

The work is presented in five manuscripts. Studies I and II are retrospective epidemiological studies dealing with the incidence and predictive factors for oral intake and pneumonia in patients with TBI. Study III defines an experience-based complex treatment approach, Facial Oral Tract Therapy, (FOTT) in a treatment manual. FOTT is used to treat neurological patients with problems in oral intake and airway protection as well as for other problems not addressed here. Study IV presents the development and validation of a measure of adherence to FOTT. Study V deals with swallowing safety - one possible outcome measure of FOTT. This study presents the development of a clinical evaluation of swallowing in relation to FOTT and test of validity and reliability.

OBJECTIVES

The purpose of the total research programme within which this PhD project has been conducted is to prove the efficacy and effectiveness of the therapeutic treatment approaches used in rehabilitation of patients with severe TBI. The framework of this thesis allowed the possibility of taking some of the necessary steps towards this ultimate goal of evidence, by exploring one "black box" in a rehabilitation programme to patients with TBI: Facial Oral Tract Therapy (FOTT).
The aims of the work were to:

- define the severity of the problem with oral intake and its duration
- define incidence of pneumonia (complication of oral intake)
- define FOTT (the treatment for problems with oral intake) in a treatment manual in an attempt to standardize FOTT and to characterize its active ingredients
- develop an adherence measure for the treatment manual
- develop an outcome measure: a standardized clinical evaluation tool for swallowing (oral intake) for FOTT; and to validate this measure using a reliable instrumental evaluation of swallowing/(a "a gold standard") (Fibreoptic endoscopic evaluation of swallowing)

Overview

Chapter 1 is a general introduction and presentation of the objectives with this study.
Chapter 2 is a brief introduction to TBI and neurorehabilitation.
Chapter 3 presents the challenges in neurorehabilitation research.
Chapter 4 presents swallowing problems and common treatments approaches.
Chapter 5 presents FOTT and how it relates to the principles of neuroplasticity.
Chapter 6 presents a discussion of the manuscripts in this thesis.
Chapter 7 presents some overall conclusions and perspectives of this work, and directions for future research in clinical rehabilitation.

TRAUMATIC BRAIN INJURY

Traumatic Brain Injury (TBI) is defined in the International Classification Of Diseases as: ”Craniocerebral trauma, specifically, an occurrence of injury to the head (arising from blunt penetrating trauma, or from acceleration/deceleration forces) that is associated with any of these symptoms attributable to the injury: decreased level of consciousness, amnesia, other neurological or neuropsychological abnormalities, skull fracture, diagnosed intracranial lesions, or death [15].

Common traumatic causes include motor vehicle and cycling accidents, falls, assaults, gunshot wounds and sport injuries[16]. TBI has been documented globally to be one of the major causes of death and disability [17]. In recent years, results show that rehabilitation of patients with TBI, (including those with very low level of functioning) is effective, and it is recommended that rehabilitation should be provided by centralized multi-disciplinary rehabilitation services[18-22].
In response to these recommendations, the Danish National Board of Health published, in 1997, a recommendation to centralise the service for severely traumatic injured patients[23]. Previously this had been provided at non-specialist departments in hospitals nearest to the patient’s homes or at nursing facilities [21, 24]. Two centres specialising in early, intensive, multidisciplinary rehabilitation of patients with severe TBI and adjacent diseases were established. One was the Traumatic Brain Injury Unit (TBIU) at Hvidovre Hospital, with an uptake area of eastern Denmark, the Faroe Islands and Greenland, while the Hammel Neuro Centre, covered western Denmark[21]. The report also recommended the establishment of a research team producing evidence of the effectiveness of treatments used by the multidisciplinary teams[23] which was established in 2005.

At the TBIU the multidisciplinary teams consist of physician, physiotherapist, occupational therapist, nurses (care-givers), social worker, neuropsychologist, speech and language therapist and educator. Interdisciplinary around-the-clock treatment and rehabilitation start on day 1 regardless of the patient’s level of consciousness. The main therapeutic principles chosen for rehabilitation are described by Affolter [7], Davies [1] and Coombes [8]. The rehabilitation approaches emphasise sensory stimulation and learning in everyday life activities, early mobilization regardless of the level of consciousness, stimulation of oral and swallowing function, optimal nutrition, contracture

prophylactic, serial casting, botolinum toxin treatment and rapid diagnosis and treatment of medical complications [21-22].

Much of the damage sustained in TBI is caused at the moment of the injury (referred to as primary injury), including diffuse axonal injury, cortical contusions, and disruption of small blood vessels. In the period after the injury development of oedema, expanding hematomas, and hydrocephalus, can exacerbate brain damage, referred to as secondary injuries. These many types of injury to the brain result in a variety of disabilities. However, as well as the location and severity of the injury, a patient's social relations and level of supportive resources influence rehabilitation and its outcome[25]. Thus, rehabilitation goals for patients with apparently similar injury may differ and require distinct interventions. Rehabilitation of patients with TBI is therefore not simply a matter of applying intervention X to treat problem Y. Rather, the therapist has to be constantly imaginative and creative in selecting, and often in constructing, the intervention with the best chance of successful outcome in a particular individual. Therefore, goals for different patients in the same rehabilitation department, such as the TBIU, can be extremely broad, ranging from sustaining a stable blood pressure during mobilisation from lying to sitting position to improving the ability to concentrate during a conversation with more than one person present.

An example of a treatment approach that manages to cover a broad span of disabilities is Facial Oral Tract Therapy (FOTT). FOTT is an integrated part of the rehabilitation program used at TBIU, and has been the base in the work contributing to this thesis as an example of the challenges met in rehabilitation research.

NEUROREHABILITATION RESEARCH

Rehabilitation professionals believe that the work they do makes a difference to the lives of the patients they treat. However, it is no longer enough to say that the treatment ”works” because it appears that the patients feel better and that they enjoy the contact with the therapist. Evidence-based practice has now become an integrated concept in rehabilitation and all professions need to have significant scientific proof that the treatments they use are effective. There is no doubt that many clinicians have theories about the mechanisms of the treatment they use and how they produce changes in patient response. However, clear evidence is still missing on whether these are actually “true”. There exists little knowledge of how the mechanism of the treatment function, which of the many components involved in the treatments are believed to be “the active ingredients”, and whether one treatment is more effective than another.

For several reasons, the demand for evidence has been met with mixed emotions from clinicians. One may be that research challenges the *status quo*. Suddenly the relevance of the treatments that have been used for many years is questioned. If it is proven ineffective or less so than another treatment, questions are raised as to whether it should ever be used again. However, there might also be other ethical and clinical issues as: what if some patients like this kind of treatment better than another even it is less effective? If a less effective treatment is easier to apply and easier to teach and perform should it still be rejected in favour of the more effective one? There are many practical, ethical and financial issues and concerns to these questions. I believe that most clinicians agree that it is beneficial that research within rehabilitation is developing since evidence-based practice is a tool to improve rehabilitation. However, there are many obstacles and challenges to such rehabilitation research.

RESEARCH IN REHABILITATION

Treatment research is designed to estimate most importantly the effectiveness of a treatment and its effectiveness relative to other treatments. It is also designed to investigate which treatment components most strongly affect outcome (active ingredients), the generalizability of a treatment across settings and the mechanisms that account for treatment effects[26]. How to investigate all these

interests related to evidence of a promising treatment takes rarely just a single study and a single type of study design to produce. Knowledge of treatment effects is built through a programme of research – developing over time, with a sequence of studies that seek to answer different questions. Programmatic research of this type is particularly crucial because it facilitates the construction of treatment theory in parallel with empirical evidence and paves the way towards evidence-based practice [27]. The work in this thesis is an example of the use of different types of research and research design, taking steps forward towards evidence-based practice.

Research in TBI rehabilitation is starting to build some evidence of the effect of rehabilitation and several reviews are available [28-32]. In their conclusions they all reach out for improved methodological quality into the research, focusing on an increase in numbers of randomized clinical trials (RCT)[33-34]. Cullen et al[31] conducted a review investigating the efficacy of rehabilitation intervention in TBI in 2007 involving studies from 1980-2005. They included 303 articles where 275 were intervention studies based on the following two inclusion criteria: 1. Studies where at least 50% of the population included patients with acquired brain injury and 2. Studies which had an evaluation of a treatment with measurable outcomes. They found that only 28% of the intervention studies were RCTs. The studies had small sample sizes; and great variety of the treatment used in the programmes and in the studied patient population; all factors making it difficult to compare the outcomes of the studies. The results of this review, however, show weak favour of:

1. Early rehabilitation is associated with better outcomes, such as shorter coma and length of stay, higher cognitive levels at discharge, better FIM scores, and a greater likelihood of discharge to home.
2. Inpatient rehabilitation improves functional outcomes in terms of improved self-care and mobility and higher FIM score.
3. Increased intensity of rehabilitation was found to reduces length of stay and improve motor recovery and functional outcome.
4. Inpatient rehabilitation results in successful return to work.
5. Community-based social and behavioural rehabilitation of at least six months results in greater level of independence, higher social activity levels, and less need for care support
6. Vocational rehabilitation results in more subjects having fair or good adjusted outcome and more than half become gainfully employed or full-time students

Nonetheless, while some positive effects of rehabilitation for individuals with TBI have been documented, it is still a great challenge, to follow the ethical recommendation only to use evidence-based treatments in the rehabilitation setting. Consequently, more research of new and ongoing treatment is needed and treatments used in clinics today cannot all be evidenced based.

RESEARCH DESIGN

Evidence-based medicine has been defined by Sackett and colleagues as the ”conscientious, explicit and judicious use of current best evidence in making decisions about the care of individual patients”[35]. To achieve this standard of practice, clinicians should act upon evidence as it becomes available, weigh the evidence in a consistent and valid manner and then change their practice appropriately. This simple description becomes very complex when used in rehabilitation treatment programmes.

Rehabilitation research aims at obtaining facts for a given problem, seeking true answers to the questions of interest. The “truth” is however influenced by several factors of uncertainty which leaves the researcher with more of a “probability of the truth”, instead of a 100% certainty. These uncertain factors are a major part of the clinical research profession and the researcher should always try at best to control for them or otherwise to be aware of them and how they might influence outcome of the study. There are many sources of components and bias that can affect treatment and outcome in therapeutic rehabilitation research as for example: selection of the study population, the state of mind of the patient or therapist on the day of the study, patient-client interaction just to mention a few. Such the estimate of the true treatment-related change can be too high or too low. The Randomized Controlled Trial (RCT) is the best research design to obtain the most optimal estimate. It is described as the gold standard in effectiveness studies [36-38] and is the most loved and hated research design in rehabilitation science, with constant debate concerning its necessity. However, many other study designs may be required before the RCT is possible to conduct and/or when the RCT might not be feasible. The work in this thesis has taken the preparatory steps towards the effectiveness study, using other types of research design in what Whyte [39] has called the “research enterprise”.

Chapter 3

A research design is a blueprint of the conduct of a study that maximizes control over factors that could possibly interfere with its desired outcome. The type of design directs the selection of a population, methods of measurement and how to conduct data collection and analysis. The choice of research design should first of all be directed by the research question and second by the available resources, contexts and expertise of the research group.

The challenge in choosing the proper designs relates to the structure of each individual treatment and the total treatment programmes, the nature of the patients studied and not least ethical consideration. Multiple interventions from different professionals are provided during the same period and the interaction of interventions may significantly influence outcomes. A relatively small, maybe non-significant, effect of a single intervention may be magnified when used in combination with other interventions, and vice versa; interventions that seem effective in isolation may act adversely when provided together.

Clinical rehabilitation research can generally be classified into two distinct categories: 1: observational and 2: experimental. Observational studies make no attempt to intervene where experimental studies do so.[40]

OBSERVATIONAL DESIGNS.

In the retrospective observational approach a phenomenon or hypothesized theory of events is investigated back in time. Retrospective studies have the advantage of being much cheaper as the data have to some extent already been collected and the outcome is not affected by bias related to gathering the data since that happened before the connection with the study was known. An example of this is the Hawthorn effect where participants experience an effect of intervention due only to the fact that he/she is part of a study. The disadvantages are that the researcher has no influence on how rigorously information was gathered. This design is used in studies I and II.

In contrary to the retrospective design, a prospective observational study gives the researcher the possibility of having greater control over the data collection process; involving what kind of data that are collected and how that should be done. However, as with the retrospective design, the researcher has little or no control over confounding variables apart from using different statistical techniques to adjust for them when analyzing the data. The advantages are:

- it is possible to observe behaviour exactly as it occurs, instead of hypothesising that it occurred as expected
- it is possible to generate new hypotheses concerning relevant factors not already known
- the results reflect the true clinical setting and have high ecological validity (i.e. that it can be generalized to real life situations).

The disadvantages are somewhat similar to the retrospective design i.e. that:

- it is not possible to estimate cause and effect, since nothing is manipulated
- there is a chance of subjective interpretation of the observations and, depending on the setup and design of the study risk, of observer influence.

This design is used in study IV.

EXPERIMENTAL DESIGNS (CLINICAL TRIALS)

The overall advantage with experimental designs is that they have the strength to test a hypothesized causal relationship and at the same time control for other confounding variables. To provide sound evidence of causal relationships between dependent and independent variables, takes a true experiment where participants are randomly assigned. Moreover the two groups of participant should be equivalent in all variables except the treatment of interest. Another important issue in investigating causal relationship is defining the hypothesized theoretical relationship. Theory provides the logical basis for causal relationship, and trying to understand the cause-effect relationship seems meaningful only when placed in the context of a theory explaining their relationship. Otherwise it may only be a statistical result rather than a clinically relevant and "practically" useful result.

There are two general categories among the different types of experimental design:

- true experimental design: This category includes more than one created group, common measured outcome(s), and random assignment. Note that individual background variables such as sex and ethnicity do not satisfy this requirement since they cannot be manipulated in this way.

- quasi-experimental design: This category is most frequently used when it is not feasible for the researcher to use random assignment.

Quasi experimental design

As stated previously, quasi-experimental designs are commonly employed when random assignment is not possible or practical. Although quasi-experimental designs need to be used commonly, they are subject to numerous interpretation problems. The following quasi-experimental designs are frequently used:

Post test only:

This design consists of administering an outcome measure to two groups receiving different treatments over a period of time. A major problem with this design is that the two groups might not necessarily be the same before any treatment takes place and may differ in important ways, influencing the rehabilitation progress and outcome.

Pre test post test:

This pre test- post test design partially eliminates a major limitation of the possible difference between the groups. At the start of the study, the researcher empirically assesses the differences in the two groups. Therefore, if the researcher finds that one group performs better than the other on the post-test, s/he can rule out initial differences (if the groups were in fact similar on the pre-test) and normal development as explanations for the differences.

Time series designs.

In time series designs, several assessments (or measurements) are obtained from the treatment group as well as from the control group. This occurs prior to and after the application of the treatment. Because measures at several points in time prior and subsequent to the treatment programme are likely to provide a more reliable picture of effect, the time series design is sensitive to trends in performance. Thus this design provides a strong picture of the outcomes of interest especially if a comparison group of similar patients is used,

One of these designs will be relevant in future studies investigating the effect of FOTT.

The "true" experiment: Randomized Controlled Trials (RCT)

This design is a traditional and highly recommended research design. The main advantage of the RCT design is that the randomization process reduces the possibility of a confounding effect from variables other than the treatment being investigated. The groups in the study are expected to differ only in exposure to the intervention; therefore the only remaining explanations for a difference in outcome between the groups are the intervention or chance. However, this design of trial may not be feasible in many clinical settings. There are several concerns in using this design in TBI research. The first of these is ethical concerns related to withholding standard treatment from a control group. If we want to investigate an already used treatment approach with nothing to compare it with, we face a design dilemma: treatment versus non treatment[37]. Alternatively, if there is another treatment to compare it with, which also may be a more beneficial design, the therapist, and maybe the patient and relatives might believe more in one of the treatments than the other and this can bias the study.

There are also practical concerns, such as the need for large sample sizes to overcome variability resulting from the multiple factors influencing outcome (e.g., pre-injury risk factors, severity); the fact there are relatively few patients with TBI at the same hospitals in different countries makes it difficult to find comparable groups of sufficient size. Another relevant issue is that an RCT study is inherently rather expensive and requires significant effort and commitment from the study personnel involved in collecting or delivering data. A final issue is that external validity might be low. The conditions and participants in an RCT study may be so straight forward that they do not reflect "real life" rehabilitation settings, making it difficult to translate the results into the everyday rehabilitation programme[40]. However, even RCT's of treatment versus no treatment are (for most problems) not feasible for programmes of severe TBI rehabilitation, it may be possible to do studies investigating different questions regarding intensity and timing.

Single subject experimental design

As discussed earlier, even when they seem very similar, impairments in patients with TBI may have quite different functional manifestations for each individual, and therefore require distinct treatment

strategies. This condition might support choosing a single subject research design. Single-subject experimental designs (SSED) are unique in providing empirical evidence of treatment effectiveness in the individual patient; they fit very closely with the clinical practice that might change constantly during one intervention or day to-day clinical practice[41]. The clinician is usually mainly interested in change in an individual patient who possesses a unique set of characteristics and circumstances[42]. There are several different types of SSED that can be used depending on research goals.
The clinical single-subject-methodology where a single individual is exposed to either one or both treatments has the advantages of providing continuous assessment and outcome information that can be used by the clinician to monitor patient progress and even adjust the treatment during the study[41]. Since therapists often adjust the treatment method throughout an intervention in TBI rehabilitation this study method seems recommendable in many ways. The disadvantages of this study design are that:

- the treatment is rarely systematically manipulated and investigated for stability of responses
- there is no control for extraneous events that might influence outcome
- it can be almost impossible to control for all related known or unknown confounding variables

These factors make it almost impossible to attribute explicitly a change in outcome to the treatment of interest.

In rehabilitation research one is interested not only in whether the treatment of interest is effective, but also in how the hypothesised active ingredients of the treatment actually work. There are at least two types of research designs that can be useful:
1. isolating the active components of an efficacious intervention and
2. examining the mechanisms of the process through which these components lead to clinical change.

Treatment component analysis design
This type of study uses a between-group design to compare the relative efficacy of the different components of a treatment package in order to determine which components are necessary and sufficient for the best possible change in patient response. Such studies can be illustrated as follows:

Treatment group 1: R O_1 X_1 O_2 O_3

Treatment group 2:	R	O_4	X_2	O_5	O_6
Comparison group:	R	O_7	X_3	O_8	O_9

(R=Random assignment, O=observation/assessment (different numbers refers to the number of assessment); X=treatment/intervention). The comparison group is receiving the full treatment ($X_{3)}$ while treatment groups 1 and 2 receive the same treatment except for the removal of one component.

Treatment mechanism design

Identifying the mechanism of change is one of the key aims of rehabilitation research[43]. Identifying some of the mechanisms behind therapeutic treatments in rehabilitation will help to discover overlaps in different treatments; increase efficiency and effect of the different treatments already used, and support the direction of new research. The methods in this type of study are very complex. An example of a diagram can be as follows:

Treatment group:	R	O/M_1	X_1	O/M_2	X_1	O/M_3	X_1	O/M_4	X_1	O/M_5	X_1	O/M_6
Comparison group:	R	O/M_7	X_2	O/M_8	X_2	O/M_9	X_2	O/M_{10}	X_2	O/M_{11}	X_2	O/M_{12}

M represents assessment of the proposed mechanism, X represents the treatment (the treatment group and comparison group are given different treatments). This research design involves several other criteria for demonstrating the mechanism of change. It is described in detail in other places and it is outside the scope of this thesis to cover those details. An example may be comparison of two different treatments for swallowing safety in which the proposed mechanism could be tongue movements or cognitive level.

Going through different possible research designs and the pros and cons of what can be accomplished within the individual methods it remains obvious that one research design will not answer all relevant questions concerning the efficacy or effectiveness of one treatment approach. Multiple studies are needed to address different questions of interest, and one may need to compromise in response to ethical and clinical concerns. Nevertheless, careful consideration of the goal with the research, the research question and hypotheses will guide the choice of study designs and so improve the outcome and what one can infer from these results.

NECESSITIES IN REHABILITATION RESEARCH

No matter what research question needs to be addressed, and irrespective of the choice of research design, all research attempting to assess the efficacy of treatment interventions must address clear definitions of three essential components: the participants in the study, the treatments of interest, and the treatment outcomes. Lack of clear definitions in these three areas is the major obstacle in rehabilitation research. The nature of the rehabilitation sphere faces complexities that are common despite the choice of research design because human beings, and here I refer to both patients and therapists, are notoriously multifaceted creatures[43]. Therefore defining the patients, therapeutic behaviour and outcomes is another major challenge in rehabilitation research.

DEFINING THE TREATMENT POPULATION:

Clear definitions of the treatment population are important for choosing the right treatment and for understanding the outcome in a research study[43]. It is critical when comparing groups of patients in pre-treatment observation. In addition, characterizing the participants is useful for:

- statistical adjustment of differences in prognosis among those receiving different treatments in any of the non-randomized studies
- using differences to predict outcome
- randomized controlled trial that has failed to achieve complete prognostic balance through randomization (most often a problem in small clinical trials) [38].

However, the question remains about how to characterize the participants most appropriately to ensure their comparability across different treatment conditions. The International Classification of Functioning, Disability and Health (ICF) offers a universal framework to classify impairments in persons with TBI[44]. However, it contains an exhaustive list of descriptions of what can be relevant for describing functioning, disability, and health and it can seem somewhat unmanageable when researchers focus on just one patient group such, as TBI, Therefore the development of a core list of

the ICF categories most relevant to patients with TBI is a recognized need [45] and is now in progress[46].

However, until this core set is prepared; no matter what framework is found most appropriate, the population should be defined in relation to the goal of the treatment, its content and outcome measures. The diagnosis of the TBI and size and localization of the lesion and injuries do not provide enough details for choosing therapeutic strategy. Therapeutic treatments are more directly related to limitation in performance of meaningful activities and the patient's ability to participate. Neither these limitations nor the performance problems can be easily defined. A functional problem such as "being able to swallow solid consistencies safely" could be a goal of a treatment within FOTT, as could: "being able to participate in a meal with others without getting food in the airway". In these examples the patient's problems are both related to swallowing, but the cause of the problems and their goals are very different. Therefore the treatment strategy and outcome measures for these two patients are also different. Such facts make it important to clearly define the patient in terms of functional problems and hypothesis of causal relationship in order to compare treatment outcome.

DEFINING THE TREATMENT APPROACH:

Perhaps the biggest challenge within TBI rehabilitation research is defining the rehabilitation approaches; many treatments may still be characterize as a "black box"[43]. One of the most essential explanations of this problem is that most therapeutic approaches in TBI rehabilitation are "complex treatments" meaning that they do not consist of a single component or set of exercises. Rather they consists of a range of components used in different combinations, with different levels of intensity and in different contexts depending on the individual patient's needs and response[9] Moreover, most of these treatment methods are experience-based. They have been developed through clinical practice by observing the patient's response to different techniques and approaches and not in the laboratory in rigorously designed experiments. The procedures used in practice involves different activating change processes, a complex treatment might use a combination of learning, coping, sensory feedback loop etc, whereas laboratory developed treatments commonly focus on one or only a few activating processes[47].

A structured way of defining rehabilitation approaches is by developing a treatment manual [9, 43, 48]. However, since the nature of the treatment is complex, separating components in a treatment manual is not straightforward. Unlike medical treatment, where a manual can specify the chemical structure of a drug, and its dose that is easy to administer to every patient involved in the study, the rehabilitation treatments are adjusted towards the patient's behaviour which makes it much more complex. The patient's behaviour and response to the treatment depends on several factors such as: the therapist delivering the treatment, the patient's mood that day, maybe even the temperature in the room (cold can increase tonus) and so on. These are just a few examples of factors that can influence the choice of components in a rehabilitation approach and explain why a strict manual is difficult if not impossible to design. Moreover this "structured variability" makes the definition of the content of the treatment a great challenge. However, this makes it no less important to characterize these treatments in terms of hypothesized active ingredients and decision processes[49].

The active ingredients should be described in a sufficient level of detail to allow the treatment to be carried out in the same way across participants, therapists, and clinical sites so it can be replicated in future research and be generalized for clinical use[50]. If it is unknown to the researcher if the treatment used involves the same active ingredients to different patients, it is very difficult to interpret the result of the study. Moreover it is not possible to either avoid using the same treatment again if the study result showed a negative treatment outcome or to replicate the treatment if the outcome turned out to be positive[43].

Defining the active ingredients in a treatment manual has several advantages both for clinicians and also for researchers: firstly, it will support the clinicians in specifying the underlying hypothesis of the treatment[43] and provide them with a tool for guidance when using complex treatment approaches. Without guidelines it can be very difficult, if not impossible, especially for less experienced therapists, to carry out such a treatment approach in a correct way. It can also be difficult to maintain the "quality" or "purity" of the treatment methods, incurring a risk that the treatment may change over time in individual settings[9]. If a treatment is not used in a standard way, either within or across sites, it is a significant problem to both internal and external validity of a study. Treatment manuals also provides the researcher with a tool that defines the components and process of the treatment studied and can support measuring if the treatment is delivered as intended (treatment adherence). Furthermore, when a treatment is defined and used in a standardized way it

is also possible to compare different therapies and to link treatment to aims and outcomes. Finally clear definition of a treatment approach will also enhance communication within professional teams involved in the rehabilitation programme as well as with the patient and their relatives. Hence, there are both clinical and scientific arguments in favour of defining rehabilitation approaches in a treatment manual, even if it is challenging.

The tradition of treatment manuals has mainly developed within behavioural therapy and psychotherapy[26]. Despite the advantages of such manuals, clinicians are not always in favour of using them and have raised critical issues. Existing manuals have been criticized on the grounds that:

- they limit the possibility of developing a therapeutic relationship with the patient
- they limit the expertise and use of experience for the individual therapist
- they restrict the possibility of individualized treatment for each patient
- they are not suitable for being implemented in clinical practice[26, 51].

Even though many of these concerns are not proven[51] it is my belief that, when a treatment manual, is being developed, some attention should be paid to this criticism since the clinicians' concerns, documented or not, can be a barrier to its implementation and use.

Developing a treatment manual should balance utility with specificity. It has to be specific enough to guide the therapist through the important components and decisions in the treatment, without being so extensive that no one will ever use it. Thus in the extreme case a manual can be outlined in so much detail that it will guide the therapist in every decision, describing exactly what to do and how to do it. Such manuals leave the therapist with no possibility of using their intuition or components from other treatments and would almost certainly be met with criticism by many therapists. Continuous adjustment of the treatment to individual needs of the patient is essential, therefore, outlining all possible adjustment methods in one manual would make the manual so extensive that it would become very difficult and time-consuming to implement in clinical practice. At the other extreme a highly flexible manual will be so "loose" that the therapist is left without enough guidance, and may result in very low internal validity[9].

There are several ways to approach this requirement to develop a manual that will lie between the two extremes and in doing so one has to decide the level of detail. This is what Whyte and Hart[43] described as the Russian doll. When you start to describe a complex rehabilitation approach there seem to be never-ending levels of layers of details that could be defined[43]. One solution can be to separate the treatment in aims and develop a manual to achieve the different aims (if such exist). Another way could be to define the decision-making process and the important decision rules in an algorithm. A decision algorithm will guide the therapist through the decision-making process in the treatment. This way of defining a treatment has so far mainly been used to classify patient-centred therapeutic activities within a single or multiple disciplines. The intention with these algorithms has not been to guide the therapist through just one rehabilitation approach but to guide the therapist through the process from assessment of the patient to the choice of treatment; not limited to one treatment approach[52-55]. However, I found the idea of using a decision algorithm[43] a solution for defining the process in one complex treatment approach. This will be discussed further in the discussion of study III.

TREATMENT ADHERENCE:

As discussed earlier, a specific treatment manual provides for measuring whether the treatment is carried out as intended[56]. Treatment adherence refers to the methodological strategies used to monitor and enhance the reliability and validity of the interventions. The overall goal in measurement of treatment adherence is to increase scientific confidence that changes in outcome are attributable to the treatment. Thus if treatment adherence is not evaluated in an outcome study, one does not know whether the result was caused by the active ingredients of the treatment or by some other unknown factors[57]. Therefore adherence should be measured during, or right after, the therapy session to ensure that the active ingredients have been provided. A high degree of adherence is needed to guarantee the possibility of study replication and generalization[9].

Even highly experienced, well-trained therapists may not always deliver an intervention perfectly when clinical circumstances, personal factors and interaction with the patient etc might disturb the “optimal” behaviour[57]. Therefore whatever the level of treatment competence, monitoring and improving delivery of the intervention are always essential. There is still no unified way to measure adherence. The measures described in the literature involve different techniques such as visual

observations, visual or audio recordings of treatment sessions, with rating scales addressed to either the patient or the therapist[58]. Treatment adherence is recommended to evaluate by coding intervention sessions (observed in vivo or video- or audiotape) according to *a priori* criteria. Either the therapist should complete process evaluation forms or behaviour checklists after each intervention session or external assessors should evaluate the video or audiotapes[57]. Hogue et al[56] specify that when choosing an adherence measure based on observations, there are several pitfalls depending on the choice of: what should be coded, who should code and what kind of coding system should be used. They recommend selection of a small section of therapy instead of targeting larger units over longer times, using non participating coders and selecting simple measures coding occurrence versus non occurrence, including frequency counts in an event-by-event coding or Likert type rating[56].

No matter what kind of measure is to be chosen, designing or defining the adherence measure has to be related to the type of treatment manual used and the level of specificity and flexibility chosen in its design. As mentioned before, the real interest when measuring adherence is whether or not the active ingredients are present as designed.

DEFINING OUTCOME MEASURES.

Specifying the outcomes of treatment is important because this is the yardstick by which efficacy is ultimately judged. The same treatment may be judged effective or ineffective depending on the outcome measures chosen [49].Similar to the choices related to defining the content of the treatment one also has to make a choice of which level of specificity the outcome measures should have; i.e. should they be addressed to narrow or broad perspective of treatment outcomes? Is it meaningful to measure the effect of just single components or does one want to measure the effectiveness of a whole treatment approach or treatment programme? Again the choice must depend on the interest of the clinician and the research question. However, the outcome measure should always be related to the treatment "target" and the treatment delivered. Moreover it should be known to the therapist using it and be valid and reliable [43]. Binary outcome measures such as death/survival are rarely specific enough or of interest in rehabilitation. Functional outcome measures, as for example the FIM instrument[59-60] are more relevant[61]. Most measures used will cover a continuum, and there is always some uncertainty in the data (the phenomenon of reliability), which may reduce the

statistical power of rehabilitation studies [61]. Another issue is that effects in rehabilitation largely reflect changes in behaviour. Changing behaviour is usually a relatively slow and complex and may not generalize from one setting to another [62].

First, one should measure and define both the immediate or short term and the long-term effects. Rehabilitation research should normally have several ‘‘primary outcome measures’’ in several different domains. For example, an exercise programme started six months after stroke might improve measures of strength (a short term goal) and also improve one or more of walking endurance, mood, fatigue, and carer stress (all long term goals) [63]. Moreover, the measures used should be clearly defined and very specific. Using general measures is more prone to generate ‘‘noise’’ from the items that bear no relation to the intervention. In contrast, specific measures are better related to the goals for the treatment and research question of interest. Existing FIM [64] and Short Form-36 [65] are widely used in preference to several more focused measures. Hence, like the process of defining the population and the treatment approach, defining treatment outcome is rather complex. Since many rehabilitation treatments affect several functions, and outcome is affected by other factors such as social relations; input and outcome may be nonlinear.

The challenges in rehabilitation argued in this section lead to the conclusion that there is a need to enhance the methodology of defining the population, the treatments and the outcome measures in rehabilitation research.

An example of such a rehabilitation approach, where the targeted population, the treatment itself and the outcome measures are vaguely or not at all defined, is Facial Oral Tract Therapy (FOTT). Despite the lack of definition and research, the occupational therapist at the TBIU uses this approach as part of the total rehabilitation programme for patients with problems in the face, mouth and swallowing/breathing. The work in this thesis has mainly focused on one of these areas: swallowing and eating.

SWALLOWING

Swallowing is critical to human survival because it involves two important procedures: airway protection and oral nutrition[66]. Swallowing problems are normally defined as problems with moving food from the mouth to the stomach[67].

NORMAL SWALLOWING PHYSIOLOGY:

In FOTT swallowing problems are defined in four phases. Other classifications of swallowing include two to six stages. However, none of these classifications recognize possible pre-oral factors operating prior to bolus preparation that may contribute to dysphagic behaviour[68]. In FOTT the first phase of the four involves anticipation in eating and drinking; defined by Kay Coombes as the oral-preparatory phase[69]. Dividing swallowing into four phases is supported by Leopold and Kagel[68] who presented a paradigm shift in describing swallowing in terms of "ingestion", where they used a five phase definition of swallowing, including a pre-oral (anticipatory) phase [68]. Including this phase when assessing and treating "swallowing problems" involves other strategies since it require other cognitive and physical abilities.

The oral preparatory phase in FOTT is where a person sees, smells, and recognizes the food before opening the mouth to take a bite or sip. This phase is crucial when working with patients with TBI. These patients have often cognitive, perceptual or motor deficits that limit their ability to prepare the food and transport it to and in the mouth. If a person is not aware or prepared to have food entering the mouth, it may not be handled properly for swallowing and can be spilled back into an unprotected airway. Moreover, recognizing the food may also start the process of saliva production, being ready to make a food bolus and ease the bolus transport[68, 70]. Once food is recognized, it is placed in the mouth and the mouth (labial area) is closed so no food or liquid falls out. (This requires a nasal airway and nasal breathing). How food is chewed and bolus is transported depends on the viscosity of the food or liquid. However, in general the food is masticated, mixed with saliva and formed into a bolus. The tongue tip is pressed against the palate just behind the front teeth and most of the bolus are located in the midline of the tongue[71]. The posterior part of the tongue elevates against the soft palate, and pushes downward to keep the bolus from escaping from the mouth- resulting in premature (non-prepared) bolus entry into the pharynx. In the Oral phase the

tongue transports the bolus into the pharynx by squeezing the bolus posteriorly against the hard palate[72]. As the tongue pushes the food or liquid toward the back of the mouth the top of the larynx begins to lift and move forward and the vocal folds closes to keep food from going into the airway (this is why breathing briefly stops when we swallow). The epiglottis also moves to help close the entrance to the airway. The soft palate lifts and retracts to close off the entrance to the nasal cavity, preventing food from coming out of the nose during a swallow and the cricopharyngeal sphincter opens to allow material to pass from the pharynx. The pharyngeal wall contraction squeezes the food through the pharynx and into the oesophagus. The Oesophageal phase involves transporting the food from the pharynx through the oesophagus to the stomach[67] by peristaltic waves.

SWALLOWING AND TBI

Swallowing problems are common in patients with TBI. Depending on how swallowing problems are evaluated and defined, and how soon after the injury they are measured, their incidence has been found to range from 26%-93%[73-77]. The causes of swallowing problems can be quite complex in patients with TBI resulting from various types of neurological injury both to the brain and to other parts of the body. Thus, swallowing problems can be caused both by neurological injury and cognitive deficits but also by, for example, injury to the jaw or face[67, 78]. Examples of deficits involved may be high muscle tone, difficulty in opening the mouth, bite reflex and loss of tongue control and thereby loss of bolus control and transport and reduced movement of the soft palate. Often there is a delay or even total absence in triggering the pharyngeal swallow. There may be reduced motor control of the pharyngeal stage of swallow when it triggers involving: reduced movement of the tongue base to generate the pressure to move the food into the pharynx; reduced laryngeal elevation because of, for example tracheostomy; or there may be reduced pharyngeal wall contraction resulting in reduced pressure generation moving food through the pharynx towards the oesophagus. Dysfunction in the airway closure can also occur, presenting a risk for swallowing safety. Cognitive and behavioural problems can influence the swallowing phases by reduced awareness, poor organization and sequencing skills, impulsivity and tendency to put too much food in the mouth, and poor understanding of the eating process and return to normal oral intake[79]. These impairments have been found to be an important predictor of swallowing problems[74, 76-77].

MEASURING SWALLOWING FUNCTION IN TBI

Both clinical and instrumental evaluation of swallowing is recommended when assessing swallowing function. I will briefly describe the advantages of each:

CLINICAL SWALLOWING EXAMINATION:

The clinical examination of swallowing is an important tool in the process of evaluating swallowing, though several studies have demonstrated that clinical swallowing assessment underestimates the frequency of silent aspiration and swallowing abnormalities and overestimates the frequency of aspiration compared with instrumental evaluation of swallowing[80-84].

Clinical examination has other advantages compared to instrumental evaluations. First it is possible to evaluate the pre-oral phase, including the patient postural prerequisites and their cognitive ability in participating in ingestion. Secondly, the therapist can carry out a thorough oral examination involving oral anatomy, function and sensation, including movements of the tongue, soft palate, and facial and labial movements[85]. Moreover respiratory function relevant for swallowing and the patient's ability to swallow over a longer time period can be examined. However, those clinical evaluations developed so far all require that the patient is able to follow a verbal command, which does not apply to many of the patients with severe TBI. The challenge is to construct a clinical evaluation tool which can easily be taught, is quickly administered and non-invasive, causes no distress to patients, can be done without the need for any qualification of the patient and produces reliable results. Until this tool is developed or discovered it is recommended to supplement the clinical evaluation with an instrumental one.

INSTRUMENTAL EVALUATION OF SWALLOWING

There are two gold standards of instrumental evaluation of swallowing: videofluoroscopy swallow study (VFSS) and fibreoptic endoscopic evaluation of swallowing (FEES).
VFSS uses fluoroscopy in video or digitized format that allows detailed analysis of the oropharyngeal swallowing process. VFSS does not diagnose the etiology of the swallowing disorder; instead, it determines the details of oropharyngeal swallow dysfunction and helps guide decisions regarding behavioural swallow therapy. It has many advantages in terms of a clear view

of the bolus flow, timing and duration time, but it also has several drawbacks: First of all the patients are exposed to radiation and because of this the evaluation is performed as fast as possible to minimize the exposure. The evaluation is often completed in 90 to 120 seconds or as little as reasonably achievable. This short duration time makes it hard to see whether there might be a risk in swallowing when the patient is tired or looses concentration. Another disadvantage is that the patient has to be transported to the radiology department, which can be impossible for some severely injured patients or constitute a safety risk that should be avoided if possible. In addition, since the observation can only be made with the presence of radium the patient has to swallow a certain amount of food or liquid in order to assess swallowing function. For patients with high suspected risk of aspiration it might be more appropriate just to evaluate swallowing of saliva. These disadvantages make this evaluation method less relevant or recommendable for patients with severe TBI.

FEES has been recommended as a superior tool for severely injured patients such as those with TBI[86]. FEES can be used at the bedside and demands less participation of the patient and the evaluation can be performed by examination only of the patient's own saliva. The evaluation is dependent on a skilled operator and it is not possible to visualize all moments of the pharyngeal swallow[87]. In the FEES procedure, a flexible fibreoptic endoscope is introduced transnasally to the patient's hypopharynx where the clinician can clearly view laryngeal and pharyngeal structures. The patient is then led through various tasks to evaluate the sensory and motor status of the pharyngeal and laryngeal mechanism. In the next step, food and liquid boluses are given to the patient so that the integrity of the pharyngeal swallow can be determined. Information obtained from this examination includes the patient's ability to:

- protect the airway
- sustain airway protection for a period of several seconds
- initiate a prompt swallow without spillage of material into the hypopharynx
- control timing and direction of movement of the bolus through the hypopharynx
- clear the bolus during the swallow, presence of pooling and residue of material in the hypopharynx
- sense the food or saliva in the pharynx (sensitivity of the pharyngeal/laryngeal structures)

Fibreoptic endoscopic evaluation of swallowing has been repeatedly demonstrated to have a high level of sensitive and specificity in determining whether a patient is demonstrating penetration or aspiration[88]. The advantages with this evaluation are that:

- it can be done over a longer period of time
- it can be done at the bedside and be repeated as many times as needed
- the only expense involved in the procedure is that of the device itself.

The disadvantage is primarily that the pharyngeal swallow cannot be viewed directly. During swallowing when the bolus enters the pharynx there is a 'white out' in the view due to pharyngeal closure. Moreover, the discomfort when the endoscope is introduced through the nasal cavity might affect the patient so that s/he responds differently in the swallowing examination compared to real life situations[89-90].

BEHAVIOURAL SWALLOWING THERAPY

As described earlier, swallowing problems are frequent in patients with TBI and are associated with an increased risk of complications, such as aspiration pneumonia, dehydration, and malnutrition. These complications are theoretically preventable by an accurate diagnosis of the swallowing disorder and appropriate intervention. The possible therapeutic intervention or behavioural swallowing therapy has been described in many different ways and called different things. One thing that seems almost consistent is that there exists very little evidence about its efficacy and effectiveness[91].

Behavioural swallowing therapy that has been tested for efficacy and described in the literature is characterized by being both compensatory and facilitative (active). However, most of the techniques investigated and described to date are single "stand-alone" methods rather than comprehensive treatment approaches.

Compensatory strategies:

Compensatory strategies impose external control on the swallowing process involving postural changes aiming at altering the dimensions of the pharynx and the direction of food flow.

Chin tuck position (head down towards the chest) is one of the most widely known postural techniques. Evidence of its effectiveness is still limited in neurological patients[92-94] and studies have also questioned consistency in its definition and standardized use[95]. The chin tuck posture is recommended for patients with delay in triggering the pharyngeal swallow, reduced tongue base retraction and reduced airway entrance closure[67].

Head rotation to the impaired side is used when the oral transit is disturbed. Logemann et al[96] is the only study evaluating the effect of this method in neurological patients. They showed that the amount of bolus swallowed almost doubled, however only five patients were involved in the study.

Side-lying is suggested as being effective in reducing the pharyngeal retention to the pharyngeal wall instead of allowing it to drop into the airway. However, this is supported by only one case study[97]

Other procedures have been described but since no studies investigated their effectiveness or efficacy they will not be considered further.

Increasing oral sensation involves increasing sensory input prior to the pharyngeal swallow. Most commonly used techniques include thermal stimulation and changing bolus viscosity. Lazarus et al[98] found that bolus volume increased, pharyngeal delay time diminished in stroke patients and Lazzara[99] found that thermal stimulation improved triggering of the swallowing reflex.

Food consistency modification

Adjusting texture of food and liquid are often used in swallowing therapy for neurological patients. Tsukada et al[100] demonstrated in a group of normal adults that thickened liquid has longer oral transit time and Logemann[101] found that a group of patients with dementia and/or Parkinson's disease had lower risk of aspiration when drinking honey-thickened liquid compared to thin liquid.

Facilitative Swallowing Manoeuvres

These manoeuvres are characterized by the need for the patient to be able to follow complex directions carefully and have good cognitive and motor function[102]. Thus, the therapist will not be able to use these techniques in many patients with severe TBI. A more relevant patient group would be oral cancer patients[103].

Mendelsohn manoeuvre:

This technique is designed to increase the extent and duration of laryngeal elevation[102]. It involves the patient squeezing the muscles of the tongue and neck for several seconds during a swallow and when the Adams apple is in its highest position.

Effortful swallow.

This technique is used to increase the posterior motion of the tongue base during the swallowing. The patient is instructed to exert more pressure on the bolus and thereby increase bolus clearings [104]

Supraglottic swallow

This is used when patients have premature spillage into the airway. The patient is instructed to take a breath and hold, and then swallow and clear the throat without inhaling.

Super-supraglottic swallow

This technique follows the same procedure as the previous one, however it involves more effortful breath-hold manoeuvre and a following voluntary cough[67], used for patients also for patients with premature spillage and reduced closure of the airway[67].

Strategies to Cognitive impairments:

This literature is also limited. Two case studies have showed that behaviour modification techniques involving structured eating environments and use of verbal cueing has positive effect on safe oral intake[105-106]

FACIAL ORAL TRACT THERAPY

Facial Oral Tract Therapy (FOTT) is a multidisciplinary approach offering a structured way of evaluating and treating patients with disturbances in swallowing and eating, oral hygiene, non-verbal communication and speech articulation caused by neurological conditions[107]. FOTT is used in Scandinavia and large parts of Europe and is also the treatment approach used at the TBIU, Hvidovre Hospital. FOTT addresses more than swallowing problems and in the following pages I will treat FOTT as a whole treatment approach; rather than "just" a behavioural swallowing therapy method.

FOTT was originally, and still are, developed by Kay Coombes, a British speech and language therapist. It is an experience-based approach, developed through clinical practice, interacting with the patients. So far only a few studies have addressed its efficacy[108]. FOTT is based on the concept, developed by the Bobaths in 1950[109], which is a problem-solving approach to the assessment and treatment of individuals with disturbances of function, movement, and tone due to a lesion of the central nervous system. Over the last 60 years it has developed by integrating new understanding of motor learning and plasticity into the concept. The goal of FOTT is to prevent the accumulation of symptoms after neurological damage. Moreover to reduce disability, avoid handicap and give the patient confidence, and become as comfortable and independent as possible. Careful attention is paid to the entire sequence of an activity from initiation, including getting ready to do something, to completion of the task[110]. FOTT can be used in all phases of neurorehabilitation. No matter what area of FOTT is used, the overall therapeutic approach should offer the patient support so that appropriate sensory feedback from the patient's own body is re-established. In FOTT treatment is adjusted to the patient and there are no demands on the patient for a threshold level of functioning to be enrolled in an FOTT programme. FOTT starts as early as possible in order to facilitate movements and prevent secondary problems such as contractures and hypersensitiveness using tactile input and guiding. Therefore it can be used with patients in coma and vegetative states, with infants and children, and patients with severe cognitive problems[13-14].

The four areas in FOTT are closely interrelated. For example the function in the face affects oral functioning and facial expression; oral functioning affects ability to take care of oral hygiene, and to eat and drink. It has been acknowledged that coordination of swallowing and breathing are critically important considering that the pharynx serves as a conduit for air going to and from the lungs and for fluids going to the stomach[111]. Moreover airflow is important for voice production. Voice is

important in communication, as is facial expression. Thus working with one area will affect another.

The decision process of when to work with what area and how to adjust the treatment to the individual patient- is essential in FOTT. The therapist starts with choosing treatment strategy, based on thorough examination of the patient in the beginning of the treatment session. This examination is fundamental for creating a hypothesis of the patient's performance problems. The strategies should however be readjusted throughout the training session on the basis of the patient responses to the treatment. The patient should only repeat exercises of which he has already demonstrated his capability, in order to learn it efficiently or if the therapist needs to evaluate his capability over time. Otherwise the therapist adjusts the treatment approach so that the patient constantly receives new information putting more demands on him. On the other hand, if the patient does not improve, or the performance decreases in response to the therapeutic strategy, the demands are lowered. This decision-making process enables adaptation of the treatment to meet the patient's needs.

Another fundamental principle in FOTT is to work with activities in the treatment sessions instead of single exercises. The activity should be meaningful for the patient and related to the goal set for the interventions. Moreover, a key ingredient in FOTT is the position for the activity. Postural control is recognized as fundamental to the ability to move and use selective normal movement patterns for all activities[112], including movements of the face and oral tract[113-115]. Therefore positioning the patient to promote postural control that is as normal as possible is an integral part of the treatment. This can be done through different approaches such as, mobilization, facilitation, positioning supported by objects as pillow etc. A normal position (in relation to) the activity provides the patient with tactile information of what is happening, a basis for normal movement, and helps normalize muscle tone.

The therapeutic treatment methods include slow, organised touch of the individual's hands, facilitating hand-to-hand and hand-to-face contact, together with specific oral stimulation, therapeutic oral hygiene routines and facilitation of swallowing. Specific therapeutic approaches involve: guiding, facilitation, elicitation, positioning and mobilization. Moreover, techniques relevant in all FOTT area are "protection of airway" and "support of swallowing". These are also mainly done using physical stimulation. The therapist most often uses his/her own hands to support

swallowing, or elicit a swallow by having the patient move forward or mobilize the patient so s/he feels the material in the throat. Thereby the patients are provided with as much support as is necessary to experience the movement as normally as possible. In FOTT the therapist does not use manoeuvres such as the Mendelssohn or supraglottic swallowing, but works with swallowing in the natural context of eating and drinking. The texture of the food and/or liquid may be adjusted according to the patient's problems. However, if the patient is not able safely to swallow material, firm consistencies may be wrapped in gauze and chewed but not swallowed. Thereby the patient can work with: chewing and tongue movements, swallow of saliva and get tactile stimulation in the mouth. Oral stimulation is another essential ingredient in FOTT. Before entering the patient's mouth the therapist might also work with preparation for swallowing or oral hygiene by oral-stimulation routines. This can also be used as a method to give the patient sensory stimulation to avoid either hypo- or hyper sensibility or a way to work with swallowing saliva.

Although it is recognized that the FOTT approach has undergone developments since its inception, not much international literature has been published to document how it has changed and how it is believed to facilitate changes. Most literature is in German and not published in international journals. Most updates, practical and theoretical, are distributed orally, and through courses which can contributes to problems in recognition of its existence and in outlining of clear testable hypothesis of its content. Moreover it provides a risk for standardize use and internal and external validity in an outcome study. Despite this, FOTT is widely used and acknowledged in clinical settings and has provided treatment to patients who previously went without it. For this reason only one could advocate that it is worth spending the time and effort to document its value in rehabilitation programmes. This uncertainty with internal and external validity has among many factors influenced the choice of methods in this PhD thesis that will be discussed in Chapter 7.

FOTT AND PLASTICITY

It is not the attention in this thesis to explore the world of neuroplasticity. However, since it is the essence in the field of neurorehabilitation I will briefly describe its principles relevant to FOTT.

Neuroplasticity is the mechanism by which patients with TBI are able to relearn lost behaviour and skills. Learning experiences, can occur, both in responses to rehabilitation programmes and also in the course of everyday life. It is noteworthy that not all learning and provoked plasticity is beneficial for increased efficiency in performance of movement and movement patterns. Over-compensation can be an unwanted outcome of neuroplasticity. A natural response of a patient with one side affected limp after brain injury is to over-compensate with the less affected limp resulting in compensating reorganization which can often interfere with relearning of normal movement-patterns using the impaired body side[4, 11]. Therapeutic treatment approaches might help prevent this phenomenon and take advantages of other principles to direct the neuroplasticity in a way which better promotes skills.
Kleim et al[4] presents a list of 10 principles that are relevant to rehabilitation outcome and here they are briefly discuss in relation to FOTT. I have condensed the 10 principles into 7 since there seemed to be some strong overlaps or close relationships in this context:

Principle 1 Use it or lose it
If a movement or movement pattern is not used or a sensory stimulus to a body part is not received, and therefore peripheral input to the sensory cortex does not take place, cortical somatosensory representation is reduced, and novel function can shift to other brain areas[1, 116] In FOTT it is hypothesized that the patient should experience the sensory and motor feedback for the lost functions even if he or she is not capable of performing the movements independently. The therapist provides the patient with as much support as is needed in order to give him/her the experience of the movement. For example, in patients who cannot safely protect the airway and are not eating, the therapist stimulates the oral cavity in order to prevent sensory deprivation of the mouth and to facilitate saliva production so the patient can swallow with assistance. Support for swallowing is done either at the floor of the mouth using the fingers to facilitate movements of the tip of the tongue up towards the palate, initiating bolus transport. It can also be done at the back of the floor of the mouth to lift the back of the tongue towards the soft palate to initiate the transport of the bolus to the pharynx and thereby initiate a swallow. These techniques aim to allow the patient to

experience a normal movement pattern of swallowing, to prevent inefficient movements such as throwing the head backwards and in that way transporting bolus to the pharynx.

Another example could be providing the patient with the possibility of swallowing even if it is not safe for them to digest solid food that has to be chewed. The therapist wraps solid food in gauze and (if possible the patient is guided to place the gauze in his/her mouth) and supported in chewing in the food. The chewing gives the patient both a sensory (tactile and taste) and a motor experience of chewing. Moreover, having food in the mouth, may also facilitate response of saliva production, and thereby enable the patient to experience swallowing in a controlled setting, without the risk of getting food in the airway. However, these are only hypothesized or experience-based activities that need to be evaluated and tested in clinical studies.

Principle 2 Use it and improve it and repetition matters (two principles comprised in one section). In line with the first "use it or lose it", several studies have shown that extended training can induce plasticity. This plasticity is especially beneficial to functioning if that training involves skill practice. However, relearned skills need to be repeated over time in order to maintain and make further improvement. This knowledge supports the hypothesis in FOTT that the therapist will continue to use a relearned movement in different settings and contexts and maybe just extend the time the patient should use this skill. However, the function will not just be used repeatedly when it is certain that the patient "knows how to do this movement" without increasing the demands on him or her. This way of challenging the patient supports the understanding that: to drive further plasticity, the "level" of performance or demands on the patient have to be increased in order to provide further learning[6, 117-118]. However, it is not clear whether the principles in FOTT of how and when to increase the demands on the patient are done with the appropriate timing and level of demand, so this will also be relevant to explore. Moreover, several studies have shown that behavioural experience can enhance behavioural performance and can advance plasticity. Also natural environment and environmental enrichment is one example that has been shown to improve functional recovery[118-120]. These results support the hypothesis of FOTT that the environment for the activity used in the treatment should be as "natural" as possible. With that precaution, a natural environment such as a "dinner with family" may involve too many stimuli for the patient to concentrate or cope with due to perceptual problems.

Principle 3: Specificity, salience and transference

The principle of specificity suggests that plasticity only occurs in the particular behaviour being trained[121], meaning that specific training of one function might not lead to improvements in another. In FOTT the therapist most often works with task-specific training. The training is almost always in a relevant activity for the performance problems. Thus if, for example, the patient has reduced movements in the mouth and lips the therapist may choose an activity like blowing into a flute. This theory links to the principle of "salience", since it seems that the more salient activity the better[122] associated to FOTT where the therapist always chooses to work with relevant activities instead of exercises. This theory may be in contradiction to the principle of transference, defined as the ability of plasticity within one part of the brain to promote plasticity in another[4]. However, our knowledge within this area in relation to swallowing is low and primarily provided by electrical stimulation of the pharynx[123-124]. Thus, in FOTT the therapist often uses the learned skill in an activity that most often demands other skills. That might induce plasticity for the other skills involved in the same activity. One testable hypothesis could be that: Oral stimulation used in FOTT as a sensory preparation for swallowing (avoiding sensory deprivation or working with hyper or hypo-sensitivity) and a way of working with swallowing of saliva, can influence a functional change.

Principle 4: Intensity Matters.

There exists some knowledge of how intense rehabilitation can enhance neuroplasticity, but the extent remains to be further explored[116]. FOTT is meant to be implemented in a total rehabilitation programme, and as such carried out by professionals who handle the patients. This can provide rather intense stimulation since it will be provided to the patients throughout the day. Rosenbek et.al[125] compared an average of 150, 300, 450 and 540 trials per week of tactile thermal stimulation on swallowing in stroke patients. None of the dosages came out as superior. So increased knowledge of the relationship of intensity is a highly relevant subject for exploration, and this may also influence the whole structure of the rehabilitation programme addressing the number of therapists and hours of therapy.

Principle 5. Time matters

Early start of behavioural training has been proposed as the best way to promote post-traumatic recovery of function. This is based on the assumption that the earlier the post-injury stimulation takes place the better the brain's plasticity would be guided towards restoration and elimination of functional deficits and "learned non use"[126-129]. In Denmark, during the first years after centralizing the rehabilitation of severe TBI patients (making possible an early start to stimulation) better

outcomes in terms of Glasgow Outcome Scale scores, FIM score, “ability to walk independently” and return to living at home under normal conditions was shown[21-22]. However, it may be more complex than that. Other studies have also showed that too early a start of intensive repetitive therapy may have detrimental consequences[130-131]. These studies involve a long-lasting immobilization, and the methods used in FOTT are not close to such intensive methods so it seems a sound recommendation to start as early as possible.

Principle 6 Age Matters

It is clear that the neuroplasticity responses are less in the aged brain[4]. FOTT can be applied to patients of all ages including infants and children.

Principle 7 Interference

Interference relates to the hypothesis that one experience can interfere with the learning of another. This has been discussed previously in relation to the development of non efficient compensatory strategies or “bad habits”; the plasticity occurring as a result of these “bad habits” may interfere with learning of more efficient strategies. Another aspect of this, demonstrated by Boyd and Winstein[132] shows that explicit (verbal directions) information given to make a patient relearn a lost task may interfere with performance and learning whereas implicit directions, such as behavioural driven experiments, promotes learning. The method in FOTT supports both strategies by working with a normal way of performance, rather than with compensatory training. The therapist uses facilitating strategies which involve the impaired body parts to be used in a “normal way” and behavioural or experience-driven directions instead of verbal instructions. However, it is still necessary to test if the method used in FOTT, supporting the “normal way” of making the patient participate in daily life and activities, is superior to the more intense Constrained Induced Movement therapy[10, 133] used in limp training[134]. This is mainly relevant for testing in the pre-oral phase where the patient participates in the preparation of the meal and ingestion rather than in the oral and pharyngeal phases of swallowing.

DISCUSSION OF THE FIVE PAPERS

This following section will briefly present the five papers of this thesis. It will discuss how they are linked together and will provide additional considerations not part of the publications.

STUDIES I AND II

Studies I and II will be discussed together since they are born out of the same study design and much of the same data.

These two studies form the background for studies III, IV and V in this thesis, by investigating the severity and consequences of swallowing problems for patients with severe TBI.
In Study I we investigated the severity of the problems with eating and drinking and the time to full recovery of oral intake. We found that swallowing problems, measured in terms of functional oral intake, are very common (93%) for patients with severe TBI admitted to a sub-acute rehabilitation department. For the patients who return to an unrestricted diet it takes a median of 28 days and maximum of 126 days. The chance of returning to total oral diet depends on the severity of the brain injury and can be predicted by levels of GCS, RLA, FIM and functional oral intake at admission.

Not many studies have dealt with this topic in TBI patients, especially not in this early phase of recovery. Mackay et al[73] found that 61% of patients with TBI had swallowing problems verified by VFSS, in line with the result of Schurr et al[135] who found that about 50% had swallowing problems also verified by VFSS. They found that 66% had problems with oral intake in a clinical evaluation of swallowing but a VFSS found that 77% of these patients had swallowing problems. One reason that our study showed an incidence of 93% could be a result of the way swallowing problems were assessed since clinical evaluations are known to overestimate the risk of aspiration and the evaluations used were tested for validity and reliability. However, the focus with this study was not only swallowing safety (aspiration and penetration) but how many patients have restrictions on their intake of food and liquid. Naturally swallowing safety is related to this but is not the only relevant

factor. Restrictions on oral intake cover a broader group of patients than just defining the group as having swallowing problems. This group of patients also involves those who, due to cognitive problems, have problems with eating and drinking. They might not understand the meaning of eating or they stuff their mouth too much. It could also be that the patient cannot swallow safely after the intake of half a meal because they become fatigued or they lose their level of concentration, affecting their swallowing function. These patients might have a normal swallowing function in an instrumental evaluation of swallowing such as VFSS or FEES that often takes place in a short time period.

A clinical evaluation of swallowing may take place over a much longer time period and in different contexts, for example social, or other more “natural” situations, and thus give valuable information not shown by instrumental swallowing information. The interest with safe swallowing is, in most situations, whether the patient can safely intake enough calories by mouth, and avoid having a feeding tube. Thus it is highly relevant to involve information from these situations in the judgement of oral intake. The difference in swallowing evaluation might be the reason for the difference in the incidence of swallowing problems. Another reason might be that we used the functional oral intake scale (FOIS) by Crary et al[136]. This scale focus on how the patient receives nutrition and not just swallowing function, where the findings from instrumental evaluations give information about anatomic and physiological impairments exclusively. This information is primarily useful for the therapist planning the treatment and less relevant for the patient and relatives. The FOIS measure the functional outcome of eating and drinking and therefore we found it more relevant to use. However, this makes the comparison with studies using aspiration as outcome measure less meaningful.

However, we do recommend that the clinical evaluation of swallowing should be standardized, and its validity and reliability investigated. There exist several studies comparing the clinical evaluation of swallowing with an instrumental evaluation[80, 82, 84, 137-139]; however the Occupational Therapist (OTs) at the TBIU uses a clinical evaluation according to the FOTT approach. As shown (later) in Study IV, FOTT differs somewhat from other swallowing therapies and therefore very interesting to evaluate the FOTT clinical evaluation of swallowing. This interest has led to Study V see discussion of this paper below.

We also showed that recovery to non restricted oral intake happened within 126 days of rehabilitation, which was very much in line with other studies as discussed in the paper. Terre R et al[140], found in a group of 26 patients with severe TBI that the number of aspirations had the most significant reduction observed in the examination made at 3 month follow up. Similar to our study the prediction of recovery was associated with the level of RLA at admission. An RLA =3 was strongly associated with the presence of aspiration one year after injury, while patients with an RLA>4 did not aspirate at this time after injury. However, we did find that chance of recovery changed when the patients had RLA=III where Terre et al[140] still found that this patient group were associated with risk of aspiration after one year. Even where this discrepancy exists it is very interesting that the level of consciousness is found to be associated with the return to safe oral intake. This finding supports the argument in favour of using the Functional Oral Intake Scale and the importance of the clinical evaluation of swallowing.

These findings in Study I are very much in line with the results we observed in Study II. Here we evaluated the incidence of pneumonia in the same group of patients. We found that 27% of the patients transferred from the Intensive Care Unit suffered from pneumonia but only 12% developed pneumonia during subacute rehabilitation at the TBIU. Higher incidence of pneumonia was found in patients with low level of consciousness, tracheotomy and tube feeding exclusively. These findings support the result that it is mainly those patients who do not have any oral intake who develop pneumonia. The cause of pneumonia can only be speculation since it is not in the nature of the two retrospective studies to conclude anything significant about this. However, it does seem very interesting that it is the patients receiving nutrition by feeding tube who develop pneumonia. This could indicate that they might have a poor oral hygiene and aspirate their own saliva with pathogen bacteria and/or gastric reflux. As discussed in the paper it can draw attention to the timing of the bolus feeding and mobilization of the patient in for example physiotherapy training sessions or occupational therapy. In the training sessions, where the patient is mobilized often from lying to sitting or standing positions, reflux can occur if there is a lot of food in the stomach. This is especially important in patients with low level of consciousness who may suffer from lower tonus in the oesophagus sphincter[141]. It may also lead to the conclusion that it is very important to be aware of the oral hygiene of this patient group since swallowing of pathogen bacteria can increase the risk of pneumonia[142].

STUDY III.

Studies I and II showed that problems with eating and drinking are frequent and can have serious complications such as the incidence of pneumonia. Therefore it is highly relevant to have an effective treatment for these problems. Since FOTT is the standard treatment at the TBIU, Hvidovre Hospital and an integrated part of the rehabilitation programme used to treat patients with problems in eating and drinking it were relevant to explore this approach. The unique feature of this approach compared to other swallowing treatments is that it combines areas related to swallowing: facial movements, oral hygiene, and breathing and speech. This breadth in the treatment approach provides the therapist with many possibilities of addressing the different problems the patient can experience in the facial oral tract area. However, this breadth of FOTT makes it difficult to define, and to know how to separate all the interrelated ingredients. Moreover there is an endless level of detailed ways to describe the components in the treatment and when they should be used. Facing these challenges; we decided to devise a rather broad manual outlining the content of the treatment by showing the "idea" behind FOTT, paying respect to the broadness and flexibility involved in this treatment approach. At the beginning of this work we paid attention mainly to separating the different ingredients and to reaching consensus on how this might be accomplished trying to satisfy the requirements of science and clinical reality, represented by the two developers.

The goal was to describe what FOTT is all about and to ensure that the therapist who follows this manual will use the active ingredients of FOTT, including following the essential decision process. Our decision algorithm accomplished that to some extent. We found a way to separate all the components in four different treatment charts and one assessment chart. The circular decision process is outlined in the treatment charts and the criteria for each decision are described in a following manual to each chart. There is still a great level of flexibility in this manual. It is not that there are many right choices in the decision process *per se*, but there are many possible solutions to the same impairment or performance problem depending on several individual factors in each patient, and that is reflected in this algorithm. However this flexibility may affect the specific guidance of therapeutic behaviour and such standardization of the FOTT approach. Whether or not this is a problem in clinical settings and future studies has to be tested.

It is important for a treatment manual that it not "only" defines the treatment of interest but also that it is possible to implement it in everyday clinical practice. The clearer the therapeutic behaviour is described the easier it might be to implement. Our manual does not describe therapeutic behaviour very specifically. However, it is not the purpose of this manual that it should be a complete guide to how to do FOTT or that it should be able to substitute for FOTT training. Hart[9], claims that one must describe at least some examples of the actual behaviours that would indicate the presence of the active ingredients to an objective observer during treatment sessions. This manual is thought to be a guideline to which components are essential in FOTT and what components belong to which area. Moreover, it should act as a guideline of when to use the different components and how they can be combined.

Though we succeeded in the major step of characterizing FOTT in a decision algorithm, there are still many issues relating to conducting a clinical trial. As described by Whyte and Hart[43], rehabilitation treatments can be characterized at different levels, micro or macro, and we could argue that our algorithm has landed somewhere in between. Hence, whether or not this algorithm is right for research may depend on the question of interest and the ultimate goal of the research. FOTT may be characterized at both broader and narrower levels than in our algorithm; we might want to consider more dimensions in which to characterize for example the use of time, exactly how to specify the chosen ingredients as location and other factors. Moreover, one of the most obvious interesting factors is to define the theory about the active ingredients. A theory will narrow the field for defining and measuring the treatment and specify what is actually hypothesized or desired to proceed in the actual treatment session. In addition, explicit defined theories also make it easier and more meaningful to compare FOTT with other forms of treatment that do not share the same theories of treatment mechanisms [27, 43]. Therefore, supplementing this manual with further theory in line with some of the points made in this thesis' section "FOTT and plasticity" seems highly relevant. However, a treatment manual has little or no value in a clinical study if one does not know whether its active ingredients are present in the treatment investigated- and that the outcome measure used in the study relates to the patients problems and treatment content. This has directed the work of Studies IV and V.

STUDY IV

In this study we developed and validated a measure of adherence to our FOTT decision algorithm. There is no unified measure of adherence and, to our knowledge, none has ever been used within swallowing therapy. Therefore, we designed our own four nominal ratings to assess adherence to our FOTT decision algorithm. We used observations of video recordings to measure whether or not the active ingredients were used in an appropriate way throughout the intervention. We hypothesized that if we found a difference in adherence, when using this coding scheme for a group of therapists specialized in FOTT and another group not familiar with it, our measure was capable of capturing the active ingredients of FOTT. As presented in the manuscript we found a difference in adherence supporting our hypothesis and we conclude that our measure is capable of capturing the active ingredients of FOTT. However, as described in Study III, development of the decision algorithm, this manual does not describe in specific ways the behaviour of the therapist that should be present in order to say if it was used appropriately. This means that measuring adherence at this point is very dependent on the knowledge of the observers. Hence, using this measure in the FOTT algorithm does require some level of knowledge of FOTT and the algorithm. This may be a subject for validity issues and if possible should be established in a different way in the future. This measure should preferably be usable by almost anybody.

The active ingredients should be described in detail in the manual and performed by the therapist in a manner which proves to an observer familiar with the manual whether the active ingredients are present as designed or not[9]. However, this falls back on the discussion in the development of the manual itself. As mentioned here, issues involved in adherence assessment are closely linked with treatment definition[58]. The flexibility and broadness in our algorithm makes it difficult always to measure specific adherence. That gave the dilemma in our study that some therapists achieved the score of “adherence” even it could have been more “FOTT true” to use another component. But it was not exactly “wrong” either to use the one they did. Such complexity and flexibility in FOTT and in this manual, also makes it difficult to have a very precise measure of adherence. Whether or not this issue gives a problem in clinical research depends on the research question of interest. If one wants to know whether or not FOTT is used appropriately in a general manner, as in this study, it seems that this measure clearly shows if the necessary components are used.

Moreover, the most interesting finding in this study might be that FOTT seems to differ significantly from the swallowing therapy method used at a rehabilitation centre in USA, which makes it even more interesting to investigate the effectiveness of FOTT.

STUDY V

The limited time did not allow all authors to get together and evaluate all FEES evaluations as described in the method section of the manuscript; such the result presented is based on the evaluation and writing of the first author only. Before submission to a scientific journal all authors will be involved as described.

This study has developed and validated swallowing safety as one of the many outcomes of FOTT. Specifying the outcomes of treatment is very important because this is the yardstick by which efficacy is ultimately judged[38] and, for patients with severe TBI and their relatives, eating and drinking is for several reasons a very important issue[143]. In this pilot study we developed a clinical evaluation of swallowing in relation to FOTT. This assessment evaluated tongue movements, breathing, breathing and swallowing coordination and the four phases of swallowing: pre-oral phase, oral phase, pharyngeal phase and oesophageal phase.
The assessment summarizes swallowing safety by rating, aspiration, penetration and retention and the recommended food consistencies. In order to know to which extent this clinical evaluation judges the patients swallowing safety in a reliable way we implemented the instrumental evaluation of swallowing: Fibreoptic Endoscopic Evaluation of Swallowing (FEES)[90] at the TBIU. We chose this instrument for swallowing examination at our department, since this method is considered to be one of the “gold standards” of swallowing assessment[86, 144], it can be used bedside for patients not able to be transported to a examination facility and can be done continuously if needed[90]. One physician was trained in this procedure together with the FOTT instructor and me. The OTs at the department was trained in the FOTT clinical evaluation of swallowing and an interrater reliability study was done showing acceptable scores. The FEES was used to test the reliability of the FOTT evaluation regarding: aspiration, penetration and retention. Results showed that the therapist were too careful judging swallowing, meaning that they found that the patients aspirated material when it was not so. This result should be interpreted with caution. First of all we had only 6 patients who aspirated during the FEES evaluation, providing little power to this result. Moreover, the clinical

evaluation and the FEES differ in the procedures that might bias this comparison. The clinical evaluation is performed in another context than the FEES. When performing the FEES, the patient is “warned” in advance that they will go through an evaluation procedure and they are aware that something “different” is going to happen. During the procedure the physician and other professionals that they might not know, are present, a device is described for them and the unknown and unpleasant examination process takes places with an endoscope put in the nose. These factors might affect that the patient is more alert and aware of what is going on (in this case swallowing) than otherwise. In contrast at a clinical evaluation of swallowing, it is a known therapist who performs the procedure and in that way the situation is more similar to “normal” FOTT treatment. There is also the issue that during FEES, the whole procedure takes place as quickly as possible, since it provides the patient with some level of discomfort. The way the patient swallows during 10 minutes in FEES and one hour during a clinical evaluation may also vary. So these factors might also influence the result of this study.

However, it still seems that there is a trend towards that the therapists at the TBIU are a little too careful when judging aspiration or the FOTT evaluation tool is not precise or good enough in its evaluation criteria. Nonetheless, this interpretation is in line with the results found in Study II showing that the patients who developed pneumonia at our department did not get any food or liquid per mouth, meaning that the onset of pneumonia is not due to too early start of oral nutrition. Swallowing safety is a central issue in FOTT, but not the only outcome of interest even when measuring the effect of the treatment in the chart “eating and drinking”. Since swallowing safety in an instrumental setting might be a different thing than swallowing safety in a social context such as a dinner with friends, or just eating for a longer time alone, this study represents the complexity of choosing an outcome measure. Just as treatments may be specified at a variety of levels from more macro to more micro, outcomes, too, may be specified at a variety of levels. This is also discussed in Study I. Here it is chosen to define “swallowing problems” in terms of how the patient receives nutrition, instead of focusing on just swallowing safety. This decision is actually supported by the issues in this paper, where the swallowing safety is just one part of the truth in judging the oral intake of the patients, and the FOIS might better reflect what is important in the “real world”. However, a validated swallowing evaluation is indeed an important issue that should be approached and definitely not neglected. This is highly supported by the findings by other studies also showing

a clear trend that the clinical evaluation of swallowing has too low specificity and sensitivity[80, 83, 145-146].

However, an important finding in this study is that we found a high level of sensitivity and specificity for the items retention and penetration. Both are relevant for swallowing safety, since this is the stage before material enters the lower airway. Since our population groups have high risk of pneumonia and pneumonia is a serious condition for these patients [147-148], one might argue that the clinical evaluation of positive retention and penetration should be the clinical indicator for a FEES examination and/or maybe the limitation of food consistencies.

Chapter 7

FUCTURE DIRECTIONS AND CONCLUSIONS

In this final chapter the main conclusions of the work described in this thesis are summarized and directions for future studies are proposed.

FUTURE DIRECTIONS

This work has produced knowledge that can immediately be useful to clinicians and implemented by them. Moreover, it also points out routes to further research.
The epidemiology studies (I and II) open up investigation into how and to what extent FOTT causes the change found in oral intake and what caused incidence of pneumonia/ and or prevented more.

The algorithm (Study III) also opened the way to several future studies:

- Implementation of the algorithm in different rehabilitation settings
- To test if the manual is capable of changing therapeutic behaviour
- Single-case experimental designs to investigate different hypotheses of treatment mechanism
- Descriptive studies investigating the differences of FOTT from other treatments
- Comparison of the outcome of FOTT with that produced by other treatments addressing the same impairments

From the results in Study V, further evaluation of outcome measures, including a more thorough study of the clinical evaluation of swallowing, is recommended. It will be obvious that this evaluation should be used in a larger study with more patients. Increased numbers of patients will also give better opportunities for investigation of predictive values for the different components included in this evaluation as tongue movements, pre-oral phase etc.

The need to collaborate with others developing methods for research in rehabilitation and/or difference types of swallowing therapy seems apparent. Indeed experiences, knowledge and ideas should be exchanged to facilitate evidence-based treatments in this area.

CONCLUSION

This thesis has explored different steps in clinical research, contributing to the process of efficacy and effectiveness studies in one of the unexplored complex treatment approaches used in neurorehabilitation.

It was found that problems with oral intake are very common within patients with severe TBI (93%), and returning to unrestricted eating and drinking can be predicted by level of consciousness and cognition over a period of 3 months. Moreover, the incidence of complications to swallowing problems, in terms of pneumonia, at the TBIU was 12%. The patients who developed pneumonia were those who received nutrition through feeding tube, exclusively. Both recovery to full oral intake and incidence of pneumonia were found to be associated with severity of injury. What could not be concluded in the epidemiological studies was what may have caused the change in oral intake and incidence and/or prevention of pneumonia. Hence these results made it interesting to explore the treatment involved in this change taking initial steps towards a clinical trial approaching either efficacy or effectiveness study in Facial Oral Tract Therapy (FOTT). These steps involved defining the active ingredients and treatment process in this complex treatment approach in a treatment manual for FOTT. The manual is made as a decision algorithm dividing and defining FOTT in 4 treatment charts, one for each area in FOTT: facial expressions, oral hygiene, swallowing and eating, and breathing voice and speech. The manual represents a balance between flexibility and specificity. It aims to capture the essential ingredients in FOTT, while allowing the possibility of continuously adjusting the treatment to the individual patient's responses (an essential component of FOTT). Moreover, another step in the rehabilitation research venture was developing and testing an adherence measure to the FOTT algorithm. We found that the measure of adherence is able to capture the active ingredients of the chart "swallowing saliva and eating and drinking". We also found that FOTT differs significantly from the swallowing therapy used at MossRehab, USA. Now, having a way to standardize the treatment of interest another necessary step is a valid outcome measure. The last study in this thesis presents the development of a new clinical evaluation of swallowing in FOTT and investigation of reliability and validity. We found a good interrater reliability test for both experienced and inexperienced FOTT evaluators. A gold standard of instrumental evaluation of swallowing (FEES) was implemented at the TBIU as part of this study and used for comparison with the clinical evaluation. It showed that the clinical evaluation overestimates the risk of aspiration, but reliably estimates penetration and retention. More studies

are needed to fully validate this evaluation tool, which seems significant since it showed promising results and is special compared to others since it can be used to patients with low level of functioning. It is highly relevant to these patients to avoid incidence of pneumonia but also to make sure that the patients receives oral nutrition safely as soon as possible.

The studies in this thesis have moved FOTT closer to being evidence based, however, further steps have to be taken before evidence of efficacy and effectiveness can be shown.

The key issue in this thesis has been to approach the challenge to do research in a complex rehabilitation treatment and to open the black box of the therapeutic method. For many reasons, clinical research is different differs from laboratory research; it makes sense to use different research methods for the two instead of trying to adjust the methods from laboratory research to clinical research. It may be time to develop a new set of research rules that are suitable or designed to the uniqueness of rehabilitation research. Whyte [39] states that "Rehabilitation research suffers from a major disadvantage compared to more mainstream biomedical research: relatively few of the treatments under consideration can be claimed as intellectual property, developed by corporations with substantial research and development budgets, or applied to vast numbers of customers to generate revenue." Even though this is true, this issue could be viewed in a slightly differently way by trying to take advantage of the reality of the world of clinical research. One major advantage is that clinicians (therapists such as: occupational therapists, physiotherapist, psychologists etc,) have spent a huge amount of time building up knowledge of the interaction of behaviour and patient response that would never be possible in a laboratory setting. This work is unique and relevant to research. Knowledge developed in the clinic before research is conducted leaves the possibility of studying real life situations by transforming them into the "laboratory". If laboratory research comes first, the theoretical hypothesis might be tested in a phase I and II study and then translated into clinical practice where it might not really fit and therefore not be useful. Studying existing therapies developed in clinical practice or "real life" already fit the clinics and is known by clinicians.

Another valuable aspect is that most clinicians are very dedicated to their jobs and to the rehabilitation process. Thus implementing new knowledge within the treatment that they already believe is the best way to help the patient, or replacing it, demands a change in their behaviour. It is

my belief, that this issue is highly underestimated in the translational process. The research enterprise should take an interest in- and acknowledge the work that is already going on in the clinics.

Going through the different possible research design previous in this thesis, it seems very clear that one study alone will not produce all the desired knowledge of one treatment approach. RCT is most likely not possible in the area of rehabilitation of patients with severe TBI and might not answer all the relevant questions. Thus, providing evidence-based knowledge in rehabilitation should be viewed as a step-wise process before conclusions can be drawn. Well-designed randomized clinical trials are to date the strongest study design for providing efficacy of rehabilitation treatments. However, as argued previously they are rarely feasible, and maybe it is time to build up a new "gold standard" of rehabilitation research design. Indeed it would have been praiseworthy if I had been able to develop such a design for rehabilitation research. Nevertheless, it is hard to imagine that this will be the work of one person; instead it might take a close collaboration of researchers working within this field sharing their knowledge, experiences and ideas.

Finally, outcome measures in this field are also an area that would benefit from further evaluation. Maybe it is time to explore more functional outcomes measure related to the interest of the patients, rather than scales more relevant to clinicians or researchers; as illustrated by the difference in using FOIS vs. aspiration and the FOTT evaluation tool vs. FEES.

Rehabilitation research is a field where methods are still developing. This thesis took some steps towards the translation of the clinical world into science and paved the way for further work in the research enterprise. The work and results are unique in the way that it has explored a complex treatment approach, already implemented in many clinics throughout Europe, and defined it in a treatment manual. This is very rarely done in the field of rehabilitation. In addition, we developed an adherence measure to this manual, essential but not common in rehabilitation research. Moreover, defining an outcome measure has also been initiated. The field of rehabilitation of swallowing problems in patients with severe TBI is still an area where much more research has to be done. This work has contributed to this research enterprise as well as to the methodology in rehabilitation science!

REFERENCES

1. Nudo RJ. Functional and structural plasticity in motor cortex: implications for stroke recovery. *Physical medicine and rehabilitation clinics of North America.* Feb 2003;14(1 Suppl):S57-76.

2. Bach-y-Rita P. Theoretical basis for brain plasticity after a TBI. *Brain Inj.* Aug 2003;17(8):643-651.

3. Robertson IH, Murre JMJ. Rehabilitation of brain damage: Brain plasticity and principles of guided recovery. *Psychol Bull.* 1999;125(5):544-576.

4. Kleim JA, Jones TA. Principles of experience-dependent neural plasticity: implications for rehabilitation after brain damage. *J Speech Lang Hear Res.* Feb 2008;51(1):S225-239.

5. Bütefisch C. Neurobiological bases of rehabilitation. *Neurological Sciences.* 2006;27(0):s18-s23.

6. Nudo RJ. Plasticity. *NeuroRx : the journal of the American Society for Experimental NeuroTherapeutics.* 2006;3(4):420-427.

7. Mogensen J, Hjortkjaer J, Ibervang KL, Stedal K, Mala H. Prefrontal cortex and hippocampus in posttraumatic functional recovery: spatial delayed alternation by rats subjected to transection of the fimbria-fornix and/or ablation of the prefrontal cortex. *Brain Res Bull.* Jun 15 2007;73(1-3):86-95.

8. Mala H, Castro MR, Knippel J, Kohler PJ, Lassen P, Mogensen J. Therapeutic effects of a restraint procedure on posttraumatic place learning in fimbria-fornix transected rats. *Brain Res.* Jun 27 2008;1217:221-231.

9. Hart T. Treatment definition in complex rehabilitation interventions. *Neuropsychol Rehabil.* Jun 18 2009:1-17.

10. Taub E, Uswatte G, Pidikiti R. Constraint-Induced Movement Therapy: a new family of techniques with broad application to physical rehabilitation--a clinical review. *J Rehabil Res Dev.* Jul 1999;36(3):237-251.

11. Mark VW, Taub E. Constraint-induced movement therapy for chronic stroke hemiparesis and other disabilities. *Restor Neurol Neurosci.* 2004;22(3-5):317-336.

12. Cheeran B, Cohen L, Dobkin B, et al. The future of restorative neurosciences in stroke: driving the translational research pipeline from basic science to rehabilitation of people after stroke. *Neurorehabil Neural Repair.* Feb 2009;23(2):97-107.

13. Coombes K. F.O.T.T. (Facial-Oral Tract Therapy). *iADH Magazine.* Vol spring, 2008: International Association For Disability and Oral Health; 2008.

14. Nusser-Müller-Busch. *Die Therapie des Facio-Oralen Trakts - F.O.T.T. nach Kay Coombes.* Vol 2. Berlin: Springer; 2007.

15. Thurman D, Sniezek J, Johnson D, al. e. Guidelines for surveillance of central nercous system injury.In: services DoHaH, ed. Atlante, Georgia: National Center for Injury Prevention and Control, Centers for Disease Control and Prevention, US; 1995.

16. Greenwald BD, Burnett DM, Miller MA. Congenital and acquired brain injury. 1. Brain injury: epidemiology and pathophysiology. *Arch Phys Med Rehabil.* Mar 2003;84(3 Suppl 1):S3-7.

17. Michaud CM, Murray CJ, Bloom BR. Burden of disease--implications for future research. *JAMA.* Feb 7 2001;285(5):535-539.

18. Braverman SE, Spector J, Warden DL, et al. A multidisciplinary TBI inpatient rehabilitation programme for active duty service members as part of a randomized clinical trial. *Brain Inj.* Jun 1999;13(6):405-415.
19. Semlyen JK, Summers SJ, Barnes MP. Traumatic brain injury: efficacy of multidisciplinary rehabilitation. *Arch Phys Med Rehabil.* Jun 1998;79(6):678-683.
20. Shiel A, Burn JP, Henry D, et al. The effects of increased rehabilitation therapy after brain injury: results of a prospective controlled trial. *Clin Rehabil.* Oct 2001;15(5):501-514.
21. Engberg AW, Liebach A, Nordenbo A. Centralized rehabilitation after severe traumatic brain injury - a population-based study. *Acta Neurol Scand.* 2006;113(3):178-184.
22. Liebach A, Nordenbo AM, Engberg AW. [Subacute intensive rehabilitation after severe traumatic brain injury. Follow-up 6 months after discharge]. *Ugeskr Laeger.* Jan 15 2007;169(3):223-227.
23. Sundhedsstyrelsen. Behandling af traumatiske hjerneskader og tilgrænsende lidelser. . In: Health) NBo, ed. København, Denmark: Komiteen for Sundhedsoplysning; 1997.
24. Engberg A.W, Teasdale TW. Traumatic brain injury in Denmark 1979-1996. A national study of incidence and mortality. *Eur J Epidemiol.* 2001;17(5):437-442.
25. Whyte J. Rehabilitation of Individuals With Traumatic Brain Injury: Status of the Art and Science. *Rehabilitation of Persons With Traumatic Brain Injury.* Vol 16: NIH Consens Statement; 1998:23-30.
26. Nezu AM, Nezu CM. *Evidence-based outcome research*. New York2008.
27. Whyte J. A Grand Unified Theory of Rehabilitation (We Wish!). The 57th John Stanley Coulter Memorial Lecture. *Archives of Physical Medicine and Rehabilitation.* 2008;89(2):203-209.
28. Cope DN. The effectiveness of traumatic brain injury rehabilitation: a review. *Brain Inj.* Oct 1995;9(7):649-670.
29. Carney N, Chesnut RM, Maynard H, Mann NC, Patterson P, Helfand M. Effect of cognitive rehabilitation on outcomes for persons with traumatic brain injury: A systematic review. *J Head Trauma Rehabil.* Jun 1999;14(3):277-307.
30. Rice-Oxley M, Turner-Stokes L. Effectiveness of brain injury rehabilitation. *Clin Rehabil.* 1999;13 Suppl 1:7-24.
31. Cullen N, Chundamala J, Bayley M, Jutai J. The efficacy of acquired brain injury rehabilitation. *Brain Inj.* Feb 2007;21(2):113-132.
32. Greenwald BD, Rigg JL. Neurorehabilitation in traumatic brain injury: does it make a difference? *Mount Sinai Journal of Medicine: A Journal of Translational and Personalized Medicine.* 2009;76(2):182-189.
33. Gordon WA, Zafonte R, Cicerone K, et al. Traumatic brain injury rehabilitation: state of the science. *Am J Phys Med Rehabil.* Apr 2006;85(4):343-382.
34. Johnston MV, Sherer M, Whyte J. Applying evidence standards to rehabilitation research. *Am J Phys Med Rehabil.* Apr 2006;85(4):292-309.
35. Sackett DL, Rosenberg WM, Gray JA, Haynes RB, Richardson WS. Evidence based medicine: what it is and what it isn't. *BMJ.* Jan 13 1996;312(7023):71-72.
36. Moses LE. Measuring effects without randomized trials? Options, problems, challenges. *Med Care.* Apr 1995;33(4 Suppl):AS8-14.
37. Powell JM, Temkin NR, Machamer JE, Dikmen SS. Nonrandomized studies of rehabilitation for traumatic brain injury: Can they determine effectiveness? *Arch Phys Med Rehabil.* 2002;83(9):1235-1244.
38. Whyte J. Clinical trials in rehabilitation: what are the obstacles? *American journal of physical medicine & rehabilitation* Oct 2003;82(10 Suppl):S16-21.

39. Whyte J. Directions in brain injury research: From concept to clinical implementation. *Neuropsychol Rehabil.* Jun 19 2009:1-17.
40. Grimes DA, Schulz KF. An overview of clinical research: the lay of the land. *Lancet.* Jan 5 2002;359(9300):57-61.
41. Perdices M, Tate RL. Single-subject designs as a tool for evidence-based clinical practice: Are they unrecognised and undervalued? *Neuropsychol Rehabil.* Jul 28 2009:1-24.
42. Tate RL, McDonald S, Perdices M, Togher L, Schultz R, Savage S. Rating the methodological quality of single-subject designs and n-of-1 trials: introducing the Single-Case Experimental Design (SCED) Scale. *Neuropsychol Rehabil.* Aug 2008;18(4):385-401.
43. Whyte J, Hart T. It's more than a black box; it's a Russian doll: defining rehabilitation treatments. *American journal of physical medicine & rehabilitation* Aug 2003;82(8):639-652.
44. (WHO) WHO. International Classification of Functioning, Disability, and Health (ICF). . [http://www.who.int/classifications/icf/en/]. Accessed October 7, 2009.
45. Koskinen S, Hokkinen EM, Sarajuuri J, Alaranta H. Applicability of the ICF checklist to traumatically brain-injured patients in post-acute rehabilitation settings. *J Rehabil Med.* Jul 2007;39(6):467-472.
46. Bernabeu M, Laxe S, Lopez R, et al. Developing core sets for persons with traumatic brain injury based on the international classification of functioning, disability, and health. *Neurorehabil Neural Repair.* Jun 2009;23(5):464-467.
47. Roland PE, Hanazawa A, Undeman C, et al. Cortical feedback depolarization waves: a mechanism of top-down influence on early visual areas. *Proc Natl Acad Sci U S A.* Aug 15 2006;103(33):12586-12591.
48. DeJong G, Horn SD, Conroy B, Nichols D, Healton EB. Opening the Black Box of Poststroke Rehabilitation: Stroke Rehabilitation Patients, Processes, and Outcomes. *Archives of Physical Medicine and Rehabilitation.* 2005;86(12):1-7.
49. Whyte J. Clinical trials in rehabilitation: what are the obstacles? *Am J Phys Med Rehabil.* Oct 2003;82(10 Suppl):S16-21.
50. Hart T. Treatment definition in experience-based rehabilitation research.
51. Addis ME, Wade WA, Hatgis C. Barriers to Dissemination of Evidence-Based Practices: Addressing Practitioners' Concerns About Manual-Based Psychotherapies. *Clinical Psychology: Science and Practice.* 1999;6(4):430-441.
52. Schenkman M, Deutsch JE, Gill-Body KM. An Integrated Framework for Decision Making in Neurologic Physical Therapist Practice. *Physical Therapy.* 2006;86(12):1681-1702.
53. Rothstein JM, Echternach JL. Hypothesis-oriented algorithm for clinicians. A method for evaluation and treatment planning. *Physical Therapy.* Sep 1986;66(9):1388-1394.
54. Dejong G, Horn SD, Gassaway JA, Slavin MD, Dijkers MP. Toward a taxonomy of rehabilitation interventions: Using an inductive approach to examine the "black box" of rehabilitation. *Archives of Physical Medicine and Rehabilitation.* Apr 2004;85(4):678-686.
55. Ballinger C, Ashburn A, Low J, Roderick P. Unpacking the black box of therapy -- a pilot study to describe occupational therapy and physiotherapy interventions for people with stroke. *Clin Rehabil.* Aug 1999;13(4):301-309.
56. Hogue A, Liddle HA, Rowe C. Treatment adherence process research in family therapy: A rationale and some practical guidelines. *Psychotherapy: Theory, Research, Practice, Training.* Sum 1996;33(2):332-345.
57. Bellg AJ, Borrelli B, Resnick B, et al. Enhancing treatment fidelity in health behavior change studies: best practices and recommendations from the NIH Behavior Change Consortium. *Health Psychol.* Sep 2004;23(5):443-451.

58. Hogue A. Adherence process research on developmental interventions: filling in the middle. *New Dir Child Adolesc Dev.* Winter 2002(98):67-74.
59. Granger CV, Hamilton BB, Linacre JM, Heinemann AW, Wright BD. Performance profiles of the functional independence measure. *Am J Phys Med Rehabil.* Apr 1993;72(2):84-89.
60. Linacre JM, Heinemann AW, Wright BD, Granger CV, Hamilton BB. The structure and stability of the Functional Independence Measure. *Arch Phys Med Rehabil.* Feb 1994;75(2):127-132.
61. Wade DT, Smeets RJ, Verbunt JA. Research in rehabilitation medicine: Methodological challenges. *J Clin Epidemiol.* Sep 27 2009.
62. Draper K, Ponsford J. Cognitive functioning ten years following traumatic brain injury and rehabilitation. *Neuropsychology.* Sep 2008;22(5):618-625.
63. Kwakkel G, van Peppen R, Wagenaar RC, et al. Effects of augmented exercise therapy time after stroke: a meta-analysis. *Stroke.* Nov 2004;35(11):2529-2539.
64. Dodds TA, Martin DP, Stolov WC, Deyo RA. A validation of the functional independence measurement and its performance among rehabilitation inpatients. *Arch Phys Med Rehabil.* May 1993;74(5):531-536.
65. Jenkinson C, Wright L, Coulter A. The SF 36 health survey questionnaire. ...if used within its limits. *BMJ.* Aug 14 1993;307(6901):449.
66. Hamdy S, Aziz Q, Thompson DG, Rothwell JC. Physiology and pathophysiology of the swallowing area of human motor cortex. *Neural Plast.* 2001;8(1-2):91-97.
67. Logemann JA. *Evaluation and treatment of swallowing disorders.* Vol 2: PRO-ED, Inc.; 1983.
68. Leopold NA, Kagel MC. Dysphagia--Ingestion or Deglutition?: A Proposed Paradigm. *Dysphagia.* 1997;12(4):202-206.
69. Coombes K. Facial oral tract therapy (F.O.T.T.). *1991-2001 Jubiläumsschrift-10 Jahre Schulungszentrum, Therapie Zentrum Burgau*: 2001.
70. Kaplan MD, Baum BJ. The functions of saliva. *Dysphagia.* 1993;8(3):225-229.
71. Dodds WJ. Physiology of swallowing. *Dysphagia.* 1989;3(4):171-178.
72. Kahrilas PJ, Lin S, Logemann JA, Ergun GA, Facchini F. Deglutitive tongue action: volume accommodation and bolus propulsion. *Gastroenterology.* Jan 1993;104(1):152-162.
73. Mackay LE, Morgan AS, Bernstein BA. Swallowing disorders in severe brain injury: risk factors affecting return to oral intake. *Archives of Physical Medicine and Rehabilitation.* Apr 1999;80(4):365-371.
74. Cherney LR, Halper, AS. Recovery of oral nutrition after head injury in adults. *J head trauma rehabil.* 1989;4(4):42-50.
75. Winstein CJ. Neurogenic dysphagia. Frequency, progression, and outcome in adults following head injury. *Phys Ther.* Dec 1983;63(12):1992-1997.
76. Hansen TS, Engberg AW, Larsen K. Functional oral intake and time to reach unrestricted dieting for patients with traumatic brain injury. *Arch Phys Med Rehabil.* Aug 2008;89(8):1556-1562.
77. Terre R, Mearin F. Prospective evaluation of oro-pharyngeal dysphagia after severe traumatic brain injury. *Brain Inj.* Dec 2007;21(13-14):1411-1417.
78. Avery-Smith W, Dellarosa DM. Approaches to treating dysphagia in patients with brain injury. *Am J Occup Ther.* Mar 1994;48(3):235-239.
79. Murdoch B, Theodoros D. *Traumatic Brain Injury: Associated Speech, Language, and Swallowing Disorders* San Diego, CA: Singular Thomson Learning 2001.

80. McCullough GH, Wertz RT, Rosenbek JC. Sensitivity and specificity of clinical/bedside examination signs for detecting aspiration in adults subsequent to stroke. *J Commun Disord.* Jan-Apr 2001;34(1-2):55-72.
81. Silva AB, Piovesana AM, Barcelos IH, Capellini SA. [Clinical and videofluoroscopic evaluation of swallowing in patients with spastic tetraparetic cerebral palsy and athetosic cerebral palsy]. *Rev Neurol.* Apr 16-30 2006;42(8):462-465.
82. Smith HA, Lee SH, O'Neill PA, Connolly MJ. The combination of bedside swallowing assessment and oxygen saturation monitoring of swallowing in acute stroke: a safe and humane screening tool. *Age Ageing.* Nov 2000;29(6):495-499.
83. Smithard DG, O'Neill PA, Park C, et al. Can bedside assessment reliably exclude aspiration following acute stroke? *Age Ageing.* Mar 1998;27(2):99-106.
84. Lim SH, Lieu PK, Phua SY, et al. Accuracy of bedside clinical methods compared with fiberoptic endoscopic examination of swallowing (FEES) in determining the risk of aspiration in acute stroke patients. *Dysphagia.* Winter 2001;16(1):1-6.
85. Logemann JA, Pauloski BR, Rademaker AW, et al. Speech and swallow function after tonsil/base of tongue resection with primary closure. *J Speech Hear Res.* Oct 1993;36(5):918-926.
86. Langmore SE, Schatz K, Olson N. Endoscopic and videofluoroscopic evaluations of swallowing and aspiration. *Ann Otol Rhinol Laryngol.* Aug 1991;100(8):678-681.
87. Hoppers P, Holm SE. The role of fiberoptic endoscopy in dysphagia rehabilitation. *J Head Trauma Rehabil.* Oct 1999;14(5):475-485.
88. Hiss SGPCCCSLP, Postma GNMD. Fiberoptic Endoscopic Evaluation of Swallowing. [Article].
89. Wu CH, Hsiao TY, Chen JC, Chang YC, Lee SY. Evaluation of swallowing safety with fiberoptic endoscope: comparison with videofluoroscopic technique. *Laryngoscope.* Mar 1997;107(3):396-401.
90. Langmore SE, Schatz K, Olsen N. Fiberoptic endoscopic examination of swallowing safety: a new procedure. *Dysphagia.* 1988;2(4):216-219.
91. Ashford J, McCabe D, Wheeler-Hegland K, et al. Evidence-based systematic review: Oropharyngeal dysphagia behavioral treatments. Part III--Impact of dysphagia treatments on populations with neurological disorders. *Journal of Rehabilitation Research & Development.* 2009;46(2):195-204.
92. Robbins J, Gensler G, Hind J, et al. Comparison of 2 interventions for liquid aspiration on pneumonia incidence: a randomized trial. *Ann Intern Med.* Apr 1 2008;148(7):509-518.
93. Shanahan TK, Logemann JA, Rademaker AW, Pauloski BR, Kahrilas PJ. Chin-down posture effect on aspiration in dysphagic patients. *Arch Phys Med Rehabil.* Jul 1993;74(7):736-739.
94. Nagaya M, Kachi T, Yamada T, Sumi Y. Videofluorographic observations on swallowing in patients with dysphagia due to neurodegenerative diseases. *Nagoya J Med Sci.* May 2004;67(1-2):17-23.
95. Okada S, Saitoh E, Palmer JB, et al. What is the chin-down posture? A questionnaire survey of speech language pathologists in Japan and the United States. *Dysphagia.* Jul 2007;22(3):204-209.
96. Logemann JA, Kahrilas PJ, Kobara M, Vakil NB. The benefit of head rotation on pharyngoesophageal dysphagia. *Arch Phys Med Rehabil.* Oct 1989;70(10):767-771.
97. Drake W, Donoghue SO, Bartram C, Lindsay J, Greenwood R. Case study Eating in side lying facilitates rehabilitation in neurogenic dysphagia. *Brain Injury.* 1997;11(2):137-142.

98. Lazarus CL, Logemann JA, Rademaker AW, et al. Effects of bolus volume, viscosity, and repeated swallows in nonstroke subjects and stroke patients. *Arch Phys Med Rehabil.* Oct 1993;74(10):1066-1070.
99. de Lama Lazzara G, Lazarus C, Logemann J. Impact of thermal stimulation on the triggering of the swallowing reflex. *Dysphagia.* 1986;1(2):73-77.
100. Tsukada T, Taniguchi H, Ootaki S, Yamada Y, Inoue M. Effects of food texture and head posture on oropharyngeal swallowing. *J Appl Physiol.* June 1, 2009 2009;106(6):1848-1857.
101. Logemann JA, Gensler G, Robbins J, et al. A randomized study of three interventions for aspiration of thin liquids in patients with dementia or Parkinson's disease. *J Speech Lang Hear Res.* Feb 2008;51(1):173-183.
102. Logemann JA. Dysphagia: evaluation and treatment. *Folia Phoniatr Logop.* 1995;47(3):140-164.
103. Lazarus C, Logemann JA, Gibbons P. Effects of maneuvers on swallowing function in a dysphagic oral cancer patient. *Head Neck.* Sep-Oct 1993;15(5):419-424.
104. Poertner LC, Coleman RF. Swallowing therapy in adults. *Otolaryngol Clin North Am.* Jun 1998;31(3):561-579.
105. Yuen HK, Hartwick JA. Diet manipulation to resume regular food consumption for an adult with traumatic brain injury. *Am J Occup Ther.* Oct 1992;46(10):943-945.
106. Tippett D, Palmer J, Linden P. Management of dysphagia in a patient with closed head injury. *Dysphagia.* 1987;1(4):221-226.
107. Nusser-Muller-busch R. *Konsensusempfehlungen 2007 zur Facio-Oralen Trakt Therapie (F.O.T.T.)*2007.
108. Seidl RO, Nusser-Muller-Busch R, Hollweg W, Westhofen M, Ernst A. Pilot study of a neurophysiological dysphagia therapy for neurological patients. *Clinical Rehabilitation.* Aug 2007;21(8):686-697.
109. Raine S. Defining the Bobath concept using the Delphi technique. *Physiotherapy research international.* Mar 2006;11(1):4-13.
110. Coombes K. Facial Oral Tract Therapy: F.O.T.T™. [http://www.arcos.org.uk/index.php?option=com_content&view=article&id=60&Itemid=66]. http://www.fott.co.uk/. Accessed September, 2009.
111. Martin BJ, Logemann JA, Shaker R, Dodds WJ. Coordination between respiration and swallowing: respiratory phase relationships and temporal integration. *Journal of Applied Physiology.* Feb 1994;76(2):714-723.
112. Nusser-Muller-Busch R, Seidl RO. Therapie- und Tracheal-kanülemangement bei neurogenen dysphagien (phase A und B) eine Fallbesceibung. *Forum logopädie.* 2004;6(18):20-26.
113. Frank JS, Earl M. Coordination of posture and movement. *Physical Therapy.* December 1, 1990 1990;70(12):855-863.
114. De Mayo T, Miralles R, Barrero D, et al. Breathing type and body position effects on sternocleidomastoid and suprahyoid EMG activity. *Journal of Oral Rehabilitation.* 2005;32(7):487-494.
115. Ayuse T, Ishitobi S, Kurata S. Effect of reclining and chin-tuck position on the coordination between respiration and swallowing. *Journal of Oral Rehabilitation.* 2006;33(6):402-409.
116. Robbins J, Butler SG, Daniels SK, et al. Swallowing and dysphagia rehabilitation: translating principles of neural plasticity into clinically oriented evidence. *J Speech Lang Hear Res.* Feb 2008;51(1):S276-300.

117. Jones TA, Chu CJ, Grande LA, Gregory AD. Motor skills training enhances lesion-induced structural plasticity in the motor cortex of adult rats. *J Neurosci.* Nov 15 1999;19(22):10153-10163.
118. Biernaskie J, Corbett D. Enriched rehabilitative training promotes improved forelimb motor function and enhanced dendritic growth after focal ischemic injury. *J Neurosci.* Jul 15 2001;21(14):5272-5280.
119. Farrell R, Evans S, Corbett D. Environmental enrichment enhances recovery of function but exacerbates ischemic cell death. *Neuroscience.* 2001;107(4):585-592.
120. Mulder T, Hochstenbach J. Adaptability and flexibility of the human motor system: implications for neurological rehabilitation. *Neural Plasticity.* 2001;8(1-2):131-140.
121. Kleim JA, Barbay S, Cooper NR, et al. Motor learning-dependent synaptogenesis is localized to functionally reorganized motor cortex. *Neurobiol Learn Mem.* Jan 2002;77(1):63-77.
122. Dimyan MA, Weinberger NM. Basal forebrain stimulation induces discriminative receptive field plasticity in the auditory cortex. *Behav Neurosci.* Aug 1999;113(4):691-702.
123. Fraser C, Rothwell J, Power M, Hobson A, Thompson D, Hamdy S. Differential changes in human pharyngoesophageal motor excitability induced by swallowing, pharyngeal stimulation, and anesthesia. *Am J Physiol Gastrointest Liver Physiol.* Jul 2003;285(1):G137-144.
124. Hamdy S, Aziz Q, Rothwell JC, et al. Explaining oropharyngeal dysphagia after unilateral hemispheric stroke. *Lancet.* Sep 6 1997;350(9079):686-692.
125. Rosenbek JC, Robbins J, Willford WO, et al. Comparing treatment intensities of tactile-thermal application. *Dysphagia.* Winter 1998;13(1):1-9.
126. Maegele M, Lippert-Gruener M, Ester-Bode T, et al. Multimodal early onset stimulation combined with enriched environment is associated with reduced CNS lesion volume and enhanced reversal of neuromotor dysfunction after traumatic brain injury in rats. *Eur J Neurosci.* May 2005;21(9):2406-2418.
127. Lippert-Gruner M, Maegele M, Pokorny J, et al. Early rehabilitation model shows positive effects on neural degeneration and recovery from neuromotor deficits following traumatic brain injury. *Physiol Res.* 2007;56(3):359-368.
128. Page S, Levine P. Forced use after TBI: promoting plasticity and function through practice. *Brain Inj.* Aug 2003;17(8):675-684.
129. Taub E, Uswatte G, Mark VW, Morris DM. The learned nonuse phenomenon: implications for rehabilitation. *Eura Medicophys.* Sep 2006;42(3):241-256.
130. Risedal A, Zeng J, Johansson BB. Early training may exacerbate brain damage after focal brain ischemia in the rat. *J Cereb Blood Flow Metab.* Sep 1999;19(9):997-1003.
131. Humm JL, Kozlowski DA, James DC, Gotts JE, Schallert T. Use-dependent exacerbation of brain damage occurs during an early post-lesion vulnerable period. *Brain Res.* Feb 9 1998;783(2):286-292.
132. Boyd L, Winstein C. Explicit information interferes with implicit motor learning of both continuous and discrete movement tasks after stroke. *J Neurol Phys Ther.* Jun 2006;30(2):46-57; discussion 58-49.
133. Taub E, Uswatte G. Constraint-induced movement therapy: bridging from the primate laboratory to the stroke rehabilitation laboratory. *J Rehabil Med.* May 2003(41 Suppl):34-40.
134. Sunderland A, Tuke A. Neuroplasticity, learning and recovery after stroke: a critical evaluation of constraint-induced therapy. *Neuropsychol Rehabil.* May 2005;15(2):81-96.

135. Schurr MJ, Ebner KA, Maser AL, Sperling KB, Helgerson RB, Harms B. Formal swallowing evaluation and therapy after traumatic brain injury improves dysphagia outcomes. *J Trauma.* May 1999;46(5):817-821; discussion 821-813.
136. Crary MA, Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. *Arch Phys Med Rehabil.* Aug 2005;86(8):1516-1520.
137. Chong MS, Lieu PK, Sitoh YY, Meng YY, Leow LP. Bedside clinical methods useful as screening test for aspiration in elderly patients with recent and previous strokes. *Ann Acad Med Singapore.* Nov 2003;32(6):790-794.
138. Leder SB, Espinosa JF. Aspiration risk after acute stroke: comparison of clinical examination and fiberoptic endoscopic evaluation of swallowing. *Dysphagia.* Summer 2002;17(3):214-218.
139. Logemann JA. Do we know what is normal and abnormal airway protection? *Dysphagia.* Fall 1999;14(4):233-234.
140. Terre R, Mearin F. Evolution of tracheal aspiration in severe traumatic brain injury-related oropharyngeal dysphagia: 1-year longitudinal follow-up study. *Neurogastroenterol Motil.* Apr 2009;21(4):361-369.
141. Ahtaridis G, Snape WJ, Jr., Cohen S. Lower esophageal sphincter pressure as an index of gastroesophageal acid reflux. *Dig Dis Sci.* Nov 1981;26(11):993-998.
142. Leibovitz A, Carmeli Y, Segal R. Effect of various antibacterial preparations on the pathogenic oral flora in elderly patients fed via nasogastric tube. *Antimicrob Agents Chemother.* Aug 2005;49(8):3566-3568.
143. Cook AM, Peppard A, Magnuson B. Nutrition considerations in traumatic brain injury. *Nutr Clin Pract.* Dec-2009 Jan 2008;23(6):608-620.
144. Langmore SE. Evaluation of oropharyngeal dysphagia: which diagnostic tool is superior? *Curr Opin Otolaryngol Head Neck Surg.* Dec 2003;11(6):485-489.
145. Logemann JA, Veis S, Colangelo L. A screening procedure for oropharyngeal dysphagia. *Dysphagia.* Winter 1999;14(1):44-51.
146. Bours GJ, Speyer R, Lemmens J, Limburg M, de Wit R. Bedside screening tests vs. videofluoroscopy or fibreoptic endoscopic evaluation of swallowing to detect dysphagia in patients with neurological disorders: systematic review. *J Adv Nurs.* Mar 2009;65(3):477-493.
147. Bronchard R, Albaladejo P, Brezac G, et al. Early onset pneumonia: risk factors and consequences in head trauma patients. *Anesthesiology.* Feb 2004;100(2):234-239.
148. Woratyla SP, Morgan AS, Mackay L, Bernstein B, Barba C. Factors associated with early onset pneumonia in the severely brain-injured patient. *Conn Med.* Nov 1995;59(11):643-647.

APPENDIX:

I) Hansen, T.S., Engberg, A.W., Larsen K. Functional Oral Intake and time to reach unrestricted dieting for patients with severe Traumatic Brian Injury. Archives of Physical Medicine and rehabilitation: 2008; 89, 8, 1556-62

II) Hansen, T.S., Larsen, K., Engberg A.W. The association of functional oral intake and pneumonia in patients with severe TBI. Archives of Physical Medicine and Rehabilitation: 2008; 89, 11, 2114-20

III) Hansen, T.S., Jakobsen, D. A decision algorithm defining the rehabilitation approach: Facial Oral Tract Therapy (FOTT) (Accepted for publication 14.december 2009 in the journal: Disability and Rehabilitation)

IV) Hansen, T.S., Jakobsen, D., Nowack, D'A, Whyte, J. Development of an adherence measure for a complex neurorehabilitation approach (submitted to the American Journal of Physical Medicine and Rehabilitation)

V) Hansen, T.S., Jakobsen, D., Westergaard, L., Speyer, R. FOTT versus FEES. A clinical evaluation of swallowing, feasible for patients with severe TBI and low level of functioning (preliminary manuscript)

Study I:

Hansen, T.S., Engberg, A.w., Larsen, K.

Functional Oral Intake and time to reach unrestricted dieting for patients with severe Traumatic Brian Injury.

Archives of Physical Medicine and rehabilitation: 2008; 89, 8, 1556-62

ORIGINAL ARTICLE

Functional Oral Intake and Time to Reach Unrestricted Dieting for Patients With Traumatic Brain Injury

Trine S. Hansen, MSci, MPH, Aase W. Engberg, DMSc, Klaus Larsen, PhD

ABSTRACT. Hansen TS, Engberg AW, Larsen K. Functional oral intake and time to reach unrestricted dieting for patients with traumatic brain injury. Arch Phys Med Rehabil 2008;89:1556-62.

Objectives: To investigate the status of functional oral intake for patients with severe traumatic brain injury (TBI) and time to return to unrestricted dieting; and to investigate whether severity of brain injury is a predictor for unrestricted dieting.

Design: Observational retrospective cohort study.

Setting: Subacute rehabilitation department, university hospital.

Participants: Patients age 16 to 65 years (N=173) with severe TBI (posttraumatic amnesia from 7d to >6mo) admitted over a 5-year period. Patients are transferred to the brain injury unit as soon as they ventilate spontaneously.

Intervention: Facial oral tract therapy.

Main Outcome Measure: Unrestricted dieting assessed by the Functional Oral Intake Scale (FOIS).

Results: We found that 93% of all patients had problems with functional oral intake at admission. Within 126 days of rehabilitation, 64% recovered to unrestricted dieting before discharge. The chance of returning to total oral diet depends on the severity of the brain injury and can be predicted by Glasgow Coma Scale (GCS; measured the day after cessation of sedation; Wald χ^2=42.78, P<.01), Rancho Los Amigos Scale (RLAS) level (Wald χ^2=11.84, P=.01), FIM instrument (Wald χ^2=44.40, P<.01), and FOIS score at admission (Wald χ^2=82.93, P<.01).

Conclusions: Impairment in functional oral intake was found to be very common for patients with severe TBI admitted to a subacute rehabilitation department. For those who recovered during hospital rehabilitation, return to unrestricted dieting happened within 126 days of rehabilitation. The chance of returning to unrestricted dieting depends on the severity of the brain injury and can be predicted by GCS score, RLAS level, FIM score, and functional oral intake at admission. These results are important when planning rehabilitation, giving information to patients and relatives, and designing efficacy studies of facial oral tract therapy, which are highly recommended.

Key Words: Brain injuries; Deglutition disorders; Rehabilitation.

PATIENTS WITH TBI are at risk of developing problems with swallowing, eating, drinking, and verbal and nonverbal communication.[1,2] These problems are often the result of a number of neurologic dysfunctions in postural tone or tone in the face, mouth, or throat, and can also be a result of cognitive and behavioral problems.[3,4] In patients with severe TBI, the incidence of swallowing problems has been reported as high as 61%,[5] verified by videofluoroscopy, and was found to be associated with injury severity.[3,5,6] Ward et al[6] recently investigated patients with TBI and dysphagia in an acute care setting and found that 55 of 117 patients achieved normal diet before discharge in an average of 22 days. Duration to the first clinical swallowing evaluation was found to be a predictor for achieving total oral intake. Results were based only on clinical evaluation of swallowing. Winstein[7] earlier reported data from patients with TBI at a later stage of rehabilitation, showing that 55% achieved normal oral diet. The average time to successful completion of 1 oral meal was 13 weeks. In recent years, the number of specialized subacute rehabilitation units for severe TBI has increased, and the positive effects of intensive interdisciplinary rehabilitation are documented.[8] In this early stage of recovery, prediction of outcome and the duration of recovery are of great importance to support clinical decision-making and provide realistic expectations to relatives. Despite this, only 1 study reported data of swallowing prognosis in a nonacute rehabilitation setting for patients with TBI.[7]

Swallowing problems are most often described in terms of swallowing physiology evaluated by videofluoroscopy and/or fiberoptic evaluation of swallowing relevant for the professionals who deal with these problems. However, cognition and level of consciousness disorders have also been found to affect oral intake.[3,9,10] From a practical point of view, in our opinion, the most important issue is how and how much the patient can eat and drink by mouth. Assessment of this is possible using a functional rating scale like the FOIS.[11] This scale measures the level of oral intake on a daily basis and has been found to be sensitive to change in oral intake over time. The duration of recovery is both a matter of quality of life for the patients and a matter of costs and time spent with swallowing therapy.[10,12] Therefore, it is interesting to investigate predictors of the duration and the chance of reaching unrestricted dieting.

List of Abbreviations

FOIS	Functional Oral Intake Scale
GCS	Glasgow Coma Scale
IQR	interquartile range
LOS	length of stay
PEG	percutaneous endoscopic gastrostomy
PTA	posttraumatic amnesia
RLAS	Ranchos Los Amigos Scale
TBI	traumatic brain injury

From the Departments of Neurorehabilitation, Brain Injury Unit (Hansen, Engberg), Occupational Therapy (Hansen), and Clinical Research Unit (Larsen), Copenhagen University Hospital Hvidovre, Hvidovre, Denmark.

No commercial party having a direct financial interest in the results of the research supporting this article has or will confer a benefit upon the authors or upon any organization with which the authors are associated.

Correspondence to Trine S. Hansen, MSci, Hvidovre Hospital, Dept of Neurorehabilitation 354, Kettegaard Allé 30, DK-2650 Hvidovre, Denmark, email: *trine.schow@gmail.com*. Reprints are not available from the author.

0003-9993/08/8908-00641$34.00/0
doi:10.1016/j.apmr.2007.11.063

The purpose of the present study was to investigate the incidence, status, and time to recovery of functional oral intake (unrestricted dieting) for patients admitted to an early subacute brain injury rehabilitation unit and to investigate brain injury severity as a predictor for return to unrestricted dieting.

METHODS

This present study is a retrospective observational study using data collected from hospital records and chart reviews. This study was approved by the Danish National Committee on Biomedical Research Ethics (the Copenhagen regional committee) and the Danish Data Protection Agency.

Participants

From October 2000, the subacute rehabilitation of all severely injured patients with TBI in Denmark was centralized to 2 units, each with geographically defined uptake areas.

The present study includes patients from the uptake area of the Copenhagen center—that is, the eastern half of Denmark, Greenland, and the Faeroe Islands, with a total of 2.4 million inhabitants. All hospitals in the uptake area, and in particular the only 2 neurosurgical clinics in the area, agreed to refer patients fulfilling the following criteria: highest priority was given to patients who, after initial treatment in a neurosurgical or other clinic, had a GCS[13] score in the range 3 to 12, 1 day after cessation of sedation. All such survivors were transferred as soon as they ventilated spontaneously. The median time spent in acute care was 15 days (range, 3–150d). The brain injury severity was confirmed by prospective assessment of the PTA period. All 177 patients age 16 to 65 years, admitted over a 5-year period, from October 2000 to December 2005, were evaluated for eligibility. We excluded patients on the basis of previously known swallowing problems because of neurologic diseases or other diagnoses. Four patients met these criteria, and 173 patients were included in the study. Patients younger than 65 years were excluded because swallowing function can be affected in older age groups of otherwise healthy people.[14]

According to the rehabilitation program of the brain injury unit,[8] all patients are enrolled in an extensive around-the-clock rehabilitation program by interdisciplinary teams starting on the day of admission. Functional rehabilitation is based on the treatment concepts: Affolter and Stricker,[15] Bobath,[16] and Coombes.[17,18] The emphasis lies on sensory stimulation, facilitating normal movements and daily activities even for patients in vegetative state. Discharge is decided on when the patient is (1) able to go home, (2) able to continue the rehabilitation in a local and less intensive setting, or (3) referred to a nursing home if no progress was made at all for a 3-month period.[8]

Swallowing Therapy

A clinical evaluation of swallowing is performed on the day of admission in line with the treatment concept of facial oral tract therapy.[17,18] This concept was developed by speech and language therapist Kay Coombes and provides a structured way to assess and treat disturbances in facial expression, movement of the jaw for eating and articulation, breathing, swallowing, and voice.[17] Treatment methods include slow, organized touch of the patient's hands, facilitating hand-to-hand and hand-to-face contact, together with specific oral stimulation, therapeutic oral hygiene routines, and facilitation of swallowing. Facial oral tract therapy does not require that the patients are capable of following instructions. Therefore, patients with a very low level of consciousness also receive facial oral tract therapy. For example, in the beginning, they will be given treatment with oral stimulation and therapeutic eating (small amounts of food given in the treatment session). In the evaluation of swallowing ad modum, Coombes covers 4 phases: the preoral phase (involves anticipatory saliva production in response to seeing and smelling food or drink, and bringing food and liquid to the mouth), the oral phase (bolus formation and transport to the back of the mouth), the pharyngeal phase (transport of bolus through pharynx from the mouth to esophagus), and the esophagus phase (transport of bolus through esophagus to the stomach).[19] All patients with impairments in the mentioned areas are enrolled in a treatment program according to facial oral tract therapy. Occupational therapists at the department are all continuously trained in facial oral tract therapy. The number of therapy sessions is determined by the patient's overall condition, severity of impairments, the patient's responses to the interventions, and/or the relatives' wishes for the rehabilitation. Each treatment is individually planned according to the evaluation of all professionals in the interdisciplinary team.

Dependent and Independent Variables

Dependent variable. FOIS[11] was assessed retrospectively by the first author on the basis of a chart review. The scale consists of 7 levels. Levels 1 through 3 relate to varying degrees of nonoral feeding; levels 4 through 7 relate to varying degrees of oral feeding without nonoral supplementation.[11] The scale was translated into Danish following recommended procedures.[20] First the scale was translated into Danish by 2 health care professionals. They both have Danish as their mother tongue and are fluent in English. They agreed on 1 Danish version, which was back-translated into English by another health care professional who is fluent in Danish and has English as her mother tongue. The original first author, Crary,[11] has given his consent that this translation sufficiently approximates the original version.

The chart review from the brain injury unit includes the following data on each patient: a transfer paper with information from acute care, documentation notes from the medical doctors at the brain injury unit (notes were made every time they consulted with the patient or made any decision regarding treatment), documentation of placement of the feeding tube, documentation notes from the occupational therapist made after every facial oral tract therapy intervention and of every clinical evaluation of swallowing, a general status of the patient noted every other week by the interdisciplinary team, and nutrition charts with information on the patient's daily diet. By combining all this information obtained from the chart review, FOIS was scored on the day of admission to the brain injury unit, every other week until discharge, and at follow-up (follow-up time was 6 months after discharge for the first 3 years and at 1 year for the patients admitted during the last 2 years). At follow-up, the patient came back to the department for 1 day but did not go through an evaluation of swallowing. The FOIS score at follow-up was therefore based on the patient's own information of daily dieting or information given from caregiver or relatives. Admission data of 60 patients were scored twice for quality control with 1 year in between.

Independent variables. GCS[13] score (measured the day after cessation of sedation), time in acute care (time from injury until admission to subacute rehabilitation brain injury unit), FIM score,[21] and RLAS level[22] were all assessed at admission. LOS (time from admission to discharge at subacute rehabilitation brain injury unit) and PTA[23] were assessed by neuropsychologists by means of the Galveston Orientation and Amnesia Test, and number of hours with swallowing therapy was assessed at discharge.

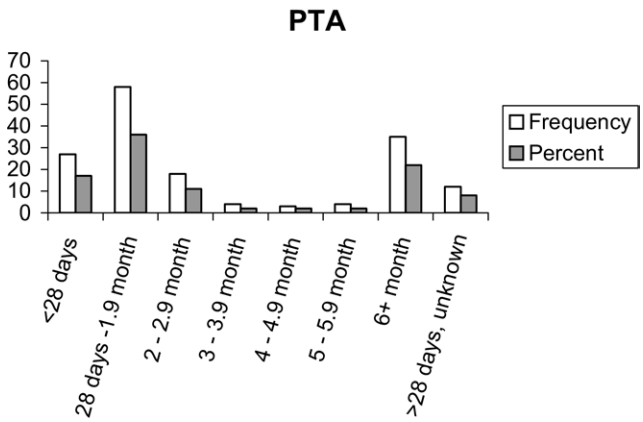


Fig 1. PTA distribution.

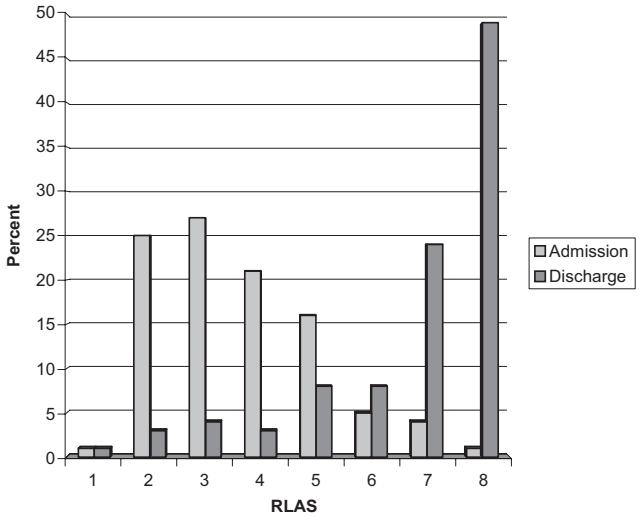


Fig 2. RLAS measured at admission and discharge.

Endpoints

The primary endpoint of this study was the time from beginning of rehabilitation (measured in number of days) until the patient had an FOIS score of 7 (total oral diet with no restrictions). Censuring events were discharge or death.

Statistical Analyses

All data were analyzed using SPSS[a] package for Windows XP.

Patient demographic variables were described by median, IQR, minimum and maximum values for continuous variables, and by number and percent for categorical variables. Further mean and SD were reported for time in acute care and LOS to allow comparison with other studies. We used the Wilcoxon signed-rank test to analyze the difference in FOIS at admission, discharge, and follow-up (the 4 patients who died at the brain injury unit are excluded in this analysis).

We used Kaplan-Meier plots to estimate the time until and the chance of reaching unrestricted dieting. Significance of difference between patients grouped by the scales were calculated using the log-rank test statistic, and we estimated the size of difference using the multiple Cox proportional hazards model.

RESULTS

Of the 173 patients in the study population, 45 were women and 168 men. They had a median age of 35 years (IQR, 24–51y; mean ± SD, 37±15y), median GCS score (measured the day after cessation of sedation) was 11 (IQR, 9–13; mean, 11±3), and 83% of all patients were in PTA more than 4 weeks. The distribution of PTA is presented in figure 1.

Median FIM score was 18.5 (IQR, 18.0–38.5), median time in acute care was 15 days (IQR, 10–24d; mean, 20±20d), and LOS at brain injury unit 86 days (IQR, 53.5–163.5d; mean, 113.5±84.0d). One patient did not respond in any manner at admission (RLAS level 1), 24% were in a vegetative state (RLAS level 2), and 27% were in a minimal consciousness state (RLAS level 3), beginning to respond adequately. Another 69 (44%) patients responded in a more stable way, and only 7 patients (4%) could cooperate relevantly in all situations (RLAS level 7–8). The RLAS levels at admission and discharge are presented in figure 2.

The median hours spent on each patient with swallowing therapy was 16 hours (IQR, 8–36h). At admission, 93% of all patients had problems with oral intake (FOIS score <7), 108 (63%) were dependent on tube feeding when admitted to brain injury unit, and 46% did not receive anything by mouth (FOIS score 1). Twenty-one percent had a tracheotomy tube during their rehabilitation, and 45% of all patients had a prolonged problem with eating and drinking and received a PEG tube.

FOIS scores at admission, discharge, and follow-up are presented in figure 3.

Of the 173 patients, 110 (64%) returned to unrestricted dieting (FOIS score 7) before discharge. Of the 63 (37%) patients with an FOIS score less than 7 at discharge, half were dependent on a PEG tube. No patients were discharged with a tracheotomy tube. Follow-up data were obtained on 142 (82%) of the 173 subjects who participated in the study. Missing data were caused by lack of information of the specific diet level at the time of follow-up, or the patient had died (4 patients), or the patient did not choose to participate in the follow-up visit. At follow-up, none of the FOIS scores had decreased from discharge, and another 16 patients had returned to unrestricted

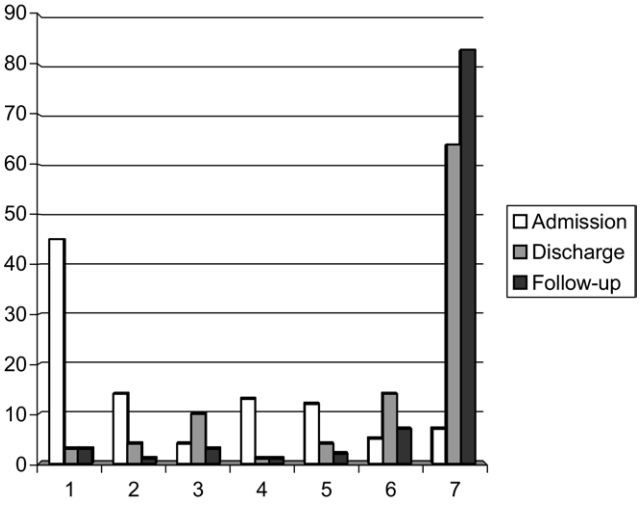


Fig 3. FOIS measured at admission, discharge, and follow-up.

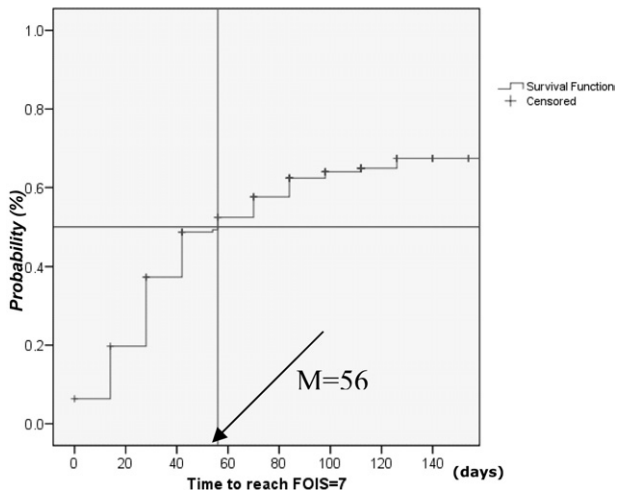


Fig 4. Kaplan-Meier plot showing the time to reach unrestricted dieting for 173 patients with TBI. Time is measured in days. Abbreviation: M, median.

dieting. The differences between FOIS at admission, discharge, and follow-up are statistically significant ($P<.01$).

Intrarater Reliability

In the intrarater reliability test, there was agreement in 55 (92%) of the 60 patients, in 4 (7%) patients there was 1 level of disagreement, and in 1 patient (2%) 2 levels. Kaplan-Meier plot (fig 4) shows that after 56 days, the chance of having reached unrestricted dieting crossed 50%. Among those achieving unrestricted dieting, the median time from admission was 28 days, and after 126 days, no more patients fully recovered despite continuing swallowing therapy. The probability of having recovered to unrestricted dieting within 126 days was 68%. This result takes account of the patients who were discharged before 126 days with restricted dieting and the patients who were still on restricted dieting and not discharged from rehabilitation. Log-rank tests comparing the chance that the patients reached unrestricted dieting before discharge, grouped by their severity of brain injury, are also illustrated by Kaplan-Meier plots (fig 5).

Patients admitted with a GCS score less than 9 had a 41% chance of returning to unrestricted dieting in subacute rehabilitation, and patients admitted with a GCS score greater than 12 had a chance of 90% (the groups were statistically significantly different; $P<.01$). The more severe the brain injury at admission to rehabilitation (low GCS score), the lower the chance of reaching an FOIS score of 7. This was also reflected in RLAS levels. Patients admitted with RLAS level 1 to 2 had a chance of only 24% of reaching unrestricted dieting at the brain injury unit, whereas 77% of the patients admitted in minimal consciousness state, RLAS level 3; 88% patients with RLAS level 4 to 5; and 100% of the patients with an RLAS level 6 to 8 reached unrestricted dieting at the brain injury unit ($P<.01$). The difference between the groups graduated by different time in acute care was rather small. Patients with time in acute care less than 24 days had a 56% chance of reaching an FOIS score of 7, and patients with time in acute care less than 7 days an 80% chance (differences between the groups are also significant; $P=.04$). Functional independence reflected in FIM shows that patients with a minimal functional independence (FIM score <19) at admission had a 50% chance of reaching unrestricted dieting, while higher levels of FIM were not very discriminative ($P<.01$). Finally, we investigated whether the patient's oral intake at admission could predict whether the patient would reach unrestricted dieting before discharge. We found a clear association showing that of the 79 patients admitted with an FOIS score of 1, 39% reached unrestricted dieting; of the patients admitted with an FOIS score of 2 to 3, 81% reached unrestricted dieting; and of the patients admitted with no feeding tube, FOIS score of 4 or higher, almost all (98%) reached unrestricted dieting before discharge ($P<.01$).

The Cox proportional hazards model is presented in table 1. Statistical significance was found in all variables, confirming that the more severe the brain injury, the lower the chance of reaching unrestricted dieting before discharge. GCS score (measured the day after cessation of sedation; Wald $\chi^2=42.78$, $P<.01$), RLAS level (Wald $\chi^2=11.84$, $P<.01$), FIM score (Wald $\chi^2=44.40$, $P<.01$), and FOIS score at admission (Wald $\chi^2=82.93$, $P<.01$) were found to be good predictors for FOIS score of 7 before discharge.

DISCUSSION

The method used in this study is a retrospective collection of data of the functional oral intake reflected in the FOIS by Crary et al.[11] It could be expected that there would be some inaccuracy when scoring the data. However, we believe the factual error is small because of the extended documentation made by the occupational therapists in their own charts, and the medical charts and the diet information charts they make for each patient. Moreover, the authors of the FOIS used retrospective chart reviews successfully.[11] None of the patients who returned to unrestricted diet had any complications in terms of aspiration pneumonia during hospitalization, and no patients went back to restricted dieting, nor were they discharged back to acute care.

This study found a very clear association between severity of brain injury and the chance of reaching unrestricted dieting before discharge. At admission, 93% of the patients had problems with eating and/or drinking, and 64% reached unrestricted dieting before discharge within a maximum time of 126 days. Our study included the most severely injured patients from a defined geographical area. The severity was confirmed by the PTA distribution, and we have found no other directly comparable group described in the literature. As mentioned, Mackay et al[5] found that 61% of patients with TBI admitted to a level I trauma center had abnormal swallowing that affected oral intake, and that the severity of swallowing impairment was associated with lower GCS scores, lower RLAS levels, presence of tracheotomy, and ventilation time longer than 2 weeks[5] (because they did not report any PTA distribution, we cannot say whether that patient group is comparable to our group). Ward et al[6] investigated predictors of oral intake in patients with TBI in acute care and found that patients with severe brain injury (GCS range, 3–8) took a longer time to reach initiation of oral intake than patients with less severe injury (GCS range, >8). Other studies support that low cognition level is associated with poor oral intake in adults with TBI[4,9] and also in children.[24] We investigated several factors focusing on different aspects, such as coma score (GCS), cognitive level (RLAS), time in acute care, level of functioning (FIM), and FOIS score at admission.

GCS score, RLAS level, FIM score, and FOIS score were all found to be statistically significant in predicting time to recovery of functional oral intake, showing that levels of consciousness, cognitive level, and functional measures can be used when predicting return to unrestricted dieting. These results are important when planning rehabilitation and giving information to the patients and relatives. Patients admitted with an RLAS

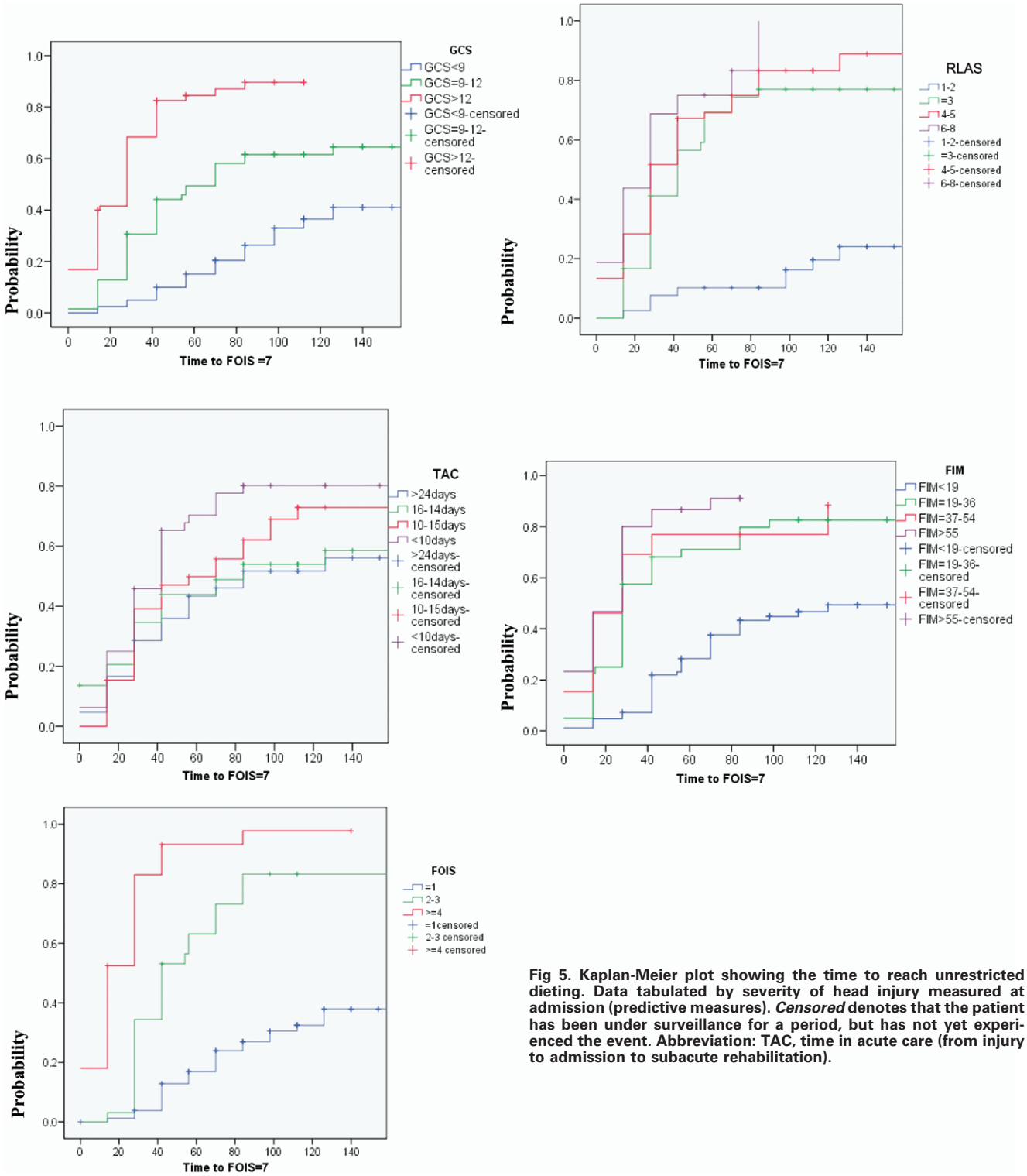


Fig 5. Kaplan-Meier plot showing the time to reach unrestricted dieting. Data tabulated by severity of head injury measured at admission (predictive measures). *Censored* denotes that the patient has been under surveillance for a period, but has not yet experienced the event. Abbreviation: TAC, time in acute care (from injury to admission to subacute rehabilitation).

level of 1 to 2 only had a 24% chance of reaching unrestricted dieting, whereas a patient with an RLAS level of 3 (patients in a minimal conscious state) had a 77% chance. Therefore, even though patients with an RLAS level of 3 are at a very low functioning state at admission, they do have a very good prognosis regarding oral diet. It could be interesting in later studies aiming to investigate the effect of swallowing therapy to use the different categorizations of brain injury severity to give the patients different treatment intensities in the different groups, in line with Carnaby et al.[25] This information could contribute to more efficient use of resources (therapist time) in planning rehabilitation of oral functioning.

Table 1: Association Between an FOIS Score of 7 and Severity of Brain Injury and Oral Intake

Scale		Wald χ^2	HR	*P*	95% CI
GCS score*		42.78		<.01	
	<9 (ref)		1.00		
	9–12		2.51	<.01	1.35–4.69
	>12		6.85	<.01	3.69–12.71
RLAS level		11.84		.01	
	≤2 (ref)		1.00		
	3		6.98	<.01	3.16–15.42
	4		7.51	<.01	3.26–17.32
	≥5		13.02	<.01	5.92–28.62
TAC (d)		7.61		.05	
	>24 (ref)		1.00		
	16–24		1.12	.71	0.63–1.99
	10–15		1.44	.22	0.81–2.55
	<10		2.00	.01	1.18–3.42
FIM score		44.40		<.01	
	<19 (ref)		1.00		
	19–36		3.07	<.01	3.07–5.03
	37–54		4.09	<.01	1.98–8.44
	>55		5.09	<.01	3.01–8.59
FOIS score		82.93		<.01	
	1 (ref)		1.00		
	2–3		3.89	<.01	2.17–6.98
	≥4		10.94	<.01	6.53–18.35

NOTE. Multiple Cox regression hazard model. All analyses adjusted for age and sex.
Abbreviations: CI, confidence interval; HR, hazard ratio; ref, reference; TAC, time in acute care (from injury to admission to subacute rehabilitation).
*Measured the day after cessation of sedation.

A more surprising result is that time in acute care was not found to be a strong predictor for recovery of oral functioning, even though time in acute care had earlier been found to predict functional outcome.[26] This could be because the time our patients stayed in neurosurgical clinics was relatively short (median, 15d), because they were admitted to a brain injury unit right after cessation of sedation. In the study by Whyte et al,[26] the patients had a median time from injury to enrollment (time in acute care) of 40.5 days. Obviously, this difference means that several aspects can influence the results, such as other medical complications in the patients and so forth. We also found that after 56 days (8wk), the chance of having reached unrestricted dieting crossed 50%, and after 126 days (18wk), no more patients returned to unrestricted dieting before discharge. Winstein[7] reported in a retrospective study of 201 patients with TBI that patients with oral intake problems returned to unrestricted dieting in an average time of 12 weeks. However, Winstein[7] did not report the severity of trauma at admission, but it can be estimated from the presented RLAS measure at admission that approximately 43% of their patient group was admitted with an RLAS level of 4 or less, in contrast with 67% in our patient group. Ward et al[6] found in their acute care setting that 47% (55 patients) returned to normal intake in a median of 22 days, but as mentioned, their patient group is not comparable to ours.

With this study, we cannot say whether the swallowing therapy (facial oral tract therapy) used at the brain injury unit has any effect on the recovery of oral functioning. Likewise, we cannot say that there is no treatment effect after 126 days. The theory of facial oral tract therapy is that stimulation of the oral cavity is important to prevent hypersensitivity and hyposensitivity, to avoid bad oral hygiene, and to prevent a decrease in oral functioning, even if the chance of returning to unrestricted dieting is low, and therefore we do not recommend that the treatment should end after 126 days until this has been investigated further.

Sixteen of our patients recovered to unrestricted dieting after discharge, and no patient had a lower level of oral intake at follow-up. Because the therapist did not perform a clinical evaluation of swallowing of each patient at follow-up, we do not know whether the follow-up results reflect improvement in the patients' oral functioning or the fact that other therapist and care practices outside the hospital use different assessments for evaluation of swallowing and/or have other criteria for when the patient can eat and drink. Despite this, it seems reasonable to conclude that in the 82% of the patients seen for follow-up, the level of function of oral intake gained during brain injury unit did not decrease over time. Implementation of a similar method for evaluation of swallowing and criteria for oral diet is recommendable in all phases of rehabilitation, as well as a description of a standardized swallowing therapy that can be used for both experienced and less experienced therapists and caregivers. We hope in the next years to publish such a guideline describing how to use facial oral tract therapy.

Results from this study are important for clinical practice in a subacute rehabilitation department. First, deficient oral intake was found in 93% of our group of patients with very severe TBI. However, functional oral intake significantly improved, so that 64% recovered to unrestricted dieting before discharge and another 9.2% had recovered at follow-up. Second, if unrestricted dieting was not reached within 126 days from admission to rehabilitation, it was not reached before discharge. Third, recovery to unrestricted dieting can be predicted using variables concerning level of consciousness (GCS), cognitive functioning (RLAS), functional ability (FIM), and FOIS, all assessed at admission to rehabilitation. Fourth, the patient group with an RLAS level of 1 to 2 at admission had a 24% chance of recovery, patients with an RLAS level of 3 had a

77% chance, and patients with an RLAS level of 7 to 8 had a 100% chance. This makes RLAS at admission a good predictor of the chance to reach unrestricted dieting in subacute rehabilitation for patients with severe TBI.

These results can be used when planning rehabilitation for patients with severe TBI, giving information about recovery to patients and relatives and designing new studies investigating the effect of high-intensity and low-intensity swallowing therapy pursuing the precise estimation of effectiveness of swallowing therapy—for example, facial oral tract therapy.

CONCLUSIONS

Impairment in functional oral intake was found to be very common, occurring in 93% of a group of 173 patients with very severe TBI admitted to a subacute rehabilitation unit. Return to unrestricted dieting occurred within a maximum of 126 days of rehabilitation. After 56 days, the chance of having reached unrestricted dieting crossed 50%. The chance of returning to total unrestricted oral dieting was found to depend on the severity of the brain injury and could be predicted in particular by RLAS level at admission, but also by GCS score, FIM score, and functional oral intake at admission. These results are important when planning rehabilitation, giving information to patients and relatives, and designing efficacy studies of facial oral tract therapy, which are highly recommended.

Acknowledgment: We thank the professionals in the interdisciplinary teams at the brain injury unit, occupational therapist, physiotherapist, neuropsychologist, nurses, speech therapist, and medical doctors for their contributions to data collection for the present study.

References

1. Schurr MJ, Ebner KA, Maser AL, Sperling KB, Helgerson RB, Harms B. Formal swallowing evaluation and therapy after traumatic brain injury improves dysphagia outcomes. J Trauma 1999; 46:817-21; discussion 821-3.
2. Avery-Smith W, Dellarosa DM. Approaches to treating dysphagia in patients with brain injury. Am J Occup Ther 1994;48:235-9.
3. Halper AS, Cherney LR, Cichowski K, Zhang M. Dysphagia after head trauma: the effect of cognitive-communicative impairments on functional outcomes. J Head Trauma Rehabil 1999;14:486-96.
4. Cherney LR, Halper AS. Swallowing problems in adults with traumatic brain injury. Semin Neurol 1996;16:349-53.
5. Mackay LE, Morgan AS, Bernstein BA. Swallowing disorders in severe brain injury: risk factors affecting return to oral intake. Arch Phys Med Rehabil 1999;80:365-71.
6. Ward EC, Green K, Morton AL. Patterns and predictors of swallowing resolution following adult traumatic brain injury. J Head Trauma Rehabil 2007;22:184-91.
7. Winstein CJ. Neurogenic dysphagia: frequency, progression, and outcome in adults following head injury. Phys Ther 1983;63: 1992-7.
8. Engberg AW, Liebach A, Nordenbo A. Centralized rehabilitation after severe traumatic brain injury: a population-based study. Acta Neurol Scand 2006;113:178-84.
9. Cherney LR, Halper AS. Recovery of oral nutrition after head injury in adults. J Head Trauma Rehabil 1989;4(4):42-50.
10. Brady SL, Darragh M, Escobar NG, O'Neil K, Pape TL, Rao N. Persons with disorders of consciousness: are oral feedings safe/effective? Brain Inj 2006;20:1329-34.
11. Crary MA, Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. Arch Phys Med Rehabil 2005;86:1516-20.
12. Logemann JA, Pepe J, Mackay LE. Disorders of nutrition and swallowing: intervention strategies in the trauma center. J Head Trauma Rehabil 1994;9(1):43-56.
13. Jennett B, Teasdale G, Braakman R, Minderhoud J, Knill-Jones R. Predicting outcome in individual patients after severe head injury. Lancet 1976;307:1031-4.
14. Hind JA, Nicosia MA, Roecker EB, Carnes ML, Robbins J. Comparison of effortful and noneffortful swallows in healthy middle-aged and older adults. Arch Phys Med Rehabil 2001;82: 1661-5.
15. Affolter F, Stricker E. Perceptual processes as prerequisites for complex human behaviour. Bern: Hans Huber; 1980.
16. Davies PM. Starting again: early rehabilitation after traumatic brain injury or other severe brain lesion. Berlin: Springer-Verlag; 1994.
17. Coombes K. Facial oral tract therapy (F.O.T.T.). In: 1991-2001 Jubiläumsschrift 10 Jahre Schulungszentrum. Burgau: Therapie Zentrum; 2001.
18. Coombes K. Von der Ernährungssonde zum Essen am Tisch. In: Lipp B, editor. Wege vom Anfang an. Frührehabilitation schwerst hirngeschädigter Patienten. Villingen-Schwenningen: Neckar Verlag GMBH; 1996. p 137-43.
19. Welter FL, Meyer-Konigsbuscher J. [Facial-oral therapy in craniocerebral diseases] [German]. Rehabilitation (Stuttg) 1998; 37:58-63.
20. Sechrest L, Fay T, Zaidi SM. Problems of translation in cross-cultural research. J Cross-Cult Psychol 1972;3:41-56.
21. Granger CV. The emerging science of functional assessment: our tool for outcomes analysis. Arch Phys Med Rehabil 1998;79:235-40.
22. Holland A. Language disorders in adults. San Diego: College Hills Pr; 1984.
23. Levin HS, O'Donnell VM, Grossman RG. The Galveston Orientation and Amnesia Test: a practical scale to assess cognition after head injury. J Nerv Ment Dis 1979;167:675-84.
24. Morgan A, Ward E, Murdoch B, Kennedy B, Murison R. Incidence, characteristics, and predictive factors for dysphagia after pediatric traumatic brain injury. J Head Trauma Rehabil 2003;18: 239-51.
25. Carnaby G, Hankey GJ, Pizzi J. Behavioural intervention for dysphagia in acute stroke: a randomised controlled trial. Lancet Neurol 2006;5:31-7.
26. Whyte J, Katz D, Long D, et al. Predictors of outcome in prolonged posttraumatic disorders of consciousness and assessment of medication effects: a multicenter study. Arch Phys Med Rehabil 2005;86:453-62.

Supplier

a. Version 13.0; SPSS Inc, 233 S Wacker Dr, 11th Fl, Chicago, IL 60606.

Study II:

II

Hansen, T.S., Larsen, K., Engberg A.W.

The association of functional oral intake and pneumonia in patients with severe TBI.

Archives of Physical Medicine and Rehabilitation: 2008; 89, 11, 2114-20

ORIGINAL ARTICLE

The Association of Functional Oral Intake and Pneumonia in Patients With Severe Traumatic Brain Injury

Trine S. Hansen, MPH, Klaus Larsen, PhD, Aase W. Engberg, DMSc

ABSTRACT. Hansen TS, Larsen K, Engberg AW. The association of functional oral intake and pneumonia in patients with severe traumatic brain injury. Arch Phys Med Rehabil 2008;89:2114-20.

Objectives: To investigate the incidence and onset time of pneumonia for patients with severe traumatic brain injury (TBI) in the early phase of rehabilitation and to identify parameters associated with the risk of pneumonia.

Design: Observational retrospective cohort study.

Setting: Subacute rehabilitation department in a university hospital in Denmark.

Participants: Patients (N=173) aged 16 to 65 years with severe TBI who were admitted during a 5-year period. Patients are transferred to the brain injury unit as soon as they ventilate spontaneously.

Interventions: Not applicable.

Main Outcome Measure: Pneumonia.

Results: Twenty-seven percent of the patients admitted to the brain injury unit were in treatment for pneumonia; pneumonia developed in 12% of the patients during rehabilitation; the condition occurred within 19 days of admission in all but 1 patient. Of these patients, 81% received nothing by mouth. Three factors identified patients at highest risk of pneumonia: Glasgow Coma Scale score less than 9 (1 day after cessation of sedation); Rancho Los Amigos Scale score less than 3 (on admission); and no oral intake on admission. Having a tracheotomy tube and/or feeding tube was also associated with a higher occurrence of pneumonia.

Conclusions: Among patients with severe TBI, 27% had pneumonia at transfer from the intensive care unit. Pneumonia developed in only 12% of the participants during rehabilitation. Patients with a low level of consciousness and patients with a tracheotomy tube or feeding tube had a higher likelihood of pneumonia.

Key Words: Brain injuries; Deglutition disorders; Pneumonia; Rehabilitation.

PATIENTS WITH TBI are at risk of developing problems with swallowing, eating, and drinking.[1,2] In recent years, the number of intensive specialized rehabilitation units for patients with TBI has increased, and positive effects of intensive interdisciplinary rehabilitation have been reported.[3-5] Problems with eating and drinking—with an incidence as high as 93%[6] in patients with severe TBI—have been found and can be potentially life-threatening by leading to malnutrition, dehydration, aspiration pneumonia,[7] and prolonged length of hospital stay.[8] Several risk factors for aspiration pneumonia have been reported, such as endotracheal intubation, mechanical ventilation, poor oral hygiene,[9] oral feeding (if the patients aspirate food or liquid),[8,10,11] and food supplementation (PEG or nasogastric tube).[12]

Patients with TBI admitted to an ICU are at high risk of contracting pneumonia.[8] In this early stage of recovery, pneumonia is found to occur within an average of 3 days of hospitalization[13] and is associated with severity of trauma.[8,14] When patients transfer to subacute rehabilitation, they often begin to be more active, and adequate nutrition is important,[15] either orally or by a supplemental tube feeding. At this stage of rehabilitation, some patients are supported with a tracheotomy tube and/or feeding tube; many have a low level of consciousness.[3] In this early phase of rehabilitation, several patient care issues become important, such as monitoring for clinical signs of aspiration, maintaining good oral hygiene, minimizing risk of reflux, managing secretions, providing tube feeding (when relevant), carefully managing tracheotomy tube (when relevant), and carefully managing initiation of oral feeding.[16]

Previous research of TBI and pneumonia often has been done during acute stages in the ICU or in neurosurgery clinics.[8,17-20] At our brain injury unit, patients with severe TBI are admitted to subacute intensive rehabilitation as soon as they ventilate spontaneously. Eighty percent of the patients are transferred directly from the neurosurgical wards.[3]

To learn more about how to prevent pneumonia in this high-risk patient group, we investigated the incidence and onset time of pneumonia and identified parameters associated with the risk of pneumonia in this early phase of rehabilitation.

METHODS

This is a retrospective observational study using data collected from hospital records and chart reviews.

List of Abbreviations

CRP	C-reactive protein
FOIS	Functional Oral Intake Scale
GCS	Glasgow Coma Scale
ICU	intensive care unit
IQR	interquartile range
LES	lower esophageal sphincter
LOS	length of stay
PEG	percutaneous endoscopic gastrostomy
PTA	posttraumatic amnesia
RLAS	Rancho Los Amigos Scale
TBI	traumatic brain injury

From the Department of Neurorehabilitation, Brain Injury Unit (Hansen, Engberg), Department of Occupational Therapy (Hansen), Clinical Research Unit (Larsen), Copenhagen University Hospital Hvidovre, Hvidovre, Denmark.

No commercial party having a direct financial interest in the results of the research supporting this article has or will confer a benefit on the authors or on any organization with which the authors are associated.

Reprint requests to Trine S. Hansen, MPH, Dept of Neurorehabilitation 354, Hvidovre Hospital, Kettegaard Allé 30, DK-2650 Hvidovre, Denmark, e-mail: *trine.schow@gmail.com.*

0003-9993/08/8911-00214$34.00/0
doi:10.1016/j.apmr.2008.04.013

Participants

In October 2000, the subacute rehabilitation of all severely injured patients with TBI in Denmark was centralized to 2 units, each with geographically defined uptake areas.

The present study includes patients from the uptake area of Copenhagen, which includes the eastern half of Denmark, Greenland, and the Faroe Islands, encompassing a total of 2.4 million inhabitants. All hospitals in the uptake area, and, in particular, the 2 neurosurgical clinics in the area, agreed to refer patients who fulfilled the following set criteria. Highest priority was given to patients who, after initial treatment in a neurosurgical or other clinic, had a GCS[21] score in the range 3 through 12 one day after cessation of sedation and patients with a GCS of 13–14 one day after cessation of sedation if they have severe focal neurologic deficits and/or are severely agitated. All such survivors were transferred as soon as they ventilated spontaneously. The brain injury severity was confirmed by prospective assessment of the duration of the PTA period. All 177 patients, age 16 to 65 years, admitted during a 5-year period from October 2000 to December 2005 were evaluated for eligibility. We excluded patients on the basis of previously known swallowing problems due to neurologic diseases or other diagnoses. Four patients met this criterion and 173 patients were included in the study. Patients older than 65 years were excluded because swallowing function can be diminished in older age groups of otherwise healthy people.[22]

According to the rehabilitation program of the brain injury unit,[3] all patients were enrolled in an extensive around-the-clock rehabilitation program by interdisciplinary teams starting on the day of admission. Functional rehabilitation was based on the principles developed by Affolter and Stricker,[23] Davies,[24] and Coombes.[25,26] The program emphasizes sensory stimulation, facilitation of normative movements, and daily activities, even for patients in a vegetative state. Patients were discharged when they were: able to go home, able to continue the rehabilitation in a local and less intensive setting, or referred to a nursing home if no progress was made for a 3-month period.[3]

Swallowing Therapy and Oral Hygiene

Examination of the mouth and a clinical evaluation of swallowing were done for all patients on the day of admission, in line with the treatment guidelines for facial oral tract therapy.[25,26] Facial oral tract therapy was developed by speech and language therapist Kay Coombes and provides a structured way to assess and treat disturbances in facial expression, movement of the jaw for eating and articulation, breathing, swallowing, and voice.[25,26] Treatment methods include slow, organized touch of the patient's hands, facilitating hand-to-hand and hand-to-face contact, along with specific oral stimulation, therapeutic oral hygiene routines, and facilitation of swallowing. Facial oral tract therapy does not require that the patients are capable of following verbal instructions. Therefore, patients with a low level of consciousness also receive facial oral tract therapy. All patients who experienced problems with oral intake, oral hygiene, breathing, and communication were enrolled in a treatment program according to facial oral tract therapy.

Patients who experienced problems with oral hygiene were treated both by the occupational therapist and caregivers. They facilitate participation in brushing teeth and cleaning the mouth. Patients who were not eating by mouth also received oral stimulation to maintain sensory input and prevent deprivation. To prevent pneumonia in patients fed exclusively by feeding tube, the patients were mobilized several times a day. In bed the patients were positioned in a 30° side-lying position (if tolerated). Supine position was avoided if possible. If the patient lay supine, the head of the bed was elevated and caregivers and therapists carefully monitored oral hygiene and accumulated secretions in the oral cavity. At our brain injury unit, a patient with a feeding tube is given bolus feedings if tolerated. Continuous feedings are given to patients with a low level of consciousness and/or patients who do not tolerate bolus feeding. All feeding begins with aspiration of the ventricle, and if more than 100mL material rests in the ventricle, clinicians caring for the patient stop the tube feeding and wait at least 1 hour before feeding is resumed. A feeding pump connected to the feeding tube monitors the feeding.

Dependent Variables

Pneumonia, an inflammation in the lung parenchyma, is defined as follows in our hospital: (1) appearance of new infiltrative changes on chest radiograph that can be explained by pneumonia and/or (2) increase in temperature to more than 38.5°C with an increase in CRP to more than 50mg/L and leukocyte count more than 9 cells/L, accompanied by respiratory symptoms such as dyspnea, coughing, and/or purulent expectoration.[27] If these criteria were absent or not documented, the diagnosis was not acknowledged and used in this study. If the same patient was diagnosed with pneumonia several times, all episodes were noted. For patients who showed signs of pneumonia as previously defined, the laboratory clinicians at the hospital routinely examined bacteria in tracheal secretions.

Independent Variables

GCS,[21] which consists of values from 3 to 15, was measured 1 day after cessation of sedation. Patients with scores less than 9 are considered to be in coma, and patients with scores of 15 are able to follow commands, are fully oriented, and have spontaneous eye opening. Time in acute care (time from injury until admission to subacute rehabilitation brain injury unit), FIM instrument[28] scores (range 18 [lowest]–126 [highest]) of level of independence, and RLAS[29] scores (range 1 [no response]–8 [purposeful and appropriate response]) were all measured at admission. FOIS was assessed retrospectively based on a chart review.[6] The scale consists of 7 levels. Levels 1 through 3 relate to varying degrees of nonoral feeding, and levels 4 through 7 relate to varying degrees of oral feeding without nonoral supplementation.[30] LOS corresponds to time from admission to discharge from subacute rehabilitation brain injury unit. Duration of PTA[31] was prospectively assessed by neuropsychologists by means of the Galveston Orientation and Amnesia Test.

Statistical Analyses

All data were analyzed using SPSS[a] package for Windows.

We described patient demographic variables by median, IQR, minimum, and maximum values for continuous variables and by number and percentage for categorical variables.

We used a Kaplan-Meier plot to estimate the time from admission to occurrence of pneumonia and the risk of contracting pneumonia and log-rank test to calculate the risk difference between patients grouped by severity of brain injury. If a patient did not contract pneumonia he/she was censored at the time of discharge in the analysis. We estimated the size of difference by a univariate Cox proportional hazards model, and analyzed covariates that changed over time using a time-dependent covariate Cox proportional hazards model.

RESULTS

Demographic Data

Patients had a median age of 35 years (IQR, 24–51y). Median GCS score measured 1 day after cessation of sedation was 11 (IQR, 9–13). For 81% of the patients, PTA duration exceeded 4 weeks. At admission to the brain injury unit, median FIM score was 18.5 (IQR, 18–38.5). Median time in acute care was 15 days (10–24) and LOS at brain injury unit 86 days (53.5–163.5d), respectively. Ninety-three percent had some degree of problems with oral intake at admission to brain injury unit (FOIS score <7). Sixty-eight percent of patients were admitted with a feeding tube, and 21% with a tracheotomy tube. Forty-five percent of patients had a prolonged problem with eating and drinking and received a PEG tube during their rehabilitation at the brain injury unit.

Pneumonia

Incidences of pneumonia are presented in table 1.

At the time that the patients were admitted to our brain injury unit, 46 (27%) were in treatment for pneumonia, and pneumonia developed in 21 (12%) at the unit. Results of the microbiologic investigations are shown in table 2 for 20 patients (data unavailable in 1 case). More than 1 species was found in secretions from 3 patients.

Of the 21 patients, 2 (10%) had pneumonia at admission, pneumonia developed once in 12 (57%), 5 (24%) had 2 episodes of pneumonia, and 4 (19%) had 3 episodes of pneumonia. Onset time of the first episode of pneumonia ranged from 1 day to 19 days after admission for all patients, except one in whom pneumonia developed after 71 days and who died after 86 days. Because this time interval differs greatly relative to the others, this patient was excluded as an outlier. In the following results, we focus only on the time until the first episode of pneumonia.

We found that 17 (81%) of the 21 patients who had pneumonia in the brain injury unit were totally dependent on tube feeding and received nothing by mouth (FOIS score of 1), 3 patients developed pneumonia when they were given minimal attempts of food and/or liquid by mouth but were still dependent on tube feeding (FOIS score range, 2–3), and pneumonia developed in 1 patient when he had a total oral diet but still needed special preparation or compensation (FOIS score of 5).

In figure 1, the Kaplan-Meier plot shows the risk of pneumonia as a function of time within the first 3 weeks after admission. The 5-day, 10-day, and 15-day rates were estimated at 6%, 8%, and 11%, respectively. After 19 days, pneumonia did not develop for the first time in any more patients. At this time the risk of pneumonia was 12%.

Kaplan-Meier plots estimating the risk that pneumonia will develop in patients in the brain injury unit, grouped by severity of brain injury, are shown in figure 2. We found that patients admitted with a low GCS score (<9) had a higher risk (25%) of pneumonia developing compared with a risk of 13% among patients with a moderate risk (GCS score range, 9–12), and a risk of 2% among those with a high GCS score (>12). Pneumonia did not develop in most patients with an RLAS score of 3 or more (ie, able to obey commands), whereas pneumonia developed shortly after admission (within the range of 1–9 days) in 23% of the patients with a lower RLAS score. Patients with an FOIS score of 1 had a 12% risk of pneumonia, patients with FOIS score of 2 or 3 had a 7% risk, and patients with an FOIS score of 4 or more had only a 1% risk of pneumonia.

Log-rank test showed that differences in risk of pneumonia among groups was statistically significant ($P<.01$) for GCS score, RLAS score, and FOIS score. Pneumonia developed in 18% of the patients admitted with low functional ability (FIM score <19), whereas patients with a high FIM score (>55) had only a 7% risk. Patients in acute care more than 24 days had a 12% risk whereas patients in acute care fewer than 7 days had only a 4% risk of pneumonia. However, there is no statistical evidence showing that the risks between the groups in FIM score and time in acute care differ ($P>.05$). Results from the Cox proportional hazards model are presented in table 3.

We found a strong association between GCS score (Wald $\chi^2=10.81$, $P<.01$), RLAS score (Wald $\chi^2=13.76$, $P<.01$), and FOIS score (Wald $\chi^2=9.3$, $P<.01$). Again, there was no statistically significant association between time in acute care and FIM.

Table 1: Incidence of Pneumonia

Incidence	n	%
No pneumonia	109	63
Pneumonia at admission	46	27
Pneumonia at BIU	21	12
No. of episodes of pneumonia		
1	12	7
2	5	3
3	4	2

Abbreviation: BIU, brain injury unit.

Table 2: Etiologic Agents From Tracheal Secretion in 20 Patients With Pneumonia

Cocci	Cases	%
Staphylococcus aureus	4	20
Coagulase-negative staphylococci	3	15
Gram-negative diplococci	1	5
Gram-negative bacilli		
Klebsiella species	4	20
Pseudomonas aeruginosa	3	15
Citrobacter species	1	5
Enterobacter cloacae	1	5
Haemophilus influenzae	1	5
No growth	6	30

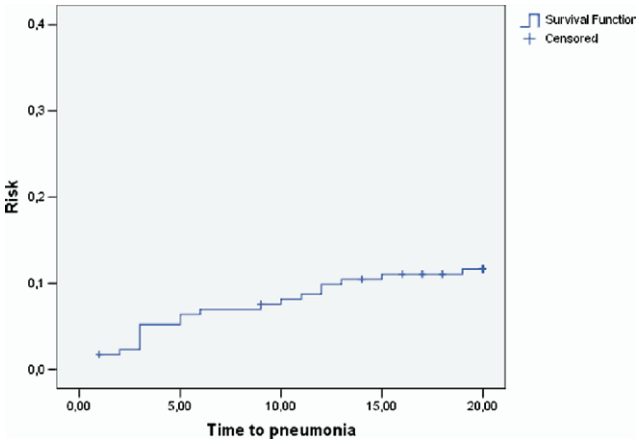


Fig 1. Kaplan-Meier plot showing the time to and risk of pneumonia for 172 patients with TBI in a subacute rehabilitation unit.

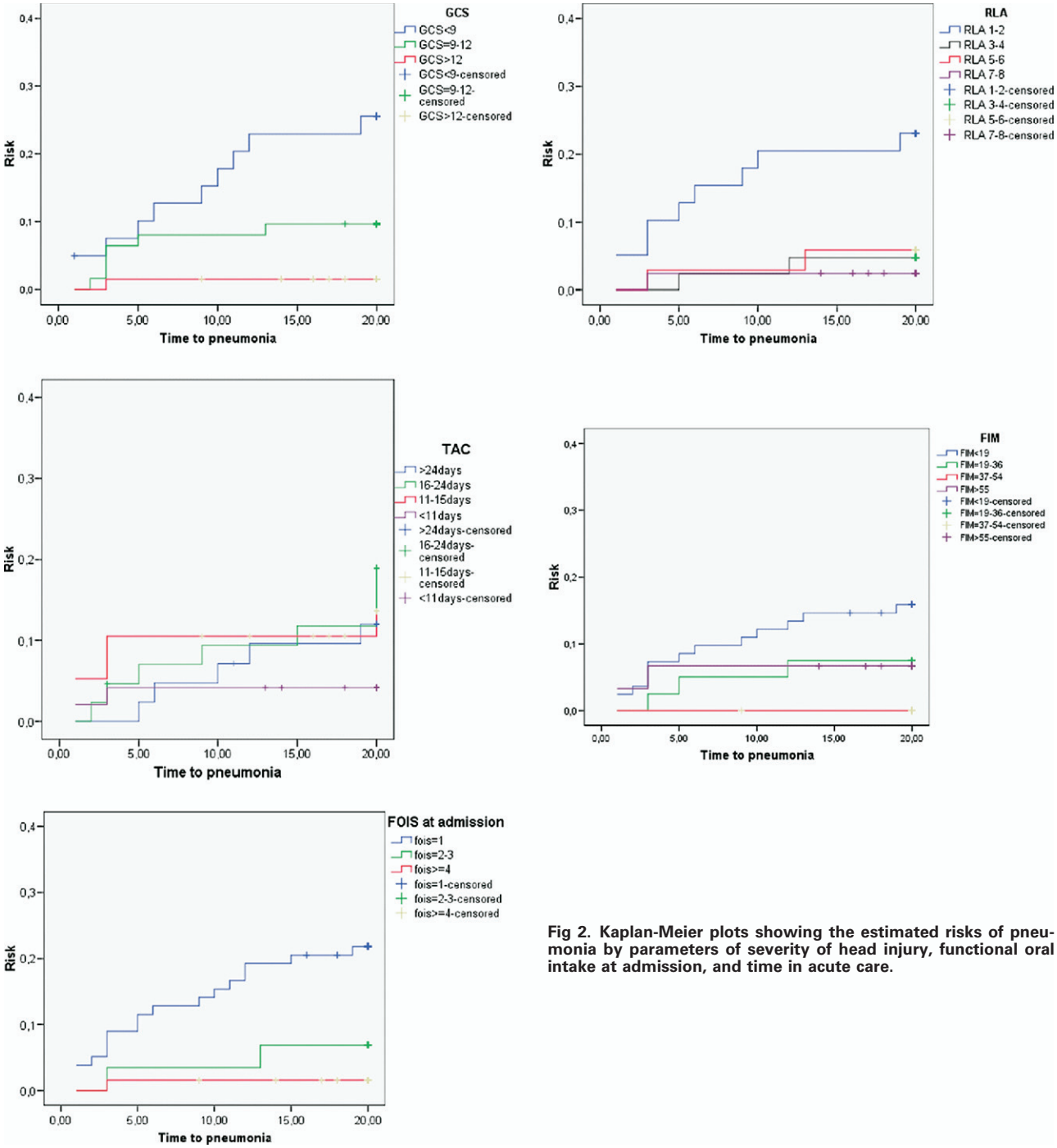


Fig 2. Kaplan-Meier plots showing the estimated risks of pneumonia by parameters of severity of head injury, functional oral intake at admission, and time in acute care.

Table 4 shows results from the time-dependent covariate model. We found a higher incidence of pneumonia in patients with tube feeding (Wald $\chi^2=8.99$, $P=.01$) and for patients with a tracheotomy tube (Wald $\chi^2=6.94$, $P=.01$).

DISCUSSION

We retrospectively summed episodes of pneumonia in a 5-year period in a subacute rehabilitation department for patients with severe TBI. Brain injury has already been documented to be associated with a risk of pneumonia.[32] In this study, we evaluated the incidence and risk factors of pneumonia in the early phase of rehabilitation for patients with severe TBI. We found that 12% of the patients developed pneumonia one time during hospitalization in a subacute specialized rehabilitation unit, and that pneumonia developed in all except one patient within 19 days after admission. GCS and RLAS scores were associated with risk of pneumonia. This is in agreement with findings from other studies.[8,14,33-35] Explanatory factors could be that reduction of consciousness leads to relaxation of muscles in the larynx and thereby reduces airway closure,[34] supported by Huxley et al,[34] who reported that 45% of healthy adults aspirated during sleep and 70% of patients with de-

Table 3: Association Between Risk of Pneumonia and Severity of Brain Injury, Functional Oral Intake at Admission, and Time in Acute Care

Scales	Wald χ^2	P*	Hazard Ratio	95% CI
GCS	10.81	<.01		
<9 (ref)			1.00	
9–12		.03	0.32	0.11–0.92
>12		<.01	0.05	0.05–0.29
RLAS	13.76	<.01		
≤2 (ref)			1.00	
3		.01	0.13	0.03–0.61
4		.02	0.16	0.03–0.77
≥5		<.01	0.07	0.01–0.55
Time in acute care (d)	3.44	.33		
>24 (ref)			1.00	
16–24		.53	1.71	0.55–5.34
10–15		.70	1.28	0.37–4.45
<10		.29	0.41	0.08–2.12
FIM	3.24	.36		
<19 (ref)			1.00	
19–36		.21	0.44	0.12–1.16
37–54		.94	0.00	0.00–0.00
>55		.14	0.32	0.07–1.44
FOIS	9.30	<.01		
1 (ref)			1.00	
2–3		.16	0.35	0.08–1.53
≥4		.01	0.06	0.01–0.42

NOTE. All analysis adjusted for age and sex; time in acute care GCS score measured 1 day after cessation of sedation; multiple Cox regression hazard model.
Abbreviation: CI, confidence interval.
*Significant at P<.05.

pressed consciousness aspirated. Moreover, Saxe et al[36] found that low GCS scores are associated with lower tone in the LES.

Studies of patients with TBI in ICUs have described the incidence of pneumonia as high as 41% to 60%.[8,13,14] Several special risk factors are related to pneumonia for patients in an ICU compared with patients in rehabilitation units. These factors include risk of aspiration in association with the trauma or accident,[13] coma, and mechanical ventilation.[37-39] However, the patients at the brain injury unit could also be at high risk due to the short time in acute care (mean, 15d). Even so, the incidence rate of pneumonia decreased to 12% in our brain injury unit compared with 27% at admission.

We found that 81% of the patients in whom pneumonia developed were fed exclusively by feeding tube. This result will not lead to advice against using feeding tubes because the only realistic alternative was intravenous nourishment. However, our results confirm that even if experienced staff takes precautions, a feeding tube does not eliminate the risk of aspiration. Our result leads to the conclusion that patients with a feeding tube either aspirate saliva, oral or laryngeal secretion, or regurgitate gastric content,[40] supported by results from other studies investigating neurologic patient groups.[41,42] Dent et al[43] found that gastroesophageal reflux is most frequent in the postprandial state and therefore more frequent in patients given continuous feeding instead of intermittent bolus feeding. Which method is preferable has been discussed in the literature[35,44,45] but no consensus has been reached. Rhoney et al[35] found that continuous enteral feeding was better tolerated in patients with acute brain injuries; however, for risk of pneumonia they only found evidence (P=.22) among nonventilated patients. On the contrary, Tejada Artigas et al[45] found that continuous enteral feeding was a risk factor for nosocomial pneumonia in patients with trauma admitted to an ICU, supported by Jacobs et al[44] in an earlier study with patients from an ICU. In our department, we give continuous feedings or very slow bolus feedings to patients with very low consciousness. One could hypothesize that a risk factor for pneumonia in our patients could be that mobilization of a patient with low tone in the LES with food in the stomach increases the risk of vomiting or reflux and thereby risk of aspiration. Cole et al[41] found in a single case study that an infusion rate more than 50mL/h increased the risk of reflux and suggested that increased feeding volume further could lead to gastric retention and distension, resulting in relaxation of the LES and leading to reflux.[41,46] This was supported by the findings of Ahtaridis et al.[46]

Some patients with severe TBI produce more saliva and secretions, which, in combination with reduced swallowing rate, could lead to risk of pneumonia. Especially in patients with poor oral hygiene, aspiration of saliva, for which feeding tubes do not offer any protection, can lead to pneumonia.[9,47-49] Oral hygiene is a problem often not recognized in critically ill patients, including patients with TBI, and can become severe due to cognitive problems, hypersensitivity in the oral cavity, impaired saliva production, nonoral feeding, decreased mobility, drugs, and intubations.[50-52]

We also found that a tracheotomy tube in our patient group was associated with a higher risk of pneumonia, which supports results from other studies.[53-57] A tracheotomy tube may be necessary for protection of the airway and protection of accumulated secretions, but it may also cause colonization of pathogens in the oropharynx,[58] absence of expiratory airflow through the larynx,[59] impaired laryngeal movement, loss of protective mechanisms such as vocal cord closure, and loss of laryngeal reflex,[60,61] all factors associated with aspiration. In addition, patients with severe TBI have weakened immune systems and therefore are at increased risk of inflammations.[62] It is not in the nature of this study to evaluate the optimal time for removal of tracheotomy tube, but we can recommend close monitoring of patients with a tracheotomy tube to prevent pneumonia.

Our findings clearly indicate that low GCS scores and presence of a tracheotomy tube or feeding tube are associated with higher rate of pneumonia in patients with severe TBI. We do not, however, present evidence that these factors have a causal relationship with pneumonia. Presence of a tracheotomy tube or feeding tube could potentially contribute to causing pneumonia, but another viable explanation is severe dysphagia,

Table 4: Relationship Between Feeding Tube and Tracheostomy Tube on Incidence of Pneumonia

Tube	Wald χ^2	P*	Hazard Ratio	95% CI
Feeding	8.99	.01		
No feeding tube (ref)			1.00	
Nasogastric		.01	18.85	2.46–144.48
PEG tube		.08	7.45	0.77–72.50
Tracheotomy	6.94	.01		
No tracheostomy (ref)			1.00	
Tracheostomy		.01	3.55	1.38–9.10

NOTE. Cox proportional hazard regression model with time-dependent covariates.
Abbreviation: CI, confidence interval.
*Significant at P<.05.

which may be why these patients need to have a tracheotomy tube or feeding tube. The aspiration pneumonia could have been caused by the dysphagia, with the tracheotomy tube and feeding tube as epiphenomena having no role in pathogenesis of the pneumonia. This possibility can be addressed in future prospective studies.

Because oral hygiene could be a potential cause of pneumonia,[9] we are now implementing a rating scale that makes it possible in future studies to address oral hygiene. Furthermore, we have started implementation of fiberoptic endoscopic evaluation of swallowing.

Study Limitations

This study method is associated with some limitations. We could not document the diagnosed pneumonia as aspiration pneumonia. However, each episode of pneumonia was specified as aspiration pneumonia in the medical files, and the spectrum of bacteria is similar to findings from earlier published studies of aspiration pneumonia.[63,64]

CONCLUSIONS

Among patients with severe TBI, 27% had pneumonia at the time of transfer from the ICU. Pneumonia developed in only 12% during subacute rehabilitation at our brain injury unit. Higher incidence of pneumonia was found in patients with low level of consciousness, tracheotomy, and exclusive tube-feeding. To prevent pneumonia, therapists and other health care professionals caring for patients with problems with oral intake should be aware of these parameters during rehabilitation.

Acknowledgments: We thank the professionals in the interdisciplinary teams at the brain injury unit: occupational therapists, physiotherapists, neuropsychologists, nurses, speech therapists, and medical doctors for their contributions with data collection. We also thank the chief physician at brain injury unit for helping with the definition of pneumonia.

References

1. Schurr MJ, Ebner KA, Maser AL, Sperling KB, Helgerson RB, Harms B. Formal swallowing evaluation and therapy after traumatic brain injury improves dysphagia outcomes. J Trauma 1999; 46:817-21; discussion 821-3.
2. Avery-Smith W, Dellarosa DM. Approaches to treating dysphagia in patients with brain injury. Am J Occup Ther 1994;48:235-9.
3. Engberg AW, Liebach A, Nordenbo A. Centralized rehabilitation after severe traumatic brain injury—a population-based study. Acta Neurol Scand 2006;113:178-84.
4. Mackay LE, Bernstein BA, Chapman PE, Morgan AS, Milazzo LS. Early intervention in severe head injury: long-term benefits of a formalized program. Arch Phys Med Rehabil 1992;73:635-41.
5. Cope DN. The effectiveness of traumatic brain injury rehabilitation: a review. Brain Inj 1995;9:649-70.
6. Hansen TS, Engberg AW, Larsen K. Functional oral intake and time to reach unrestricted dieting for patients with traumatic brain injury. Arch Phys Med Rehabil. 2008;89:1556-62.
7. Mackay LE, Morgan AS, Bernstein BA. Swallowing disorders in severe brain injury: risk factors affecting return to oral intake. Arch Phys Med Rehabil 1999;80:365-71.
8. Woratyla SP, Morgan AS, Mackay L, Bernstein B, Barba C. Factors associated with early onset pneumonia in the severely brain-injured patient. Conn Med 1995;59:643-7.
9. Terpenning MS, Taylor GW, Lopatin DE, Kerr CK, Dominguez BL. Aspiration pneumonia: dental and oral risk factors in an older veteran population. J Am Geriatr Soc 2001;49:557-63.
10. John S, Marlene H, Kathryn H, Michael R. Videofluoroscopic evidence of aspiration predicts pneumonia and death but not dehydration following stroke. Dysphagia 1994;9:7-11.
11. Martin BJ, Corlew MM, Wood H, et al. The association of swallowing dysfunction and aspiration pneumonia. Dysphagia 1994;9:1-6.
12. Nishiwaki S, Araki H, Goto N, et al. Clinical analysis of gastroesophageal reflux after PEG. Gastrointest Endosc 2006;64:890-6.
13. Hsieh AH, Bishop MJ, Kubilis PS, Newell DW, Pierson DJ. Pneumonia following closed head injury. Am Rev Respir Dis 1992;146:290-4.
14. Rodriguez JL, Gibbons KJ, Bitzer LG, Dechert RE, Steinberg SM, Flint LM. Pneumonia: incidence, risk factors, and outcome in injured patients. J Trauma 1991;31:907-12; discussion 912-4.
15. Denes Z. The influence of severe malnutrition on rehabilitation in patients with severe head injury. Disabil Rehabil 2004;26:1163-5.
16. Langmore SE. Dysphagia in neurologic patients in the intensive care unit. Semin Neurol 1996;16:329-40.
17. Ramsey DJ, Smithard DG, Kalra L. Early assessments of dysphagia and aspiration risk in acute stroke patients. Stroke 2003;34: 1252-7.
18. Abele-Horn M, Dauber A, Bauernfeind A, et al. Decrease in nosocomial pneumonia in ventilated patients by selective oropharyngeal decontamination (SOD). Intensive Care Med 1997;23: 187-95.
19. Andermahr J, Greb A, Hensler T, et al. Pneumonia in multiple injured patients: a prospective controlled trial on early prediction using clinical and immunological parameters. Inflamm Res 2002; 51:265-72.
20. Lazarus C, Logemann JA. Swallowing disorders in closed head trauma patients. Arch Phys Med Rehabil 1987;68:79-84.
21. Jennett B, Teasdale G, Braakman R, Minderhoud J, Knill-Jones R. Predicting outcome in individual patients after severe head injury. Lancet 1976;307:1031-4.
22. Hind JA, Nicosia MA, Roecker EB, Carnes ML, Robbins J. Comparison of effortful and noneffortful swallows in healthy middle-aged and older adults. Arch Phys Med Rehabil 2001;82: 1661-5.
23. Affolter F, Stricker E. Perceptual processes as prerequisites for complex human behaviour: a theoretical model and its application to therapy. Bern: Huber; 1980.
24. Davies PM. Starting again: early rehabilitation after traumatic brain injury or other severe brain lesion. Berlin: Springer-Verlag; 1994.
25. Coombes K. Facial oral tract therapy (F.O.T.T.). In: 1991-2001 Jubiläumsschrift 10 Jahre Schulungszentrum. Burgau: Therapie Zentrum; 2001.
26. Coombes K. Von der Ernährungssonde zum Essen am Tisch. In: Lipp B, Schlagel W, Wege vom Anfang an. Frührehabilitation schwerst hirngeschädigter Patienten. Villingen-Schwenningen: Neckar Verlag GMBH; 1996:137-43.
27. Niederman MS, Mandell LA, Anzueto A, et al. Guidelines for the management of adults with community-acquired pneumonia. Diagnosis, assessment of severity, antimicrobial therapy, and prevention. Am J Respir Crit Care Med 2001;163:1730-54.
28. Granger CV. The emerging science of functional assessment: our tool for outcomes analysis. Arch Phys Med Rehabil 1998; 79:235-40.
29. Holland A, Language disorders in adults. San Diego: College Hills Pr; 1984.
30. Crary MA, Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. Arch Phys Med Rehabil 2005;86:1516-20.
31. Levin HS, O'Donnell VM, Grossman RG. The Galveston Orientation and Amnesia Test. A practical scale to assess cognition after head injury. J Nerv Ment Dis 1979;167:675-84.
32. Warren JB. Pulmonary complications associated with severe head injury. J Neurosurg Nurs 1983;15:194-200.

33. Nercelles P, Herrera R, Peirano L. Interventional program in nosocomial pneumonia associated to depressed consciousness in a Chilean teaching hospital, 7-year follow-up [abstract]. Am J Infect Control 2004;32:E48.
34. Huxley EJ, Viroslav J, Gray WR, Pierce AK. Pharyngeal aspiration in normal adults and patients with depressed consciousness. Am J Med 1978;64:564-8.
35. Rhoney DH, Parker D Jr, Formea CM, Yap C, Coplin WM. Tolerability of bolus versus continuous gastric feeding in brain-injured patients. Neurol Res 2002;24:613-20.
36. Saxe JM, Ledgerwood AM, Lucas CE, Lucas WF. Lower esophageal sphincter dysfunction precludes safe gastric feeding after head injury. J Trauma 1994;37:581-4; discussion 584-6.
37. Magnotti LJ, Croce MA, Fabian TC. Is ventilator-associated pneumonia in trauma patients an epiphenomenon or a cause of death? Surg Infect (Larchmt) 2004;5:237-42.
38. Richardson CJ, Rodriguez JL. Identification of patients at highest risk for ventilator-associated pneumonia in the surgical intensive care unit. Am J Surg 2000;179:8-11.
39. Chevret S, Hemmer M, Carlet J, Langer M. Incidence and risk factors of pneumonia acquired in intensive care units. Results from a multicenter prospective study on 996 patients. European Cooperative Group on Nosocomial Pneumonia. Intensive Care Med 1993;19:256-64.
40. Finucane TE, Bynum JP. Use of tube feeding to prevent aspiration pneumonia. Lancet 1996;348:1421-4.
41. Cole MJ, Smith JT, Molnar C, Shaffer EA. Aspiration after percutaneous gastrostomy. Assessment by Tc-99m labeling of the enteral feed. J Clin Gastroenterol 1987;9:90-5.
42. Hassett JM, Sunby C, Flint LM. No elimination of aspiration pneumonia in neurologically disabled patients with feeding gastrostomy. Surg Gynecol Obstet 1988;167:383-8.
43. Dent J, Dodds WJ, Friedman RH, et al. Mechanism of gastroesophageal reflux in recumbent asymptomatic human subjects. J Clin Invest 1980;65:256-67.
44. Jacobs S, Chang RW, Lee B, Bartlett FW. Continuous enteral feeding: a major cause of pneumonia among ventilated intensive care unit patients. JPEN J Parenter Enteral Nutr 1990;14:353-6.
45. Tejada Artigas A, Bello Dronda S, Chacon Valles E, et al. Risk factors for nosocomial pneumonia in critically ill trauma patients. Crit Care Med 2001;29:304-9.
46. Ahtaridis G, Snape WJ Jr, Cohen S. Lower esophageal sphincter pressure as an index of gastroesophageal acid reflux. Dig Dis Sci 1981;26:993-8.
47. Bartlett JG, Finegold SM. Anaerobic infections of the lung and pleural space. Am Rev Respir Dis 1974;110:56-77.
48. Finegold SM. Aspiration pneumonia. Rev Infect Dis 1991; 13(Suppl 9):S737-42.
49. Terpenning M, Shay K. Oral health is cost-effective to maintain but costly to ignore. J Am Geriatr Soc 2002;50:584-5.
50. Zasler ND, Devany CW, Jarman AL, Friedman R, Dinius A. Oral hygiene following traumatic brain injury: a programme to promote dental health. Brain Inj 1993;7:339-45.
51. Munro CL, Grap MJ. Oral health and care in the intensive care unit: state of the science. Am J Crit Care 2004;13:25-33; discussion, 34.
52. Talbot A, Brady M, Furlanetto DL, Frenkel H, Williams BO. Oral care and stroke units. Gerodontology 2005;22:77-83.
53. Varela JE, Varela K, Doherty J, Fishman D, Salzman S, Merlotti G. Incidence and predictors of aspiration after prolonged intubation in trauma patients. J Am Coll Surg 2004;199:76.
54. Kofteridis DP, Papadakis JA, Bouros D, et al. Nosocomial lower respiratory tract infections: prevalence and risk factors in 14 Greek hospitals. Eur J Clin Microbiol Infect Dis 2004;23:888-91.
55. Spray SB, Zuidema GD, Cameron JL. Aspiration pneumonia: incidence of aspiration with endotracheal tubes. Am J Surg 1976; 131:701-3.
56. Deshpande AA, Millis SR, Zafonte RD, Hammond FM, Wood DL. Risk factors for acute care transfer among traumatic brain injury patients. Arch Phys Med Rehabil 1997;78:350-2.
57. Alp E, Güven M, Yildiz O, Aygen B, Voss A, Doganay M. Incidence, risk factors and mortality of nosocomial pneumonia in intensive care units: a prospective study. Ann Clin Microbiol Antimicrob 2004;3:17.
58. Cross AS, Roup B. Role of respiratory assistance devices in endemic nosocomial pneumonia. Am J Med 1981;70:681-5.
59. Seidl RO, Nusser-Müller-Busch R, Ernst A. The influence of tracheotomy tubes on the swallowing frequency in neurogenic dysphagia. Otolaryngol Head Neck Surg 2005;132:484-6.
60. Shaker R, Milbrath M, Ren J, Campbell B, Toohill R, Hogan W. Deglutitive aspiration in patients with tracheostomy: effect of tracheostomy on the duration of vocal cord closure. Gastroenterology 1995;108:1357-60.
61. Buckwalter JA, Sasaki CT. Effect of tracheotomy on laryngeal function. Otolaryngol Clin North Am 1984;17:41-8.
62. Leder SB, Espinosa JF. Aspiration risk after acute stroke: comparison of clinical examination and fiberoptic endoscopic evaluation of swallowing. Dysphagia 2002;17:214-8.
63. Leibovitz A, Carmeli Y, Segal R. Effect of various antibacterial preparations on the pathogenic oral flora in elderly patients fed via nasogastric tube. Antimicrob Agents Chemother 2005;49:3566-8.
64. Reza Shariatzadeh M, Huang JQ, Marrie TJ. Differences in the features of aspiration pneumonia according to site of acquisition: community or continuing care facility. J Am Geriatr Soc 2006; 54:296-302.

Supplier

a. Version 13.0; SPSS Inc, 233 S Wacker Dr, 11th Fl, Chicago, IL 60606.

Study III

Hansen, T.S., Jakobsen, D.

III

A decision algorithm defining the rehabilitation approach: Facial Oral Tract Therapy (FOTT)

Accepted for publication 14.december 2009 in the journal: Disability and Rehabilitation

Title

A decision algorithm defining the rehabilitation approach: 'Facial Oral Tract Therapy' ®

Authors:

Trine Schow Hansen, M.Sci. Public Health, Daniela Jakobsen, OTR.

Affiliation:

Department of Neurorehabilitation, Brain Injury Unit, Department of Occupational Therapy, Copenhagen University Hospital Hvidovre, Hvidovre, Denmark

Correspondence to:

Trine Schow Hansen, Hvidovre Hospital, Department of Neurorehabilitation 354, Kettegaard Allé 30, DK-2650 Hvidovre Denmark

Phone work: +45 3632 6353

Fax number: + 45 3632 3312

Email: trine.schow@gmail.com

Phone home: +4522516782

Abstract

Title

A decision algorithm defining the rehabilitation approach: 'Facial Oral Tract Therapy' ®

Authors

Trine Schow Hansen, M.Sci. Public Health, Daniela Jakobsen, OTR.

Department of Neurorehabilitation, Brain Injury Unit and Department of Occupational Therapy, Copenhagen University Hospital Hvidovre, Hvidovre, Denmark

Purpose

To describe and define the rehabilitation approach: 'Facial Oral Tract Therapy' (F.O.T.T.)®

Method

We defined the content and process of the rehabilitation approach (F.O.T.T.)® in a decision-algorithm supported by a manual with supplementary material. The algorithm was developed by a research occupational therapist and an F.O.T.T.® senior instructor. We used an inductive approach combining existing knowledge from: F.O.T.T.® instructors, therapists trained in using the F.O.T.T.® approach and existing literature. A group of F.O.T.T.® instructors and the originator of the treatment approach Mrs. Kay Coombes has given comments to and approved the algorithm.

Result

The algorithm consist of 5 flowcharts: 'one assessment' chart guiding the therapist in the examination of the patient and four 'treatment charts', one for each of the four areas of F.O.T.T.®: Swallowing and eating; Oral hygiene; Breathing, voice and speech articulation; Facial expression, giving guidance on interventions. The algorithm outlines all important components in the treatment that the therapist should decide to use or not to use in the intervention. The algorithm is supported by a manual with criteria of when to use which components.

Conclusion

This algorithm is designed to be a practical guideline to therapists using F.O.T.T.® in clinical practice and in educational settings. The use of this algorithm may support standardization of F.O.T.T.® and thereby promote and maintain the quality in the treatment. This in turn will facilitate research that addresses F.O.T.T.® and outcomes.

1. Introduction

Over the last decade there has been a call for studies to investigate the effectiveness and efficacy of treatment approaches used in neurorehabilitation. However, systematic characterization and definition of the rehabilitation interventions involved are obstacles for such research.[38, 43] Such definitions are important for understanding the active ingredients of the treatment methods, for reproduction of the treatment and for generalization of research results[9, 43]. In this article, we present a simplified way to define the content and process of the rehabilitation approach Facial Oral Tract Therapy (F.O.T.T.®)[69]) in a decision-algorithm. Today, there exists no evidence for its efficacy or effectiveness. Before this can be tested, it's 'active ingredients' must be distilled into some replicable form, and one must have the ability to determine whether an individual clinician is delivering those ingredients to a patient. Such the development of a decision making algorithm is one step of many toward producing evidence of the efficacy and effectiveness of the F.O.T.T.

F.O.T.T.® is one of the approaches widely used in neurorehabilitation today[149], despite the low numbers of studies addressing its effectiveness or efficacy[108]. It is an inter-professional multidisciplinary approach and offers a structured way of evaluation and treatment of patients with disturbances in swallowing and eating, oral hygiene, non-verbal communication and speech articulation caused by neurological conditions[107]. These problems are very common in patients with injury to the central nervous system[73, 150-152].

The treatment approach used in F.O.T.T.® is special because it can be used to very severely injured patients, even patients in vegetative and minimal consciousness state. The patient does not need to be able to follow a verbal instruction in contrast to other treatment methods where the patient must have some level of functioning and should be able to follow instructions.

In F.O.T.T.® the therapist does not use a fixed sequence of exercises, but uses consistent principles to choose between different components (approaches) in order to support the patient in performing movement patterns as normally as possible[69]. The components are adjusted and structured so that patients constantly receive new information reinforcing the organization of new neural networks for motor control[120]. They are used in different combinations and with different intensities depending on the patient's responses and progress. Therefore the therapist continuously needs to analyze and (re) evaluate the patient's performance throughout the intervention to decide what components to

use and how to adjust them. This decision-making process enables adaptation of the treatment to meet the patient's needs, supporting neural plasticity and motor learning.[120] However, since F.O.T.T. ® is a multifaceted intervention involving several kinds of activation processes of recovery such as, learning, coping, adaptation and several constructs denoting neural or behavioral plasticity, it is by nature a complex treatment to use and define.[9, 48, 54] This complex approach, together with the lack of treatment manuals, can result in both a high level of inter-therapist and intra-therapist (inter-subject) variability, making it a technical challenge to carry out a research study.[153] However, defining the treatment process and components in a treatment manual could help therapists to follow a similar decision-making process and select appropriately from a list of components identified for patients with similar symptoms. This will assist the standardization of F.O.T.T.® and help reduce the inter-, and intra therapist variability in both clinical practice and research.[43, 153] Although the advantages of a treatment manual can seem obviously, stringent use of manuals has met with a lot of criticism from the clinicians[51]. One issue is that the manuals are often designed for prototypical patients[154] but since the variation of the patients situations (specially in severe TBI) and the complexity of clinical practice are immense it is difficult if not impossible to make a strict manual that is equally applicable to all patients[39]. Moreover, therapists have criticized manuals for reducing their ability to use intuition and adjust the treatment according to the patients needs. Therefore they will not follow a treatment manual in a strict way but strive to tailor each intervention to the individual patient [43, 51]. Henry et al. also found that dictating specific therapist behaviors so they follow a treatment manual very strictly may interfere with treatment outcome[155]. To overcome these challenges, one possibility is to define the treatment in a decision algorithm, where the specific choices of behavior are outlined, dictated by the patient's response[39, 43]. Algorithms have previously been used in defining types of interventions[156] or ways to navigate between different types of e.g. physical therapy treatments[157]. The use of a decision algorithm has the potential to provide greater flexibility allowing the therapist to individualize the treatment to the patients needs. This is valuable in neurorehabilitation since few patients are limited by a single impairment[39]. Still such a tool will provide the therapist with a guideline to specific treatments[43], and thereby balance the critical dimension of flexibility and specificity that are the great challenge in manuals for complex rehabilitation approaches[9, 158]. F.O.T.T. is used to patients with a broad range of impairments and performance problems and involves a broad range of components and decision processes. Making a strict manual for F.O.T.T. would limit the possibility to continuously adjust the treatment approach to the patients needs, which is an important component of this

treatment. Therefore we believe that a decision algorithm could be a possible way to make a useful manual within this complex rehabilitation approach.

The objective of this study was to develop a tool that would define the content and process of F.O.T.T.® in a systematically and simple way where the different components and the range of variations in their application in therapy are included. First we will describe more details of the F.O.T.T.® concept.

1.1. Facial Oral Tract Therapy (F.O.T.T.®)

F.O.T.T.®, developed by speech and language therapist Kay Coombes [159], is based on the Bobath concept.[160-161] The theoretical assumptions were originally derived from the principles of neurophysiology [109] and have evolved with subsequent knowledge of neuroplasticity and motor learning.[1, 6, 162-163] F.O.T.T.® covers 4 areas: Swallowing; Oral hygiene; Breathing/Voice production and speech articulation; Non-verbal communication[69]. These areas often influence each other. The patient is provided with structured input to promote experience of posture and movements that are as normal as possible[13]. There is little or no use of verbal instructions because the theoretical assumption is that motor learning occurs through successful performance[164]. The F.O.T.T.® approach is used with patients who have (severe) sensory-motor, perceptual and cognitive problems. In assessment and treatment of the patients, everyday life activities are used whenever possible to exploit relevant context[14]. The therapist may use techniques of oral stimulation, tongue mobilization, facilitation of swallowing and work to establish therapeutic routines of oral hygiene. In F.O.T.T.® postural control is recognized as fundamental to selective normal movement patterns for all activities, including movements of the face and oral tract.[113-115] Therefore positioning the patient to promote postural control that is as normal as possible is an integral part of the treatment. F.O.T.T.® differs from other swallowing therapies. It is an integrated treatment and assessment for swallowing, speech, breathing and facial expressions combined in one approach, which is unique compared to other. Moreover, in contrast to other treatments F.O.T.T.® uses functional activities and objects from everyday life where the therapist provides the patient with tactile information in order to facilitate movement that is as normal as possible instead of using verbal instructions mainly for exercises[14]. To our knowledge there is no other dysphagia- rehabilitation concept with a similar approach. In other behavioral therapeutic approaches the patient has to have sufficient perceptive, cognitive and sensomotorical prerequisites in order to perform strategies or maneuvers[165] like the

Mendelssohn maneuver[67], supra glottis swallowing [67] and the chin tuck maneuver [165-166] These strategies are focusing on airway protection, strengthening of muscles and compensation maneuvers where in F.O.T.T.® the therapist will strive for the patient to perform a movement or a movement pattern (eg chewing, drinking from a cup) as normal as possible and involve the patient as much as possible[14]. Since F.O.T.T.® is used in many countries, in many different neurorehabilitation settings, several courses is held in most parts of Europe every year [149], and the problems F.O.T.T.® addresses is very common[73, 76] it is highly relevant to explore the efficacy of this approach.

The treatment manual is intended to be useful for both clinical practice and research. We wanted it to be practical and contain all the information required to guide the therapist through the decision making process without being so detailed that it would be too cumbersome for anyone to use. Striving to achieve this balance resulted in a decision algorithm. The algorithm navigates through the different steps in the F.O.T.T.® intervention, leading the therapist to the important decisions and components in the therapeutic approach.

2. Development of the algorithm

The algorithm was developed by a research occupational therapist and an F.O.T.T.® senior instructor. We used an inductive approach combining existing knowledge from:

- F.O.T.T.® instructors
- Therapists trained in using the F.O.T.T. ® –approach
- Existing literature (books, articles[167-169], web sites [149, 159, 170] and F.O.T.T.® course material)

The structure in the algorithm follows the F.O.T.T.® model (figure 1). Using this model, the therapist begins with setting a goal for the patient based on the examination of the patient's abilities and problems and a hypothesis for the underlying causes. Then he/she chooses a strategy of how to reach that goal. The strategy includes choosing: a) an activity to work with, b) a of location of where the activity should take place and what furniture and objects to use, c) a therapeutic approach meaning how the therapist will work with the patient. Available components to each choice are outlined in the decision-algorithm separately to each area of F.O.T.T.®. While working with the patient according to the chosen strategy the therapist continuously observes the patient respond to the treatment and analyze if the strategy should be changed and how. To make the charts of the algorithm simple we did not include the decision rules in the charts, they are outlined in a

supplementing manual to each chart. The therapist continues analyzes patient response and adjusting approach accordingly to the responses until the session ends. Then she evaluates the choices she made in relation to the goal and the hypothesis of underlying causes to the patients problems.

The classifications of the elements in the therapeutic strategy follows the model of International Classification of Functioning, Disability and Health (ICF)[171]. We also used the 'person-environment occupational model' by Law et. al [172] used in occupational therapy. This model illustrates the system of occupational performance as an interaction of three elements: The activity (occupation), the person performing the activity and the environment.

Insert figure 1 approximately here

The algorithm has been presented to a group of F.O.T.T.® instructors [149]). An F.O.T.T.® instructor is certified by the originator of F.O.T.T.® Mrs. Kay Coombes to be qualified to arrange and teach at F.O.T.T.® courses. They reached consensus towards adding more components in the algorithm and we revised it taking account of their comments. It was presented at a F.O.T.T.® symposium in Bellikon, Switzerland and in Hamburg, Germany for Mrs. Kay Coombes and other therapists with special interest and experience in F.O.T.T. ®. Here we present an overview of the algorithm and illustrative examples since comprehensive details of each technique are too extensive to include in this paper. Complementation material is contained in a supporting manual (not published here).

3. The Algorithm

The content of the algorithm covers how to work with the patient; the supporting manual includes criteria for when to use various applications or therapy components. The algorithm consists of five charts: one assessment chart and four treatment charts.

Chart 1: What to look for (assessment chart)

Insert figure 2 approximately here

3.1. Choosing an area

- Before examining and treating the patient, important information about their condition and medical history is gathered. It is fundamental to begin with observing overall posture and how it influences the patient's function in the four F.O.T.T.® areas, for example in a first hand observation of the patient. This information is used to decide in which area to begin the examination; there is no pre-defined order, the choice depends on the individual patient's problems. Common problems in the four areas could be:
    - Oral Hygiene: Hypersensitive responses or bite reflex [173]
    - Breathing/voice and speech: Disturbed coordination between breathing and swallowing or disturbed voice and articulation[174]
    - Swallowing: Problems with eating and/ or drinking[175] or problems with coughing, including inefficient protection of the airway
    - Facial expressions: Reduced spontaneous facial movements, lack of selective movements of the head, jaw, arms and shoulders needed in nonverbal communication

The method of examining the patient is briefly described here and some of the intervention that may be indicated is outlined. It is fundamental to examine how disturbed tone, sensation and perception influences the patient's performance, and how the therapist can enable motor-sensory learning of normal movements and normal movement patterns and to find a way to support carry over in everyday life.

3.2. Swallowing

The purpose of examining swallowing is to investigate:

- If swallowing of saliva is effective and safe

  For example: protection of the airway in case of penetration and/or aspiration e.g. by spontaneous coughing, followed by a swallow.

The patient's ability to swallow saliva can be clinically examined by a visual and tactile examination of the mouth. The therapist will observe e.g. if the patient swallows spontaneously and if saliva is accumulated in the mouth.

- If the sequence of swallowing in eating and drinking is effective and safe.

(The term 'the swallowing sequence in eating and drinking' has been described by Kay Coombes since the 1970s emphasizing the importance of readiness and the preparatory or anticipatory pre-oral phase production e.g. on smelling food).

In F.O.T.T® the swallowing sequence is divided into four phases:

1. The pre-oral phase involves preparation and transport of food to the mouth. Preparation includes anticipation of the meal, coordination of movements of the eyes, arms and hands together with the movements of the trunk, head and jaw.

2. The oral-phase comprises:

- Forming of the bolus by biting, chewing and mixing food with saliva and
- Transport of the bolus through the oral cavity.

3. The normal pharyngeal phase involves transport of the bolus safely from the mouth through the pharynx and into the oesophagus with protection of the airway

4. The oesophageal phase comprises transport of the food through the oesophagus into the stomach

The therapist uses a visual and tactile examination of the mouth to look for causes to the patient's swallowing problems e.g. how does the patient manage the food, chew, transport it in the mouth, does the patient swallow spontaneously and protect airway. When relevant supplemented by an instrumental evaluation like Fiberoptic Endoscopic Evaluation of Swallowing (FEES).[90]

3.3. Oral hygiene

The purpose of examining oral hygiene is to investigate two aspects:

1) How the patient take care of oral hygiene
2) The status of the patients oral hygiene

The therapists will observe:

- The competence of the patient's spontaneous cleaning movements
- Sensation and tongue movements necessary for detection and removing remains of food in the oral cavity
- The patient's sensory-motor, perceptual and cognitive abilities for carrying out oral hygiene, e.g. brushing the teeth, rinsing, using products such as dental floss
- Investigate how the patient relearn necessary movements for oral hygiene by the therapist e.g. acts as a visual model or uses facilitation

Oral hygiene can be examined by e.g. visual and tactile examination of the mouth or by carrying out the tooth brushing involving the patient as much as possible and observing spontaneous performance.

3.4. Breathing/voice/speech articulation

The purpose of examining breathing/voice/speech articulation is to investigate:

- Location of movements e.g. upper chest breathing or intercostal-diaphragmatic breathing
- The rate, e.g. is the rate normal and does it change adequately when e.g. moving the patient
- Coordination of breathing and swallowing, which is important for protection of the airway [176-177]
- Thoracic and laryngeal coordination for making sound (vocalisation and breathing) and laryngeal and oral movement for articulated speech [178]
- Coordination of breathing and speech with active movements e.g. walking and talking at the same time

To examine this area the therapist e.g. uses their hands on the patient's chest to monitor breathing, listens to any spontaneous voice or tries to elicit sound in voiceless patients.

3.5. Facial expressions

The purpose of examining facial expressions is to investigate:

- Spontaneous facial movements
- The patient’s ability to use spontaneous, selective facial movements in different positions to verbal and non-verbal communication.

Examining facial expression is carried out e.g. by a visual and tactile examination of the face.

3.6. Performance problems?

During the examination the therapist observes if the patient has any performance problems and must at this stage make hypotheses of the underlying causes of these problems. Outlining the hypotheses provides a rationale for the treatment and guides the therapist in the intervention process. It also encourages the therapist to adjust the treatment individually to the patient needs, instead of using routine treatment approaches.[179]

If the therapist does not observe any performance problems the answer at this step in the algorithm is NO and the therapist will either choose to analyze another area or, if all areas have already been analyzed and no performance problems found, will conclude that the patient probably has no problems relevant for F.O.T.T.®.

If the answer is YES the therapist will choose the relevant area to work with:

3.7. Choice of chart:

It is not required at this stage to select the action chart with the same heading as the area just examined.

4. The four treatment-charts:

The four charts have the same design and decision flow. Each chart guides the therapist through the decision-making process within each area and through all the different components involved. Arrows on the left of the chart highlight the different steps. The different levels and approaches are combined and used according to the patient's needs. We will briefly describe each step in the charts (heading numbering follows the charts).

Insert figure 3 approximately here

1. Goal(s)

Goal setting is directed by the patient's problems and goals (if able to communicate them). In this algorithm the goal is expected to be attainable within a short timescale (days maybe up to 2 weeks; a short-term goal) and it must be clear and measurable. It should be associated with an activity where the level of participation of both the therapist and the patient is specified.

An example could be:

- To enable the patient to eat 100ml of purée or soft food in a sitting position, safely twice a day with assistance from nurse or therapist (assisted eating)

2. Strategy

2.a: Activity

The therapist chooses, if possible together with the patient, an activity for the intervention. The activity must be related to the goal and be meaningful for the patient.

An example could be:

- To eat small amounts of apple puree (3-5 teaspoons) safely

2.b: Environmental factors

The chosen environment should enable the patient to perform the activity as normally as possible:

- *2.b.1.Location*

  There can be several factors that determine the best place for the intervention. The ideal would be a room that is a normal place for the chosen activity. This will facilitate the patient's understanding and recognition of the situation and thereby also the movements that enables him to carry out the activity. Moreover, the therapist should consider if the situation might involve other people in the room. If the patient has problems with attention, concentration or perception, much auditory and visual stimulation may be deliberately avoided.

- *Furniture*

  The chosen furniture should be as relevant as possible to support the patient's recognition and understanding of the situation and at the same time support their postural posture. Sometimes the therapist has to make a compromise between these two challenges. In the example of eating small amounts of apple puree, the therapist positions the patient on a plinth with an adjustable table in front. This position helps the patient to come forward with his trunk while supporting the arms and gives the therapist the possibility to give manual support from behind. The sitting position at a table is quite normal for the activity 'eating'.

- *Objects and aids*

  Objects used in the treatment must again be normal for the activity. Special aids are used if they can help the patient to:

  - Move more normally than without aids
  - Use the less affected side of his body
  - Be more independent without increasing associated reactions, tonus and abnormal movement patterns

  Examples of object and aids can be: Packs to help when position the patient, Gauze to wrap in food the patient can chew on, special cup, special spoon (Cheyne spoon) reducing bite reaction, toothbrush with a thicker grip than normal, child toothbrush e.g.

2.c: Therapeutic intervention

Therapeutic intervention concerns the way the therapist supports the patient in the activity. The therapist has different approaches and working levels to choose between in the four areas. The methods are elaborated in the manual for each action chart, together with the criteria of when to use which level or approach. We will here outline the general approach available in all areas and highlight the differences.

The box: Working approach includes a list of therapeutic techniques relevant for all areas in F.O.T.T.®. This box is identical in each of the four action charts. It shows how to support the patient by:

- Positioning:
  Positioning means that the patient is brought to a certain position such as lying on the side or sitting, with support from the therapist and/or pillows or duvets as necessary. The goal is to normalize the patient's tonus and perception and get him into the best possible alignment for the activity/treatment. Positioning is used when the patient suffers from neuromuscular, musculoskeletal and perceptive problems that influence his postural control and the possibility of using selective movements. The patient is positioned before starting the activity, and the position is adjusted as needed during the intervention.
- Mobilization:
  Mobilization is applied to body parts or structures (muscles, joints or neural structures), which cannot move freely. The therapist can mobilize parts of the patient's body, e.g. the upper trunk or mobilize specific structures (like joints, muscles or nerves) to achieve a wider range of movement, more normal alignment or more normal tone.
- Guiding (principles from the Affolter concept[180]):
  Guiding is applied to patients with perceptual problems. The therapist uses physical guiding in problem-solving activities[180-181]. Guiding provides the patient with tactile-kinaesthetic experiences stimulating development and reconstruction of disordered performance[180].
- Elicit:
  Elicit means to bring about a response or reaction e.g. the therapist acts as a visual model of frowning to elicit movement of the patient’s forehead and eyebrows or the therapist moves the patient to another position to enable voice production in order to elicit a swallow.
- Facilitating:

Facilitation in an activity involves assisting the patient in the process of problem-solving so that movements become possible. It “requires manual contact to activate sensory and proprioceptive afferents, activate muscles or guide movement….”[182] and should result in change in motor behaviour. Facilitation can be adjusted according to the patient's responses. The therapist has 'hands on' until the patient responds and continues independently the movement- then the therapist takes 'hands off'. Facilitation can be addressed to support different sequences in the activity, as well as to single movements e.g. the therapist supports the jaw and floor of the mouth to facilitate tongue movement

The box with working levels is different in each area and includes more specific techniques defined in the F.O.T.T.® course material and in the manual. Moreover the box with protection of the airway is present in all charts.
We go briefly through them here:

Swallowing of saliva and eating

- The goal is to enable the patient to swallow his saliva safely and/or eat and drink safely by working on the phases of the swallowing sequence. The working levels range from using no food at all (oral stimulation) to offering OR use different amounts and consistencies of food in therapeutic eating. The therapist can use the different working approaches described earlier, either singly or in combination to enable the patient to swallow his saliva or eat/drink safely and as normally as possible. This chart also has a specific box for tongue movements, since the tongue is important in bolus forming and transport, and in swallowing, and a box for protection of the airway, because airway protection is always essential when working with swallowing and eating.

Insert figure 4 approximately here

Oral hygiene

- The main points in oral hygiene are to achieve and/or maintain a healthy mouth and enable the patient to learn the movements necessary for cleaning the mouth. The patient is involved in the whole sequence starting with preparing the requisites needed in the oral hygiene process. The box working levels outlines the different levels of use of requisites as tooth brush, dental floss

etc. Again it is important to support the patient in protecting the airway if relevant and the same box is included here.

Insert figure 5 approximately here

Breathing/voice and speech articulation

- Here the therapist has the possibility to combine working with breathing, voice and speech articulation with different positions or active movements in order to enable the patient to communicate and protect the airway. Levels of these methods are described in a box for supporting breathing and a box of using position at different levels. Again it is important to support the patient in protecting the airway if relevant and the same box is included here

Insert figure 6 approximately here

Facial expressions

- Facial expressions convey emotion. Abnormal muscle tone can disrupt facial expression (and eating) and the therapist can choose to work with passive or active facial movements in different positions depending on the patient's abilities.

3. Evaluating patient responses

After choosing the strategy the therapist applies it, monitors and adapts it and immediately evaluates (analyses) the patients responds to the intervention to update the analysis. The therapist must analyze whether the patient responds to the treatment in such a way that the activity is performed more normally.

4. Choose new strategy

If the patient performs the activity in a more normal way then the answer is YES and the therapist then may reduce the level of support or change to a more challenging activity. If the patient cannot perform the activity in a more normal way the answer is NO and the therapist might increase the level of support and/or change the activity to reduce . This re-evaluation process continues throughout the intervention.

5. Evaluate goal

At the end of each intervention the therapist evaluates whether or not the goal has been reached. The hypotheses made during the examination are combined with the different decisions and hypotheses made during the intervention. If the goal has been reached a new goal is set demanding a higher level of function of the patient, if it is not reached, and is unattainable in the near future, a new goal requiring a lower level of function will be set instead.

5. Discussion of the algorithm and utility

We have developed a therapeutic tool in the form of a decision algorithm to the rehabilitation concept Facial Oral Tract Therapy®. It outlines the various components of F.O.T.T.® and guides the therapist through the decision making process in this complex treatment approach.

The inductive method we used involving one researcher and one senior F.O.T.T.® instructor was very beneficial. The combination of clinical and theoretical background has made it possible to develop this tool. The theoretical and structural knowledge makes it possible to separate the components so they can be defined individually; the therapeutic knowledge is of course essential for developing a tool that reflects the treatment's content. This approach has been helpful bridging the gap between the clinical work and research.

The 'Person-environment occupation model'[172] illustrates the way the occupational therapist analyses occupational performance and structures their intervention, with a focus on using activities, environment and therapeutic support to improve performance, which we found matches the structure in F.O.T.T.®. The ICF model [183] offered an existing framework for classification and will at first sight make parts of the algorithm familiar for professionals seeing, which might ease understanding and adherence.

This algorithm provides a guideline through the different steps in F.O.T.T.® but the therapist is still left with a high level of flexibility when making the choices of which treatment approach to use. It has been suggested that the ability to individualize the treatment to each patient may be the active ingredient itself [29]. Though if outcome is to some extent influenced by treatment components, a high level of flexibility can influence the possibility to replicate treatment activities [9, 158]. In clinical practice several factors constantly affect and change the setting for the treatment session and we find this flexibility necessary to maintain the individually adjusted approach. Restriction of clinical innovation and clinical expertise of the therapist is also one issue where other treatment manuals

have been criticized[51, 184]) Though to follow our algorithm in the "right way" or how to make the "most appropriate choices" we recommend that the therapist gets an introduction to the F.O.T.T.® concept (preferable a F.O.T.T.® course) and follows the manual for each chart. As described by Calhoun et al.[154], it is not the case that a treatment manual is sufficient to learn the technique; additional training is required of the therapists to achieve necessary competence. Other studies [157, 185] using decision algorithms or treatment manuals to guide the therapist in using complex treatments have not defined each component in a manual. Instead they has used a patient centered approach, where the therapeutic activities are classified relating to one single impairment like risk of falling[186] or they combine several professions, like occupational therapy and physiotherapy in a more broad guideline to rehabilitation of stroke patients[55]. Thus the intension has not been to guide the therapist through just one rehabilitation approach but to guide the therapist through the process from assessment of a patient to the choice of treatment[52-55]. In contrast our intention was to outline and define F.O.T.T.® by itself. The treatment manual should provide an overview of the content and process of this approach and be used both in research and in the clinic. This algorithm is to guide both the experienced and the inexperienced therapist through the same decision-making process, and to work in a goal-oriented manner which hopefully will support a more standardized practice of F.O.T.T.®. Moreover this tool can be useful in educational settings and in communication in interdisciplinary teams. The weakness with such a model as the algorithm is that it cannot capture all details of a complex treatment approach. However, we do believe that therapists using the algorithm will treat patients in a more adequate way because they have a guideline in the process of examination and treatment. Of course this has to be tested in future studies, starting by developing and testing an adherence measure[26], which is in process. As mentioned earlier a treatment manual serves many purposes. The development of a manual has been suggested to be a step wise process, where each successive step may lead to more complex clinical issues. Carroll & Nuro [187] suggested a parallel stage model aiming at supporting the development of "clinical-friendly" manual that would facilitate greater use of empirically supported treatments in clinical practice This stage model goes from stage I (where the critical role of the manual is to define the treatment in broad strokes for preliminary evaluation of feasibility and efficacy) to stage II (where the manual can be used as the basis for training therapists and linking process to outcome) to stage III (where the manual may be used to for example replications of clinical trials in other settings and ultimately to serve as a component of clinical care standards as well as a tool used in training of clinicians)[187] This F.O.T.T.® decision algorithm is in its early development at stage I. It still needs further descriptions

of the theoretical constructs and how theory relates to each hypothesized active ingredient [9]. Moreover there is limited specific information of how the therapists exactly should perform or deliver the active ingredients in the right context and appropriate manner (stage II), which might need to be made as a practical handbook that relates to the decision algorithm since incorporating this in the algorithm's flowchart or supporting manual will make an extensive tool. The next step with this treatment algorithm will for example be to implement it in clinical practice and investigate its capability to structure therapist behavior so they use F.O.T.T.® in a more standardized way. Then it might be taken to the next levels in the stage model and be used in efficacy and effectiveness clinical trials.

One of the first ideas by creating this algorithm was to open one of the 'black boxes' in neurorehabilitation and define the components of F.O.T.T.® making it possible to outline and evaluate the hypothesized active ingredients. However, this algorithm still leaves several candidates to be active ingredients. What we have is a framework outlining all components. Depending on the specific research question, future studies need to specify parts of this algorithm in greater detail. Doing this one needs to outline the theory describing which components that are hypothesized to change a functional deficit (performance problem) and how. Defining the treatment in greater details can serve to narrow the different variables that are hypothesized to exert effect as suggested by Whyte and Hart [43] supporting both research and clinical practice. In order to follow this algorithm the therapists already needs to make a hypothesis based on a theory of the desired change. There are many components and theories to be tested in F.O.T.T.® and this algorithm makes it more clear how to test them. However, the high level of flexibility might affect the possibility to establish internal validity. The clearer and more specific the treatment manual is the more likely the treatment as practiced will reflect the intension and actual mechanism of the treatment and outcome, but if it is too specific it might not reflect the treatment that actually goes on in real clinical settings and thereby fail to establish external validity. Moreover, many clinicians are concerned of the use with manuals that do not provide any flexibility. Thus, such a manual can have a negative impact on therapeutic alliance (or adherence to the algorithm). We made this algorithm as a "therapist-friendly" manual reflecting the complexity of F:O.T.T. by focusing on describing the essential key decisions and active ingredients of the therapy[26]. But how this manual balance the trade off of internal validity (is it specific enough to guide therapeutic behavior in a clinical trial) and external validity (can it be implemented in real rehabilitation setting[43]) is still to be evaluated.

Another research perspective could be to investigate the decision rules as the active ingredients which again should be supported by theoretical assumptions[43]. These rules are not explicit in this algorithm, but are supported in the manual.

6. Conclusion

We have developed a decision algorithm that systematically characterizes and defines the content and process of the rehabilitation concept Facial Oral Tract Therapy (F.O.T.T.®). We believe that this tool provides the therapist with a guideline to the variety of components and decision processes in F.O.T.T.®, still leaving the therapist with the flexibility to adjust the treatment to the patient's needs and responses. We hope it will be used in clinical practice and educational settings so that the quality and outcome of the treatment are maintained and standardized. Finally we hope that it will support outlining the theoretical hypothesis and thereby facilitate the necessary efficacy studies.

Acknowledgements

The authors would like to thank Professor John Whyte for inspiration and discussion during the development of this algorithm. We are also very grateful to the FOrmaTT group for their constructive comments on the algorithm, as well as the occupational therapists at our Department, and to Ingrid Poulsen Med dr. for critical reading of this paper. We are also very pleased that Ms Kay Coombes, the originator of F.O.T.T.® has been very supportive of our work and we thank her for critical reading of this paper together with Margaret Walker.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

References

1. Nudo RJ. Functional and structural plasticity in motor cortex: implications for stroke recovery. *Physical medicine and rehabilitation clinics of North America.* Feb 2003;14(1 Suppl):S57-76.

2. Bach-y-Rita P. Theoretical basis for brain plasticity after a TBI. *Brain Inj.* Aug 2003;17(8):643-651.

3. Robertson IH, Murre JMJ. Rehabilitation of brain damage: Brain plasticity and principles of guided recovery. *Psychol Bull.* 1999;125(5):544-576.

4. Kleim JA, Jones TA. Principles of experience-dependent neural plasticity: implications for rehabilitation after brain damage. *J Speech Lang Hear Res.* Feb 2008;51(1):S225-239.

5. Bütefisch C. Neurobiological bases of rehabilitation. *Neurological Sciences.* 2006;27(0):s18-s23.

6. Nudo RJ. Plasticity. *NeuroRx : the journal of the American Society for Experimental NeuroTherapeutics.* 2006;3(4):420-427.

7. Mogensen J, Hjortkjaer J, Ibervang KL, Stedal K, Mala H. Prefrontal cortex and hippocampus in posttraumatic functional recovery: spatial delayed alternation by rats subjected to transection of the fimbria-fornix and/or ablation of the prefrontal cortex. *Brain Res Bull.* Jun 15 2007;73(1-3):86-95.

8. Mala H, Castro MR, Knippel J, Kohler PJ, Lassen P, Mogensen J. Therapeutic effects of a restraint procedure on posttraumatic place learning in fimbria-fornix transected rats. *Brain Res.* Jun 27 2008;1217:221-231.

9. Hart T. Treatment definition in complex rehabilitation interventions. *Neuropsychol Rehabil.* Jun 18 2009:1-17.

10. Taub E, Uswatte G, Pidikiti R. Constraint-Induced Movement Therapy: a new family of techniques with broad application to physical rehabilitation--a clinical review. *J Rehabil Res Dev.* Jul 1999;36(3):237-251.

11. Mark VW, Taub E. Constraint-induced movement therapy for chronic stroke hemiparesis and other disabilities. *Restor Neurol Neurosci.* 2004;22(3-5):317-336.

12. Cheeran B, Cohen L, Dobkin B, et al. The future of restorative neurosciences in stroke: driving the translational research pipeline from basic science to rehabilitation of people after stroke. *Neurorehabil Neural Repair.* Feb 2009;23(2):97-107.

13. Coombes K. F.O.T.T. (Facial-Oral Tract Therapy). *iADH Magazine.* Vol spring, 2008: International Association For Disability and Oral Health; 2008.

14. Nusser-Müller-Busch. *Die Therapie des Facio-Oralen Trakts - F.O.T.T. nach Kay Coombes.* Vol 2. Berlin: Springer; 2007.

15. Thurman D, Sniezek J, Johnson D, al. e. Guidelines for surveillance of central nercous system injury. . In: services DoHaH, ed. Atlante, Georgia: National Center for Injury Prevention and Control, Centers for Disease Control and Prevention, US; 1995.

16. Greenwald BD, Burnett DM, Miller MA. Congenital and acquired brain injury. 1. Brain injury: epidemiology and pathophysiology. *Arch Phys Med Rehabil.* Mar 2003;84(3 Suppl 1):S3-7.

17. Michaud CM, Murray CJ, Bloom BR. Burden of disease--implications for future research. *JAMA.* Feb 7 2001;285(5):535-539.

18. Braverman SE, Spector J, Warden DL, et al. A multidisciplinary TBI inpatient rehabilitation programme for active duty service members as part of a randomized clinical trial. *Brain Inj.* Jun 1999;13(6):405-415.

19. Semlyen JK, Summers SJ, Barnes MP. Traumatic brain injury: efficacy of multidisciplinary rehabilitation. *Arch Phys Med Rehabil.* Jun 1998;79(6):678-683.

20. Shiel A, Burn JP, Henry D, et al. The effects of increased rehabilitation therapy after brain injury: results of a prospective controlled trial. *Clin Rehabil.* Oct 2001;15(5):501-514.

21. Engberg AW, Liebach A, Nordenbo A. Centralized rehabilitation after severe traumatic brain injury - a population-based study. *Acta Neurol Scand.* 2006;113(3):178-184.

22. Liebach A, Nordenbo AM, Engberg AW. [Subacute intensive rehabilitation after severe traumatic brain injury. Follow-up 6 months after discharge]. *Ugeskr Laeger.* Jan 15 2007;169(3):223-227.

23. Sundhedsstyrelsen. Behandling af traumatiske hjerneskader og tilgrænsende lidelser. . In: Health) NBo, ed. København, Denmark: : Komiteen for Sundhedsoplysning; 1997.

24. Engberg A.W, Teasdale TW. Traumatic brain injury in Denmark 1979-1996. A national study of incidence and mortality. *Eur J Epidemiol.* 2001;17(5):437-442.

25. Whyte J. Rehabilitation of Individuals With Traumatic Brain Injury: Status of the Art and Science. *Rehabilitation of Persons With Traumatic Brain Injury.* Vol 16: NIH Consens Statement; 1998:23-30.

26. Nezu AM, Nezu CM. *Evidence-based outcome research*. New York2008.

27. Whyte J. A Grand Unified Theory of Rehabilitation (We Wish!). The 57th John Stanley Coulter Memorial Lecture. *Archives of Physical Medicine and Rehabilitation.* 2008;89(2):203-209.

28. Cope DN. The effectiveness of traumatic brain injury rehabilitation: a review. *Brain Inj.* Oct 1995;9(7):649-670.

29. Carney N, Chesnut RM, Maynard H, Mann NC, Patterson P, Helfand M. Effect of cognitive rehabilitation on outcomes for persons with traumatic brain injury: A systematic review. *J Head Trauma Rehabil.* Jun 1999;14(3):277-307.

30. Rice-Oxley M, Turner-Stokes L. Effectiveness of brain injury rehabilitation. *Clin Rehabil.* 1999;13 Suppl 1:7-24.

31. Cullen N, Chundamala J, Bayley M, Jutai J. The efficacy of acquired brain injury rehabilitation. *Brain Inj.* Feb 2007;21(2):113-132.

32. Greenwald BD, Rigg JL. Neurorehabilitation in traumatic brain injury: does it make a difference? *Mount Sinai Journal of Medicine: A Journal of Translational and Personalized Medicine.* 2009;76(2):182-189.

33. Gordon WA, Zafonte R, Cicerone K, et al. Traumatic brain injury rehabilitation: state of the science. *Am J Phys Med Rehabil.* Apr 2006;85(4):343-382.

34. Johnston MV, Sherer M, Whyte J. Applying evidence standards to rehabilitation research. *Am J Phys Med Rehabil.* Apr 2006;85(4):292-309.

35. Sackett DL, Rosenberg WM, Gray JA, Haynes RB, Richardson WS. Evidence based medicine: what it is and what it isn't. *BMJ.* Jan 13 1996;312(7023):71-72.

36. Moses LE. Measuring effects without randomized trials? Options, problems, challenges. *Med Care.* Apr 1995;33(4 Suppl):AS8-14.

37. Powell JM, Temkin NR, Machamer JE, Dikmen SS. Nonrandomized studies of rehabilitation for traumatic brain injury: Can they determine effectiveness? *Arch Phys Med Rehabil.* 2002;83(9):1235-1244.

38. Whyte J. Clinical trials in rehabilitation: what are the obstacles? *American journal of physical medicine & rehabilitation* Oct 2003;82(10 Suppl):S16-21.

39. Whyte J. Directions in brain injury research: From concept to clinical implementation. *Neuropsychol Rehabil.* Jun 19 2009:1-17.

40. Grimes DA, Schulz KF. An overview of clinical research: the lay of the land. *Lancet.* Jan 5 2002;359(9300):57-61.

41. Perdices M, Tate RL. Single-subject designs as a tool for evidence-based clinical practice: Are they unrecognised and undervalued? *Neuropsychol Rehabil.* Jul 28 2009:1-24.

42. Tate RL, McDonald S, Perdices M, Togher L, Schultz R, Savage S. Rating the methodological quality of single-subject designs and n-of-1 trials: introducing the Single-Case Experimental Design (SCED) Scale. *Neuropsychol Rehabil.* Aug 2008;18(4):385-401.

43. Whyte J, Hart T. It's more than a black box; it's a Russian doll: defining rehabilitation treatments. *American journal of physical medicine & rehabilitation* Aug 2003;82(8):639-652.

44. (WHO) WHO. International Classification of Functioning, Disability, and Health (ICF). . [http://www.who.int/classifications/icf/en/]. Accessed October 7, 2009.

45. Koskinen S, Hokkinen EM, Sarajuuri J, Alaranta H. Applicability of the ICF checklist to traumatically brain-injured patients in post-acute rehabilitation settings. *J Rehabil Med.* Jul 2007;39(6):467-472.

46. Bernabeu M, Laxe S, Lopez R, et al. Developing core sets for persons with traumatic brain injury based on the international classification of functioning, disability, and health. *Neurorehabil Neural Repair.* Jun 2009;23(5):464-467.

47. Roland PE, Hanazawa A, Undeman C, et al. Cortical feedback depolarization waves: a mechanism of top-down influence on early visual areas. *Proc Natl Acad Sci U S A.* Aug 15 2006;103(33):12586-12591.

48. DeJong G, Horn SD, Conroy B, Nichols D, Healton EB. Opening the Black Box of Poststroke Rehabilitation: Stroke Rehabilitation Patients, Processes, and Outcomes. *Archives of Physical Medicine and Rehabilitation.* 2005;86(12):1-7.

49. Whyte J. Clinical trials in rehabilitation: what are the obstacles? *Am J Phys Med Rehabil.* Oct 2003;82(10 Suppl):S16-21.

50. Hart T. Treatment definition in experience-based rehabilitation research.

51. Addis ME, Wade WA, Hatgis C. Barriers to Dissemination of Evidence-Based Practices: Addressing Practitioners' Concerns About Manual-Based Psychotherapies. *Clinical Psychology: Science and Practice.* 1999;6(4):430-441.

52. Schenkman M, Deutsch JE, Gill-Body KM. An Integrated Framework for Decision Making in Neurologic Physical Therapist Practice. *Physical Therapy.* 2006;86(12):1681-1702.

53. Rothstein JM, Echternach JL. Hypothesis-oriented algorithm for clinicians. A method for evaluation and treatment planning. *Physical Therapy.* Sep 1986;66(9):1388-1394.
54. Dejong G, Horn SD, Gassaway JA, Slavin MD, Dijkers MP. Toward a taxonomy of rehabilitation interventions: Using an inductive approach to examine the "black box" of rehabilitation. *Archives of Physical Medicine and Rehabilitation.* Apr 2004;85(4):678-686.
55. Ballinger C, Ashburn A, Low J, Roderick P. Unpacking the black box of therapy -- a pilot study to describe occupational therapy and physiotherapy interventions for people with stroke. *Clin Rehabil.* Aug 1999;13(4):301-309.
56. Hogue A, Liddle HA, Rowe C. Treatment adherence process research in family therapy: A rationale and some practical guidelines. *Psychotherapy: Theory, Research, Practice, Training.* Sum 1996;33(2):332-345.
57. Bellg AJ, Borrelli B, Resnick B, et al. Enhancing treatment fidelity in health behavior change studies: best practices and recommendations from the NIH Behavior Change Consortium. *Health Psychol.* Sep 2004;23(5):443-451.
58. Hogue A. Adherence process research on developmental interventions: filling in the middle. *New Dir Child Adolesc Dev.* Winter 2002(98):67-74.
59. Granger CV, Hamilton BB, Linacre JM, Heinemann AW, Wright BD. Performance profiles of the functional independence measure. *Am J Phys Med Rehabil.* Apr 1993;72(2):84-89.
60. Linacre JM, Heinemann AW, Wright BD, Granger CV, Hamilton BB. The structure and stability of the Functional Independence Measure. *Arch Phys Med Rehabil.* Feb 1994;75(2):127-132.
61. Wade DT, Smeets RJ, Verbunt JA. Research in rehabilitation medicine: Methodological challenges. *J Clin Epidemiol.* Sep 27 2009.
62. Draper K, Ponsford J. Cognitive functioning ten years following traumatic brain injury and rehabilitation. *Neuropsychology.* Sep 2008;22(5):618-625.
63. Kwakkel G, van Peppen R, Wagenaar RC, et al. Effects of augmented exercise therapy time after stroke: a meta-analysis. *Stroke.* Nov 2004;35(11):2529-2539.
64. Dodds TA, Martin DP, Stolov WC, Deyo RA. A validation of the functional independence measurement and its performance among rehabilitation inpatients. *Arch Phys Med Rehabil.* May 1993;74(5):531-536.
65. Jenkinson C, Wright L, Coulter A. The SF 36 health survey questionnaire. ...if used within its limits. *BMJ.* Aug 14 1993;307(6901):449.
66. Hamdy S, Aziz Q, Thompson DG, Rothwell JC. Physiology and pathophysiology of the swallowing area of human motor cortex. *Neural Plast.* 2001;8(1-2):91-97.
67. Logemann JA. *Evaluation and treatment of swallowing disorders.* Vol 2: PRO-ED, Inc.; 1983.
68. Leopold NA, Kagel MC. Dysphagia--Ingestion or Deglutition?: A Proposed Paradigm. *Dysphagia.* 1997;12(4):202-206.
69. Coombes K. Facial oral tract therapy (F.O.T.T.). *1991-2001 Jubiläumsschrift-10 Jahre Schulungszentrum, Therapie Zentrum Burgau*:
; 2001.
70. Kaplan MD, Baum BJ. The functions of saliva. *Dysphagia.* 1993;8(3):225-229.
71. Dodds WJ. Physiology of swallowing. *Dysphagia.* 1989;3(4):171-178.
72. Kahrilas PJ, Lin S, Logemann JA, Ergun GA, Facchini F. Deglutitive tongue action: volume accommodation and bolus propulsion. *Gastroenterology.* Jan 1993;104(1):152-162.
73. Mackay LE, Morgan AS, Bernstein BA. Swallowing disorders in severe brain injury: risk factors affecting return to oral intake. *Archives of Physical Medicine and Rehabilitation.* Apr 1999;80(4):365-371.
74. Cherney LR, Halper, AS. Recovery of oral nutrition after head injury in adults. *J head trauma rehabil.* 1989;4(4):42-50.
75. Winstein CJ. Neurogenic dysphagia. Frequency, progression, and outcome in adults following head injury. *Phys Ther.* Dec 1983;63(12):1992-1997.
76. Hansen TS, Engberg AW, Larsen K. Functional oral intake and time to reach unrestricted dieting for patients with traumatic brain injury. *Arch Phys Med Rehabil.* Aug 2008;89(8):1556-1562.
77. Terre R, Mearin F. Prospective evaluation of oro-pharyngeal dysphagia after severe traumatic brain injury. *Brain Inj.* Dec 2007;21(13-14):1411-1417.
78. Avery-Smith W, Dellarosa DM. Approaches to treating dysphagia in patients with brain injury. *Am J Occup Ther.* Mar 1994;48(3):235-239.
79. Murdoch B, Theodoros D. *Traumatic Brain Injury: Associated Speech, Language, and Swallowing Disorders* San Diego, CA: Singular Thomson Learning 2001.
80. McCullough GH, Wertz RT, Rosenbek JC. Sensitivity and specificity of clinical/bedside examination signs for detecting aspiration in adults subsequent to stroke. *J Commun Disord.* Jan-Apr 2001;34(1-2):55-72.

81. Silva AB, Piovesana AM, Barcelos IH, Capellini SA. [Clinical and videofluoroscopic evaluation of swallowing in patients with spastic tetraparetic cerebral palsy and athetosic cerebral palsy]. *Rev Neurol.* Apr 16-30 2006;42(8):462-465.
82. Smith HA, Lee SH, O'Neill PA, Connolly MJ. The combination of bedside swallowing assessment and oxygen saturation monitoring of swallowing in acute stroke: a safe and humane screening tool. *Age Ageing.* Nov 2000;29(6):495-499.
83. Smithard DG, O'Neill PA, Park C, et al. Can bedside assessment reliably exclude aspiration following acute stroke? *Age Ageing.* Mar 1998;27(2):99-106.
84. Lim SH, Lieu PK, Phua SY, et al. Accuracy of bedside clinical methods compared with fiberoptic endoscopic examination of swallowing (FEES) in determining the risk of aspiration in acute stroke patients. *Dysphagia.* Winter 2001;16(1):1-6.
85. Logemann JA, Pauloski BR, Rademaker AW, et al. Speech and swallow function after tonsil/base of tongue resection with primary closure. *J Speech Hear Res.* Oct 1993;36(5):918-926.
86. Langmore SE, Schatz K, Olson N. Endoscopic and videofluoroscopic evaluations of swallowing and aspiration. *Ann Otol Rhinol Laryngol.* Aug 1991;100(8):678-681.
87. Hoppers P, Holm SE. The role of fiberoptic endoscopy in dysphagia rehabilitation. *J Head Trauma Rehabil.* Oct 1999;14(5):475-485.
88. Hiss SGPCCCSLP, Postma GNMD. Fiberoptic Endoscopic Evaluation of Swallowing. [Article].
89. Wu CH, Hsiao TY, Chen JC, Chang YC, Lee SY. Evaluation of swallowing safety with fiberoptic endoscope: comparison with videofluoroscopic technique. *Laryngoscope.* Mar 1997;107(3):396-401.
90. Langmore SE, Schatz K, Olsen N. Fiberoptic endoscopic examination of swallowing safety: a new procedure. *Dysphagia.* 1988;2(4):216-219.
91. Ashford J, McCabe D, Wheeler-Hegland K, et al. Evidence-based systematic review: Oropharyngeal dysphagia behavioral treatments. Part III--Impact of dysphagia treatments on populations with neurological disorders. *Journal of Rehabilitation Research & Development.* 2009;46(2):195-204.
92. Robbins J, Gensler G, Hind J, et al. Comparison of 2 interventions for liquid aspiration on pneumonia incidence: a randomized trial. *Ann Intern Med.* Apr 1 2008;148(7):509-518.
93. Shanahan TK, Logemann JA, Rademaker AW, Pauloski BR, Kahrilas PJ. Chin-down posture effect on aspiration in dysphagic patients. *Arch Phys Med Rehabil.* Jul 1993;74(7):736-739.
94. Nagaya M, Kachi T, Yamada T, Sumi Y. Videofluorographic observations on swallowing in patients with dysphagia due to neurodegenerative diseases. *Nagoya J Med Sci.* May 2004;67(1-2):17-23.
95. Okada S, Saitoh E, Palmer JB, et al. What is the chin-down posture? A questionnaire survey of speech language pathologists in Japan and the United States. *Dysphagia.* Jul 2007;22(3):204-209.
96. Logemann JA, Kahrilas PJ, Kobara M, Vakil NB. The benefit of head rotation on pharyngoesophageal dysphagia. *Arch Phys Med Rehabil.* Oct 1989;70(10):767-771.
97. Drake W, Donoghue SO, Bartram C, Lindsay J, Greenwood R. Case study Eating in side lying facilitates rehabilitation in neurogenic dysphagia. *Brain Injury.* 1997;11(2):137-142.
98. Lazarus CL, Logemann JA, Rademaker AW, et al. Effects of bolus volume, viscosity, and repeated swallows in nonstroke subjects and stroke patients. *Arch Phys Med Rehabil.* Oct 1993;74(10):1066-1070.
99. de Lama Lazzara G, Lazarus C, Logemann J. Impact of thermal stimulation on the triggering of the swallowing reflex. *Dysphagia.* 1986;1(2):73-77.
100. Tsukada T, Taniguchi H, Ootaki S, Yamada Y, Inoue M. Effects of food texture and head posture on oropharyngeal swallowing. *J Appl Physiol.* June 1, 2009 2009;106(6):1848-1857.
101. Logemann JA, Gensler G, Robbins J, et al. A randomized study of three interventions for aspiration of thin liquids in patients with dementia or Parkinson's disease. *J Speech Lang Hear Res.* Feb 2008;51(1):173-183.
102. Logemann JA. Dysphagia: evaluation and treatment. *Folia Phoniatr Logop.* 1995;47(3):140-164.
103. Lazarus C, Logemann JA, Gibbons P. Effects of maneuvers on swallowing function in a dysphagic oral cancer patient. *Head Neck.* Sep-Oct 1993;15(5):419-424.
104. Poertner LC, Coleman RF. Swallowing therapy in adults. *Otolaryngol Clin North Am.* Jun 1998;31(3):561-579.
105. Yuen HK, Hartwick JA. Diet manipulation to resume regular food consumption for an adult with traumatic brain injury. *Am J Occup Ther.* Oct 1992;46(10):943-945.
106. Tippett D, Palmer J, Linden P. Management of dysphagia in a patient with closed head injury. *Dysphagia.* 1987;1(4):221-226.
107. Nusser-Muller-busch R. *Konsensusempfehlungen 2007 zur Facio-Oralen Trakt Therapie (F.O.T.T.)*2007.

108. Seidl RO, Nusser-Muller-Busch R, Hollweg W, Westhofen M, Ernst A. Pilot study of a neurophysiological dysphagia therapy for neurological patients. *Clinical Rehabilitation.* Aug 2007;21(8):686-697.

109. Raine S. Defining the Bobath concept using the Delphi technique. *Physiotherapy research international.* Mar 2006;11(1):4-13.

110. Coombes K. Facial Oral Tract Therapy: F.O.T.T™. [http://www.arcos.org.uk/index.php?option=com_content&view=article&id=60&Itemid=66]. http://www.fott.co.uk/. Accessed September, 2009.

111. Martin BJ, Logemann JA, Shaker R, Dodds WJ. Coordination between respiration and swallowing: respiratory phase relationships and temporal integration. *Journal of Applied Physiology.* Feb 1994;76(2):714-723.

112. Nusser-Muller-Busch R, Seidl RO. Therapie- und Tracheal-kanülemangement bei neurogenen dysphagien (phase A und B) eine Fallbesceibung. *Forum logopädie.* 2004;6(18):20-26.

113. Frank JS, Earl M. Coordination of posture and movement. *Physical Therapy.* December 1, 1990 1990;70(12):855-863.

114. De Mayo T, Miralles R, Barrero D, et al. Breathing type and body position effects on sternocleidomastoid and suprahyoid EMG activity. *Journal of Oral Rehabilitation.* 2005;32(7):487-494.

115. Ayuse T, Ishitobi S, Kurata S. Effect of reclining and chin-tuck position on the coordination between respiration and swallowing. *Journal of Oral Rehabilitation.* 2006;33(6):402-409.

116. Robbins J, Butler SG, Daniels SK, et al. Swallowing and dysphagia rehabilitation: translating principles of neural plasticity into clinically oriented evidence. *J Speech Lang Hear Res.* Feb 2008;51(1):S276-300.

117. Jones TA, Chu CJ, Grande LA, Gregory AD. Motor skills training enhances lesion-induced structural plasticity in the motor cortex of adult rats. *J Neurosci.* Nov 15 1999;19(22):10153-10163.

118. Biernaskie J, Corbett D. Enriched rehabilitative training promotes improved forelimb motor function and enhanced dendritic growth after focal ischemic injury. *J Neurosci.* Jul 15 2001;21(14):5272-5280.

119. Farrell R, Evans S, Corbett D. Environmental enrichment enhances recovery of function but exacerbates ischemic cell death. *Neuroscience.* 2001;107(4):585-592.

120. Mulder T, Hochstenbach J. Adaptability and flexibility of the human motor system: implications for neurological rehabilitation. *Neural Plasticity.* 2001;8(1-2):131-140.

121. Kleim JA, Barbay S, Cooper NR, et al. Motor learning-dependent synaptogenesis is localized to functionally reorganized motor cortex. *Neurobiol Learn Mem.* Jan 2002;77(1):63-77.

122. Dimyan MA, Weinberger NM. Basal forebrain stimulation induces discriminative receptive field plasticity in the auditory cortex. *Behav Neurosci.* Aug 1999;113(4):691-702.

123. Fraser C, Rothwell J, Power M, Hobson A, Thompson D, Hamdy S. Differential changes in human pharyngoesophageal motor excitability induced by swallowing, pharyngeal stimulation, and anesthesia. *Am J Physiol Gastrointest Liver Physiol.* Jul 2003;285(1):G137-144.

124. Hamdy S, Aziz Q, Rothwell JC, et al. Explaining oropharyngeal dysphagia after unilateral hemispheric stroke. *Lancet.* Sep 6 1997;350(9079):686-692.

125. Rosenbek JC, Robbins J, Willford WO, et al. Comparing treatment intensities of tactile-thermal application. *Dysphagia.* Winter 1998;13(1):1-9.

126. Maegele M, Lippert-Gruener M, Ester-Bode T, et al. Multimodal early onset stimulation combined with enriched environment is associated with reduced CNS lesion volume and enhanced reversal of neuromotor dysfunction after traumatic brain injury in rats. *Eur J Neurosci.* May 2005;21(9):2406-2418.

127. Lippert-Gruner M, Maegele M, Pokorny J, et al. Early rehabilitation model shows positive effects on neural degeneration and recovery from neuromotor deficits following traumatic brain injury. *Physiol Res.* 2007;56(3):359-368.

128. Page S, Levine P. Forced use after TBI: promoting plasticity and function through practice. *Brain Inj.* Aug 2003;17(8):675-684.

129. Taub E, Uswatte G, Mark VW, Morris DM. The learned nonuse phenomenon: implications for rehabilitation. *Eura Medicophys.* Sep 2006;42(3):241-256.

130. Risedal A, Zeng J, Johansson BB. Early training may exacerbate brain damage after focal brain ischemia in the rat. *J Cereb Blood Flow Metab.* Sep 1999;19(9):997-1003.

131. Humm JL, Kozlowski DA, James DC, Gotts JE, Schallert T. Use-dependent exacerbation of brain damage occurs during an early post-lesion vulnerable period. *Brain Res.* Feb 9 1998;783(2):286-292.

132. Boyd L, Winstein C. Explicit information interferes with implicit motor learning of both continuous and discrete movement tasks after stroke. *J Neurol Phys Ther.* Jun 2006;30(2):46-57; discussion 58-49.

133. Taub E, Uswatte G. Constraint-induced movement therapy: bridging from the primate laboratory to the stroke rehabilitation laboratory. *J Rehabil Med.* May 2003(41 Suppl):34-40.

134. Sunderland A, Tuke A. Neuroplasticity, learning and recovery after stroke: a critical evaluation of constraint-induced therapy. *Neuropsychol Rehabil.* May 2005;15(2):81-96.

135. Schurr MJ, Ebner KA, Maser AL, Sperling KB, Helgerson RB, Harms B. Formal swallowing evaluation and therapy after traumatic brain injury improves dysphagia outcomes. *J Trauma.* May 1999;46(5):817-821; discussion 821-813.

136. Crary MA, Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. *Arch Phys Med Rehabil.* Aug 2005;86(8):1516-1520.

137. Chong MS, Lieu PK, Sitoh YY, Meng YY, Leow LP. Bedside clinical methods useful as screening test for aspiration in elderly patients with recent and previous strokes. *Ann Acad Med Singapore.* Nov 2003;32(6):790-794.

138. Leder SB, Espinosa JF. Aspiration risk after acute stroke: comparison of clinical examination and fiberoptic endoscopic evaluation of swallowing. *Dysphagia.* Summer 2002;17(3):214-218.

139. Logemann JA. Do we know what is normal and abnormal airway protection? *Dysphagia.* Fall 1999;14(4):233-234.

140. Terre R, Mearin F. Evolution of tracheal aspiration in severe traumatic brain injury-related oropharyngeal dysphagia: 1-year longitudinal follow-up study. *Neurogastroenterol Motil.* Apr 2009;21(4):361-369.

141. Ahtaridis G, Snape WJ, Jr., Cohen S. Lower esophageal sphincter pressure as an index of gastroesophageal acid reflux. *Dig Dis Sci.* Nov 1981;26(11):993-998.

142. Leibovitz A, Carmeli Y, Segal R. Effect of various antibacterial preparations on the pathogenic oral flora in elderly patients fed via nasogastric tube. *Antimicrob Agents Chemother.* Aug 2005;49(8):3566-3568.

143. Cook AM, Peppard A, Magnuson B. Nutrition considerations in traumatic brain injury. *Nutr Clin Pract.* Dec-2009 Jan 2008;23(6):608-620.

144. Langmore SE. Evaluation of oropharyngeal dysphagia: which diagnostic tool is superior? *Curr Opin Otolaryngol Head Neck Surg.* Dec 2003;11(6):485-489.

145. Logemann JA, Veis S, Colangelo L. A screening procedure for oropharyngeal dysphagia. *Dysphagia.* Winter 1999;14(1):44-51.

146. Bours GJ, Speyer R, Lemmens J, Limburg M, de Wit R. Bedside screening tests vs. videofluoroscopy or fibreoptic endoscopic evaluation of swallowing to detect dysphagia in patients with neurological disorders: systematic review. *J Adv Nurs.* Mar 2009;65(3):477-493.

147. Bronchard R, Albaladejo P, Brezac G, et al. Early onset pneumonia: risk factors and consequences in head trauma patients. *Anesthesiology.* Feb 2004;100(2):234-239.

148. Woratyla SP, Morgan AS, Mackay L, Bernstein B, Barba C. Factors associated with early onset pneumonia in the severely brain-injured patient. *Conn Med.* Nov 1995;59(11):643-647.

149. FOrmaTT. FOrmaTT GmbH. [http://www.formatt.org/].

150. Lazarus C, Logemann JA. Swallowing disorders in closed head trauma patients. *Archives of Physical Medicine and Rehabilitation.* Feb 1987;68(2):79-84.

151. Smithard DG, O'Neill PA, Parks C, Morris J. Complications and outcome after acute stroke. Does dysphagia matter? *Stroke.* Jul 1996;27(7):1200-1204.

152. Harrington R. Communication for the aphasic stroke patient: assessment and therapy. *Journal of the American Geriatrics Society.* Jun 1975;23(6):254-257.

153. Nudo RJ, Dancause N. Neuroscientific basis for occupational and physical therapy interventions. In: Zasler ND, Katz DI, Zafonte RD, eds. *Brain injury medicine.* Vol 1. 1 ed. New York: Demos Medical Publishing, LLC; 2007:913-928.

154. Calhoun KS, Moras K, Pilkonis PA, Rehm LP. Empirically supported treatments: implications for training. *J Consult Clin Psychol.* Feb 1998;66(1):151-162.

155. Henry WP, Strupp HH, Butler SF, Schacht TE, Binder JL. Effects of training in time-limited dynamic psychotherapy: changes in therapist behavior. *J Consult Clin Psychol.* Jun 1993;61(3):434-440.

156. Tinetti ME, McAvay GJ, Fried TR, et al. Development of a tool for eliciting patient priority from among competing cardiovascular disease, medication-symptoms, and fall injury outcomes. *J Am Geriatr Soc.* Apr 2008;56(4):730-736.

157. Rothstein JM, Echternach JL, Riddle DL. The Hypothesis-Oriented Algorithm for Clinicians II (HOAC II): a guide for patient management. *Physical Therapy.* May 2003;83(5):455-470.

158. Moncher FJ, Prinz RJ. Treatment fidelity in outcome studies. *Clinical Psychology Review.* 1991;11(3):247-266.

159. Ibita. Theoretical assumptions. [http://www.ibita.org]. 2004. Accessed 6´th of April, 2005.

160. Davies PM. *Starting again : Early rehabilitation after traumatic brain injury or other severe brain lesion*. Berlin Heidelberg Springer-Verlag; 1994.
161. Bobath B. *Hemiplegia. Evaluation and treatment.* . London William Heinemann Medical Books Ltd; 1978.
162. Mulder T, Geurts A. Recovery of motor skill following nervous system disorders: a behavioural emphasis. *Baillieres Clinical Neurology.* Apr 1993;2(1):1-13.
163. Mayston M. Setting the scene. In: Edwards S, ed. *Neurological Physiotherapy - a Problem Solving Approach*. 2 ed: Churchill Livingstone; 2001.
164. Alaverdashvili M, Foroud A, Lim DH, Whishaw IQ. "Learned baduse" limits recovery of skilled reaching for food after forelimb motor cortex stroke in rats: A new analysis of the effect of gestures on success. *Behavioural Brain Research.* 2008;188(2):281-290.
165. Miller RM, Chang MW. Advances in the management of dysphagia caused by stroke. *Phys Med Rehabil Clin N Am.* Nov 1999;10(4):925-941, x.
166. Welch MV, Logemann JA, Rademaker AW, Kahrilas PJ. Changes in pharyngeal dimensions effected by chin tuck. *Arch Phys Med Rehabil.* Feb 1993;74(2):178-181.
167. Kjaersgaard A. *Ansigt, Mund og Svaelg*. Copenhagen: Fadl; 2005.
168. Nusser-Muller-busch R. *Die Therapie des Facio-Oralen Trakts.* . 1 ed. Berlin: Nachdruck Springer; 2004.
169. Davies PM. *Steps to Follow*. 2 ed. NY & Germany:: Springer Verlag 2000.
170. ARCOS. Facial - Oral Tract Therapy : F.O.T.T.™. [http://www.arcos.org.uk/]. Accessed 4'th of april 2006, 2006.
171. WHO. International Classification of Functioning, Disability and Health: Available from: http://www.who.int/classifications/icf/en/; 2006.
172. Law M, Cooper BA, Strong S, Stewart D, Rigby P, Letts L. The Person-Environment-Occupational Model: a transactive approach to occupational performance. *Canadian Journal of Occupational Therapy.* 1996;63(1):9-23.
173. Talbot A, Brady M, Furlanetto DL, Frenkel H, Williams BO. Oral care and stroke units. *Gerodontology.* Jun 2005;22(2):77-83.
174. Rozet I, Domino KB. Respiratory care. *Best Practice & Research Clinical Anaesthesiology.* 2007;21(4):465-482.
175. Hansen T, Larsen K, A.W E. Functional oral intake and time to reach unrestricted dieting in patients with severe traumatic brain injury. *Archives of Physical Medicine and Rehabilitation.*In press.
176. Ward EC, Green K, Morton AL. Patterns and predictors of swallowing resolution following adult traumatic brain injury. *Journal of head trauma rehabilitation.* May-Jun 2007;22(3):184-191.
177. Martin BJ, Logemann JA, Shaker R, Dodds WJ. Coordination between respiration and swallowing: respiratory phase relationships and temporal integration. *J Appl Physiol.* Feb 1994;76(2):714-723.
178. Sataloff RTRT, Heman-Ackah YDYD, Hawkshaw MJMJ. Clinical anatomy and physiology of the voice. *The Otolaryngologic clinics of North America.* 2007;40(5):909-929, v.
179. Rothstein JM, Echternach JL, Riddle DL. The Hypothesis-Oriented Algorithm for Clinicians II (HOAC II): A Guide for Patient Management. *PHYS THER.* May 1, 2003 2003;83(5):455-470.
180. Affolter F, Bischofberger W. *Nonverbal perceptual and cognitive processes in children with language disorder. Toward a new framework for clinical intervention*. Mahwah, New Jersey: Lawrence Erlbaum Associates; 2000.
181. Affolter F. *Perception, Interaction and Language*. Berlin Heidelberg1991.
182. Ibita. Theoretical Assumptions and Clinical Practice. [http://www.ibita.org/]. 2007. Accessed December, 2007.
183. Organisation WH. International Classification of Functioning, Disability and Health: Available from: http://www.who.int/classifications/icf/en/; 2006.
184. Kathleen M. Carroll, Kathryn F. Nuro. One Size Cannot Fit All: A Stage Model for Psychotherapy Manual Development. *Clinical Psychology: Science and Practice.* 2002;9(4):396-406.
185. Echternach JL, Rothstein JM. Hypothesis-oriented algorithms. *Phys Ther.* Jul 1989;69(7):559-564.
186. Tinetti ME. Instability and falling in elderly patients. *Semin Neurol.* Mar 1989;9(1):39-45.
187. Carroll KM, Nuro KF. One Size Cannot Fit All: A Stage Model for Psychotherapy Manual Development. *Clinical Psychology: Science and Practice.* 2002;9(4):396-406.
188. Luborsky Lester , Diguer Louis , Seligman David A. , et al. The Researcher's Own Therapy Allegiances: A "Wild Card" in Comparisons of Treatment Efficacy. *Clinical Psychology: Science and Practice.* 1999;6(1):95-106.
189. Schoenwald SK, Henggeler SW, Brondino MJ, Rowland MD. Multisystemic therapy: monitoring treatment fidelity. *Fam Process.* Spring 2000;39(1):83-103.

190. Terré R, Mearin F. Prospective evaluation of oro-pharyngeal dysphagia after severe traumatic brain injury. *Brain Injury.* 2007;21(13/14):1411-1417.
191. Frymark T, Schooling T, Mullen R, et al. Evidence-based systematic review: Oropharyngeal dysphagia behavioral treatments. Part I--Background and methodology. *Journal of Rehabilitation Research & Development.* 2009;46(2):175-183.
192. Elmstahl S, Bulow M, Ekberg O, Petersson M, Tegner H. Treatment of dysphagia improves nutritional conditions in stroke patients. *Dysphagia.* Spring 1999;14(2):61-66.
193. Landis JR, Koch GG. The measurement of observer agreement for categorical data. *Biometrics.* Mar 1977;33(1):159-174.
194. Bülow M, Olsson R, Ekberg O. Videoradiographic Analysis of How Carbonated Thin Liquids and Thickened Liquids Affect the Physiology of Swallowing in Subjects with Aspiration on Thin Liquids. *Acta Radiol.* 2003;44(4):366-372.
195. Fink TA, Ross JB. Are We Testing a True Thin Liquid? *Dysphagia.* Feb 21 2009.
196. Park C, O´Neill P. Management of neurological dysphagia. *Clin Rehabil.* 1994;8:166-174.
197. Denes Z. The influence of severe malnutrition on rehabilitation in patients with severe head injury. *Disabil Rehabil.* Oct 7 2004;26(19):1163-1165.
198. Langmore SE. Dysphagia in neurologic patients in the intensive care unit. *Semin Neurol.* Dec 1996;16(4):329-340.
199. Splaingard ML, Hutchins B, Sulton LD, Chaudhuri G. Aspiration in rehabilitation patients: videofluoroscopy vs bedside clinical assessment. *Arch Phys Med Rehabil.* Aug 1988;69(8):637-640.
200. Seidl R, Nusser-Muller-busch R. *Der Berliner Dysphagie Index.* Berlin: Unfallkrankenhaus 2001.
201. Rosenbek JC, Robbins JA, Roecker EB, Coyle JL, Wood JL. A penetration-aspiration scale. *Dysphagia.* Spring 1996;11(2):93-98.
202. Hansen TS, Jakobsen D. A decision algorithm defining the rehabilitation approach: 'Facial Oral Tract Therapy' ® *Under review.*
203. Hansen TS, Jakobsen D, Nowack D, Whyte J. Development of an adherence measure for a complex neurorehabilitation approach: "Facial Oral Tract Therapy" ® *Under review.*
204. McCullough GH, Wertz RT, Rosenbek JC, Mills RH, Ross KB, Ashford JR. Inter- and intrajudge reliability of a clinical examination of swallowing in adults. *Dysphagia.* Spring 2000;15(2):58-67.
205. Butler SG, Stuart A, Markley L, Rees C. Penetration and Aspiration in Healthy Older Adults as Assessed During Endoscopic Evaluation of Swallowing. *Annals of Otology, Rhinology & Laryngology.* 2009;118(3):190-198.
206. Trapl M, Enderle P, Nowotny M, et al. Dysphagia bedside screening for acute-stroke patients: the Gugging Swallowing Screen. *Stroke.* Nov 2007;38(11):2948-2952.
207. Eisenhuber E, Schima W, Schober E, et al. Videofluoroscopic assessment of patients with dysphagia: pharyngeal retention is a predictive factor for aspiration. *AJR Am J Roentgenol.* Feb 2002;178(2):393-398.
208. Daniels SK, Ballo LA, Mahoney MC, Foundas AL. Clinical predictors of dysphagia and aspiration risk: outcome measures in acute stroke patients. *Archives of Physical Medicine and Rehabilitation.* Aug 2000;81(8):1030-1033.
209. Warms T, Richards J. "Wet Voice" as a predictor of penetration and aspiration in oropharyngeal dysphagia. *Dysphagia.* Spring 2000;15(2):84-88.

Figure Legends

Figure 1. F.O.T.T.® model by Davies J, Coombes K 1987

Figure 2. Assessment chart: What to look for

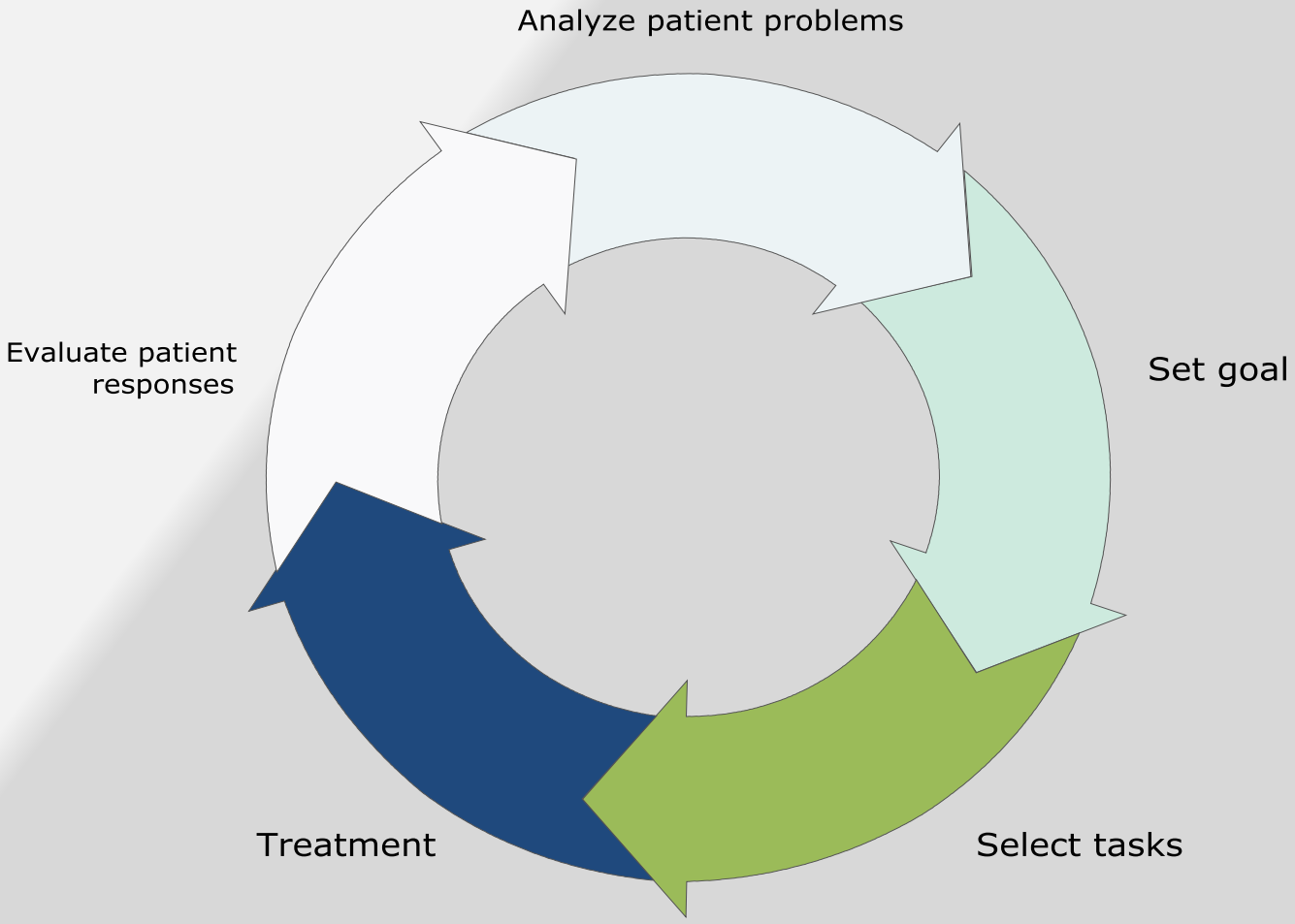
Figure 3. Treatment chart: Swallowing of saliva and eating

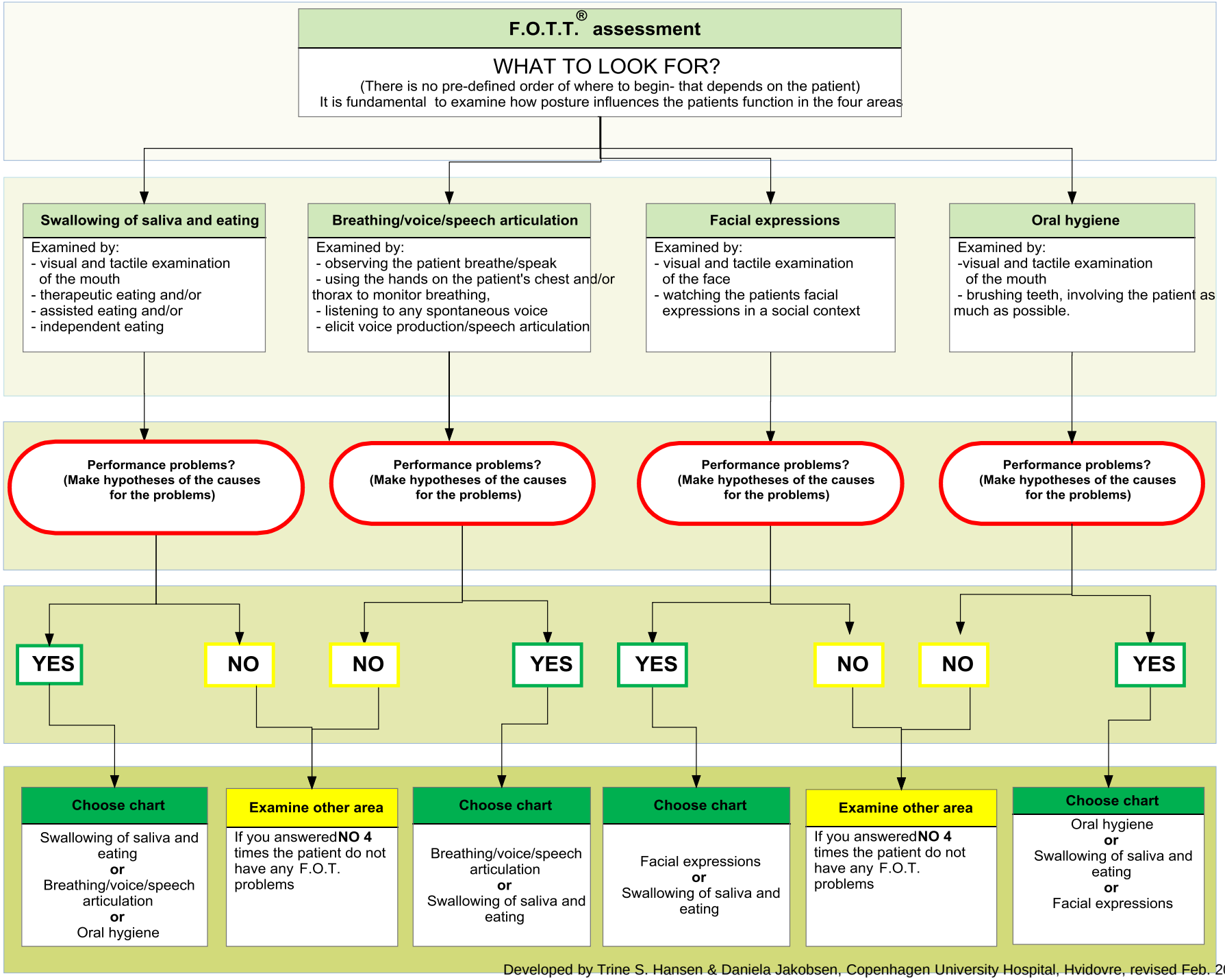
Figure 4. Treatment chart: Oral hygiene

Figure 5. Treatment chart: Breathing/ voice and speech articulation

Figure 6. Treatment chart: Facial expressions

F.O.T.T. Model


Analyze patient problems
Set goal
Select tasks
Treatment
Evaluate patient responses



Developed by Trine S. Hansen & Daniela Jakobsen, Copenhagen University Hospital, Hvidovre, revised Feb. 2009

Swallowing of saliva and eating

1 Goal

2 Choose a strategy for the intervention

3 Evaluate performance

4 Choose new Strategy/behavior or new activity

5 Evaluate goal

1. Set a goal for theF.O.T.T. intervention

Select a clear, measurable "short-term" goal for the F.O.T.T. intervention , directed by the patient's problems and "wishes"

2.a. Activity

Select an activity for the intervention (if possible an everyday life activity) Remember observations of postural control

2.b. Environmental factors

Chose the environment for the activity and the patient

2.b.1. Products & tech-nology in daily living

- chose location
- chose furniture
- chose objects or use aids

2.c. Therapeutic intervention

Therapeutic interventions for task orientated movements and postural control

2.c.1. Working levels for preparation to swallowing

- 1 supported oral stimulation
- 2 partly supported stimulation
- 3 independent oral stimulation

2.c.2. Working levels for swallowing saliva and eating/drinking

- 1 swallowing saliva
- 2 therapeutic eating
- 3 assisted eating
- 4 independent eating

Food consistencies

- 1 puree
- 2 chopped
- 3 normal with restrictions
- 4 normal food

Liquid

- 1 thickened
- 2 thin
- 3 All liquids

2.c.3 Working approach

- move and position the patient
- guide the patient
- mobilize the patient
- elicit
- facilitate the patient:
- *1 whole time*
- *2 in between the movement/function/activity*
- *3 initiate in the beginning*

2.c.4 Tongue movements

- 1 tongue is moved by therapist
- 2 therapist moves the tongue to one or several positions and the patient holds it there
- 3 patients moves the tongue actively
- patient moves and repeats movements of the tongue in a functional context

3.a. Does the patient perform the task in a more normal manner?/ to better quality?

Yes

No

4.a. Reduce support

Change intervention strategy, reduce support or set a new goal

Yes

No

4.b. Improvement?

End of treatment

4.a. Increase support

Change intervention strategy, increase support or set a new goal

2.c.5 Working with tactile support for swallowing saliva

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

2.c.6 Working with protection of airway

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

5.a. Is the goal reached?

Developed by Trine S. Hansen & Daniela Jakobsen, Copenhagen University Hospital, Hvidovre, revised Feb. 2009

Oral hygiene

1 Goal

1. Set a goal for theF.O.T.T. intervention

Select a clear, measurable "short-term" goal for the F.O.T.T. intervention , directed by the patient's problems and "wishes"

2 Choose a strategy for the intervention

2.a. Activity

Select an activity for the intervention (if possible an everyday life activity) Remember observations of postural control

2.b. Environmental factors

Chose the environment for the activity and the patient

2.b.1. Products & technology in daily living

- chose location
- chose furniture
- chose objects or use aids

2.c. Therapeutic intervention

Therapeutic interventions for task orientated movements and postural control

2.c.1. Working levels for oral stimulation & preparation to therapeutic oral hygiene

- 1 supported oral stimulation
- 2 partly supported stimulation
- 3 independent oral stimulation

2.c.2.Working levels

- 1. Therapeutic oral hygiene using alternatives to toothbrushes and other normal requisites
- 2. Therapeutic oral hygiene without rinsing
- 3. Therapeutic oral hygiene with rinsing
- 4. Assisted oral hygiene

2.c.3. Working approach

- move and position the patient
- guide the patient
- mobilize the patient
- elicit
- facilitate the patient:
  - 1 whole time
  - 2 in between the movement/function/activity
  - 3 initiate in the beginning

3 Evaluate performance

3.a. Does the patient perform the task in a more normal manner? / to better quality?

Yes / No

2.c.4. Working with tactile support for swallowing saliva

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

2.c.5. Working with protection of airway

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

4 Choose new Strategy/behavior or new activity

4.a. Reduce support

Change intervention strategy, reduce support or set a new goal

Yes / No

4.b. Improvement?

4.a. Increase support

Change intervention strategy, increase support or set a new goal

End of treatment

5 Evaluate goal

5.a. Is the goal reached?

Developed by Trine S. Hansen & Daniela Jakobsen, Copenhagen University Hospital, Hvidovre, revised Feb. 2009

Breathing/ voice/ speech articulation

1 Goal

2 Choose a strategy for the intervention

3 Evaluate performance

4 Choose new Strategy/behavior or new activity

5 Evaluate goal

1. Set a goal for theF.O.T.T. treatment

Select a clear, measurable "short-term" goal for the F.O.T.T. intervention , directed by the patient's problems and "wishes"

2.a. Activity

Select an activity for the intervention (if possible an everyday life activity). Remember observations of postural control

2.b. Environmental factors

Chose the environment for the activity and the patient

2.b.1. Products & tech-nology in daily living

- chose location
- chose furniture
- chose objects or use aids

2.c. Therapeutic intervention

Therapeutic interventions for task orientated movements and postural control

2.c.1. Working levels

- 1 support breathing
- 2 support breathing-voice coordination
- 3 support breathing-voice and articulation
- 4 support breathing and speech

2.c.2. Working approach

- Move and position the patient
- Guide the patient
- Mobilize the patient
- Elicit
- Facilitate the patient:
- 1 whole time
- 2 in between the movement/function/activity
- 3 initiate in the beginning

2.c.3. Working with position and movements

- 1 Lying
- 2 Lying in combination with movements
- 3 Sitting
- 4 Sitting in combination with movements
- 5 Standing
- 6 Standing in combination with movements

2.c.4. Working with tactile support for swallowing saliva

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

2.c.5. Working with protection of airway

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

3.1. Does the patient perform the task in a more normal manner?/ to better quality?

Yes

No

4.a. Reduce support

Change intervention strategy, reduce support or set a new goal

Yes

No

4.b. Improvement?

4.a. Increase support

Change intervention strategy, increase support or set a new goal

End of treatment

5.a. Is the goal reached?

Developed by Trine S. Hansen & Daniela Jakobsen, Copenhagen University Hospital, Hvidovre, revised Feb. 2009

Facial expressions

1 Goal

2 Choose a strategy for the intervention

3 Evaluate performance

4 Choose new Strategy/behavior or new activity

5 Evaluate goal

1. Set a goal for theF.O.T.T. intervention

Select a clear, measurable "short-term" goal for the F.O.T.T. intervention , directed by the patient's problems and "wishes"

2.a. Activity

Select an activity for the intervention (if possible an everyday life activity). Remember observations of postural control

2.b. Environmental factors

Chose the environment for the activity and the patient

2.b.1. Products & technology in daily living

- chose location
- chose furniture
- chose objects or use aids

2.c. Therapeutic intervention

Therapeutic interventions for task orientated movements and postural control

2.c.1. Working levels for preparation to facial movements

- 1 supported oral stimulation
- 2 partly supported stimulation
- 3 independent oral stimulation

2.c.2. Working levels in facial movements

- 1 use passive facial movements
- 2 use active facial movements
- 3 use facial movements in a functional activity

2.c.3. Working approach

- move and position the patient
- guide the patient
- mobilize the patient
- Elicit
- facilitate the patient
- 1 whole time
- 2 in between the movement/function/activity
- 3 initiate in the beginning

2.c.4. Working with tactile support for swallowing saliva

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

2.c.5. Working with protection of airway

- support coughing
- support clearing the throat
- support subsequent swallow
- support using voice
- support mouth clearing
- support posture forward

3.a. Does the patient perform the task in a more normal manner?/ to better quality?

Yes / No

4.a. Reduce support

Change intervention strategy, reduce support or set a new goal

Yes / No

4.b. Improvement?

4.a. Increase support

Change intervention strategy, increase support or set a new goal

End of treatment

5.a. Is the goal reached?

Developed by Trine S. Hansen & Daniela Jakobsen, Copenhagen University Hospital, Hvidovre, revised Feb. 2009

Study IV

Hansen, T.S., Jakobsen, D., Nowack, D'A, Whyte, J.

IV

Development of an adherence measure for a complex neurorehabilitation approach

Submitted for publication: the American Journal of Physical Medicine and Rehabilitation

Title

Development of an adherence measure for a complex neurorehabilitation approach: "Facial Oral Tract Therapy" ®

Authors:

Trine Schow Hansen OTR, M.Sci. Public Health[1], Daniela Jakobsen, OTR.[2], D'Anna Nowack SLP[3], John Whyte, MD, PHD[4]

Affiliation:

Department of Neurorehabilitation, Brain Injury Unit, Department of Occupational Therapy, Copenhagen University Hospital Hvidovre, Hvidovre, Denmark[1, 2]

Center for Communication Disorders, Drucker Brian Injury Center, MossRehab Hospital, Elkins Park, Pennsylvania, USA[3]

Moss Rehabilitation Research Institute, Albert Einstein Healthcare Network, Elkins Park, Pennsylvania, USA[4]

Correspondence to:

Trine Schow Hansen, Hvidovre Hospital, Department of Neurorehabilitation 354, Kettegaard Allé 30, DK-2650 Hvidovre Denmark

Phone work: +45 3632 6353, Fax number: + 45 3632 3312

Email: trine.schow@gmail.com

Author disclosures:

We certify that no party having a direct interest in the results of the research supporting this article has or will confer a benefit on us or on any organization with which we are associated. This study was funded by Hvidovre Hospital, Department for Occupational Therapy and Elsass foundation.

Abstract

Objectives

To develop and validate a measure of adherence to the rehabilitation approach ”Facial Oral Tract Therapy®” (FOTT)

Design

We videotaped two independent groups of therapists applying swallowing therapy to patients with brain injury (BI), one group highly skilled in FOTT (TBIU) and one not familiar with this approach (Moss). 9 patients were videotaped in each group and coded independently by two observers. Inter observer agreement was analyzed with Cohen’s Kappa and percent agreement and difference in adherence between centers with the Mann Whitney *U* test.

Result

Inter-observer agreement in the sessions from TBIU had a mean Kappa across the 9 components of 0.64-0.95 (percent agreement 97-99%) and at Moss 0.28-1.00 (88-99%). Adherence was significantly higher at TBIU in 7 out of the 9 components. There was a significant difference between centers in the total number of components and choices used in an FOTT-adherent way.

Conclusion

In this study we developed an adherence measure that was found to be capable of capturing the essential components of FOTT; a complex treatment approach used for disturbances in the face, mouth and throat. This measure makes it possible to verify adherence to FOTT in future comparative trials or to use degree of adherence to FOTT as a measure in observational studies.

Keywords: Adherence measure, Rehabilitation research, Brain injury, Observational study

Introduction

There are still gaps in our understanding of the efficacy and effectiveness of the therapeutic approaches used within neurorehabilitation. Obstacles to and methodological challenges of rehabilitation research have been thoroughly discussed in recent years and a clear definition of the treatment involved, in terms of its hypothesized active ingredients is one of the important issues identified as necessary to understand what generates patient outcomes[9, 43, 48]. However, many therapeutic rehabilitation methods are still "black boxes," defined grossly in terms of hours of treatment by a discipline, or in terms of the intended goal of the treatment (e.g., "gait training") rather than the actual critical components (active ingredients) of the treatment. This, in turn, limits the ability to compare them to other treatments, or to disseminate them to future generations of clinicians. Thus, there is an ongoing need for more research on the content of treatment programmes[43].

For treatments with single components (e.g., strength training of the upper extremity), defining the hypothesized active ingredient is relatively easily achieved, but it is much more complicated in many of the complex treatment *approaches* typical of rehabilitation programmes. Treatment approaches typically involve a complex decision process (often implicit) that specifies under what conditions each specific ingredient should be delivered [38, 43]. Thus, in such approaches, the therapist should continuously adjust the treatment to the specific problems and response of each individual. This "structured variability" makes the definition of the content of the treatment a great challenge, since one can't look for invariant components across patients and sessions to define and characterize the active components that are hypothesized to exert the treatment effects of interest.

A treatment manual or treatment algorithm can help in the process of defining complex treatment approaches, by specifying the underlying logic by which treatment decisions are made with respect to specific patient characteristics and patient responses to prior treatment steps. Defining treatment approaches in this way serves a number of important clinical and scientific purposes. First, in the developmental stages of research, creating such a manual forces researchers and clinicians to articulate their underlying hypotheses and beliefs about the mechanisms of treatment effects[43]. Second it provides the clinician with a tool that can guide them in how to use such a complex treatment approach. It is particularly challenging for less experienced therapists, who lack implicit decision rules, to use a poorly defined treatment in the right way. Such manuals also strengthen the

ability to compare different therapies, by ensuring standardized treatment procedures in the experimental group, and by ensuring that the comparison treatment lacks these key components. Thus, treatment manuals and algorithms help to link the treatment to the clinical aims and the outcomes observed[43].

A treatment manual with clear definitions of the essential components also provides the researcher with a tool that makes it possible to assess treatment adherence. A measure of adherence will tell the researcher whether the therapy was applied as intended [56], which is vital to maintain internal validity[158], to be able to replicate the study and to tell if the conclusion of a study investigating treatment outcome is reliable[188-189]. Measures of treatment adherence can also support observational studies by allowing assessment of the relationship between *degree of adherence* and strength of the treatment effect. In clinical practice, assessment of adherence can be used in therapist training and supervision. Unless treatment adherence is investigated, it will remain unclear to what extent the hypothesized mechanisms of the treatment components being tested are the primary mechanisms of the study outcome [26, 57] and it is therefore a necessary step in rehabilitation research.

Dysphagia treatment for patients with traumatic brain injury (TBI) is one such treatment domain where multiple specific treatment components are typically applied. However, in spite of the fact that the incidence of dysphagia is reported to be as high as 61-93% [73, 76, 190] in TBI, and can have serious consequences for rehabilitation progress, research within dysphagia management is still limited [191]. Many factors, both neuromuscular and cognitive [165], contribute to swallowing difficulties, and therefore swallowing treatments and components of each treatment approach vary in nature depending on what kind of problems they are targeting. Therapeutic strategies include both compensatory and restorative techniques [192]. However, most of the techniques investigated and described to date are single”stand-alone” components rather than comprehensive treatment approaches. Even these individual components are sometimes defined in different ways and named differently. For example, Okada et al[95], showed poor agreement between Speech Language Pathologists (SLPs) in Japan and the USA about the meaning of the commonly used chin tuck method and found multiple definitions of this method in the available literature. These discrepancies make it difficult both to use and investigate such an intervention[95].
Facial Oral Tract Therapy (FOTT)[14] is an example of a broad rehabilitation approach, covering problems within the area of facial movements, oral hygiene, eating and drinking, and breathing and

communication. It is widely used throughout Scandinavia and much of Europe in neurological rehabilitation programmes[108]. FOTT is multidisciplinary and relates to several different professions with different backgrounds. It deals with multiple interrelated impairments and employs multiple interrelated treatment components. Essential to this approach is the concept that therapists continuously evaluate and adjust the treatment to the patient's responses. Presumably, due to this complexity, FOTT has not been described and defined in a structured way and no evidence exists of its efficacy or effectiveness. Defining and measuring the delivery of the hypothesized active ingredients of FOTT is one step on the way to opening a "black box" in rehabilitation in the area of swallowing therapy, and can also serve as an example of how to approach challenging research problems of this type.

In the course of this research, we developed a treatment manual for FOTT (Hansen T, resubmitted discuss). To include the essential dynamic interplay between choice of therapeutic approach and patient response we defined the components of the treatment in terms of a decision algorithm. The decision algorithm characterizes the decision process in FOTT and defines the essential components. It has been approved by the developer of FOTT, K. Coombes[170]. To determine if therapists adhere to this decision algorithm a method had to be developed to test whether or not the most important components are used as intended[58].

To our knowledge no studies have investigated treatment adherence in swallowing therapy. Thus, the aim of this study was to develop and test a method to measure adherence to the FOTT approach. More specifically, we sought to assess inter-observer reliability of this adherence measure, and to apply it to treatment sessions that were intended to embody FOTT principles vs. sessions that employed a different therapeutic approach, in order to assess the measure's specificity to the FOTT approach.

Method:

We used an observational method in this study. We videotaped swallowing therapy sessions to be able to observe therapeutic behavior and determine if the therapy was performed as intended according to the FOTT algorithm. We videotaped two independent groups of therapists applying swallowing therapy to patients with BI - one group that was thoroughly trained in FOTT and one group not familiar with this approach. We reasoned that a difference in adherence between therapists trained and not trained in the FOTT approach would support the hypothesis that FOTT

differs from another approach in its actual components, not merely in its name, and furthermore, that the adherence measure that we developed was sensitive to these differences in components. On the other hand, no detected difference in adherence could reflect either the insensitivity of our adherence measure and/or the fact that two different groups of therapists used very similar approaches under different names.

Subjects:
Two rehabilitation hospitals were chosen for this study, one in Denmark and one in the USA.
1: Traumatic Brain Injury Unit (TBIU), Copenhagen University Hospital, Hvidovre, Denmark. The occupational therapists use FOTT on a daily basis and an FOTT instructor continuously teaches and supervises the other therapists. All therapists starting at the department receive an introductory course in FOTT, and most of them have attended either the certified FOTT basic or advanced course.
2: MossRehab Hospital, Elkins Park, USA. The speech and language pathologists treat patients with dysphagia but they are not familiar with the FOTT approach.

The subjects for this study were therapists treating patients with brain injury, aged 18-80 years, FIM at admission < 52 (total) with swallowing disorders. Consent for participation in the study and video recording was obtained from both therapists and patients. If the patients were not able to give consent themselves, it was obtained from an appropriate surrogate.

Procedure:
The FOTT algorithm is divided into four treatment charts. In this study we chose to measure adherence to the treatment chart “swallowing of saliva and eating”. The other three are named: “facial expressions”; “oral hygiene” and “breathing, voice and speech”. This “swallowing of saliva and eating” chart was chosen for two reasons: 1) it simplified the study as only one chart was investigated at a time and 2) this chart describes the treatment used for patients with problems in swallowing and eating so the methods defined in this chart could be compared to dysphagia treatment used in other swallowing therapy rehabilitation programmes.

9 sessions of swallowing therapy, involving 9 different patients, 1 session each, were recorded at each centre. Treatment was performed by 3 different therapists at Moss and 7 therapists at TBIU,

respectively. The swallowing therapy session was videotaped by a person not involved in the session. The handheld camera was directed so that both the therapist and the patient could be observed at the same time. The normal procedures for swallowing therapy at both centers were followed. The two originators of the algorithm watched the videos independently and rated adherence to the algorithm.

The videotapes were coded each minute for adherence to 9 pre-specified treatment components for up to 15 minutes of swallowing treatment. For every minute we coded whether the therapist's choice of treatment in the previous minute was, or was not, an appropriate choice according to the FOTT algorithm and whether or not they chose to use the component or not and if they used it in an appropriate way.

Since in FOTT the therapist constantly treats and evaluates the patient's responses, the algorithm demonstrates a circular decision process instead of a decision tree where "yes" and "no" choices lead to a new step. In the FOTT algorithm the therapist can choose to use or not to use the same approach several times during one treatment session. There is no right or wrong in regard to how many times they should make a different choice; the important issue is that the therapist evaluates their choices according to patient response and adjusts their approach by making a choice to continue the same approach or choose a different approach.
An example of the coding system is shown in table 1

Insert table 1 approximately here

Adherence to the FOTT approach was defined as the number of choices that were "reasonable" divided by the total choices i.e. those that were reasonable plus those that were not.

Table 2 gives an overview of the most important components of FOTT. It is not required that the therapist should use each of these nine components within each minute of the treatment session however they should make a choice whether or not it is appropriate to use them.

Insert Table 2 approximately here

Data Analysis:

Session length was compared between centers on the possibility that adherence might change across the treatment session, and therefore whether systematic differences in session length at the 2 sites might confound the comparison of adherence rates. The Mann-Whitney *U exact* test was used to test the significance of the difference between centers in session length and in patients' admission FIM score.

Inter-observer agreement was analyzed by calculating, for each videotape and each component, percent agreement between observers, as well as Cohen's kappa. Cohen's kappa assesses agreement between the two observers adjusted for chance. We used the criteria of Landis and Koch[193] for interpretation: a kappa value below 0.20 constitutes to no or slight agreement, 0.21 to 0.40 is fair agreement, 0.41 to 0.60 is moderate agreement, 0.61 to 0.80 is substantial agreement, and 0.81 to 1.00 is almost perfect agreement. The 4 category rating system for measuring adherence: 2 measures for adherence and 2 for non adherence (see Table 1) were used for this calculation. Comparisons of adherence between centers and categories were done after collapsing this matrix to only two categories, one for adherence and one for non adherence. Differences in adherence between therapists at the 2 sites were analyzed with the Mann-Whitney *U exact* test. Likewise we used the Mann-Whitney *U exact* test to investigate the difference between the total number of components used within a treatment session between the two centers and the number of components used in a correct way according to FOTT.

An alpha level of 0.05 (2 tailed) was used for all analysis.

Ethics

This study was approved by The Danish National Committee on Biomedical Research Ethics, (the Copenhagen regional committee) and the Institutional Review Board of the Albert Einstein Healthcare Network. All participants or surrogates provided informed consent.

Results

We videotaped 9 different patients (4 females) and 3 different therapists (all females) at Moss and 9 different patients (3 females) and 8 different therapists (all females) at TBIU, respectively. Patients at TBIU had a mean age of 38 years (range: 20-61) and a median FIM score at admission of 23 (23-23) 8 patients had TBI and 1 nontraumatic subarachnoid hemorrhage. Patients at Moss were similar with a mean age of 35years (range: 19-61) but with a somewhat higher admission FIM score (median=30 (range=23-51; $U = 18$, $P = .02$). 8 patients suffered from TBI and 1 from encephalitis.

Duration of the observed swallowing therapy sessions:

The median duration of swallowing therapy did not differ significantly: 13 minutes at TBIU and 10 minutes at Moss (U= 25.50; p= 0.18(two tailed)), respectively.

Inter-observer agreement

It was not possible to compute kappa values for all components in all sessions because in some sessions one or both observers used only one rating category for a given component throughout the session. In these cases only percent agreement is presented. See table 3.

Insert Table 3 approximately here

In the sessions from the TBIU, the mean Kappa value across the 9 components ranged from 0.64-0.95, with agreement in the substantial range for 4 components and in the almost perfect range for 5 components. In the sessions from Moss, the mean Kappa ranged from 0.28-1.00 with 1 in the fair range, 3 in the moderate to substantial range and 5 in the almost perfect range. Mean percent agreement ranged from 0.87-0.99 across all components in both centers, suggesting that the lowest Kappa values, as well as the ones that could not be calculated, were related to the infrequent use of certain coding categories (and hence the increased possibility of agreement by chance for the codes used), rather than frequent disagreement.

Adherence patterns:

Since the agreement between coders was generally high, we used an average of their individual adherence rates for further analysis. Adherence differed significantly in 7 out of the 9 components between centers, with greater adherence at TBIU. The two components which did not differ were: “swallowing of saliva” and “tongue movements/other”. There was a trend in the same direction for these 2 components. (See table 4)

Insert Table 4 approximately here

The grand mean adherence (across components and sessions) for the therapists at TBIU was 92% and at Moss 38%).

At TBIU the highest adherence was seen in “swallowing of saliva” and lowest in “positioning”. The same picture was seen at Moss. The greatest differences in adherence between coders were seen in

the components: Location (diff= 84%), position (diff=75%), therapeutic approach (diff=70%) and support to swallow (diff=73%). The number of different components used per session differed significantly: median TBIU= 8 (range 6-9) and median Moss=5 (U=6.5; p<0.01 (two tailed)). An even greater difference was seen in the number of components per session used appropriately as defined in the algorithm: median TBIU: 8 (range 6-9), median Moss=1 (range 0-3) (U=0; p<0.01), respectively. (See table 5)

Insert table 5 approximately here

Discussion

We developed a treatment manual for a complex rehabilitation approach used in neurorehabilitation to treat patients with dysphagia and related problems. The treatment manual describes the decision processes used in FOTT and operationalizes the delivery of the hypothesized active ingredients in a decision algorithm. In this study we developed and validated a measure of adherence to this manual by comparing therapists' observed decisions with the essential components of the algorithm.

Several factors besides FOTT may have had an impact on adherence as measured in this study. First, some of the 9 essential components of FOTT tend to be highly FOTT-specific whereas others are more generic. Specific published evidence-based components of swallowing therapy may be more likely to be generally adopted by all therapeutic approaches providing a greater chance that these components are used in the same way in both centers, giving similarly high adherence. An example of such a generic approach is food modification (part of the component "eating and drinking" in FOTT), where the therapist adjusts the food consistencies according to the nature of the patient's swallowing problems. It is a generally acknowledged technique, with some evidence of its effect [101, 194-195]. Other components such as eliciting a swallow by moving or mobilizing the patient or facilitating a swallow by moving the patients tongue back and upwards pressing the fingers at the floor of the mouth, which are not evidence-based, are more specific to FOTT and therefore the chance that a therapist at Moss will adhere to these components is lower.

Another factor that might affect adherence as measured is the clinical presentation of the patient. Some patients may not require the choice of a particular treatment component and, therefore, it would remain unclear whether the therapist would have used that component in the appropriate

context. An example of this is the component, “swallowing of saliva”. If all the patients being treated are safe to work with various food consistencies, then it will rarely be relevant to work with “swallowing of saliva”. The non-use of this component would be coded as adherent but provides no information about whether the therapist would treat a patient with more severe impairments by FOTT principles.

Finally, another factor affecting adherence is that some treatment components involve more frequent different choices than others. For example, with the component, “Location/furniture,” the therapist often will make a choice of what location or furniture to use within the first minute of intervention and rarely make a change within the treatment session, giving a bimodal distribution of adherence (adherent in all coded minutes vs. no coded minutes). In contrast, components that require frequent choices to adjust treatment, such as “Guiding” or “Facilitation”, may result in a more continuous distribution of adherence, since a therapist can be adherent one minute but not the next.

Our data showed a significant difference in adherence between centers in the predicted direction in seven out of the nine components. However, adherence at MOSS was largely the result of appropriately choosing not to use some of the FOTT treatment components, since Moss therapists rarely (an average of 1 component per session) used the components in an FOTT adherent way. There were two components where we did not find a significant difference in adherence between Moss and TBIU, namely: “Swallowing of saliva” and “Tongue movements/other.” The component “Swallowing of saliva” was not relevant for most patient at Moss, as discussed above, because of their significantly higher FIM scores and better swallowing function. Thus the therapist always used the relatively generic component “Eating and drinking,” which was coded as adherent but still leaves uncertainty whether the therapists would have ever chosen to work with “swallowing of saliva” when appropriate. However, adherence was still not 100% at Moss for this component. In a few cases with tracheostomies “Eating and Drinking” was coded as non-adherent, because according to FOTT it is rarely appropriate to let these patients eat a whole meal or drink a cup of thin liquids because they cannot protect their airway effectively, and an FOTT-trained therapist would work with “Swallowing of saliva” instead, or only with small amounts of food. The other variable which showed no significant difference in adherence was “Tongue movements/other.”

Again, the use of this technique is more relevant to severely impaired patients, so the choice to not use it was often coded as adherent.

Therapists at Moss had a low level of adherence to the components: “Location/furniture” and “Position” reflecting the bimodal distribution discussed previously; they tended to make a “non appropriate” choice in the beginning of the session, and make no further changes. However they also had low adherence in “Therapeutic approach,” for a different reason, making several new decisions throughout the treatment session, but not ones that were appropriate according to FOTT-specific principles. Similarly, “Support of swallowing” and “Protection of airway,” also appear to be FOTT-specific since the times the therapists at Moss were coded as adherent were when they chose *not* to use them.

In addition, we found that the therapists at TBIU generally used more different treatment components in a session than the therapists at Moss. This is another characteristic feature of the FOTT approach and very much in line with the circular process defined in the FOTT algorithm, where the therapist is supposed to continuously analyze and readjust their choice of strategies. And maybe more important the therapist at TBIU also used more components in an appropriate way according to FOTT. This pattern is consistent with the use of a larger set of techniques in a single session at TBIU and different contexts for use of some of the same techniques between the two centers. Together this suggests that FOTT is a definable and distinctive therapeutic approach that can be contrasted with other approaches in efficacy and effectiveness research.

Study limitations

This study found almost perfect percent agreement between coders but with somewhat more variable Kappa values. Kappa can vary drastically for the same observed agreement in relation to the value of expected agreement. Thus, when one coding category was always or almost always used, we can’t rule out the possibility that some of the high agreement was due to chance. Patients at Moss had significantly higher FIM scores that, in some cases, might affect the choices of treatment components. However, despite the average difference in FIM scores, the Moss patients had a broad range of functioning, but still saw clear adherence differences between sites.

Another limitation in this study is the choice of video observation. Not every response of the patient that directs the decision of therapeutic techniques is observable on videotape. There might be some

reasons for the therapeutic behavior that the observers did not recognize, e.g. an unobserved change in facial tone, movements of the soft palate or tongue, etc. This issue however, applies to the therapists at both centers and may be assumed to be equally biasing the study. Also, we only observed 3 different therapists at Moss and 7 at the TBIU, and only 9 patients at each site, limiting the ability to generalize our adherence findings to all FOTT-trained therapists treating all kinds of patients vs. therapists trained in other approaches. Finally, an obvious limitation of this study is that the originators of the algorithm were also the persons rating the videos and thereby testing the validity of the adherence measure to "their own manual"[56]. This was dictated by the fact that a detailed knowledge of the FOTT algorithm, not yet widely shared, was required for coding. However, the substantial minute-to-minute agreement of the 2 coders mitigates this concern to some degree. In a future study, the validity of this adherence measure may benefit of being repeated using other observers also trained in the algorithm and FOTT.

Despite the limitations, this measure of adherence shows promises in capturing whether or not a therapist is delivering the intended components of FOTT. In attempting to define the active ingredients of a complex treatment approach like FOTT, the development of a treatment manual was the first step, and the adherence measure the next. Future studies should address implementing the decision algorithm in clinical settings and investigating this manual's ability to structure therapeutic adherence. The manual and adherence measure can be used in efficacy and effectiveness studies involving either experimental or observational study designs.

Conclusion:

We designed a treatment manual for FOTT, a complex therapeutic approach used widely for swallowing therapy throughout Europe, defining the most essential components that are hypothesized to enhance behavioral change. In this study we developed and tested an adherence measure and showed that this measure is able to capture whether or not the therapist uses these components as intended. An important outcome of this study was that swallowing therapy in Denmark and the United States differs in content and not merely in name. This makes it highly relevant to investigate the impact of these components of FOTT and compare their effectiveness to other treatment methods with different proposed mechanisms. This manual and adherence measure paves the way to future efficacy and effectiveness studies.

References

1. Hart, T., *Treatment definition in complex rehabilitation interventions.* Neuropsychol Rehabil, 2009: p. 1-17.
2. Whyte, J. and T. Hart, *It's more than a black box; it's a Russian doll: defining rehabilitation treatments.* American journal of physical medicine & rehabilitation 2003. **82**(8): p. 639-52.
3. DeJong, G., et al., *Opening the Black Box of Poststroke Rehabilitation: Stroke Rehabilitation Patients, Processes, and Outcomes.* Archives of Physical Medicine and Rehabilitation, 2005. **86**(12): p. 1-7.
4. Whyte, J., *Clinical trials in rehabilitation: what are the obstacles?* American journal of physical medicine & rehabilitation 2003. **82**(10 Suppl): p. S16-21.
5. Hogue, A., H.A. Liddle, and C. Rowe, *Treatment adherence process research in family therapy: A rationale and some practical guidelines.* Psychotherapy: Theory, Research, Practice, Training, 1996. **33**(2): p. 332-345.
6. Moncher, F.J. and R.J. Prinz, *Treatment fidelity in outcome studies.* Clinical Psychology Review, 1991. **11**(3): p. 247-266.
7. Luborsky Lester , et al., *The Researcher's Own Therapy Allegiances: A "Wild Card" in Comparisons of Treatment Efficacy.* Clinical Psychology: Science and Practice, 1999. **6**(1): p. 95-106.
8. Schoenwald, S.K., et al., *Multisystemic therapy: monitoring treatment fidelity.* Fam Process, 2000. **39**(1): p. 83-103.
9. Nezu, A.M. and C.M. Nezu, *Evidence-based outcome research*, ed. O.U. Press. 2008, New York.
10. Bellg, A.J., et al., *Enhancing treatment fidelity in health behavior change studies: best practices and recommendations from the NIH Behavior Change Consortium.* Health Psychol, 2004. **23**(5): p. 443-51.
11. Terré, R. and F. Mearin, *Prospective evaluation of oro-pharyngeal dysphagia after severe traumatic brain injury.* Brain Injury, 2007. **21**(13/14): p. 1411-1417.
12. Mackay, L.E., A.S. Morgan, and B.A. Bernstein, *Swallowing disorders in severe brain injury: risk factors affecting return to oral intake.* Archives of Physical Medicine and Rehabilitation, 1999. **80**(4): p. 365-71.
13. Hansen, T.S., A.W. Engberg, and K. Larsen, *Functional oral intake and time to reach unrestricted dieting for patients with traumatic brain injury.* Arch Phys Med Rehabil, 2008. **89**(8): p. 1556-62.
14. Frymark, T., et al., *Evidence-based systematic review: Oropharyngeal dysphagia behavioral treatments. Part I--Background and methodology.* Journal of Rehabilitation Research & Development, 2009. **46**(2): p. 175-183.

15. Miller, R.M. and M.W. Chang, *Advances in the management of dysphagia caused by stroke.* Phys Med Rehabil Clin N Am, 1999. **10**(4): p. 925-41, x.
16. Elmstahl, S., et al., *Treatment of dysphagia improves nutritional conditions in stroke patients.* Dysphagia, 1999. **14**(2): p. 61-6.
17. Okada, S., et al., *What is the chin-down posture? A questionnaire survey of speech language pathologists in Japan and the United States.* Dysphagia, 2007. **22**(3): p. 204-9.
18. Nusser-Müller-Busch, *Die Therapie des Facio-Oralen Trakts - F.O.T.T. nach Kay Coombes*. Vol. 2. 2007, Berlin: Springer.
19. Seidl, R.O., et al., *Pilot study of a neurophysiological dysphagia therapy for neurological patients.* Clinical Rehabilitation, 2007. **21**(8): p. 686-97.
20. ARCOS. *Facial - Oral Tract Therapy : F.O.T.T.™*. [http://www.arcos.org.uk/] [cited 2006 4'th of april 2006].
21. Hogue, A., *Adherence process research on developmental interventions: filling in the middle.* New Dir Child Adolesc Dev, 2002(98): p. 67-74.
22. Landis, J.R. and G.G. Koch, *The measurement of observer agreement for categorical data.* Biometrics, 1977. **33**(1): p. 159-74.
23. Logemann, J.A., et al., *A randomized study of three interventions for aspiration of thin liquids in patients with dementia or Parkinson's disease.* J Speech Lang Hear Res, 2008. **51**(1): p. 173-83.
24. Bülow, M., R. Olsson, and O. Ekberg, *Videoradiographic Analysis of How Carbonated Thin Liquids and Thickened Liquids Affect the Physiology of Swallowing in Subjects with Aspiration on Thin Liquids.* Acta Radiol, 2003. **44**(4): p. 366-372.
25. Fink, T.A. and J.B. Ross, *Are We Testing a True Thin Liquid?* Dysphagia, 2009.

Acknowledgements

The authors wish to thank Tara Smith and Caron Morita for their help in subject recruitment. We also want to thank the occupational therapists at TBIU and speech and language therapists at Moss for agreeing to be subjects in this study and for helping with patient recruitment.

Table 1. Examples of a coding scheme for adherence measure to the FOTT decision algorithm: Facilitating a swallow by pressing at the floor of the mouth

	Reasonable	**Not reasonable**
Did the therapist make a decision to use this approach?	**1)** The therapist supports the patient swallowing, by pressing the fingers at the floor of the mouth, in order to move the tongue upwards and backwards when the patient does not initiate a swallow independently.	**2)** The therapist supports the patient swallowing by pressing the fingers at the floor of the mouth even when the patient swallows independently and efficiently.
Did the therapist make a decision NOT to use this approach?	**3)** The therapist stops facilitating swallowing, if the patient starts swallowing at the right time (spontaneously and efficiently)	**4)** The therapist stops facilitating a swallow, even the patient do not initiate a swallow by him self or does it later than normal, and is of risk of aspiration or penetration.

*Codes of 1 to 3 refers adherence and 2 an 4 to non adherence

Table 2. Nine components (active ingredients) in the FOTT swallowing therapy manual

Key points	Definitions
1. Location and furniture	Involves choosing the appropriate place and furniture for the intervention. They should be as normal as possible for the activity used in the treatment session but should also be adapted to the patient's capacity of, for example, level of concentration and perceptual problems.
2. Objects and aids	Involves choosing objects and aids that relates to the activity and patients impairments, supports the patient to use more normal way of performing the activity.
3. Position of the patient	The position of the patient should be as normal as possible for the activity and support normalizing muscle tone.
4. Swallowing of saliva	Involves working with oral stimulation as a preparation for working with eating and drinking or as a way of working with swallowing of saliva
5. Eating and drinking	Working with eating and drinking with different levels of support and different consistencies of food and liquid
6. Therapeutic approach	Involves choosing how to support the patient in performing the activity as normal as possible using: mobilization, facilitation (physical), guiding, elicitation.
7. Tongue movements or other area	Working with movements of the tongue, when relevant or another area as breathing or facial movements
8. Supporting swallowing	The therapist can choose facilitating swallowing by: pressing with fingers by the mouth floor, make the patient breath harder or use the voice, mobilize the patient body or parts of the body for example. shoulder, neck to elicit a swallow
9. Protection of airway	Involves different techniques to make the patient clear the airway (mainly by physical facilitation): supporting coughing, facilitate the patient clearing the throat, facilitating use of the voice, or facilitate the patient to come forward or cleaning the patients mouth for rests of food

Table 3. Inter-observer agreement in the two centers for all nine variables

	Kappa			Percent agreement	
Patient	***N**	**Mean/median**	**Min/max**	**Mean/median**	**Min/max**
HH Location	8	0.95/1.00	0.63-1.00	99/100	93-100
HH Objects	9	0.64/0.67	-0.11-100	87/87	73-100
HH Position	9	0.83/0.83	0.69-1.00	93/93	86-100
HH Swallow saliva	9	0.78/.079	0.58-1.00	93/92	90-100
HH eating/ drinking	3	0.83/0.83	0.64-1.00	95/100	80-100
HH Therapeutic approach	6	072/0.74	0.55-0.89	90/92	77-100
HH Tongue mov, other	4	0.87/0.84	0.80-1.00	95/93	92-100
HH- Support to swallow	9	0.86/0.85	0.63-1.00	94/93	86-100
HH Protection of airway	6	0.72/0.75	0.44-0.85	94/92	87-100
MOSS Location	7	1.00/1.00	1.00-1.00	99/100	93-100
MOSS Objects	7	0.84//0.85	0.70-1.00	90/87	75-100
MOSS Position	3	0.64/1.00	-0.07-1.00	97/100	87-100
MOSS Swallow saliva	2	0.28/0.28	-0.17-0.72	90/93	71-100
MOSS eating/ drinking	7	0.88/100	0.58-1.00	95/100	75-100
MOSS Therapeutic approach	6	0.48/0.67	-0.71-0.72	90/93	71-100
MOSS Tongue mov, other	4	0.97/1.00	0.87-1.00	96/100	86-100
MOSS- Support to swallow	7	0.77/0.80	0.55-1.00	88/87	71-100
MOSS- Protection of airway	8	0.84/0.84	0.55-1.00	93/93	75-100

*Numbers of sessions where Kappa could be calculated (N=9 for each center for calculation of percent agreeement)

Table 4: Percent of choices adherent to the Facial Oral Tract Therapy approach in USA(Moss) and Denmark (TBIU)

	*Moss	*TBIU	Mann-Whitney *U*	Z	Exact sign 2-tailed
Location/ furniture	16.03	100	5.00	-3.42	<0.01
Object	40.60	91.74	7.5	-2.97	<0.01
Position	1.45	76.80	0.00	-3.74	<0.01
Swallowing saliva	91.65	100	24.00	-1.74	0.11
Eating/drinking	47.87	98.89	6.5	-3.14	<0.01
Therapeutic approach	11.08	81.45	0.00	-3.59	<0.01
Tongue movements/ other	80.24	97.57	30.50	-0.94	0.36
Support swallowing	20.87	93.91	0.00	-3.62	<0.01
Protection of airway	29.62	86.92	2.50	-3.38	<0.01

*Numbers are an average percent measure between the 9 treatment sessions at each center

Table 5. Number of components where the therapist made a right choice to use it at least once throughout a treatment session.

	Number of "Right Choices" to use the components		**Total number of choices where a com**	
Patient	**N TBIU**	**N Moss**	**N TBIU**	**N Moss**
1	7	3	7	5
2	9	0	9	6
3	8	1	8	5
4	8	1	8	4
5	9	0	9	3
6	6	1	7	5
7	8	1	9	4
8	6	2	6	6
9	6	3	6	7
Total mean/median (min-max)	7.4/8 (6-9)	1.33/1 (0-3))	7.75/8 (6-9)	5/5 (3-7)

Study V

Hansen, T.S., Jakobsen, D., Westergaard, L., Speyer, R.

FOTT versus FEES. A clinical evaluation of swallowing, feasible for patients with severe TBI and low level of functioning

V

Preliminary manuscript. This manuscript contains all the planned analysis. However, due to the time limitation, not all FEES evaluation has been done by all evaluators as mentioned in the method section. Before this is done the manuscript cannot be submitted. This will be done as soon as possible.

FOTT versus FEES

A clinical swallowing evaluation feasible for patients with severe TBI and low level of functioning

Abstract

Swallowing evaluation for patients with severe Traumatic Brain Injury (TBI) involving low level of consciousness is a challenging procedure since such patients often cannot follow a verbal command. At our department for patients with TBI we use an evaluation in line with the treatment approach known as Facial Oral Tract Therapy (FOTT), however the clinical evaluation has never been standardized. The purpose of this study was to develop and validate a FOTT assessment tool of swallowing using Fibreoptic endoscopic evaluation of swallowing (FEES) as a gold standard.
First we did an inter-rater reliability test where a pair of therapists, both highly experienced in FOTT, evaluated the same 11 patients simultaneously and filled out the FOTT evaluation tool independently. Another pair of therapists, less experienced in FOTT, underwent the same test. Following this, the same pairs if therapists evaluated another 20 patients with the FOTT evaluation tool followed by FEES within a maximum of 48 hours determining retention, penetration and aspiration status.
The inter-rater reliability test had a kappa value in the “substantial to almost perfect” agreement range for 97% of the 30 items for the experienced therapists and 92% for the inexperienced therapists.
Only 6 patients out of the 20 patients in the validation study suffered from aspiration. Sensitivity of the FOTT evaluation was 83% for retention, 79% for penetration and 67% for aspiration; specificity was100% for retention, 83% for penetration and 50% for aspiration, respectively. The false positive rate for retention was= 0%, penetration=17% and aspiration=50%.
We found a high level of inter-rater reliability on most items of this evaluation for two different food consistencies. When compared with FEES evaluation we found a high sensitivity and specificity for retention and penetration but somewhat lower score for aspiration. Since very few patients showed aspiration on the FEES evaluation, we recommend more studies using this FOTT evaluation. The clinical FOTT evaluation tool showed promising results for all other aspects.

Keywords: Dysphagia, swallowing safety, dysphagia evaluation

Introduction

Dysphagia after severe traumatic brain injury (TBI) may lead to undernutrition, dehydration and aspiration pneumonia [73, 138, 196], which can have serious consequences for the rehabilitation progress, and can indeed be life threatening. The neurological impairments can affect both the physical, cognitive and behavioural processes involved in the pre-oral, oral, pharyngeal, and oesophageal phases of swallowing.

When patients with severe TBI continue to subacute rehabilitation, it is very important that they receive adequate nutrition [197], either orally or by a supplemental tube. Assessment of dysphagia and aspiration are essential to provide the right treatment and prevent the risk of pneumonia[198].

Fibreoptic endoscopic evaluation of swallowing (FEES) and videoflouroscopy has been recognized as the two most valid and objective instrumental tools for the assessment of swallowing and aspiration. However patients with severe TBI are not always able to cooperate in these evaluation methods and a clinical evaluation of swallowing is needed. A clinical evaluation can be repeated several times, at different times during the day and in different contexts. Moreover, evaluating the patient's ability to participate in the whole process of eating, from anticipation to the actual swallowing phase, generates essential knowledge when planning the rehabilitation programme. However, clinical evaluation has been found to miss up to 40%[199] of patients who suffer from silent aspiration.

At our department clinical bedside evaluation follows the concept of Facial Oral Tract Therapy (FOTT). This concept is widely used in neurorehabilitation today, despite the low numbers of studies addressing its effectiveness or efficacy[108]. It offers a structured way of evaluation and treatment of patients with disturbances in swallowing and eating, oral hygiene, non-verbal communication and speech articulation caused by neurological conditions[107]. The clinical evaluation approach used in FOTT is special because, in contrast to other assessments, the patient does not need to be able to follow a verbal command; the therapist performs the evaluation by physical facilitation of the different movements involved. Therefore this evaluation is also applicable to very severely injured patients. The evaluation includes assessment of: tongue movements, the quality of spontaneous swallow of saliva, the coordination of breathing and swallowing, the patient’s anticipation in eating and drinking including the ability to transport the food to and into the mouth (pre-oral phase), bolus formation and transport (oral phase), transport of food through the larynx (pharyngeal phase) and to the stomach (oesophageal phase). The FOTT

evaluation method is presumably similar across different clinics and rehabilitation settings but the data documentation has not yet been standardized.

In this study we wanted to develop an assessment form to structure the data retrieved from the FOTT evaluation of swallowing since it is important to have a clinical evaluation method that is standardized and can also be used for severely injured patients.

Objectives

The aim of this study was to develop and validate a FOTT assessment tool of swallowing using the FEES as a golden standard.

Methods

Development of the FOTT swallowing assessment form

We selected items for the initial FOTT assessment form based on the material from the basis FOTT course and existing literature describing FOTT The assessment is divided in four parts: 1. Examination of the oral cavity; including movements of the tongue (10 items) and soft palate (1 item) 2. Swallowing of saliva (4 items). 3. Swallowing of food and liquid including both anticipation in eating/drinking and safety (9 items to each consistency). 4. Presents of retention, penetration, aspiration and severity of dysphagia (4 items). Each item can be rated from 0-3, corresponding to: 0= no problem, 1=slightly disturbed, 2=severe, need of assistance 3=no function. Retention, aspiration and penetration are scored as Present, Yes or No

We have developed a manual with rating criteria for each item. After pilot application in our own department, unused items were omitted. The assessment form was then presented to an international group of FOTT instructors and adjusted according to their comments. No item demands that the patient should be able to follow a verbal command. The therapist always tries to judge the movement in a functional context. Movements are rated in relation to effectiveness and sufficiency and whether the patient needs support to perform the movement, se Appendix 1. The evaluation starts with a stimulation of the oral cavity (a routine in the FOTT approach) where movements of the tongue, soft palate and swallow of saliva are observed. Then, if the therapist finds it safe to

continue (the patient can safely swallow own saliva), the patient's ability to swallow food/liquid is assessed with the following consistencies: thickened liquid, puréed food, solid food and thin liquid. (The order of which consistencies are presented is not always the same. Depending on the individual, the patients can have difficulties with different consistencies, which do not necessarily reflect the severity of the problems but more their nature). The FOTT tool final items are: retention, penetration and aspiration.

- Retention is scored when some food or liquid remains in the back of the oral cavity after swallowing
- Penetration is scored when food/liquid were in the larynx, below the epiglottis but above the true vocal folds. This is clinical observed by wet voice, sounds during inspiration and exhalation, slight cough spontaneously followed by clearing swallow (compared with information of bolus transport and collection of bolus, dysfunction of the soft palate, late or missing clearing swallow, retention and/or reduced laryngeal movement
- Aspiration is scored when there was food/liquid below the true vocal folds. This was observed by: severe coughing followed by clearing swallow or leakage of bolus, impaired breathing. These symptoms were compared with information of bolus transport and collection of bolus, dysfunction of the soft palate, late or missing clearing swallow, retention, reduce laryngeal movement.

  Suspected silent aspiration was scored if the patient: makes sounds when breathing, has a wet voice without reaction from the patient, has very low swallowing frequency, drools in sitting position, never swallows spontaneously and never coughs when swallowing.

First we performed the inter-rater reliability test of the FOTT evaluation tool to test the utility and reliability of the test (study 1) and then we did a test of accuracy of detecting swallowing safety (study 2).

Study 1: Inter-rater reliability test of the FOTT swallowing assessment form

Subjects

The subjects in studies 1 and 2 are patients with severe TBI admitted to our subacute intensive rehabilitation department. Eighty percent of the patients are transferred directly from the neurosurgical wards. Patients are admitted as soon as they ventilate spontaneously[21]. Inclusion

criteria to both studies were age 15-75 years, some degree of swallowing problems and ability to complete a clinical evaluation of swallowing. Informed consent was obtained from the closest relatives since the patients were not able to give it themselves. Exclusion criteria were unstable medical condition such as pneumonia or other severe infection.
All items were tested for inter-rater reliability by four Occupational Therapists (OTs) performing the inter-rater test in two pairs. Two OTs did the swallowing assessment together with the patient and filled out the assessment form independently afterwards. Each pair tested 10 patients. One pair was very experienced in FOTT (the basic course and more than 1 year of experience in FOTT) while the other pair was less experienced (had an introductory course and no more than one year's experience).

Study 2. Accuracy of the FOTT swallowing assessment tool.
A prospective observational study.

FEES

Equipment

The equipment consisted of a 3.2 mm diameter flexible fibreoptic rhinolaryngovideoscope (Olympus ENF-V2), Imaging and Light Source system (Olympus OTV-SI) and Colour Monitor (Olympus OEV-191).

Procedures

An occupational therapist and a physician experienced in FEES procedure assessed whether the patients with problems eating/drinking were able to participate in a FEES.
A physician performed FEES together with two occupational therapists. The FEES were performed according to the description by Langmore[90] with slightly modifications. The fibreoptic laryngoscope was passed transnasally to the hypopharynx, where the mucosa, soft palate, pharyngeal wall, base of the tongue, sinus piriformis and larynx were viewed. Next, integrated swallowing function was observed beginning with spontaneous swallows (swallow of saliva) followed by swallowing of food and liquid with the different consistencies. The evaluation was stopped if there was any severe risk to the patient: Approximately 5 ml bolus was given with three trials per consistency in the following order: thickened liquid, puree food, solid food and thin liquid (all coloured with blue food dye). The FEES results were documented using the modified BDI scoring paper[200] and the penetration aspiration scale[201]. The videos were reviewed and examined by

four persons experienced in FEES. Consensus was reached for all items. All were blinded for the results of the clinical evaluation.

In both the clinical evaluation and the FEES the patients were mobilized in the best possible position for swallowing. They were positioned in a chair if possible or with the upper part of the body being elevated in the bed. We tried to ensure that the patients were positioned in an active sitting position, with equal weight bearing on both sides of the body-midline. Furthermore the head and body were symmetric in relation to midline and limbs and the neck was "long" and not abducted.

Subjects

Twenty patients were included in the study period of one year. It was not the intention to investigate all patients with dysphagia admitted at the department so the number of patients does not reflect the number meeting the inclusion criteria. There were several practical reasons why we did not have the opportunity to include more patients in this study period such as: the practical issue that the one physician who performed the FEES and the only OT should be present at the department at the same time, that it was possible to schedule that the clinical bedside evaluation and FEES could be done within the 48 hours. Additionally, in this study period there was a strike at the department for several weeks and several patients had Methicillin-resistant staphylococcus aureus (MRSA) and could not be in contact with the FEES device.

FOTT evaluation of swallowing (clinical evaluation)

Two occupational therapists performed the clinical evaluation and filled out the FOTT assessment form together within 48 hours of the FEES examination. The procedure of clinical evaluation was the same as described above except that the two occupational therapists doing the evaluation rated the items together.

Statistics:

The FOTT evaluation form was tested for inter-rater reliability using Cohen's kappa with 95% confidence interval and percent of agreement between raters. Cohen’s kappa assesses agreement between the two observers adjusted for chance. We used the criteria of Landis and Koch[193] for interpretation: a kappa value below 0.20 constitutes no or slight agreement, 0.21 to 0.40 is fair

agreement, 0.41 to 0.60 is moderate agreement, 0.61 to 0.80 is substantial agreement, and 0.81 to 1.00 is almost perfect agreement.
The Mann-Whitney *U exact* test was used to test the significance of the difference between FIM score at the time of the swallowing examination in the two groups.

The sensitivity and specificity, positive and negative predictive value and false negative- and positive rate were computed by taking the FEES as objective measure of retention, penetration and aspiration.

Ethics:
This project was accepted by The Danish National Committee on Biomedical Research Ethics (the Copenhagen regional committee) and The Danish Data Protection Agency. All participants or surrogates provided informed consent.

Results
Inter-rater reliability study:
Patients evaluated by the experienced raters (Group 1) had a mean age of 47 years (range: 25-69) and a median FIM score at the time of the swallowing assessment of 23 (23-52). 9 patients had TBI and 2 non-traumatic subarachnoid haemorrhage. Patients evaluated by the inexperienced raters (Group 2) were similar with a mean age of 47 years (range: 23-62) but with a somewhat higher FIM score at time of assessment (median=64 (range=23-103; U = 36, P = .09). 7 patients suffered from TBI, 2 from anoxia and 2 from non-traumatic subarachnoid haemorrhage.

Inter-rater reliability
The percentage of agreement is presented for all items with Cohen's kappa and statistical significance ($p<0.05$). It was not possible to compute kappa values for all items if one or both observers used only one rating category. In these cases only percent agreement is presented. See table 1

Insert table 1 approximately here

In group 1 kappa value ranged from 0.56- 1.00, with agreement in the moderate range for 1 component (eye-hand contact for puréed food); in the substantial range for 5 components and in the

almost perfect range for 24 components. In Group 2, the kappa ranged from 0.23-1.00 with 2 in the fair range, 3 in the moderate range, 7 in the substantial range and 18 in the almost perfect range. Percent agreement ranged from 0.82-100 across all components in Group 1 and 62-100 in Group 2. Agreements for aspiration, penetration and retention were 100%, kappa 1.00 for the Group 1 and 91-100% (kappa 0.62-1.00) for group 2.

FEES results

Twenty patients were included in this study. There were 9 female and 11 males. 11 patients suffered from TBI, 5 from anoxia cerebri and 4 from haemoragia. Patient characteristics are presented in table 2.

Insert table 2 approximately here

All of the 20 FEES were done within 24 hours of the clinical examination. 19 (95%) were able to drink thickened liquid, 17 (85%) received purée consistency, 15 (75%) got chopped food, 14 (70%) received thin liquid and 10 (50%) normal (firm) consistency.

Only 6 (30%) patients had aspiration risk identified with FEES evaluation and 14 (70%) experienced penetration and 18 (90%) retention, respectively.

Table 3 shows the actual numbers from the evaluations and table 4 shows sensitivity and specificity, positive and negative predictive value and false negative and positive rate for aspiration risk, penetration and retention as determined by comparing the clinical evaluation with FEES.

Insert Table 3 and 4 approximately here

The FOTT evaluation identified 11 out of the 20 patients with risk of aspiration, where 4 were correctly identified and seven incorrectly when compared with FEES. Moreover two patients were wrongly identified as having no risk of aspiration. This gives sensitivity for aspiration = 67%; a specificity=50%, a false negative rate=33% and a false positive rate=50%; a positive predictive value=36% and a negative predictive value= 78% (kappa 0.13; percent agreement 55%). For penetration the FOTT evaluation correctly identified 11 patients as suffering from penetration and 1 patient when he/she did not. The FOTT evaluation failed to identify 3 patients as suffering from penetration. This results provide a sensitivity= 79%, specificity= 83%, false negative rate= 21%, false positive rate= 17%, positive predictive value= 92% and a negative predictive value= 63% (kappa= 0.57; percent agreement 80%). Finally the FOTT evaluation correctly identified 15 patients

as having retention after swallowing and no patients were identified as having retention if they did not. 3 patients were identified as having no retention on the FOTT evaluation when they had it in the FEES evaluation (kappa= 0.5 and percent agreement= 85%).

Discussion

We have developed a clinical bedside evaluation for patients who are not able to follow a verbal command and investigated inter-rater reliability and validity to identify patients' swallowing safety in a pilot study of 20 patients.

In both groups inter-rater reliability was high for most items. For the experienced therapists, only 1 item (eye-hand coordination) was in the moderate range. For the inexperienced therapists 5 items were below substantial range. Items with lowest Kappa value were: tongue ROM out of the mouth and in the mouth, movements of the soft palate and hand-hand coordination for both food consistencies. Since two therapists performed the evaluation at the same time, it might be difficult for them to have equal view to all movements. This could explain why tongue movements have a low reliability score. The inexperienced therapists were asked about the disagreement on hand-hand coordination after the evaluations and it showed that they interpreted the manual for this item differently. The manual will be clarified further according to these comments. It was expected that the inexperienced therapists did not have as high agreement as the experienced ones since FOTT is a quite challenging and complex approach to learn and use[202-203]. Hence, the fact that 88% of all items were conducted with substantial to almost perfect agreement is promising for our swallowing evaluation tool.

McCullough et al.[204] investigated inter-rater- and intra-rater- reliability of clinical features typically used in clinical evaluation of swallowing and found that fewer than 50% of the measures were rated with sufficient reliability. The reason why our clinical evaluation found high inter-rater reliability may be that the clinicians in FOTT had gone through a very thorough training in this method. Even the therapist categorized as inexperienced had had some specific introduction in this method, where others may have different experience picked up from different methods throughout their professional career.

We found that our clinical evaluation underestimate a risk of aspiration for some patients who are at risk and that the therapists overestimated aspiration risk in patients who did not exhibit it. These results are in agreement with investigations from other population groups [137-138]. The clinical evaluation tool showed better outcomes for both penetration and retention.

A recent review paper of bedside screening tests vs. VFFS or FEES conclude that a water test combined with pulse oximetry is the most promising results as a screening tool [146]. However, the water-swallowing test does not include consistencies other than thin liquid. Since thin liquid does not approximate their diet throughout the day it is recommended to evaluate more consistencies than only one[146]. Moreover, some patients may be able to swallow thin liquid safely, but not other consistencies, due for example to decreased tongue movements. On the other hand, some patients may aspirate thin liquids and not purée consistencies. Butler et al[205] showed significantly more aspiration in healthy adults on thin liquids than on purée or solid food[205].

The Gugging Swallowing Screen[206], also used different food consistencies and demonstrated higher sensitivity and specificity for aspiration than our study. However, this scale uses cut off scores both to evaluate aspiration and to decide which patients can participate in the swallowing evaluation. Due to the differences of the swallowing problems found in patients with severe TBI and low level of functioning, it does not seem appropriate to use such a cut -ff score for this population group. Almost none of the patients in this study would have succeeded in swallowing any material according to the Gugging Swallowing Screen with the result of nil per mouth to all patients. In comparison we had “only” 30% with aspiration.

Retention was the component that was most correctly identified by the clinical evaluation of swallowing in our study. This parameter is rarely used in other studies, however Eisenhuber et al [207] found that 65% of the patients who had retention had post deglutive overflow resulting in aspiration. Since our patient group has a high risk of pneumonia[148] it could be appropriate to be a little more precautious than in other population groups and recommend that the clinical evaluation of retention should be an indication for referring the patient to a FEES. However, we also found high sensitivity and specificity for penetration[146] and so this could also be the FEES indicator.

We found that the patients evaluated by the inexperienced group of therapists had a significant higher FIM score than the experienced group. One might argue that a lower level patient may in some cases be harder to evaluate than one with a higher level of functioning. However, despite the average difference in FIM scores, the patients in the inexperienced group still had a wide range of FIM scores that did not affect kappa values systematically. A great limitation of this study was that the number of participants was low and of whom only 30% suffered from aspiration. This provides little power to investigate the predictive values of swallowing safety for the different items in the clinical evaluation of swallowing; especially the predictive value of aspiration. Several studies has investigated the predictive value of different clinical features such as dysphonia[208], wet voice[83, 209], coughing[83-84], choking[83-84, 137], abnormal gag[139], however one feature does not seem to be strong enough to rely on in order to predict aspiration[146]. Since there are many different causes of swallowing problems, the fact that only one feature cannot predict aspiration seems very reasonable. In future studies we would like to investigate the pharyngeal phase of swallowing in combination and investigate if problems in the pre-oral phase of swallowing or the oral phase could be strong enough to predict penetration/aspiration.

Another limitation with this study design is, that while the FEES evaluation is performed over a rather short period of time, the clinical evaluation of swallowing may take 30 – 60 minutes. During this long time, the therapist may have information about how the patient reacts when starting to get tired and perhaps lose concentration. Moreover, there may be an effect on the patient's arousal and concentration when the doctor approaches the room with a device involving having the endoscope entered through the nose. These factors might influence the patient's swallowing ability so that it is less in the FEES compared to the clinical evaluation. Hence it could be that some patients were not found to aspirate during the relatively short examination period during the FEES but actually did so during the clinical evaluation, biasing the result of this study.

In conclusion we have developed a clinical evaluation tool for the FOTT swallowing evaluation. The special feature for this evaluation compared to others presented in the literature is that it can be used for patients who are unable to follow a verbal command and that it involves a pre-oral phase of swallowing including the time from the patient handling the food to transporting it towards and in the mouth. We found a high level of inter-rater reliability on most items of this evaluation for two different food consistencies. When compared with FEES evaluation we found a high sensitivity and

specificity for retention and penetration but somewhat lower score for aspiration. Since this evaluation method shows promising results we find it highly relevant to investigate further in a study with more participants.

Acknowledgement

The professionals in the interdisciplinary teams at the BIU, occupational therapist's physiotherapists, neurophysiologists, nurses, speech therapists and medical doctors are thanked for their contributions with data collection for the present study.

References

1. Mackay, L.E., A.S. Morgan, and B.A. Bernstein, *Swallowing disorders in severe brain injury: risk factors affecting return to oral intake.* Archives of Physical Medicine and Rehabilitation, 1999. **80**(4): p. 365-71.
2. Leder, S.B. and J.F. Espinosa, *Aspiration risk after acute stroke: comparison of clinical examination and fiberoptic endoscopic evaluation of swallowing.* Dysphagia, 2002. **17**(3): p. 214-8.
3. Park, C. and P. O´Neill, *Management of neurological dysphagia.* Clin Rehabil, 1994. **8**: p. 166-174.
4. Denes, Z., *The influence of severe malnutrition on rehabilitation in patients with severe head injury.* Disabil Rehabil, 2004. **26**(19): p. 1163-5.
5. Langmore, S.E., *Dysphagia in neurologic patients in the intensive care unit.* Semin Neurol, 1996. **16**(4): p. 329-40.
6. Splaingard, M.L., et al., *Aspiration in rehabilitation patients: videofluoroscopy vs bedside clinical assessment.* Arch Phys Med Rehabil, 1988. **69**(8): p. 637-40.
7. Seidl, R.O., et al., *Pilot study of a neurophysiological dysphagia therapy for neurological patients.* Clinical Rehabilitation, 2007. **21**(8): p. 686-97.
8. Nusser-Muller-busch, R., *Konsensusempfehlungen 2007 zur Facio-Oralen Trakt Therapie (FOTT).* 2007.
9. Engberg, A.W., A. Liebach, and A. Nordenbo, *Centralized rehabilitation after severe traumatic brain injury - a population-based study.* Acta Neurol Scand, 2006. **113**(3): p. 178-184.
10. Langmore, S.E., K. Schatz, and N. Olsen, *Fiberoptic endoscopic examination of swallowing safety: a new procedure.* Dysphagia, 1988. **2**(4): p. 216-9.
11. Seidl, R. and R. Nusser-Muller-busch, *Der Berliner Dysphagie Index.* 2001, UnfallkrankenhausBerlin. p. 1-35.
12. Rosenbek, J.C., et al., *A penetration-aspiration scale.* Dysphagia, 1996. **11**(2): p. 93-8.
13. Landis, J.R. and G.G. Koch, *The measurement of observer agreement for categorical data.* Biometrics, 1977. **33**(1): p. 159-74.
14. Hansen, T.S. and D. Jakobsen, *A decision algorithm defining the rehabilitation approach: 'Facial Oral Tract Therapy'* ® Under review.
15. Hansen, T.S., et al., *Development of an adherence measure for a complex neurorehabilitation approach: "Facial Oral Tract Therapy"* ® Under review.
16. McCullough, G.H., et al., *Inter- and intrajudge reliability of a clinical examination of swallowing in adults.* Dysphagia, 2000. **15**(2): p. 58-67.
17. Chong, M.S., et al., *Bedside clinical methods useful as screening test for aspiration in elderly patients with recent and previous strokes.* Ann Acad Med Singapore, 2003. **32**(6): p. 790-4.
18. 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.
19. Butler, S.G., et al., *Penetration and Aspiration in Healthy Older Adults as Assessed During Endoscopic Evaluation of Swallowing.* Annals of Otology, Rhinology & Laryngology, 2009. **118**(3): p. 190-198.

20. Trapl, M., et al., *Dysphagia bedside screening for acute-stroke patients: the Gugging Swallowing Screen.* Stroke, 2007. **38**(11): p. 2948-52.
21. Eisenhuber, E., et al., *Videofluoroscopic assessment of patients with dysphagia: pharyngeal retention is a predictive factor for aspiration.* AJR Am J Roentgenol, 2002. **178**(2): p. 393-8.
22. Woratyla, S.P., et al., *Factors associated with early onset pneumonia in the severely brain-injured patient.* Conn Med, 1995. **59**(11): p. 643-7.
23. Daniels, S.K., et al., *Clinical predictors of dysphagia and aspiration risk: outcome measures in acute stroke patients.* Archives of Physical Medicine and Rehabilitation, 2000. **81**(8): p. 1030-3.
24. Warms, T. and J. Richards, *"Wet Voice" as a predictor of penetration and aspiration in oropharyngeal dysphagia.* Dysphagia, 2000. **15**(2): p. 84-8.
25. Smithard, D.G., et al., *Can bedside assessment reliably exclude aspiration following acute stroke?* Age Ageing, 1998. **27**(2): p. 99-106.
26. Lim, S.H., et al., *Accuracy of bedside clinical methods compared with fiberoptic endoscopic examination of swallowing (FEES) in determining the risk of aspiration in acute stroke patients.* Dysphagia, 2001. **16**(1): p. 1-6.
27. Logemann, J.A., *Do we know what is normal and abnormal airway protection?* Dysphagia, 1999. **14**(4): p. 233-4.

Table 1. Inter-rater agreement for a group of FOTT- experienced and - inexperienced therapists

	Experienced raters (Group 1)			**Inexperienced raters (Group 2)**		
Categories	Percent of agreement (%)	Cohen's Kappa	Significance	Percent of agreement (%)	Cohen's Kappa	Significance
Tongue tonus right	100%	1.00	>0.01	100%	1.00	>0.01
Tongue tonus left	100%	1.00	>0.01	100%	1.00	>0.01
Tongue ROM in the mouth	91%	0.74	0.01	82%	0.42	0.09
Tongue ROM out the mouth	100%	1.00	>0.01	73%	0.23	0.43
Soft palate	91%	0.74	0.01	82%	0.54	0.07
Swallow of saliva						
Quality of swallowing	100%	1.00	>0.01	100%	1.00	>0.01
Breathing	91%	0.62	0.03	91%		
Coordination of breathing and swallowing	100%	1.00	>0.01	100%	1.00	>0.01
Protection of airway	100%	1.00	>0.01	100%	1.00	>0.01
Swallow of food and liquid						
Thickened liquid						
Eye contact with food/drink	91%	0.81	>0.01	91%		
Eye-hand coordination	82%	0.69	>0.01	91%	0.74	0.01
Hand-hand coordination	91%	0.76	>0.01	64%	0.29	0.30
Hand-mouth coordination	91%	0.83	>0.01	91%	0.62	0.03
Bolus formation	100%	1.00	>0.01	91%	0.62	0.03
Bolus transport	100%	1.00	>0.01	91%	0.74	0.01
Pharyngeal response	100%	1.00	>0.01	100%	1.00	>0.01
Clearing swallow	100%	1.00	>0.01	100%	1.00	>0.01
Transport to the stomach	100%	1.00	>0.01	100%		
Puree consistency						
Eye contact with food/drink	91%	0.84	>0.01	100%	1.00	>0.01
Eye-hand coordination	82%	0.56	0.01	100%	1.00	>0.01
Hand-hand coordination	100%	1.00	>0.01	73%	0.53	0.03
Hand-mouth coordination	91%	0.85	>0.01	100%	1.00	>0.01
Bolus formation	91%	0.85	>0.01	91%	0.76	>0.01
Bolus transport	91%	0.85	>0.01	91%	0.76	>0.01
Pharyngeal response	91%	0.85	>0.01	100%	1.00	>0.01
Clearing swallow	100%	1.00	>0.01	100%	1.00	>0.01
Transport to the stomach	100%	1.00	>0.01	100%	1.00	>0.01
Retention	100%	1.00	>0.01	91%	0.62	0.03
Penetration	100%	1.00	>0.01	100%	1.00	>0.01
Aspiration	100%	1.00	>0.01	100%	1.00	>0.01

*For Group 1 calculations on thickened liquid are based on 10 patients and purée on 9 patients. For Group 2 thickened liquid is based on 11 patients and purée on 10 patients.

Table 2. Patient characteristic for the FEES study population

	Median (IQR)	Mean	Min	Max
Age (mean)	47 (37-55)	47	20	72
GCS admission at BIU	7 (5-9)	7.7	4	14
FIM admission at BIU	23 (23-23)	23	23	23
FIM at assessment	23 (23-37)	38	23	141

Table 3. Number of subjects identified with retention, penetration and aspiration in the clinical evaluation of swallowing and FEES

	Retention present with FEES	No retention present with FEES	Penetration present with FEES	No penetration present with FEES	Aspiration risk with FEES	No aspiration risk with FEES
Positive FOTT evaluation	15	0	11	1	4	7
Negative FOTT evaluation	3	2	3	5	2	7

Table 4. Comparison of aspiration, penetration and retention between FOTT clinical evaluation of swallowing and fibreoptic endoscopic evaluation of swallowing

	Sensitivity	Specificity	False positive rate	False negative rate	Predictive value		Kappa (percent agreement)
					Positive	Negative	
Retention	83%	100%	0%	17%	100%	40%	0.5 (85%)
Penetration	79%	83%	17%	21%	92%	63%	0.57 (80%)
Aspiration	67%	50%	50%	33%	36%	78%	0.13 (55%)