MASTICATION
IN CHILDREN WITH CEREBRAL PALSY

Lianne Remijn
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For reasons of consistency within this thesis, some terms have been standardized throughout the text. Consequently, the text may differ in this respect from the articles that have been published.
MASTICATION IN CHILDREN WITH CEREBRAL PALSY

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Voor Lauren, Nathan en Rogier
dat zij hun eigen talenten zullen ontplooien
en hun dromen mogen realiseren
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Introduction
Eating and drinking is one of our daily activities, not only necessary for energy and nutrition intake but also for participating in social events with family and friends. However, many children suffer from problems with these activities, such as refusing food, coughing or gagging while eating or drinking, prolonged mealtimes, excessive drooling, frequent spitting up or vomiting. Unfortunately, for many children and their parents, family mealtimes go along with feeding and swallowing problems, which result in daily stressful situations.

The American Speech-Language-Hearing Association (ASHA) described feeding and swallowing problems (also dysphagia) as: “difficulties gathering food and getting ready to suck, chew, or swallow it”. This includes “difficulty with any step of the feeding process from accepting foods and liquids into the mouth to the entry of food into the stomach and intestines”, resulting in “developmentally atypical eating and drinking behaviors, such as not accepting age-appropriate liquids or foods, being unable to use age-appropriate feeding devices and utensils, or being unable to self-feed”.

Feeding and swallowing problems are estimated to occur in 25-45% of children in the general population and in up to 90% of children with medical or neurological disorders. The prevalence of feeding and swallowing problems is increasing due to improved survival rates among children born prematurely with low birth weight and with complex medical conditions. However, the nature and severity of feeding and swallowing problems within the pediatric population are very heterogeneous.

Speech-language therapists (SLTs) are involved in feeding and swallowing problems through the assessment of and intervention with disabilities associated with the client’s situation. The ultimate goal is improving the client’s health-related quality of life. In clinical practice, however, there are only limited uniform systems or guidelines for describing or assessing feeding and swallowing problems, and the results of interventions, especially in children, are rarely described. This thesis focuses on the development and validation of an observation assessment for mastication, particularly used on children with cerebral palsy (CP). In addition, the feasibility of the use of quantitative methods for daily clinical practice using valid and reliable measures of mastication are investigated, and well-defined objectives for subsequent intervention will be formulated.

This chapter starts then with a description of the factors related to feeding and swallowing problems in children, to be followed by a discussion on mastication and assessments of mastication, especially those used for children with CP. Finally, this chapter concludes with an outline of the thesis.
**FACTORS RELATED TO FEEDING AND SWALLOWING PROBLEMS**

Feeding and swallowing problems in children are caused by various medical (e.g., neurological impairments, gastrointestinal problems, oropharyngeal dysfunction), developmental (e.g., prematurity), and behavioral factors, such as neophobia (i.e., rejection of foods that are novel or unknown to a child). Besides the daily symptoms of feeding and swallowing problems, these children may be at risk of problems in later childhood, including dehydration or poor nutrition status, decreased height, delayed progression of oral feeding skills, recurrent respiratory diseases, and/or isolation in social situations involving eating. Moreover, the parents of these children often exhibit low self-esteem, as well as parental stress and problems in parent-child interaction related to their child’s food intake. The text box below presents a case study on feeding and swallowing problems from my clinical practice that is followed throughout the introduction section.

**Case Joey (part 1)**

Joey is a 2-year-old boy who was born prematurely and suffers epileptic insults. Despite problems with drinking from a bottle and accepting spoon feeding, he developed well in his first months of life. When his growth and weight gain stopped at the age of 9 months, he received tube feeding. Around that time, he experienced several choking incidents. His intake of oral food decreased and he refused solid foods more frequently. His current oral intake varies between 10-150 grams of pureed food per meal. He does not participate in family mealtimes and his parents are increasingly hesitant to offer him solid foods, due to a fear of choking. Moreover, Joey is not accepted in child care facilities due to tube feeding.

In the literature, most classifications of feeding and swallowing problems in children are based on abnormal behavior, medical characteristics, or an interaction between these factors. The framework of the World Health Organization’s (WHO) International Classification of Functioning, Disability and Health, Child and Youth version (ICF-CY) offers a conceptual framework for describing the functioning of children and youth by considering their physical, social, and psychological development, and can be used to describe interacting factors that play a role in eating and drinking (Figure 1). Functioning is described from three different perspectives: the body (functions and structures), the individual (activities), and society (participation). The ICF framework also includes environmental and personal factors that influence individual functioning, so this is more extensive than behavioral or medical classifications. Figure 1 shows a diagram of the interaction of various components of the ICF-CY.
Figure 1. Diagram representing the various components of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY)\textsuperscript{19} (p.17).

According to the WHO\textsuperscript{19}, body structures are anatomical parts of the body, such as the mouth (teeth, gums, palate, tongue). Body functions concern functions of body systems (e.g., biting, chewing, manipulating food in the mouth, swallowing) and psychological functions (e.g., attention and cognition, mental functions, psychomotor functions). Activity is the execution of a task by an individual, such as focusing attention and eating various food textures. Body structures and functions and the acquisition of activities change during childhood and are associated with growth, maturation, and learning: for example, processing more and various foods during infancy. Participation is defined as a person’s involvement in a life situation (e.g., participating in mealtimes at kindergarten, at school or with peers). Functioning is the umbrella term for body structures and functions and activities and participation, whereas the term disability is associated with impairments of body structures and functions, limitations in activities and restrictions in participation. Environmental factors make up “the physical, social and attitudinal environment in which people live and conduct their lives”\textsuperscript{19}(p.17), whereas personal factors are “the particular background of an individual’s life and living, and comprise features of the individual that are not directly part of a health condition of health status”\textsuperscript{19}(p.44), such as character, habits, coping style, temper, and age.\textsuperscript{19}

A person’s functioning and disability are conceived as a dynamic interaction between health conditions (e.g., diseases, disorders, traumas) and contextual factors (e.g., environmental and personal), which can be supportive or hampering. In ICF terminology, feeding and swallowing problems are a combination of impairments in functions and structures and limitations in activities which may lead to restrictions in participation at mealtimes. Additional environmental factors may positively or negatively influence eating and drinking situations (e.g., parental stress during mealtimes and the quantity of exposure to different textures and tastes). The case of Joey (see Text box) illustrates this interaction.
between an impaired sensorimotor function that leads to an inability to chew, which results in limited food intake and problems with participation, combined with contextual factors.

**Case Joey (part2)**

Within the ICF-CY framework, the following was observed for Joey: disability in chewing (body function), limited food intake (activities) and denied access to childcare facilities due to tube feeding (participation). Moreover, important environmental factors related to the parents include anxiety related to their loss of another baby and the mother’s subsequent emotional problems. As for personal factors, Joey is a strong-willed boy who uses food refusal to seek attention from his parents.

Within the ICF framework, a selection of some relevant domains related to feeding and swallowing problems in a large population of children can be given in terms of body functions and structures, activities and participation items, and environmental factors on the first level (Table 1). The codes mentioned between brackets in Table 1 relate to the second-level categories of the ICF-CY within the selected chapters. Personal factors are not classified in the ICF.

**Table 1.** Relevant chapters and second-level categories of the ICF-CY for feeding and swallowing (derived from WHO, 2007).19

<table>
<thead>
<tr>
<th>ICF code and name of codes</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Body Functions (b)</strong></td>
</tr>
<tr>
<td>b1 Global mental functions: dispositions and intra-personal functions (b125), temperament and personality functions (b126), energy and drive (b130), perceptual functions (b156)</td>
</tr>
<tr>
<td>b2 Sensory functions and pain; taste function (b250), sensory functions related to temperature and other stimuli (b270)</td>
</tr>
<tr>
<td>b5 Functions related to the digestive system: ingestion functions (b510) (biting, chewing, manipulation of food in the mouth, salivation, swallowing), digestion functions (b515) (transport of food through stomach and intestines, tolerance of food), sensations associated with the digestive system (b535)</td>
</tr>
<tr>
<td>b7 Neuromusculoskeletal and movement-related functions: muscle power functions (b730), muscle tone functions (b735), motor reflex functions (b750), control of voluntary movement functions (b760)</td>
</tr>
<tr>
<td><strong>Body structures (s)</strong></td>
</tr>
<tr>
<td>s3 Structures involved in voice and speech: structure of mouth (s320) (teeth, gums, palate, tongue), structure of pharynx (s330)</td>
</tr>
<tr>
<td>s5 Structure related to digestive, metabolic and endocrine system: structure of salivary glands (s510), structure of esophagus (s520)</td>
</tr>
<tr>
<td><strong>Activities &amp; participation (d)</strong></td>
</tr>
<tr>
<td>d1 Learning and applying knowledge: focusing attention (d160)</td>
</tr>
<tr>
<td>d2 General tasks and demands; Handling stress and other psychological demands (d240)</td>
</tr>
<tr>
<td>d5 Self-care: eating (d550), drinking (d560)</td>
</tr>
<tr>
<td><strong>Environmental Factor (e)</strong></td>
</tr>
<tr>
<td>e3 Support and relationships: immediate family (e310)</td>
</tr>
<tr>
<td>e4 Attitudes: individual attitudes of immediate family members (e410)</td>
</tr>
<tr>
<td>e5 Services, systems and policies: health services, systems and policies (e580)</td>
</tr>
</tbody>
</table>
Eating and drinking are complex activities that are influenced by several factors which cannot be covered within one thesis. The current research is largely dedicated to the body function ‘chewing’, frequently referred to as ‘mastication’.

**MASTICATION**

Mastication is described by Logemann as the process of oral intake via food processing and bolus formation and rhythmic oral-motor activity to prepare for swallowing. However, the definition of ‘masticate’ in the Merriam-Webster Online Dictionary is “to grind or crush (food) with the teeth and to soften or reduce to pulp by crushing or kneading”. This narrow definition only extends to the motor process used to reduce a piece of food. In this thesis, Logemann’s broader description is used to understand the human mastication process. An object-oriented framework is described for mastication, consisting of one section describing the mastication process, one the masticatory apparatus, and one the neuromuscular control of mastication.

**Mastication process**

The mastication process is divided into three main oral stages: [1] oral transport stage I, [2] food processing, and [3] oral transport stage II. These stages are preceded by the [0] oral preparatory stage and followed by the [4] pharyngeal stage.

In the [0] oral preparatory stage (ingestion), the mouth opens as the tongue drops down to make space for the entering bite. At the time of ingestion, the jaws are maximally open. As soon as the food is deposited, the bite is placed on the anterior-middle tongue surface. Next, the body of the tongue shifts backward in the oral cavity and rotates to deliver the food to the chewing surface of the molars.

In [2] food processing, the food is softened and reduced in size, manipulated and mixed with saliva and liquids derived from the food itself. Mandible movements are associated with the cyclic movements of the tongue and the hyoid bone. The food is processed on the occlusal surface by a combination of rhythmic tongue-pushing and cheek-pushing actions.

In the [3] oral transport stage II, the bolus is formed into an acceptable size for swallowing. Portions of the food bolus are propelled through the faucial pillars into the oropharynx, where it is stored in anticipation of swallowing. Cyclic motions of the jaw and tongue continue in a linked pattern; in some chewing cycles food is pulled backward on the surface of the tongue, and in other cycles, the tongue pushes the food against the palate and squeezes it back along the palate into the oropharynx. During this phase, a combination of reflexive and voluntary mechanics is responsible for the mastication process.
In the [4] pharyngeal stage, the pharyngeal swallow begins and the food bolus in the oropharynx is propelled through the hypopharynx into the esophagus. Bolus propulsion is primarily produced by the backward thrust of the tongue into the oropharynx, followed by pharyngeal wall action that moves the bolus into the esophagus. 

Swallowing occurs intermittently in a chewing sequence. Adults need at least two swallows per bolus, even with one bite of food. “Most of the food is swallowed in the first swallow, and any residual food is aggregated by the tongue into a bolus and then swallowed in the last swallow”.25(p.172)

Masticatory apparatus

The masticatory apparatus consists of four major components: bones, muscles, teeth and soft tissues.30 Bones involved in mastication are the maxilla (upper jaw) and mandible (lower jaw). The movements of the jaws are bound by two mutually linked temporomandibular joints.31

Masticatory movements are executed using muscles connected to the maxilla and mandible. Figures 2 and 3 provide an overview of the masticatory muscles responsible for jaw movements. (1) The temporalis, masseter, and medial pterygoid muscles are responsible for the occlusion of the mouth (elevators). (2) The digastric, mylohyoid and geniohyoid muscles are responsible for opening the mouth (depressors). (3) The lateral pterygoid muscles assist in opening the mouth and draw the mandible forward. (4) The posterior fibers of the temporalis muscle retract the mandible. (5) The lateral and medial pterygoid muscles are responsible for the mandible’s lateral movements.30,32 The masticatory muscles participate in complex actions and the amount of muscle activity depends on the texture of food; harder food needs more muscle activity.24,31

The teeth are important in the masticatory system for fragmentation of different foods. The fragmentation depends on the total occlusal area, number of teeth24, and forces in different directions.31

Soft tissues, such as the tongue, lips, and cheeks, are important for manipulating food to maximize chewing efficiency and bolus control in the oral cavity.33,34 The tongue moves food distally through the oral cavity, collects food particles to form a bolus and then transports it to the pharynx for swallowing.26 During chewing, the coordinated actions of tongue muscles result in the tongue a constantly changing shape and position29, and a range of horizontal and vertical movements.35 The intrinsic muscles, arranged along the length of the tongue, are responsible for its lengthening, shortening and curling, and the flattening and rounding of its surface.36,37 The extrinsic muscles of the tongue (genioglossus, geniohyoid, styloglossus, and hyoglossus muscles) are connected with the mandible, hyoid, and cranial base (Figure 3). The genioglossus and geniohyoid muscles form the main body
of the tongue and are tongue protruders, whereas the styloglossus and hyoglossus muscles are responsible for elevating, retracting and depressing the tongue. Moreover, the tongue is essential for sensory input related to food, such as recognizing texture and volume.

Lips and cheeks are important to maximizing chewing efficiency and ensuring bolus control in the oral cavity. The movements of the lips are executed by the orbicularis oris muscle, which encircles the mouth and is responsible for closing and protruding the lips (Figure 3); lip pressure prevents loss of food and saliva. The movements of the cheeks are mainly made by the buccinator muscle and push the food between the molars (Figure 3).
Coordination of the complex feeding, grinding and swallowing process
The neuromuscular regulatory system is designed to coordinate mastication. The trigeminal nerve (Cranial Nerve V) is responsible for sensations in the face and for motor functions, such as biting and chewing. The hypoglossal nerve (Cranial Nerve XII) provides the motor innervations to the muscles of the tongue. The medial branch of the hypoglossal nerve innervates the extrinsic protruders (genioglossus and geniohyoid muscles) and their homologous intrinsic muscles, whereas the lateral branch innervates the extrinsic retractors (styloglossus and hyoglossus muscles) and their homologous intrinsic muscles. The facial nerve (Cranial Nerve VII) controls the muscles for facial expressions and taste sensations from the anterior two-thirds of the tongue and oral cavity.

The motor program of the coordination of mastication is controlled by the masticatory central pattern generator, located in the brainstem, which results in basic rhythmic stimulation of the jaw muscles. The motor program adapts to the physical properties of the food being chewed in response to oral receptors that provide textual, size and taste information through peripheral feedback.

DEVELOPMENT OF NORMAL CHEWING IN CHILDREN
Throughout their first year, infants shift from being able to only suckle, swallow and take in liquid foods to being able to chew as their mouth, tongue, and digestive tract mature. Movements of mouth, lips, and tongue transform from undifferentiated movements in infants to the differentiated and refined movements that toddlers and young children require for biting, chewing, and bolus formation and propulsion. In typically developing infants, the transition to processing foods usually begins around 4 to 6 months with the introduction of smooth pureed fruit and vegetables. Around that age, food is mashed by upward-downward movements of the tongue. From 8 months onwards, food is crushed by raising and lowering the mandible, without a rotary component, called ‘munching’. At this age, pieces of soft chewable foods are introduced.

Chewing becomes increasingly efficient and coordinated; the tongue becomes more mobile and independent of the mandible, resulting in increased control of food manipulation. Lateral tongue movement is developed from 10 months on. At the age of 12 months, the coordination of chewing refines; typically developing infants can move food from the center of the tongue to the side, chew it and then move it back to the center again. At that age, children can eat various foods and textures. The oral sensorimotor function provides information about whether food is ready for swallowing. If food pieces are still too large, pieces are moved back to the side of the mouth for more chewing.

By the time infants are 24 to 36 months old, circulatory jaw rotations are present, resulting in food moving from one side of the mouth to the other in an easy, smooth chewing
pattern.\textsuperscript{2,30} Gradually, infants develop better coordination and strength that enables them to eat increasingly more complex foods and a combination of textures that require simultaneously chewing and liquid swallowing.\textsuperscript{45,46} Moreover, children are increasingly able to process solid foods in various mealtime contexts, such as at school or an outdoor picnic. As such, a typically developing pre-school child is assumed to be ready to participate safely and efficiently in school eating activities with peers with age-appropriate support.\textsuperscript{2}

The developmental mastication process between 2 and 4 years of age is influenced by teeth eruption and refining control of lips and tongue.\textsuperscript{30} Chewing effectiveness continues to improve at least until children reach the age of 8 years.\textsuperscript{30} Literature reports a decrease in chewing duration and number of chewing cycles in children from 6 months to 8 years of age\textsuperscript{47,48}, which is interpreted as a continuous improvement with age.

It is suggested that critical and sensitive periods exist in the development of normal feeding behavior. During such periods, the development of skills depends on offering adjusted experiences with foods. The critical period for introduction of chewable foods is 9-12 months.\textsuperscript{49} The longer the delay in the introduction of solids after the critical period, the more difficult it is for many children to accept food that requires chewing. Offering products that are well adapted to a child’s mastication ability can facilitate the acceptance of new textures and tastes.\textsuperscript{50,51}

\textbf{Case Joey (part 3)}

After birth, Joey was too premature to have mature oral reflexes and started with tube feeding. Pressure was put on the mother to feed him orally and to gain weight to the threshold for discharge from the hospital. This increased the stress round feeding moments. When he reached the age of three months gestational age, he could be discharged. He frequently vomited and had trouble gaining weight. He was given pureed food at six months (corrected for gestational age). However, problems with the amount of food that he would take, delayed the introduction of solid foods. At the age of 1 year, he mainly had tube feeding and pureed food, so he missed the critical period for chewing and learning from experience with solid foods.

\textbf{DISRUPTION OF THE NORMAL DEVELOPMENT IN CHEWING}

The development of mastication is a complex process of anatomic, physiological and neurological development, influenced by personal or environmental factors. Normal motor development of mastication, which includes motor control and motor learning, may be disrupted by neurological diseases or impairments (e.g., CP, neuromuscular disease, intellectual impairments), genetic syndromes (e.g., Down syndrome, autism
spectrum disorder), or anatomic/structural etiologies (e.g., cleft palate).\textsuperscript{28,52,53} Moreover, the development of mastication can also be impeded by a lack of experience of eating and drinking within the normally expected age range or sensitive period. This could be due to a poor medical or physical condition. Mastication is also affected by impairments in attention functions, sensory perception, and behavioral aspects.\textsuperscript{46} Children who have problems with mastication exhibit loss of food and fluid from the mouth, choking, prolonged processing of solid foods, swallowing large pieces of food and so forth.\textsuperscript{54,55}

**Casus Joey (part 4)**

His insufficient growth resulted in restarting the tube feeding and thus decreasing his oral intake. In general, Joey refused solid foods. He experienced several choking incidents and his parents were anxious about giving him solid foods. Joey accepted a biscuit, manipulated it with his hands and licked it several times before giving it back. Joey was unable to perform isolated tongue movements and showed an oral dyspraxia. Moreover, he only made non-specific speech sounds and gross motor development was very slow. After a neurological assessment at the age of 1 year, cerebral palsy was diagnosed.

For intervention purposes, understanding the mechanism of normal mastication is essential to interpreting the characteristics of disrupted mastication. This thesis focused on children with cerebral palsy (CP) as a group with potential mastication problems.

**CEREBRAL PALSY**

The most widely accepted definition of cerebral palsy or CP was put forward by Rosenbaum: “Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitations, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. Beside the motor impairments, sensation, perception, cognition, communication, behaviour, epilepsy and secondary musculo-skeletal problems are also accompanied to cerebral palsy”.\textsuperscript{56(p.6)} The worldwide prevalence of CP is approximately 2-2.5 per 1000 live births and it is the most frequent cause of motor disability in childhood.\textsuperscript{57,58} Over the last 20 years, the incidence of CP has remained relatively stable. Although improved perinatal care has resulted in fewer children born with neurological damage, the higher survival rates of low-weight premature babies have resulted in a stable prevalence.\textsuperscript{59} Preterm infants are at the highest risk of developing CP. The causes of CP can be classified into congenital (e.g., developmental, malformations, syndromic) or acquired (e.g., traumatic, infectious, hypoxic, ischemic, infections) factors.\textsuperscript{52,60}
The predominant form of CP is spasticity; other forms are dyskinesia (dystonia or choreoathetosis) and ataxia.\textsuperscript{58,61} CP can be classified according to the topographic distribution of motor impairments: 25% of children with spastic CP exhibit hemiplegia, 37.5% exhibit quadriplegia and 37.5% exhibit diplegia.\textsuperscript{61} The motor impairments of children with CP can affect all movements and postures, which may change over time and range from subtle limitations to severe limitations with complete dependence on others for care.\textsuperscript{62-64} These sensory and motor impairments are caused by a central nervous system disorder in which muscle tone is variable, primitive reflexes may be strong and persistent, and coordination of muscles is inadequate. This not only affects gross and fine motor functions, but may also result in difficulty with speech and feeding.\textsuperscript{2,65,66}

The prevalence of feeding and swallowing problems in individuals with CP is unclear. Estimates range from 45%\textsuperscript{6} to 90%\textsuperscript{7}, depending on the type of CP, the definition of feeding and swallowing problems and the assessment tools used.\textsuperscript{1} The prevalence of feeding and swallowing problems is highly related to the severity of motor impairments\textsuperscript{54}, although eating and drinking difficulties also occur in individuals with mildly affected gross motor functions.\textsuperscript{15} Feeding and swallowing problems in children with CP result into significant health implications, inadequate weight and growth, and eventually malnutrition.\textsuperscript{12,54,67-69} Children with severe forms of CP and neurologic impairments.\textsuperscript{15} Oral-motor impairments in children with CP may include difficulties, such as poor lip closure, impaired tongue movements, restricted tongue lateralization and primitive or disturbed oral reflexes, including suck-swallow reflex, tonic bite, and impaired swallow reflex.\textsuperscript{55} Limited knowledge is available about interventions to treat mastication problems in children with CP. There is no high-quality evidence from (quasi-)randomized controlled trials to provide conclusive results about the effectiveness of any oral-motor therapy for children with neurological impairments.\textsuperscript{53} Some studies with small groups of children have shown positive results for interventions. Clawson and colleagues reported improvement of oral intake in eight children with spastic diplegic CP by means of a combination of behavioral and oral-motor intervention.\textsuperscript{70} Gisel noted an improvement in sensorimotor treatment and functional training in spoon feeding, biting and chewing in 11 children with CP.\textsuperscript{71} This result was confirmed in a more recent study by Baghbadorani et al.\textsuperscript{72} Mastication has also been found to improve using an intra-oral appliance in 20 children with CP.\textsuperscript{73} Since most of these studies had different inclusion criteria, it is difficult to compare the methods of assessment, intervention and outcome variables, and study results, or to generalize the findings to other groups of children with CP.
ASSESSMENT OF MASTICATION

The assessment of mastication in children is likely to encompass multiple dimensions, including: medical and feeding history, health and nutritional status, somatic growth, neuro-developmental status, orofacial structures, environmental issues, feeding and swallowing examination, and instrumental assessment.\(^5\) The outcome of any assessment should therefore focus on three goals:

- detecting children with mastication problems;
- gaining insight into the factors underlying the mastication problems necessary to make decisions for intervention, care instruction, and, in some cases, adaptations, such as avoiding solid foods;
- evaluation of changes in mastication performance over time due to development and/or intervention.

The choice of measurement method is driven by the clinical question and will result in different measurement outcomes. For example, assessment of caloric food intake differs from detailed analyses of motor performance of mastication. Norm-referenced or criterion-referenced tests can be used in assessments. In norm-referenced tests, individual performance is compared to the performance of typically developing peers in the same age group; in criterion-referenced tests, performance is compared to a predefined set of specific skills.\(^74\) Moreover, the score on a criterion-referenced test is based on absolute standards; variability of scores is not obtained because perfect or near-perfect scores are desired, and the results enable clinicians to plan an intervention program or determine its effectiveness by examining an individual child’s performance.\(^75\) The diversity of children with mastication problems supports the use of criterion-referenced tests.

In clinical practice, observational assessments are most common. The advantages of these observational instruments are that they are non-invasive, easy to use and inexpensive. An observational assessment provides an opportunity to observe children in mealtime settings and determine the efficacy of the mastication process. The disadvantages, however, are that the psychometric characteristics of most pediatric feeding and swallow assessments cannot be guaranteed.\(^54,76\) Moreover, some instruments include only a few items related to mastication and are therefore inappropriate for a detailed mastication assessment. Some of these instruments also focus on testing a broader range of oral motor problems and combine the assessment of speech and oral-motor tasks. Other instruments use a dichotomous scale and lack nuance that can differentiate in the performance during development and learning, which seems relevant when evaluating over time. Therefore, a clinical observational tool with detailed scoring may be desirable for assessing mastication and determining eventual intervention goals.

An overall disadvantage of observational assessment in mastication is the difficulty of examining intra-oral movements or the position and variable shape of the tongue,
even in slow-motion video recording. For research purposes, different instrumental measures are used to (indirectly) quantify mastication efficiency, such as food bolus characterization (particle-size distribution), muscle activity measurement (bite force and electromyography), fluorography, ultrasound, and jaw movement tracking. Most of these measurements are mainly used in adults. To quantify tongue movements, researchers have also used a ‘marker’ pellet technique. This technique with glued tongue markers is effective for speech purposes, but is less appropriate for exercises with food and fluid due to the interference of glue with saliva or foods. Moreover, it may interrupt the subject’s habitual chewing pattern by changing sensory input from the pellets.

Ultrasound creates images of soft tissue in real time by using sound waves with a frequency too high to be audible to humans. Ultrasonic images are made by sending sound waves into soft tissues by using a transducer (also known as a probe). Submental recordings show the shape changes of the tongue surface, although the presence of air under the tongue can prevent imaging. The method is non-invasive for the subjects and the submental-placed transducer hardly interrupts natural chewing. Depending on the transducer used, tongue movements can be measured in both the sagittal and coronal planes.

Surface electromyography (sEMG) provides information about the properties of the neuromuscular system and allows us to identify the processes of mastication. Electrical signals emitted from muscle cells are recorded to obtain information about muscle activation time, activity, strength, and fatigue. This method is used for studies in mastication in both healthy children and adults and those with CP. The relationship between the muscle fiber conduction velocity, frequencies and the force of the muscle contractions give insight into the adequacy of the muscle coordination.

The use of 3D kinematics by high-speed digital cameras to record movements of chin markers in three dimensions provides accurate and detailed information about mandibular movement displacement, speed, and performance variability during chewing. This technology has been used with children to detect gains in mandibular control in early child development.

The above-mentioned instrumental studies have contributed to the understanding of mastication, but differences in measurement protocols and systems have resulted in a variety of outcome variables with related but distinct definitions. Standardized, validated and reliable outcome measures of the mastication characteristics in children with CP are needed to assess mastication and to develop interventions in this field.
AIMS AND OUTLINES OF THIS THESIS

This thesis intends to guide SLTs in clinical decision making related to choosing measurements of feeding and swallowing, especially for mastication in children with CP. From this perspective, the following specific aims are addressed in this thesis:

1. to describe feeding and swallowing problems, including masticatory problems in children, and their impact on daily life in adolescence and young adulthood in individuals with CP;
2. to develop and validate an observation instrument for mastication;
3. to evaluate quantitative instruments for measuring mastication and to establish the contrasts with the observation instrument;
4. to test the feasibility of using different instruments to distinguish differences in mastication between children with CP and typically developing children.

Chapter 2 describes an in-patient intervention program in a heterogeneous group of young children with feeding problems along the ICF classification. The children are divided into three groups: i) children with tube feeding, ii) children with selective food refusal, and iii) children with behavioral food refusal. The results of the intervention are related to the nutritional intake.

Chapter 3 describes the exploration of the perception of feeding and swallowing disabilities in daily life based on interviews with adolescents and young adults with CP.

Chapters 4 and 5 describe the development of the Mastication Observation and Evaluation (MOE) instrument, to detect difficulties managing solid or lumpy foods. Firstly, we establish the oral-motor movements that should be considered, based on the literature and the clinical experience of experts. Secondly, we verify whether these items can be identified in young typically developing children (aged 6 to 24 months) and children with CP (chapter 4). Based on the results of this preliminary study, we determine the psychometric characteristics of the MOE instrument with typically developing children and children with CP. The intra-rater, inter-rater reliability, construct validity, ceiling effect and sensitivity of the MOE are calculated (chapter 5).

In chapter 6, we accept the challenge to develop a method for analyzing dynamic tongue movements using ultrasound during mastication in adults with CP and adults without neurological dysfunction.

Chapter 7 determines the reproducibility of different measurement variables using 3D kinematics and sEMG to evaluate the mastication process of healthy adults and estimate the smallest detectable differences of these variables.

Chapter 8 describes a study to explore the feasibility of the MOE instrument, ultrasound and 3D kinematics to distinguish differences in mastication in typically developing children and in children with spastic CP with respect to different mastication mechanisms. Moreover,
we will compare the results from the MOE instrument with the quantitative measurements.

**Chapter 9** contains the general discussion of the findings and methodical considerations of the above-mentioned studies. Furthermore, a model for decision making in cases of mastication problems in children and recommendations for clinical practice of speech language therapists are presented.
REFERENCES


34. Chen J. Food oral processing; some important underpinning principles of eating and sensory perception. Food Struct 2014; 2: 91-105.


Chapter 1


Multiple case study to describe influencing factors on effectiveness of an interdisciplinary in-patient intervention for feeding problems in children

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R Speyer
PCM Holtus
J van Limbeek
MWG Nijhuis-van der Sanden
Chapter 2

ABSTRACT

Underlying diagnoses and physical, cognitive, and behavioral impairments vary among children with chronic feeding problems. In addition to these variables, we hypothesize that personal and environmental factors contribute to the success of intervention for feeding problems. This exploratory study describes the effectiveness and influencing factors of an intensive, multidisciplinary child and parent-centered intervention on calorie intake and solid food consumption.

The intervention included a behavioral program, oral-motor training, parental coaching, and dietary support. The children participating in the intervention could be separated into three groups, based on characteristics of the food intake: (i) tube-fed (n=12), (ii) selective food refusal by texture (n=6), and (iii) unpredictable food refusal (n=11). For each group, we present a descriptive representative case study. Outcome measures were calorie intake and the amount of solid food consumed. The average duration of the in-patient feeding intervention was 4.3 weeks (SD 1.4 weeks). Three months after discharge, 50% of the children receiving tube feeding had a complete oral intake.

Children with selective food refusal by texture made small progress during the intervention, but the solid food intake had increased at follow-up. Children with unpredictable food refusal increased their oral intake already during the intervention and maintained these gains at home. The intensive interdisciplinary intervention showed increased calorie and oral intake in most children and reduced tube feeding but was less successful in children with metabolic dysfunction. As a group, recovery time was longest in the tube-fed group, but results varied considerably per child. A successful feeding intervention needs to consider a child’s underlying physical and behavioral and environmental factors.
INTRODUCTION

The clinical manifestation of feeding problems in children varies from selective food refusal to dysphagia. With an incidence of 25 to 35 percent, minor feeding problems are common in early childhood for otherwise healthy children; for children with chronic medical problems, however, the incidence is 40 to 80 percent. The impact of feeding problems on a child’s health ranges from mild to considerable and the relationship between food consumption and long-term health outcomes has become increasingly evident. Early feeding experiences are related to dietary preferences in later life and modulate food intake and nutritional status.

Authors suggest an interaction between oral-motor factors, behavioral issues, and environmental factors. A child’s refusal to eat results in an inadequate development of oral-motor skills (e.g. chewing) and this prevents the child from advancing to food textures appropriate to the child’s age. Moreover, inadequate oral-motor abilities cause reduced food intake resulting in a significant proportion of malnourished children with neurodevelopmental disorders. A child’s feeding disorder also has implications on the child’s family. Parents of children with feeding problems report more stress with regard to social isolation and self-perception than parents of healthy children. Meals and meal preparation take considerable time and result in less time for social activities or for the parent to fulfill developmental activities with the child.

Several interdisciplinary interventions for children with feeding problems exist. Although behavioral-based interventions shown positive results in children with developmental disabilities, they have not succeeded in improving oral-motor skills in such a way that all food consistencies could be eaten. Likewise, oral-motor based programs do not address the behavioral aspect of the feeding disorder. Therefore, it seems appropriate to combine the two approaches in a behavioral-based intervention with oral-motor based elements. To do this, a systematic problem analysis should be the basis for designing a tailored intervention plan.

Such a systematic problem analysis should be based on both child specific problems as well as problems in the environmental conditions. The International Classification of Functioning, Disability and Health version Child and Youth (ICF-CY) of the World Health Organization provides a framework to describe feeding difficulties along four dimensions: (1) Body Function/Structures [gastrointestinal conditions, respiratory status and/or neuromuscular conditions]; (2) Activity and Participation [eating, drinking, learning]; (3) Environmental Factors [parental reaction to the child’s food refusal], and (4) Personal Factors [developmental issues, age]. This framework is helpful in both clinical practice and in research to analyze the complexity of feeding problems in children with feeding problems with various etiologies.
The purpose of this multiple-case report is to describe the characteristics of children with feeding problems using the ICF framework and to gain insight into the influencing factors and the effectiveness of the interdisciplinary analysis and in-patient intervention to improve the calorie intake and the solid food consumption.

**METHODS**

**Participants**

Children aged one to six years were eligible for the intervention when (i) feeding difficulties had existed for a period of at least six months and (ii) feeding difficulties were related to physical etiology, and/or were affecting the child’s health status. Children with diagnosed psychiatric problems or severe developmental disabilities or children in palliative care were excluded from this study. We categorized the children into three groups depending on how the parents characterized the feeding problem. Of the 29 children, 41% were categorized as ‘Tube feeding’ (n=12; 9 nasogastric tube and 3 gastrostomy tube), 21% were categorized as ‘Selective food refusal by texture’ (n=6) and 38% were categorized as ‘Unpredictable food refusal’ (n=11). The characteristics of the participants are listed in Table 1.

<table>
<thead>
<tr>
<th>Table 1. Characteristics of the participating children.</th>
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</thead>
<tbody>
<tr>
<td>n (%)</td>
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<tr>
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</tr>
<tr>
<td><strong>n (%)</strong></td>
</tr>
<tr>
<td><strong>Age in years Mean (SD)</strong></td>
</tr>
<tr>
<td><strong>Gender</strong></td>
</tr>
<tr>
<td>Boys (n)</td>
</tr>
<tr>
<td>Girls (n)</td>
</tr>
<tr>
<td><strong>Weight (SD)</strong></td>
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<td>Mean (SD)</td>
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<tr>
<td>Range</td>
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<tr>
<td><strong>Height (SD)</strong></td>
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<td>Mean (SD)</td>
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<tr>
<td>Range</td>
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<tr>
<td><strong>Weight for Height (SD)</strong></td>
</tr>
<tr>
<td>Mean (SD)</td>
</tr>
<tr>
<td>Range</td>
</tr>
</tbody>
</table>

In each subsection of this article we present a representative case study per group.
‘Tube-feeding’
B. was a 1.6 years old girl with multiple respiratory problems, kidney dysfunction, and delayed psychomotor development. At birth, she presented with severe feeding problems and a nasogastric tube was used for feeding. Vomiting occurred at least twice a day. B. refused food by screaming and holding her breath. B.’s parents ceased feeding when B. started crying or vomiting. Because of the dependence on tube feeding the parents were unable to enrol B. in a childcare facility and the mother had to resign from her job, which resulted in financial problems for the family. Anthropometric data: SD of age-appropriate height was -2.9, SD of age-appropriate weight was -2.2, and SD weight for height was -0.3.

‘Selective food refusal by texture’
J. was a 5.5 years old boy with unilateral cerebral palsy and mild developmental problems. He was referred for intervention to optimize his intake of solid and lumpy foods. He took medication for constipation. His parents reported long mealtimes, especially diners, where J. was often angry and went into a rage. Parents reported that family meals were stressful for both them and their three children. J’s parents also reported that they were inconsistent in their use of incentives to encourage J. to eat solid foods.
J. showed hyposensitivity and hypotonicity of his mouth. He could make limited discrete movements with his tongue and movements were asymmetric. When observing him eating bread, he showed minimal chewing and he gagged when swallowing the piece of bread. Chewing a piece of hamburger took a long time and in the end J. removed the meat out of his mouth. Although his daily calorie intake was appropriate for his age, his diet consisted of excessive milk and sugar products and limited fiber. J. had normal anthropometric data.

‘Unpredictable food refusal’
M. was a 2.5 years old girl with no medical problems except multiple middle ear infections during her first year of life. Her food intake was supplemented with a drink and a powder for 50% of her daily intake. Feeding sessions took a large part of the day. M. was the only child in the family. Her parents reported not knowing how to cope with M.’s food refusal and that M. slept twice a day for three hours at a time. In addition to food refusal M. showed limited interaction with peers and had an aversion to being affectionate.
Despite food supplements, M.’s fluid intake was only 40% and her calorie intake was 80% of the recommended amount. M. was pale and thin and had blue circles under her eyes. Anthropometrics data; SD of age-appropriate height was -0.77, SD of age-appropriate weight was -2.7, and SD weight for height was -2.0.
SETTING AND MATERIALS

In this exploratory multiple case study data was obtained from 29 children participating in an in-patient feeding intervention at the rehabilitation center of the Sint Maartenskliniek (Nijmegen, the Netherlands). Therapy sessions lasted 45-60 minutes (inclusive parental instructions) and were held three or four times a day based on the intervention of Clawson et al.\textsuperscript{25} The weekdays feeding intervention varied per child between four to six weeks depending on individual factors (e.g. intervention goal, child’s physical condition, progression in oral intake, parent-child interaction, and personal or environmental factors). The feeding sessions were held in a room by the psychological trainer or speech-language therapist (SLT). Between therapy sessions, the children played with peers or participated in homely activities.

Materials used in the feeding sessions were plastic children’s tableware, regularly available foods and drinks for children, calorie supplements, and interactive toys or internet applications with sound and moving elements.

PROCEDURE

Clinical assessment

After enrolling in the center, parents were requested to keep a diary of their child’s oral food intake for seven days and to make a video recording of a representative mealtime with the family. At the assessment, a pediatrician, psychologist, SLT, and dietician collected data related to each child’s physical and cognitive development, feeding history, current oral-motor skills, and current food intake. This data was obtained by using three questionnaires (Child Behavior Checklist, Caregiver-Teacher Report Form and Sensory Profile-NL), analysis of the diary, and physical examination by the pediatrician and SLT. The Child Behavior Checklist for ages 1½ - 5 years is a questionnaire for parents with 99 items concerning behavioral and emotional problems of their child.\textsuperscript{27} The Caregiver-Teacher Report Form for ages 1½ - 5 years has 99 items concerning behavioral and emotional problems of the child.\textsuperscript{28} The Sensory Profile-NL contains 125 items on processing sensory stimulus in everyday situations.\textsuperscript{29} Anthropometric data (weight and height) were also collected and compared with the growth curves for diagnose, gender, and cultural background.

Parents were also interviewed and asked about their thoughts and expectations about the intervention. The mealtime recording was analyzed by the psychologist in terms of the child’s feeding-related behavior and family members’ reactions to the child’s behavior. Each child’s assessment was distributed over a single day. After data analysis, the team met and used the ICF framework to determine the body functions, activities, and environmental and personal factors. This information was used to formulate a hypothesis on the reasons for the feeding problem and the factors continuing the feeding problem. An overview of this analysis in terms of the ICF framework is illustrated in Table 2.
Multiple case study to describe influencing factors on effectiveness of an interdisciplinary in-patient intervention

Table 2. Overview of signs and symptoms classified conform the International Classification of Functioning, Disability and Health (ICF) per feeding group.

<table>
<thead>
<tr>
<th>Description</th>
<th>‘Tube feeding’</th>
<th>‘Food refusal by texture’</th>
<th>‘Unpredictable food refusal’</th>
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</tr>
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<tr>
<td><strong>Body function</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>b117/b147 Intellectual functions/ psychomotor function</td>
<td>4/12</td>
<td>2/6</td>
<td>1/11</td>
<td>7/29</td>
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<tr>
<td>b122 Global psychosocial function</td>
<td>1/12</td>
<td>2/6</td>
<td>3/11</td>
<td>6/29</td>
</tr>
<tr>
<td>b125 Dispositions and intra-personal functions</td>
<td>4/12</td>
<td>1/6</td>
<td>9/11</td>
<td>14/29</td>
</tr>
<tr>
<td>b126 Temperament and personality functions</td>
<td>6/12</td>
<td>3/6</td>
<td>4/11</td>
<td>13/29</td>
</tr>
<tr>
<td>b130 Energy and drives functions</td>
<td>6/12</td>
<td>1/6</td>
<td>1/11</td>
<td>8/29</td>
</tr>
<tr>
<td>b134 Sleep functions</td>
<td>1/12</td>
<td></td>
<td>6/11</td>
<td>7/29</td>
</tr>
<tr>
<td>b140 Attention functions</td>
<td>1/12</td>
<td>3/6</td>
<td>2/11</td>
<td>6/29</td>
</tr>
<tr>
<td>b152 Emotional functions</td>
<td>5/12</td>
<td>3/6</td>
<td>7/11</td>
<td>15/29</td>
</tr>
<tr>
<td>b250/b270 Taste/Sensory function related to temperature and other stimuli</td>
<td>7/12</td>
<td>1/6</td>
<td>9/11</td>
<td>17/29</td>
</tr>
<tr>
<td>b280 Pain</td>
<td>3/12</td>
<td>1/11</td>
<td>4/29</td>
<td></td>
</tr>
<tr>
<td>b435 Immunological system function</td>
<td>5/12</td>
<td>4/11</td>
<td>9/29</td>
<td></td>
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<tr>
<td>b510 Ingestion functions</td>
<td>6/12</td>
<td>6/6</td>
<td>7/11</td>
<td>19/29</td>
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<tr>
<td>b515 Digestive functions</td>
<td>4/12</td>
<td>3/11</td>
<td>7/29</td>
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<tr>
<td>b525 Defecation</td>
<td>3/12</td>
<td>3/6</td>
<td>4/11</td>
<td>10/29</td>
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<tr>
<td>b735 Muscle tone function</td>
<td>1/12</td>
<td>1/6</td>
<td>2/29</td>
<td></td>
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<tr>
<td>b760 Control of voluntary movement functions</td>
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<td>3/6</td>
<td>5/29</td>
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<tr>
<td>b761 Spontaneous movement functions</td>
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<td><strong>Body structures</strong></td>
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<tr>
<td>s250 Structure of middle ear</td>
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<td></td>
<td></td>
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<tr>
<td>s330 Structure of pharynx</td>
<td>1/12</td>
<td>2/6</td>
<td>4/11</td>
<td>7/29</td>
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<tr>
<td><strong>Activities/participation</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>d250 Handling psychological demands</td>
<td>2/12</td>
<td>1/6</td>
<td>7/11</td>
<td>10/29</td>
</tr>
<tr>
<td>d330 Speaking</td>
<td>3/12</td>
<td>2/6</td>
<td>1/11</td>
<td>6/29</td>
</tr>
<tr>
<td>d550 Eating</td>
<td>7/12</td>
<td>4/6</td>
<td>7/11</td>
<td>18/29</td>
</tr>
<tr>
<td>d560 Drinking</td>
<td>4/12</td>
<td>1/6</td>
<td>1/11</td>
<td>6/29</td>
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<tr>
<td>d710 Basic interpersonal interactions</td>
<td>1/12</td>
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### Environmental factors

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<th>‘Food refusal by texture’</th>
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<tbody>
<tr>
<td>Products or substances for personal consumption</td>
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<td>5/11</td>
<td>8/29</td>
<td></td>
</tr>
<tr>
<td>Immediate family</td>
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<td>1/6</td>
<td>8/11</td>
<td>12/29</td>
</tr>
<tr>
<td>Individual attitudes of immediate family members</td>
<td>8/12</td>
<td>2/6</td>
<td>10/11</td>
<td>20/29</td>
</tr>
<tr>
<td>Health services, systems and policies</td>
<td>12/12</td>
<td>2/6</td>
<td>1/11</td>
<td>15/29</td>
</tr>
</tbody>
</table>

### Personal factors

- age/ negative experiences/ not feeling well /refusal behavior  

<table>
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<tr>
<th>Description</th>
<th>‘Tube feeding’</th>
<th>‘Food refusal by texture’</th>
<th>‘Unpredictable food refusal’</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>age/ negative experiences/ not feeling well /refusal behavior</td>
<td>9/12</td>
<td>1/6</td>
<td>10/11</td>
<td>20/29</td>
</tr>
</tbody>
</table>

### ‘Tube feeding’

Team’s conclusion: B. was a girl with a complicated medical history resulting in tube feeding but at presentation was in a healthy state. She was small and thin but had a sufficient height and weight for her age. The girl had missed the critical period for oral feeding\(^{10,30,31}\) and lacked the ability to eat a variety of tastes and consistencies. B. was used to getting attention from her family in periods of distress. B.’s parents were anxious about giving her food because of previous vomiting and breath holding.

The goal of the 4-week behavioral intervention therapy was to reduce B.’s food refusal behavior and reduce the amount of tube feeding.

### ‘Selective food refusal by texture’

Team’s conclusion: J. was a boy with partial food refusal caused by oral-motor problems related to neurological disease and inconsistent use of strategies by J.’s parents to encourage J. to eat.

The goals of the 4-week oral-motor therapy were to teach J. to chew on soft and hard foods, to structure the mealtimes by using behavioral techniques and to optimize J.’s intestines by changing the quality of his food intake.

### ‘Unpredictable food refusal’

Team’s conclusion: M. was a malnourished girl with no energy level. M. had no impairments of body functions. The strong-willed girl had learned to avoid food and drinks and her parents were unable to cope with her behavior during feeding.

The goal of the 4-week therapy was to improve M.’s nutritional status using a maximum of food supplements and to reduce M.’s food avoidance by using the behavioral intervention.
Multiple case study to describe influencing factors on effectiveness of an interdisciplinary in-patient intervention

Feeding intervention

The intervention consisted of a behavioral program given by the psychologist, an oral-motor program given by the SLT, or a combination of both programs given by both the psychologist and the SLT. The behavioral program was based on various studies[^13][^15][^24] and included positive reinforcement, tangible rewards (non-contingent access to preferred items), extinction of inappropriate behaviors, and the use of shaping and fading techniques. During the behavioral therapy, the child sits on the therapist’s lap and is given a small amount of food just below the child’s current acceptance level. When the child accepts this given amount of food over three consecutive sessions, the amount is increased by 25 grams (if acceptance level was below 100 grams) or 50 grams (if acceptance level was above 100 grams). The child received a tangible reward (e.g. music toy) and/or verbal reward (‘well done’) after every bite in a standardized order.

The focus of the oral-motor program was to practice the oral skills of processing different food consistencies. The child receives standardized verbal instructions with visual and tactile support during the session. Regular contact was maintained between the psychologist, SLT, and the dietician to ensure each child received optimal and adequate nutrition. Weekly meetings were held with the intervention team and the parents to discuss progress and child nutrition. Parents were taught feeding techniques and strategies on how to manage their child’s food refusal behavior.

Outcome measures

Food intake was derived from the 7-day feeding diary. Average food intake was compared with the recommended amount of calories and fluid based on child gender, age, weight, height, activity level, disease factor, and catch-up growth and this was calculated by means of the Schofield formula[^32]. Data on calorie intake (kcal) was computed at three measurement moments: the week leading up to the assessment day ($t_1$), the week prior to discharge ($t_2$) and three months after discharge ($t_3$). In addition to calorie intake, we also collected group-specific data. For the ‘Tube feeding’ group, we separated calorie consumption into oral intake and tube feeding; for the ‘Unpredictable food refusal’ group, we separated calorie consumption into normal oral intake and supplements; and for ‘Selective food refusal by texture’ group, we separated calorie consumption into the amount of solid, puree and liquid intake (kcal).

Data analysis

Descriptive statistics were used to describe characteristics of the participating children and to describe the outcome measures of the total group and subgroups. To allow comparison
between evaluation moments and between participants, we transformed the change in calorie intake per group into z-scores. All statistics were performed using SPSS (version 17.1).

RESULTS

Treatment frequency and duration
The mean duration of the intervention was 4.3 weeks (60.7 therapy sessions). The ‘Tube feeding’ group received the most therapy sessions (mean 66.9 sessions) and the ‘Selective food refusal by texture’ group had the fewest number sessions (mean 51.0 sessions). Frequency data for the ‘Unpredictable food refusal’ group are skewed as two children had an extended stay in the in-patient intervention due to family factors (inappropriate home environment and parental psychiatric problems). The parental coaching sessions are included in the count but sessions with the dietician are not included.

Tube feeding
Before the intervention, 12 children required tube feeding for 20 to 100% of their dietary needs. At discharge two of these children were no longer receiving tube feeding and at follow-up another four children had ceased receiving tube feeding. The remaining five children (follow-up data of one child is missing) received (partial) tube feeding at follow-up (range= 30 - 90%). Two of them decreased tube feeding by 50 and 75% compared to pre-intervention data. Both these two children had co-morbid medical problems (cardiac defect; multiple congenital malformations of the skeleton, muscles, heart, and kidneys). The maximum duration of the feeding sessions was 30 minutes and these two children were often too fatigued to eat the necessary amount. The oral meals were completed with tube feeding to prevent stressful feeding sessions and malnutrition. Three children were not successful in decreasing the tube feeding; they had metabolic dysfunctions and gastrointestinal problems with episodes of extreme vomiting. Figure 1 presents the evaluation of the oral feeding.

Figure 1. Food intake in percentage of the recommended amount of calories based on gender, age, weight for height, activity level, disease factor, and catch-up growth of each child at t₁ (n=12), t₂ (n=12) and t₃ (n=11) for the ‘Tube feeding’ group.
Multiple case study to describe influencing factors on effectiveness of an interdisciplinary in-patient intervention

**Calorie intake**

Figure 2 presents the information on calorie intake for children with unpredictable food refusal for the period from \( t_1 \) to \( t_3 \). In addition to an insufficient calorie intake, some children had a one-sided and incomplete menu at \( t_1 \). Three months after discharge almost all children had a sufficient calorie intake. However, in eight cases supplemental calories in powder form were added to food or drinks. The children with selective food refusal by texture generally had a sufficient calorie intake and they showed no significant change in calorie-intake.

![Figure 2. Food intake in percentage of the recommended amount of calories based on gender, age, weight for height, activity level, disease factor, and catch-up growth of each child at \( t_1 \) (n=11), \( t_2 \) (n=11) and \( t_3 \) (n=10) for the ‘Unpredictable food refusal’ group.](image)

The ‘Tube feeding’ group had the largest improvement in calorie intake during the treatment \((z\text{-score } \Delta t_1-t_2 = 2.3; \Delta t_2-t_3 = 1.1)\). The ‘Unpredictable food refusal’ group increased their calorie intake after the treatment more than during the treatment \((z\text{-score } \Delta t_1-t_2 = 0.7; \Delta t_2-t_3 = 1.3)\). Results at follow-up suggest that parents of both groups were generally able to maintain and even increase the calorie intake at home.

**Age appropriate oral intake**

Children with selective food refusal by texture could be divided into those who did not accept solid food \((n=4)\) and those who ate solid and lumpy food but had poor chewing skills due to neurological problems (cerebral palsy, prematurity; \( n=2)\). The therapy for all children in this group was focused on developing chewing technique rather than on increasing the amount of solid food consumed. As a result, the amount of solid food intake during treatment for this group decreased but increased after discharge. At follow-up, the amount of solid food being eaten by the 6 children was more than double the pre-intervention amount (from 115 to 310 grams per day).
ICF framework

We shared the children into three groups based on their specific food intake characteristics. Using the ICF classification we noticed differences between the three groups and their environment. The children with tube feeding had physical problems and as result of multiple medical treatments they were anxious about eating. Two of the children in this group were not able to enroll at childcare because of the tube feeding and/or vomiting. The children with food refusal by texture showed more motor and attention problems and they had difficulties with defecation whereas children in the other groups showed more vomiting. The children with unpredictable food refusal had difficulties with psychosocial function, dispositions and accommodation and sleeping. Approximately 30% of these children had multiple ear inflammations, enlarged tonsils or food intolerance. Most parents of children in this group were unable to give clear structure and instructions or provide positive rewards for eating. This resulted in children not participating in family mealtimes and the child having his or her own ritual for eating. In three cases, we were concerned about the child’s social emotional development and we referred the child for further assessment.

‘Tube feeding’

B. received 78 sessions during 4.2 weeks of intervention. At t₁, B. was 100% dependent on tube feeding and received 800 ml per day via tube feeding, which meant a lack of 100 kcal. B.’s first feeding therapy sessions started with 10 grams of custard and fruit puree. After three successful sessions, the amount was doubled. Her meals were extended with pureed vegetables. After two weeks of the intervention, B.’s parents became involved in the training. The parents were taught how to use firm instructions and how to cope with B.’s refusal behavior. During the weekdays parents were coached in the feeding sessions. In the weekend’s, they made video recordings to discuss with the psychologist.

At the end of the intervention, the tube feeding was reduced to 25% and she ate 5 homogenized meals a day. The mealtimes took a maximum of 30 minutes. Three weeks after discharge the feeding tube was permanently removed. B. still required calorie supplements. At follow up, the mealtimes were reasonable in terms of time demands and child behavior, but drinking and non-homogenized consistencies caused substantial refusal behavior. A three-month outpatient intervention with therapy sessions once a week with the SLT resulted in adequate calorie and fluid intake in mealtimes.
‘Selective food refusal by texture’
J. received 60 sessions (30 with the SLT; 30 with the psychologist) over a period of 4 weeks. At t1, J.’s calorie intake was sufficient but the amount of fluids consumed was 200 ml below the daily recommendation.
Starting with crunchy crackers and using visual and auditory feedback, the SLT taught J. how to chew. He then learned how to bite off pieces of food, move and extend his tongue laterally and to form a bolus and control a swallow. Due to J.’s oral hyposensitivity, we adapted the foods (e.g. baked instead of boiled potatoes and easy to chew meat). He was rewarded when he finished a meal within 20 minutes. J.’s parents understood the relationship between cerebral palsy and oral-motor problems and they accepted the need for J. to have easy-to-chew food. After 4 weeks of intervention, J. ate a cracker before bread and succeeded in finishing his meal within a given time. His consumption of liquids was adequate and the digestive medication had ceased being given.
Three months after discharge J.’s parents reported that mealtimes were now cozy and were no longer stressful.

‘Unpredictable food refusal’
M. received 76 sessions over a period of 4 weeks. At first, she showed extreme food refusal behavior and tangible rewards were not an effective strategy. M. started with preferred food during three therapy sessions a day. Therapy focused on consuming calorie enriched juices. M. learned to eat custard and puree and to drink 700 ml a day divided over 6 portions. M. often appeared unhappy during the sessions. M. showed limited affection towards her parents. During the inpatient intervention, we discussed our observations with the parents. At follow-up M.’s physical condition had improved but mealtimes remained stressful. Parents agreed on referring M. to the psychiatrist for further investigation.

DISCUSSION AND CONCLUSION
In this study, we described a 4-6-week in-patient multidisciplinary intervention including a behavioral program based on theories of operant conditioning combined with oral-motor training, parental coaching, and dietary support to be effective for children with feeding problems. For children with severe metabolic disorders the intervention had limited success in increasing oral food intake, however parents reported that their child vomited less and that parents had more insight into their child’s oral intake pattern.

The average length of 4-6-week intervention in our study was shorter than in other studies; however, the number of therapy sessions required to reduce tube feeding is in line with these studies.12,22,24 Parents were unanimously positive about the duration of the intervention, possibly because of the high frequency of intervention sessions. Studies report
positive results when parents are given a more prominent role in the intervention\textsuperscript{25,33}, and this fits with our findings. We did not systematically analyze changes in levels of parental stress, but we hypothesize that providing parental training to transfer intervention elements to the home setting was a key factor for the success of our intervention. The in-patient setting enabled us to observe characteristics of both children and parents in addition to the feeding sessions. Parents were taught how to offer their child food, how to reward their child for food acceptance and how to ignore their child’s refusal behavior. By changing the parents’ perception and interpretation of their child’s behavior, parents gained insight into their child’s temperament and how to cope with the conflicts during feeding and during sleeping. Therefore, the primary goal for children with tube feeding was not eliminating the tube feeding as fast as possible, but rather focused on the capacity of parents to handle the refusal behavior of their child. Success was not limited to type or presence of chronic illness but also to the personal factors of the children, that is, the child’s character and coping style as illustrated in the three cases.

Of the 29 children in the present study, 26 improved their qualitative as well as their quantitative food intake. The dietician-designed individual menus (including food supplements) meant parents had clear recommendation on their child’s food intake and this resulted in reduced parental stress. In our study, 67% of the children receiving tube feeding at admittance to the intervention had reduced the amount of tube feeding by the end of the intervention. Based on our findings, it seems that a total reduction of tube feeding in such a heterogeneous group is not to be expected especially in cases with complicated medical issues, such as in metabolic illness.

Although we did not quantify the degree of improvement in oral-motor skills in children with ‘Selective food refusal by texture’, we found that the average amount of solid food consumed almost trebled for the group at follow-up. The subgroup of children with neurological impairments was able to improve chewing technique, which led to a safer handling of solid and lumpy foods. This is in line with the results published by Clawson et al.\textsuperscript{25} Parents learned to stimulate good chewing and to adapt the meals to the oral-motor abilities of their child.

Definite conclusions on the success of the intervention need to be postponed. We tried to handle the heterogeneity of the group in this study by defining subgroups based on the feeding problem instead of based on the diagnosis, which seemed appropriate as the ratio of behavioral and oral-motor program components was linked to the goal of the intervention. This, however, was not independent of the subgroup profile. This study offers insight into the barriers and strengths of the designed intervention and provides a solid basis of information for a future study with a larger cohort.
ACKNOWLEDGEMENTS

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REFERENCES


chapter 3

Experiences with eating and drinking among adolescents and young adults with cerebral palsy: A qualitative study

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Clinical Rehabilitation submitted
ABSTRACT

The impact of difficulties with eating and drinking in adolescents and young adults with cerebral palsy (CP) is unknown. The purpose of this study is to gain insight into experiences with these activities and perceived disabilities among this group in the daily practice. Ten participants with spastic CP (aged 15 - 23 years) participated in individual interviews. Meaningful segments from each interview were selected and then organized into clusters and synthesized to specific themes.

All participants reported experiencing eating and drinking problems (e.g., choking or not processing all food textures). Four main themes were extracted from the interviews: (i) feelings (e.g., shame, frustration, fear, and distress); (ii) coping strategies (e.g., adaptation or food avoidance); (iii) the influence of the social and physical context (e.g., the accessibility of restaurants or assistance); and (iv) concerns about the future. One striking finding was that all but one of the participants had not recently received either monitoring or intervention for feeding skills.

This study shows that adolescents and young adults with CP experience many restrictions in eating and drinking situations, leading to negative feelings and lower participation levels, while little attention is directed towards these problems. Regular multidisciplinary rehabilitation programs should include evaluation, advice, and intervention with regard to eating and drinking ability in order to increase participation. Such programs should draw on the latest insights of research and technological developments, and they should involve socially acceptable and age-appropriate food adaptation. Moreover, timely intervention could prevent choking and nutritional deficiency.
INTRODUCTION

Cerebral palsy (hereinafter CP) is one of the most common disabling motor disorders of childhood. In general, CP is regarded as a stable condition that starts in childhood and persists through adulthood. In the Netherlands 1.51 of every 1000 inhabitants are diagnosed with CP. Most individuals with CP are diagnosed with spasticity, muscle weakness and/or coordination problems, and levels of functioning range from mild effects to profound deficits in all domains.

Between 50% and 80% of all individuals with CP experience oral-motor disabilities, which result in problems with feeding and swallowing. Such problems are defined as disorders of eating and drinking activities (dysphagia), with feeding being associated with activities, such as sucking, spoon feeding, chewing, or drinking from a cup. Individuals with feeding and swallowing problems are at risk for nutrition-related health implications, including lung problems, limited caloric intake, malnutrition, and poor growth. Furthermore, eating and drinking problems influence a person’s dignity, self-esteem, and the quality of mealtime experiences.

Although the brain injury that initially causes CP is not progressive, the effects of CP manifest differently throughout the life span due to physical growth, as well as to cognitive and social-emotional development. Moreover, several factors could decrease physical activities, including increased spasticity and/or contractures, and deterioration of condition and muscle strength. In one study, Krakovsky and colleagues observed a decrease in eating ability among 30 participants with CP (age range 11 - 30 years). This decrease was due to difficulty in handling oral secretions, and it resulted in reduced social contacts. Other researchers have also reported a gradual worsening in swallowing and mealtime capabilities among adults with CP from as early as 30 years of age. In contrast, a literature survey reveals no systematic intervention studies on the maintenance or improvement of eating and drinking abilities in adults with CP.

Life satisfaction is described in terms of subjective well-being – the ways in which individuals evaluate their lives emotionally and cognitively – which can be influenced by personality dispositions, health, affections, adaptation, and possibilities. In this light, social contacts during mealtimes in various environmental settings (e.g., with family or friends, in known or unknown places) could thus also influence feelings of well-being. Although studies have demonstrated that young people with CP (aged 8 - 18 years) are capable to consider their perceptions on activities of daily living and emotional and social functioning, most studies concerning perceptions and views are based on those of parents or adults with CP. In a survey conducted by Sleigh, parents reported that their children with CP (aged 2.5 - 15 years) experienced limited gross and fine motor ability and problems with feeding and nutrition activities. Parents (mostly mothers) expressed anxiety with regard to ensuring that their children would receive a well-balanced diet in small volumes. In addition, they
mentioned feelings of concern, frustration, guilt, and despair related to mealtimes.\textsuperscript{22,23} In another study, adults with CP (n=279) reported gradually increasing difficulties with eating and drinking, as well as an increase in the avoidance of foods.\textsuperscript{13} They also reported reduced social interaction during meals and persistent dependence on others for eating and drinking, along with emotional responses to these changes and difficulties in their relationships with their families at mealtimes.\textsuperscript{13}

To our knowledge, no studies have explored the experiences and perceptions of adolescents and young adults with CP regarding their eating and drinking abilities. We also do not know whether and how the age of young individuals with CP influences their perceptions of these activities. The purpose of this study is to generate insight into the types of perceived disabilities, and experiences with eating and drinking of adolescents and young adults with CP. The study concludes by discussing the implications of the findings for the quality of life of individuals with CP and by presenting recommendations for care.

\section*{METHOD}

\subsection*{Participants}

The participants in this study were 10 individuals with CP (4 male, 6 female), with a mean age of 18.5 years (range 15 - 23 years). Inclusion criteria were as follows: diagnosis of spastic CP, at least 50% oral feeding, and the ability to communicate with strangers about eating and drinking ability and personal concerns. The nature and frequency of the feeding and swallowing problems of the participants varied among the members of the group (Table 1). The participants were recruited through a social media group for adolescents with CP, organized by a national organization for individuals with congenital physical impairments (BOSK).

\subsection*{Interview guide development}

We developed a semi-structured interview guide on the topic of this study. First, a set of preliminary questions was developed, based on aspects involved in eating and drinking. The interview guide was then tested in a pilot interview with a 26-old man with quadriplegic CP and moderate oral problems. His input led to further refinement of the questions and additional questions. The final interview guide was structured in four parts concerning general personal information, gross and fine motor abilities, eating and drinking abilities, and perceptions regarding mealtimes in social life. All interviews started with the following request: “Tell me something about yourself.” After a while, the interview turned to the question, “Could you rate your eating and drinking skills on a scale from 1 to 10?” Finally, personal values and wishes were asked.
Method
Ethical approval was obtained from the Ethics Committee at Slotervaart Hospital and Reade Rehabilitation Center in Amsterdam (Netherlands) (study number P1529). Each participant took part in an individual semi-structured interview with the lead researcher (LR), in the presence of two students of speech-language therapy (SLT). Prior to the interview, all participants signed a written informed consent, as did the parents of participants younger than 18 years. All participants (and/or parents) agreed to the use of anonymized personal statements for research presentations and publications.

The face-to-face interviews were held in an environment that was familiar to the participants, and each lasted 30-45 minutes. The conversations were kept as natural as possible and adapted to the conversational level of the participant. The order of questions depended on the participant’s responses. In three interviews, the participant’s mother was also present to support the child’s communication, if necessary, in case of severe dysarthria.

All interviews were recorded with a digital voice recorder and transcribed in Word 2013 (using Windows media player) by one SLT, who was present at the interview. All transcriptions were reviewed by another SLT student, who was not present at the interview. To improve accuracy and completeness, the interview transcriptions were sent to the participants for a member check, possibly with parental assistance. The interviews were conducted and analyzed in Dutch. All quotations in this article were translated by a native speaker to ensure that they accurately reflect the intentions of the interviewees.

Design
Interviewing is regarded as a valuable qualitative method for learning about expressions, perceptions, and contexts. Such qualitative studies are not intended to establish frequencies, but to determine the diversity of particular topics of interest within a given population, thereby identifying meaningful variations within that population. Theoretical saturation is the point at which no new insights are obtained, no new themes are identified, and no issues arise regarding a category of data.

Data collection and analysis
After the transcription, all interviews were analyzed with the ATLAS-ti software program (version 7.0.73). Following an initial reading of the interview transcript, the lead researcher (LR) and the three SLT students independently highlighted and coded each relevant phrase (meaningful segment). Finally, inductive analysis was used to organize meaningful segments with similar meanings into clusters, which were then synthesized to particular themes. In the next phase, the project team, consisted of the lead researcher (LR) and two senior researchers who had not been involved in the data collection and coding procedures (LE, MN), used the themes to create an affinity diagram. During two meetings, each team
member independently assembled codes into categories and overall themes. The final diagram was discussed until consensus was achieved and no new categories or themes were generated. The results are presented as narrative descriptions.

RESULTS

All participants affirmed that the interview transcripts accurately reflected their physical situations, views, feelings, and experiences. The characteristics of the 10 participants are presented in Table 1.

Table 1: Overview of the participants.

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Age in years</th>
<th>Type and severity of CP in GMFCS*</th>
<th>Daily activity and living situation</th>
<th>Parental support</th>
<th>Rating of eating and drinking skills</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>F</td>
<td>20</td>
<td>GMFCS V Quadriplegic</td>
<td>Employed, living with parents</td>
<td>No</td>
<td>7</td>
</tr>
<tr>
<td>2</td>
<td>F</td>
<td>22</td>
<td>GMFCS IV Quadriplegic</td>
<td>Student; assisted living</td>
<td>No</td>
<td>8</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>22</td>
<td>GMFCS II Hemiplegic right</td>
<td>Student; living with parents</td>
<td>No</td>
<td>7</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>15</td>
<td>GMFCS V Quadriplegic</td>
<td>Special education; living with parents</td>
<td>Yes</td>
<td>8</td>
</tr>
<tr>
<td>5</td>
<td>M</td>
<td>23</td>
<td>GMFCS V Quadriplegic</td>
<td>Sheltered employment setting</td>
<td>No</td>
<td>6</td>
</tr>
<tr>
<td>6</td>
<td>F</td>
<td>16</td>
<td>GMFCS I Hemiplegic social-emotional problems</td>
<td>Student; living in a care center</td>
<td>Yes</td>
<td>-</td>
</tr>
<tr>
<td>7</td>
<td>M</td>
<td>15</td>
<td>GMFCS IV Quadriplegic</td>
<td>Student; living with parents</td>
<td>Yes</td>
<td>-</td>
</tr>
<tr>
<td>8</td>
<td>M</td>
<td>16</td>
<td>GMFCS III Hemiplegic left</td>
<td>Special education; living with parents</td>
<td>No</td>
<td>9</td>
</tr>
<tr>
<td>9</td>
<td>F</td>
<td>19</td>
<td>GMFCS II Quadriplegic</td>
<td>Social work setting</td>
<td>No</td>
<td>8</td>
</tr>
<tr>
<td>10</td>
<td>M</td>
<td>17</td>
<td>GMFCS II Hemiplegic left</td>
<td>Student; living with parents</td>
<td>No</td>
<td>8</td>
</tr>
</tbody>
</table>

*GMFCS = Gross Motor Function Classification System

Five quadriplegic participants were fully dependent on electric wheelchairs. All others were able to walk or used wheelchairs only to bridge long distances. In general, the participants consumed a variety of products for meals. In the morning, they had hot cereal \((n=4)\) or bread \((n=6)\), sandwiches at lunch time \((n=10)\), and a varied supper in the evening \((n=10)\). All participants reported consuming a variety of foods, with the exception of foods that are difficult to process foods, such as steak, salads, chunky vegetable soup, hard candy. Two participants received high-energy powder or drinks each day to complete their nutritional needs. Only one of the participants had recently had an intervention to improve mastication. None of the others had received any monitoring or intervention for their
eating and drinking abilities for years.

Eight of the 10 participants rated their eating and drinking ability on a 10-point scale. Two young participants (aged 16 and 17 years) were unable to rate their feeding ability. The ratings of the other participants varied between 6 and 9. The older participants described both internal and external influences on their eating and drinking ability (e.g., adaptations to foods), whereas the younger participants mentioned only external influences (e.g., assistance).

The meaningful narrative quotes could be synthesized into four themes, additional to the perceived eating and drinking problems. The eating and drinking problems were interacted with feelings, coping strategies, context variables, and concerns about the future. The manner in and the intensity with which the participants experienced these themes differed, as did the way in which they expressed. We extracted some analogue issues per theme. A graphic overview of the themes and mentioned issues is presented in Figure 1.

![Figure 1](image)

**Figure 1.** Overview of the interacting themes and mentioned issues related to the perceived eating and drinking disabilities of adolescents and young adults with cerebral palsy.

We followed the International Classification of Functioning Disability and Health (ICF) in describing the eating and drinking problems mentioned in the interviews according to body functions/structures, activities, participation, and personal and environmental factors. These topics are discussed below, with quotations from interviews provided to support the stated interpretations. The bracketed numbers refer to the numbers assigned to the participants.

**Eating and drinking problems**

Although all participants were complete oral feeders, they expressed a variety of eating and drinking problems. With regard to functions, most participants mentioned at problems with swallowing and mastication, which were influenced by food textures. One man reported a problem with the force required to swallow when eating solid food: “When
swallowing, I really have to work at it. It’s quite hard for me to get it down.” [10]. Regarding sensibility, a woman with hemiplegic CP observed the following: “Well, I don’t have any feeling on my left side. No, so if a piece of pasta is there, I can’t feel it.” [6]. The following quotation illustrates choking events: “Choking; when it happens, and that’s at least twice a week... And it sometimes happens if a piece is a bit too big.” [4].

As described by the participants, these impairments in functions led to consequences for activities. Six of the interviewees mentioned restrictions in the time available to them for consuming meals: “It’s just that, yeah, when we have lunch at work, almost everyone finishes before me.” [1]; “I sometimes think, ‘Oh, I feel like an apple’, but it takes so long to eat.” [3]. All participants mentioned having problems in processing various food textures. As described by one participant, “Cake, but sometimes other things as well, like white bread or wraps; they get stuck on the roof of my mouth.” [9]. Half of the participants experienced difficulties with eating hard or lumpy foods. As recounted by one man, “...really hard things, like hard candy or stuff like that. I just can’t get it down.” [5]. Three participants mentioned problems with eating meat, “But even if someone else says it’s not tough, it seems tough to me, because I keep on chewing it until it’s a little ball. And I can’t get it down.” [9]. Two participants described problems with carbonated drinks: “For example, I’ve always had trouble because it’s really hard for me to drink carbonated drinks. It can take me an hour to finish one glass because I can’t swallow it.” [1].

Five participants described problems with handling utensils, peeling or cutting, using a glass, or eating food by hand. As observed by one man, “I can’t hold a fork properly in my left hand... When asked: “Do you have trouble, say, bringing food to your mouth?”, he answered: “That is hard. Either I do it with my right hand, or it’s really hard.” [8].

Three participants mentioned that their eating and drinking abilities had decreased over time. On the question: “And now you can see it’s getting a bit worse in all respects? Motor skills, fine motor skills, with eating and drinking.”, a woman noted: “...Yeah, slowly. But we never know whether it’s getting worse because of stopping the therapy or whether it’s just coincidental.” [1]. One participant recounted, “Grapes are okay, and I used to eat tangerines and then it got harder, but now I don’t eat them anymore.” [9].

Two participants also mentioned problems with starting up in the morning: “In the morning, then yes, my muscles are really stiff, but I’m really weak at the same time. So yeah, then I can hardly eat at all. So, I eat yogurt or custard, but even that can be a problem if it has chunks in it. My mouth is so sensitive that I can barely get it down.” [1]. Another woman reported a nearly identical problem: “In the morning, I eat hot cereal. At that time, I’m a bit stiff...I can chew, but it takes a long time.” [4].

The four themes that emerged regarding the eating and drinking problems (feelings, coping, context, and concerns) are depicted in Figure 1 and described in the following sections.
Feelings
All the participants exhibited a wide range of emotions during the interviews, both verbally and non-verbally, including shame, frustration, fear, and distress. One woman mentioned feelings of shame: “Sometimes, my mother yells, ‘You’ve still got food lying there!’ That makes me feel uncomfortable, because everyone looks at me, wondering what’s going on.” [3]. One man noted, “And if you really have trouble swallowing, everyone sees you sitting there half spastic and stretching [imitating a choking event].” [5]. Some participants also mentioned feelings of shame about the necessary adaptations. When the interviewer asked: “Do you also take that cup and straw to school?”, one participant answered: “I just take that aqua-blue stuff. It’s one of those sugary drinks. But at any rate, the bottle has a sport cap, so it’s less obvious.” [9].

Most of the participants mentioned feelings of frustration: “The break was already short, and now it’s even shorter. I was really annoyed about that. Now I can only eat, and there’s no time left for fun things.” [4]. When asked if she could eat an apple, one woman replied: “Yes, but – I don’t know why – I often eat it in pieces, because I have the feeling it would take forever if I were to just start biting into it. Because of that, I don’t eat them as often. Sometimes I think, ‘Oh, never mind.’” [1].

Fear was mentioned by six participants, each time in relation to choking. The mother of one participant recounted the following: “She’s usually able to cough it up on her own. And sometimes, we really have to…, we don’t really panic, but we think, ‘Oh, no. You really can’t get it…, it’s stuck.’” [4]. One man noted, “Because of that, eating alone, really being alone to eat, that can be scary as well.” [5].

The eating and drinking problems reported by the participants also led to feelings of distress. As described by one woman, “When I was still at school, we would often have whole days without a break. On those days, my classmate would grab a quick bite, but I couldn’t do that. No, I wasn’t able to do that, so I just didn’t eat. Then I would get home around four o’clock, and I hadn’t eaten anything all day.” [1]. Another woman recounted: “Some days, I don’t eat and drink enough. And sometimes, I eat only half a meal in the evening. I’m still hungry, but it’s just too much trouble.” [2]. One young man expressed sadness because he was unable to participate in social situations with his peers: “And last week, xxx went out to eat with friends. Yeah, I’d like to do that too.” [7]. Stated even more strongly, “But there are also times I think, ‘I’m so glad it [the mealtime] is over.’” [5].

Coping
Defined as “constantly changing cognitive and behavioral efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person,” coping strategies are completely dependent on how successfully individuals cope with their disabilities. The most commonly mentioned coping strategies were avoidance
or adaptation. One woman reported that she avoided her favorite foods due to chewing problems: “For example, I really like bacon, but I only eat it at home, not around others.” [1]. Some participants reported that fear of choking also led them to avoid certain foods; “My class went out to a restaurant with all-you-can-eat spareribs. And spareribs are one of my favorite things to eat. But I thought, ‘I’d better not risk it. I’ll just have spaghetti.’” [9].

Some participants mentioned adaptation at mealtimes. For example: “You’ve adapted how you eat bread. Right, because I don’t put anything on my bread. I do eat cheese, cucumbers, or other things, but on the side...instead of on the bread. It’s easier that way...” [1]. One man noted, “…over the years, you do learn tricks; you teach yourself tricks for doing things on your own, doing things differently, or asking for help sooner.” [5].

Other coping strategies included acceptance, which led some participants to keep on trying or to accept help. As related by one woman; “…that I really can’t do much about it right now. Except, with drinking as well, trying to take a few sips at one time, to keep practicing that, so to speak.” [1].

Context
The context variables included both physical issues, such as adaptations and the accessibility of restaurants and bars, and social issues, such as assistance. Almost all participants reported that they needed devices or adaptations to facilitate eating and drinking. Commonly used devices included anti-slip mats (n=2), adapted utensils (n=5), and drinking straws or cups (n=6). The participants noted that these devices were old, that they had been recommended a long time ago, and that they experienced feelings of shame when using them in unfamiliar environments. Going out to dinner in a wheelchair was relatively rare; “…because when we were on vacation in xxx, we had to search for an hour and a half before we found a restaurant that could accommodate three wheelchair users.” [2]. Moreover, menu choices were determined primarily by texture, and not by taste preferences. These aspects reflect severe limitations to participation in large social events.

Participants noted that, within familiar environments, assistance at mealtimes tends to be based on familiarity and anticipation. As explained by one woman: “If we’re having something that’s a bit hard to cut, my mother always puts a special, sharper knife on the table. Then I make sure that the pieces are very small.” [3]. Participants noted that they often receive no assistance or understanding from unfamiliar individuals, particularly in work settings. Illustrations from the previous examples of limited lunch times at school and work can be complemented by the comments of one participant regarding various types of assistance: “They think, ‘Well, it would be good to alternate giving things.’ While xxx and xxx also think, ‘Okay, I’ll eat this first, and then that.’ So, you have to be careful it’s not too hot.” [4]. One man concluded, “But actually, you always need someone to be able to eat.” [9]. Such continued dependence on others is likely to lead to problems, frustration, and concerns.
Concerns

Concerns expressed by participants largely had to do with new circumstances, the future, or food intake. One man worried about mealtimes in unfamiliar environments: “Will these people be able to do it, and will they dare to give me food?” [10]. Another participant was concerned “…because, as you age, the problems associated with CP are more likely to increase than decrease. So, I’m convinced that, in the future, I might need to use more assistive devices to eat, or they might have to cut it in even smaller pieces.” [5]. As noted by another participant, “All in all, it’s fine, although I can’t eat all types of meat. So, I’ll forget how, I think, if I decide to stop eating something.” [9]. One woman mentioned being concerned “that, some days, I don’t eat and drink enough. And sometimes in the evening, I only eat half a meal.” [2].

Half of the participants told us that they would like to improve their eating and drinking abilities. They had noticed that feeding ability and nutritional status was not included in their regular physical check-ups and that they did not receive information about new adaptations or interventions.

DISCUSSION

This qualitative study examines the perceptions of 10 adolescents and young adults with CP (aged 15 - 23 years) regarding their eating and drinking activities, as well as factors related to these perceptions. Despite the relatively high ratings that they assigned to their eating and drinking skills, all participants reported having at least some problems with eating and drinking (e.g., processing various food textures, choking on fluids and solids, and handling utensils). This discrepancy is likely due to the tendency of participants to assume that their abilities are simply a part of them and that they cannot do anything about them. Moreover, these problems have not received enough attention from health care professionals. The perceptions of the adolescents and young adults with CP about avoiding foods, reduced social interaction during meals, and dependence on others during mealtimes were similar to those described by adults with CP in the study by Balandin and colleagues. [13]

The transition to adulthood is a challenging period for young people with CP, as argued by Björquist and colleagues. [30] Young people with multiple handicaps, including CP, often lose or regress in their functional ability as they age. [17] Moreover, physical and facial growth (e.g., during the growth spurt in puberty) may lead to changes in motor patterns and the processing of various foods. Advancing age is accompanied by a desire for greater independence and adaptation to the social and cultural environment. [30] Experiences with eating and drinking are likely to be a delicate issue for these young people. In fact, at a certain age, they are likely to prefer avoiding certain foods or beverages to having to rely on assistance. The younger interviewees (aged 15 - 17 years) described fewer negative
experiences than the older interviewees (aged 19 - 23 years), probably due to the more protected living conditions in the parental home, lower social-emotional development, and/or fewer physical changes associated with the growth spurt in late puberty.

Participants described experiencing a variety of emotions in mealtime situations, including feelings of shame (e.g., drawing attention due to their manner of eating), frustration (e.g., due to avoiding some of their favorite foods, missing social participation during mealtimes, and/or being unable to use utensils), fear, and distress. Fear was expressed most commonly in relation to choking, which was identified as a serious problem. All of these feelings were comparable to those mentioned by parents in a study by Sleigh. In our study, even individuals with only mild disabilities described a large range of negative perceptions, most likely because they were more highly integrated into society.

One serious consequence of eating and drinking problems has to do with the risk of qualitative and quantitative deficiencies in food intake, as illustrated by participants who reported having inadequate food intake due to limited force, insufficient time for consuming meals, or a tendency to avoid certain foods. Children and adolescents with CP are at risk for malnutrition and nutritional deficiencies (e.g., skeletal maturation). Both overweight and underweight could be observed among our participants. We recommend devoting attention to optimal food intake, balanced with attention to physical activity and growth throughout the life course.

Individuals with CP benefit from adaptations to eating and drinking equipment that can improve the mealtimes. For the participants in this study, however, most adaptations had been recommended many years ago, with regard to other levels of developments or under other circumstances, and they had not been adjusted to new life situations, physical conditions, or insights. The participants, therefore, continued to rely on outdated solutions, which sometimes resulted in feelings of shame. Attention to self-management, application, and technology for this age-group is needed, and it should be an issue in rehabilitation. In this respect, we recommend the use of devices to simplify meal preparation, as well as for eating and drinking, in addition to encouraging individuals with CP to seek assistance. Moreover, regarding to self-management, this could help individuals with CP to become more inventive in compensating for practical impediments, including in mealtime settings and asking for adequate assistance.

Despite the small sample size addressed in this study, saturation of the meaningful segments was reached. Due to selection bias, however, it is difficult to draw general conclusions. Given that the participants were self-selected, their opinions may not reflect those of all adolescents and young adults with CP. All participants in our study had 100% oral feeding, although two also received food supplements. In contrast, it is known that some proportion of all individuals with CP receive supplementary tube feeding. Although four participants had severe disabilities and were dependent on electric wheelchairs, only...
one had severe oral-motor problems and severe dysphagia. Moreover, all participants were native Dutch. Individuals of other nationalities could be expected to encounter different cultural issues. For this reason, future studies should include larger samples and quality-of-life questionnaires aimed at obtaining objective information on eating and drinking situations. Such studies could enrich the available data and provide more details concerning the perceptions of adolescents and young adults with CP about eating and drinking problems.

**IMPLICATIONS FOR PRACTICE**

This study raises several important points relating to the abilities needed to increase the independence of adolescents and young adults with CP in eating and drinking. Based on the information obtained from the interviews, disabilities in eating and drinking can pose ongoing problems, and they should be a point of attention for caregivers. Healthcare providers should devote sufficient time and effort to addressing activities associated with eating and drinking in various social environments, thereby helping young people with CP to make the transition to adulthood and to function independently in society. This recommendation is consistent those proposed by Donkervoort and colleagues. Regular multidisciplinary involvement in evaluation, advice, and interventions related to eating and drinking ability is needed in order to create optimal conditions for eating and drinking throughout the life course. These efforts should be adapted to changing living conditions and advancing insight, as well as to new adaptations and new foods. In adults with CP, interventions have yielded positive effects on strength training for purposes of mobility, and these results might also be relevant to eating and drinking abilities. Health professionals are challenged to identify evidence-based interventions that could enable adolescents and young adults to maintain or improve their functional abilities or to adapt to or compensate for difficulties. Moreover, timely intervention could prevent choking and nutritional deficiency. At the same time, we advocate providing advice and/or education on how to manage these issues in different environments (e.g., workplace), in order to ensure ongoing safety and optimal well-being in this domain. In the interest of improving advice and enhancing self-management, parents, teachers, employers, colleagues are in need of knowledge about conditions relating to eating and drinking for individuals with CP. We also recommend instructing the staff of restaurants and catering services to offer more menu choices and adaptations for disabled guests.
CLINICAL MESSAGE

- Adolescents and young adults with cerebral palsy experience difficulties with eating, drinking, and swallowing, and they encounter difficulties in participating in mealtimes with family and friends.
- Adolescents and young adults with cerebral palsy exhibit a variety of coping strategies for their eating and drinking disorders.
- Although adolescents and young adults with cerebral palsy rated their eating and drinking skills as reasonable, they reported feelings of shame, frustration, fear, distress, and concerns.
- Regular multidisciplinary involvement with eating and drinking is needed for purposes of evaluation, advice, and intervention throughout the life course, adjusted to living conditions and the latest insights.
- Adolescents and young adults with cerebral palsy showed limited initiative in asking for tailored assistance in eating and drinking activities.
ACKNOWLEDGEMENTS

The authors are grateful to the participants and parents who agreed to be interviewed and to share their experiences with us. We would also like to thank Mandy ten Have, Emma van den Manacker, and Marieke Mocking, SLT students at HAN University of Applied Sciences in Nijmegen, for analyzing the interviews.
REFERENCES


Assessment of mastication in healthy children and children with cerebral palsy: a validity and consistency study

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ABSTRACT

The aim of this study was to develop the Mastication Observation and Evaluation instrument for observing and assessing the chewing ability of children eating solid and lumpy foods. This study describes the process of item definition and item selection and reports the content validity, reproducibility, and consistency of the instrument. In the developmental phase, 15 experienced speech-language therapists assessed item relevance and descriptions over three Delphi rounds. Potential items were selected based on results from a literature review. At the initial Delphi round, 17 potential items were included. After three Delphi rounds, 14 items regarded as providing distinctive value in the assessment of mastication (consensus > 75%) were included in the Mastication Observation and Evaluation instrument.

To test item reproducibility and consistency, two experts and five students evaluated video recordings of 20 children (10 children with cerebral palsy aged 29 - 65 months and 10 healthy children aged 11 - 42 months) eating bread and a biscuit. Reproducibility was estimated by means of the intraclass correlation coefficient (ICC). With the exception of one item concerning chewing duration, all items showed good to excellent intra-observer agreement (ICC students: 0.73 - 1.0). With the exception of chewing duration and number of swallows, inter-observer agreement was fair to excellent for all items (ICC experts: 0.68 - 1.0 and ICC students: 0.42 - 1.0).

Results indicate that this tool is a feasible instrument and could be used in clinical practice after further research is completed on the reliability of the tool.
INTRODUCTION

Mastication, or chewing, is a rhythmic oral-motor activity in the feeding process. A central pattern generator, located in the pons and medulla, is thought to regulate the rhythm and coordination of the mouth, oral muscles, and joints.\(^1,2\) An efficient masticatory process requires repetitive vertical excursions of the jaw in combination with rotary movements of the tongue and motor activity of the lips and cheeks.\(^3-5\) Tongue movements reduce the size of the bolus and transform bolus consistency by mixing the bolus with saliva and releasing fluid in the food.\(^6\) Maintaining lip closure prevents loss of food and saliva.\(^7,9\)

The normal development of mastication has been well described. At the age of six months, infants can move their tongue laterally when the food is placed inside the mouth on the left or right side and by eight months, infants can move their tongue from the center of the mouth to the sides of the mouth.\(^10,11\) Chewing soft food occurs at the mean age of 9.42 months (SD 1.79) and chewing and swallowing firmer food without choking is achieved at 12.17 months (SD 2.28).\(^12\) The increase in SD reflects the inter-individual differences in developmental stages. Green et al.\(^8\) reported that at 12 months of age, infants have developed a basic chewing pattern that they refine over the next three years. By the time the child is two years old, the child can transfer the bolus over the midline of the tongue.\(^12\) Most studies report that an infant begins a transition period towards mature mastication after the child’s first 12 months when, in addition to vertical excursions of the jaw, horizontal jaw movements emerge. Researchers are inconsistent on when rotary patterns of the mandible are established.\(^5,13\)

As many children with developmental, medical or oral-motor disabilities have difficulties processing solid or lumpy foods, assessing a child’s chewing skills is important for diagnostic purposes and for identifying intervention goals. The assessment of chewing skills is usually based on clinical judgment. In most Dutch settings, during the diagnostic phase, a child’s oral-motor skills are observed and described in a semi-standardised manner. As there is no standardized protocol, there is considerable between-observer and within-observer variation. This limits whether the data can be used to compare chewing skills between children or groups of children or even whether a child’s skills can be monitored over time (i.e. to evaluate the effect of an intervention). A well-designed mastication assessment is necessary to determine initial conditions of mastication and the results could be helpful in generating hypotheses in the diagnostic phase, identifying intervention goals and monitoring changes in mastication.

Several instruments are available for assessing oral-motor skills for feeding. Instruments with only 1-3 items on chewing and swallowing solids are too broad for detailed chewing assessment, e.g. Dysphagia Disorder Survey (DDS)\(^14\), Modified Functional Feeding Assessment (FFAm)\(^1,15\), and Oral Motor Assessment Scale (OMAS)\(^16\). Instruments combining assessment of speech and oral-motor tasks\(^15,17-19\) concentrate on testing a broad range of
oral motor skills\textsuperscript{15,17,19}, whereas we were interested in a detailed and standardized manner of assessing chewing skills in everyday eating tasks.

Although the Schedule of Oral Motor Assessment (SOMA) is a detailed, valid and reliable instrument\textsuperscript{20-22}, there are four drawbacks to its use: (i) in clinical practice we observe that poor item description leads to decreased reliable for some items, (ii) instrument training is currently not available in the Netherlands, (iii) by scoring on a dichotomous scale it is not possible to differentiate between stages of skill development and learning, and (iv) the instrument does not contain an item on lateral tongue movement for the subscale ‘cracker’, which is an important aspect of mastication\textsuperscript{23,24}. New research into the (developmental) masticatory process making use of electromyography (EMG)\textsuperscript{25-27} and ultrasound\textsuperscript{28,29} has yet to be integrated into clinical application. These studies have contributed to the understanding of mastication (e.g. the importance of lateral tongue movements during mastication).

To assess the individual chewing skills required of a child for the safe consumption of solid and lumpy foods, we developed a concept version of the Mastication Observation and Evaluation instrument (MOE-concept). Our goal was to develop a valid and reliable instrument in which the five stages of mastication as listed by Hiiemae et al.\textsuperscript{30} could be evaluated and which allowed differentiation between normal and abnormal chewing. The initial list of MOE-concept items was drawn from the SOMA\textsuperscript{20-22}, FFAm\textsuperscript{7,15}, research papers concerning mastication\textsuperscript{18,19,24-27}, and our clinical experience.

The purpose of this study was to determine the validity and consistency of the MOE-concept based on the quality criteria for measurement properties of health status questionnaires developed by Terwee et al.\textsuperscript{32} in particular (i) content validity (aim of the instrument, target population, theoretical foundation, item selection and item reduction), (ii) reproducibility (intra-observer agreement), and (iii) consistency (inter-observer agreement), also defined in the Consensus-based Standards for Measurements of health status Instruments (COSMIN) checklist.\textsuperscript{31} The study was designed in two parts so we could identify which items were important for the clinical assessment of mastication in healthy young children and children with cerebral palsy (CP) (part 1) and investigate whether these items could be reliably scored (part 2).

**MATERIALS AND METHODS**

**Part 1. Content validity study**

*Delphi technique*

We used Delphi rounds to achieve consensus on the relevance, terminology, and description of the distinctive items for processing solid or lumpy foods in children. The Delphi method is a structured communication process with three characteristics: anonymity, consecutive
rounds of questions with controlled feedback and a statistical group outcome measure. This method is suitable for item selection and item definition of a test. Figure 1 displays the steps of the Delphi rounds. At each Delphi round, participants could suggest the removal, addition or revision of an item.

![Figure 1. Steps in the Delphi method.](image)

**Participants**
Speech-language therapists (SLTs) were eligible for this study if they (i) currently worked in a rehabilitation center or (university) medical center or as a lecturer in the field of feeding difficulties in young children, (ii) were available to participate in all Delphi rounds, and (iii) had experience with feeding problems in young children and children with CP. Twelve SLTs from the Sint Maartenskliniek, the Radboud University Medical Center and Groot Klimmendaal and three speech therapy lecturers participated in the Delphi rounds.

**Procedure**
The first Delphi round took place in the form of a group meeting. This meeting lasted three hours and was run by the first author and an independent moderator. Prior to the group meeting, the participants received a document with a description of the aim of the instrument, terminology, and results of recent research concerning mastication.
and background information about the chosen items. The MOE-concept had nine items assessing discrete movements (e.g. of lips, tongue and mandible) and eight items assessing integrated movements (e.g. food transport and coordination of chewing). The instrument was designed so it could be used for assessing both a soft, dissolvable consistency (bread) and a crispy, dissolvable consistency (biscuit). We selected these consistencies as they are everyday consistencies that can be swallowed safely by children of a young age and the process of mastication differs between both consistencies.\textsuperscript{36,37}

Each of the 17 MOE-concept items was explained by the first author and illustrated with examples (recordings of children with sufficient and insufficient skills). After discussing the items, the participants answered a questionnaire. The questionnaire consisted of two tasks per item: (i) score the relevance of the item on a 4-point scale (very important, important, unimportant and very unimportant) and (ii) score the clarity of the item description on a 4-point scale (very clear, clear, unclear, and very unclear). Participants could propose improvements for item description and response options.

During the first Delphi round, the panel decided a 4-point ordinal scale varying from very inappropriate to very appropriate should be used for the items of the MOE. Moreover, the panel also decided to adapt some items (e.g. frequency or range of motion). Based on the discussion from the first Delphi round, a mastication evaluation protocol was developed for instruction concerning the observation procedure, the feeding conditions, and the scoring system. The two follow-up Delphi rounds were conducted in written form and participants judged the improved description of the original and added items and the response options.

Data analysis

All written responses were anonymously processed by the first author before responses were discussed with the last author (MN). Proposed improvements were discussed within the research group. After each Delphi round, participants received a letter with the scores on item relevance and item description. To avoid influencing the participants at the next Delphi round, comments and feedback were not included in this letter.

We defined consensus as the proportion of participants who rated item relevance as ‘very important’ or ‘important’ per item. The cut-off point (75%) for rejecting an item was set before the study. Items with a consensus below 60% were removed from the list and items with a consensus from 60 to 75% were retained for further discussion at the next Delphi round. Participant remarks were used to improve item descriptions and the response options for the next Delphi round. Both item descriptions (original and adapted) were judged in the next rounds.
Part 2. Consistency study

Participants
Two groups of children participated in this study. Group 1 included 10 children (6 boys) with bilateral CP aged 29 - 65 months (SD 13 months) recruited from the Sint Maartenskliniek rehabilitation center in Nijmegen. CP was classified on the Gross Motor Function Classification System (GMFCS) as being mildly to moderately disabled. Group 2 included 10 healthy children (6 boys) aged 11 - 42 months (SD 10 months) recruited from day-care centers in the Nijmegen region. All parents received written information about the study and provided written informed consent.

The study was approved by the Slotervaart Hospital’s Medical Ethics Committee and READE in Amsterdam (dossier number NL40472.048.12) and was carried out in accordance with the Helsinki Declaration.

Assessment procedure
All feeding sessions were carried out in a quiet environment and were recorded using a digital hard disk camcorder (Sony DCR-SR90E). The camera was placed on a stand at a distance of 1.5 meters from the child and at an angle of 30° to obtain a semi-profile view of the child’s face and neck. In all cases, the research SLT faced the child.

During an assessment, each child was offered three pieces of wheat bread with chocolate spread (1.5 x 1.5 cm) and three pieces of a sweet crunchy biscuit. The SLT held a piece of bread or biscuit in front of the child’s mouth. The biscuit had to be bitten off by the child. The child’s mouth had to be empty before another piece of food was offered.

Rating Procedure
Two experienced SLTs from part one of the study and five speech therapy students assessed all video recordings. Prior to assessing the recordings, all observers completed a three-hour training session on the items and the response options in the MOE-concept (determined in the Delphi rounds). The recordings of ten children from each group were presented to the observers in a randomized order and did not include any information relating to a child’s age, gender or diagnosis. All observers assessed the recordings independently of each other and scored three bites per child and per consistency. Hence, in total six chewing activities were assessed per child. The five students rescored the recordings after an interval of two weeks for calculating the intra-observer agreement.

Statistical analysis
We used the intraclass correlation coefficient (ICC) (absolute agreement, two-way random, average measurements) to measure the inter-observer and intra-observer agreement.
The ICC defines the strength of the relationship among multiple observations of the same variables. ICCs above 0.80 are considered to reflect almost perfect agreement and values from 0.60 to 0.80 indicate good to excellent agreement. Items with an ICC exceeding 0.60 were considered reliable.

The response options per item varied from 1 to 4: the scores 1 and 2 represent an inappropriate oral-motor movement and the scores 3 and 4 represent an appropriate oral-motor movement. The difference between score 1 and 2, and score 3 and 4 is the degree of (in)appropriateness. The lowest score of three bites per item for each child and observer was used for calculating the ICC. All analyses were performed with SPSS (version 17.0).

RESULTS

Part 1. Content validity study
All 15 SLTs participated in the first two Delphi rounds. In the third round, one panel member was not able to participate. In the first round, 17 items were included. Tables 1 and 2 display the consensus on item relevance and item descriptions after the Delphi rounds. At the first Delphi round, the consensus on item relevance was more than 75% for 11 of the 17 items. The item categories had good average scores for movements of the tongue (90.5%), movements of the jaw (78.6%), and integrated movements (80.9%). The score was poor for movements of the lips (66.3%) and activity of the cheeks (50.0%). Qualitative analysis was used to improve item description and the response options (see Table 2). Improvements included eliminating words, rewriting sentences and splitting an item to separate the oral and pharyngeal stages.

After the first Delphi round, four items were removed from the list. Three items because less than 60% of the participants judged these items as relevant. This concerned items 3 (sucking of the lower lip), 11 (chewing on both sides), and 12 (activity of the cheeks). For item 11, the qualitative analysis showed that the reason for the low consensus was that this item did not have a discriminative function. A similar finding was reported by Mioche et al. The panel made recommendations for item 8 (trunk and head stability). The authors discussed these recommendations and decided to remove this item from the list because stability could be a consequence of the basic tonus and would, therefore, not necessary be related to mastication. Items 14 (gagging) and 15 (transport of the bolus) were both split into two items (oral stage and pharyngeal stage) because of the panel’s recommendations.
Table 1. Results of Delphi rounds 1, 2, and 3. Agreement presented as percentage of a dichotomous scale (very clear/very important or clear/important) on item relevance and item description.

<table>
<thead>
<tr>
<th>Item</th>
<th>Relevance (% important)</th>
<th>Description (% clear)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Lip closure</td>
<td>78.6</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>57.1</td>
<td>86.6</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td></td>
</tr>
<tr>
<td></td>
<td>80</td>
<td></td>
</tr>
<tr>
<td>2. Active lip movements *</td>
<td>64.3</td>
<td>66.7</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>71.4</td>
<td>73.3</td>
</tr>
<tr>
<td></td>
<td>n.a.</td>
<td></td>
</tr>
<tr>
<td>3. Sucking pattern of the lower lip *</td>
<td>53.8</td>
<td>rejected</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>53.8</td>
<td>n.a.</td>
</tr>
<tr>
<td></td>
<td>n.a.</td>
<td></td>
</tr>
<tr>
<td>4. Tongue protrusion</td>
<td>85.7</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>57.1</td>
<td>93.3</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>73.3</td>
</tr>
<tr>
<td>5. Lateral tongue movements</td>
<td>100</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>78.6</td>
<td>60.0</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>73.3</td>
</tr>
<tr>
<td>6. Munching</td>
<td>85.7</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>38.5</td>
<td>73.3</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>86.7</td>
</tr>
<tr>
<td>7. Controlled bite</td>
<td>85.7</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>57.1</td>
<td>66.7</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>80</td>
</tr>
<tr>
<td>8. Jaw stability *</td>
<td>78.6</td>
<td>rejected</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>42.8</td>
<td>n.a.</td>
</tr>
<tr>
<td></td>
<td>n.a.</td>
<td></td>
</tr>
<tr>
<td>9. Jaw movement</td>
<td>92.9</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>50.0</td>
<td>66.7</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>53.3</td>
</tr>
<tr>
<td>10. Chewing duration</td>
<td>78.6</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>46.2</td>
<td>73.3</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>73.3</td>
</tr>
<tr>
<td>11. Chewing on both sides *</td>
<td>53.9</td>
<td>rejected</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>46.2</td>
<td>n.a.</td>
</tr>
<tr>
<td></td>
<td>n.a.</td>
<td></td>
</tr>
<tr>
<td>12. Activity of cheeks *</td>
<td>46.1</td>
<td>rejected</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>42.9</td>
<td>n.a.</td>
</tr>
<tr>
<td></td>
<td>n.a.</td>
<td></td>
</tr>
<tr>
<td>13. Food or saliva loss</td>
<td>92.9</td>
<td>accepted</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>100</td>
<td>100</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>73.3</td>
</tr>
<tr>
<td>14. Gagging/ disgust/ coughing/ aspiration</td>
<td>92.9</td>
<td>a. gagging/ disgust</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>69.2</td>
<td>93.3</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>80</td>
</tr>
<tr>
<td></td>
<td>b. coughing/ aspiration</td>
<td>69.2</td>
</tr>
<tr>
<td></td>
<td></td>
<td>93.3</td>
</tr>
<tr>
<td></td>
<td></td>
<td>73.3</td>
</tr>
<tr>
<td>15. Transport of the bolus</td>
<td>85.7</td>
<td>a. oral transport</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>92.9</td>
<td>100</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>80</td>
</tr>
<tr>
<td></td>
<td>b. pharyngeal transport</td>
<td>92.9</td>
</tr>
<tr>
<td></td>
<td></td>
<td>100</td>
</tr>
<tr>
<td></td>
<td></td>
<td>80</td>
</tr>
<tr>
<td>16. Number of swallows</td>
<td>61.5</td>
<td>78.7</td>
</tr>
<tr>
<td></td>
<td>Round 1</td>
<td>Round 2</td>
</tr>
<tr>
<td></td>
<td>69.2</td>
<td>85.7</td>
</tr>
<tr>
<td></td>
<td>Round 3</td>
<td>66.7</td>
</tr>
<tr>
<td>17. Fluency and coordination</td>
<td>71.4</td>
<td>73.3</td>
</tr>
<tr>
<td></td>
<td>30.8</td>
<td>80.0</td>
</tr>
<tr>
<td></td>
<td>66.7</td>
<td></td>
</tr>
</tbody>
</table>

*Item deleted in the final version of the MOE.

n.a. = not applicable
**Table 2.** Qualitative analyses of rater’s comments on the definition (def.) and answer options (ans.) after the three Delphi rounds. The number of times that a note has been made is in parentheses.

<table>
<thead>
<tr>
<th>Item</th>
<th>Textual remarks</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Lip closure</td>
<td>Def. Use a clear description.; delete the word “normally”.</td>
</tr>
<tr>
<td>Ans.</td>
<td>Use terminology analogue to ICF terminology (mostly, sometimes). (2); Delete the word “adequate”. (3); Prefer indication in percentage instead of mostly/sometimes. (2); Lip closure is only relevant in case of food loss.</td>
</tr>
<tr>
<td>4. Tongue protrusion</td>
<td>Def. Add; unless food is moved from the lips. (3) Make a differentiation of the tongue beyond the lips and beyond the teeth.</td>
</tr>
<tr>
<td>Ans.</td>
<td>Describe the difference between “frequent” and “several times”. (4) Tongue movement beyond the teeth and beyond the lips are used interchangeably remove “the lips”.</td>
</tr>
<tr>
<td>5. Lateral tongue movement</td>
<td>Def. Add: unilateral puff out of the cheeks. Remove “When the lateral movement is not visible”</td>
</tr>
<tr>
<td>Ans.</td>
<td>Add the description of the sufficient movement.</td>
</tr>
<tr>
<td>6. Munching</td>
<td>Def. The definition is not clear.</td>
</tr>
<tr>
<td>Ans.</td>
<td>Use same hierarchy from insufficient to sufficient. “Normally none” change to “none”.</td>
</tr>
<tr>
<td>7. Controlled bite</td>
<td>Def. Improve the definition of help in biting. (2)</td>
</tr>
<tr>
<td>9. Jaw movement</td>
<td>Def. The description of the movement of the jaw; w shaped or rotated. Change “jaw” into “mandible”. (3)</td>
</tr>
<tr>
<td>10. Chewing duration</td>
<td>Def.</td>
</tr>
<tr>
<td>Ans.</td>
<td></td>
</tr>
<tr>
<td>13. Loss of food or saliva</td>
<td>Def.</td>
</tr>
<tr>
<td>Ans.</td>
<td>The difference between scores 1 and 2 is difficult</td>
</tr>
<tr>
<td>14. Gagging/disgust/coughing/aspiration</td>
<td>Def. Delete the part with reaction as result of food aversion</td>
</tr>
<tr>
<td>Ans.</td>
<td></td>
</tr>
<tr>
<td>15. Transport of the bolus</td>
<td>Def.</td>
</tr>
<tr>
<td>Ans.</td>
<td>What is the difference in score 2 and 3?</td>
</tr>
<tr>
<td>16. Number of swallows</td>
<td>Def. How to score a piece meal deglutition.</td>
</tr>
<tr>
<td>Ans.</td>
<td></td>
</tr>
<tr>
<td>17. Fluency and coordination</td>
<td>Def. It is good to give an overall score for the quality of mastication, because it is impossible to give an objective view. (2) Clarify the definition.</td>
</tr>
<tr>
<td>Overall remarks</td>
<td>Def. Add an item of formation of the bolus.</td>
</tr>
<tr>
<td>Ans.</td>
<td></td>
</tr>
</tbody>
</table>
### Qualitative remarks

Healthy children might chew with open mouth;
Possibility to close lips?
Is lip closure required for chewing? (2)

Difficult to indicate more than 50% closed lips; Percentage is clearer than terms, such as mostly; Use a timer for this item.

Swallowing during the oral chewing stage cannot be distinguished from the swallowing stage.
For evaluating chewing the swallow stage is not relevant.
This is only visible when the lips are open.
Suggestion: make more items for tongue movements.
Make a differentiation for age.

The lateral tongue movement is more important than placing the bolus between the molars.
The score concerns the result of the tongue movement. The importance is the movement itself.
An asymmetric movement of the mouth corner might be caused by the combination of activity of cheeks and lips without tongue movement.
Difference in score 3 and 4 is not clear.

Jaw and tongue move integrated.
Munching is also a part of mature mastication.
Use a 2-point scale for this item.
Depends on food consistency and age.

What is the definition of a controlled bite? (4)
How to judge a hyper mobility of the jaw

Unclear item: the result of the chewing is more important than the direction of motion of the jaw. (2)
The mandible might compensate a limited tongue movement.
In case of fixed food consistency and size of the bite give an indication of the duration. (4)
Motivation for food or distraction influences the duration.
Is the munching included in the duration?
How to count the duration while a child takes a new bite when the mouth is not emptied.

Use a normal value. What is long/short?
Too long or too short is both not okay, but one is not worse than the other. Use a 2 or 3-point scale for this item. (4)

Loss of food and saliva might occur independently.
Use a 2 or 3-point scale for this item.
Split this item into two items because of different stages of the mastication process (8)

A head movement could also be a habitual behaviour.
Split this item into two items because of different stages of the mastication process (8)

Swallowing is not a part of mastication (2)
Formation of the bolus is more important than swallowing.
Score 3 and 4 are both adequate. Could you make a differentiation
How to score when a bite is rhythmical but not fluent?

Clarify the difference between “frequent “and several times” (3)
A 4-point scale gives the possibility to make nuances in scoring. (10)
In the second Delphi round, items 2 (active lip movements), 16 (number of swallows), and 17 (fluency and coordination) were re-presented for reviewing item relevance, as consensus was 60-75% in the first Delphi round. After the second Delphi round, item 2 (active lip movements) received a consensus on relevance below 75% and was removed from the list. In the third round the panel was asked to confirm the removal of the five items from the initial list. More than 73% (range 73.3 - 93.3 %) of the panel agreed on definite item removal.

Consensus on the clarity of the item descriptions increased from the first to subsequent rounds with a mean of 58.4% (SD 19.4) to 83.3% (SD 13.5) (see Table 1). Although the description of the items was clear for most participants, the description of the response options was improved based on the recommendations made by the participants. If consensus on the descriptions in the final Delphi round was lower than in the second round, we used the description of the response options with the best consensus in the final version (e.g. tongue protrusion, jaw movement, loss of food or saliva loss). As seen in the consensus scores, the definitions of most items were clear for the participants. Concerning the response options, participants attained consensus for all items except item 9 (jaw movement). After three Delphi rounds, 14 items to be scored on a 4-point scale were included in the MOE (see Appendix Chapter 5 for the final list).

Some of the participants provided feedback not related to the performance of mastication or the purpose of the study (e.g. food preference of the child, frequency of offered solids). Only feedback related to mastication was included for the discussion part of the Delphi rounds.

### Part 2. Consistency study

**Intra-observer agreement**

Table 3 lists the intra-observer agreement for each MOE item. The observers were fairly consistent assessing whether an item was present but they were less consistent regarding the degree of occurrence. Therefore, we recoded the responses into a dichotomous scale. The results indicate an almost perfect intra-observer agreement with ICC’s varying from ≥0.88 for bread and ≥0.73 for biscuit. With the exception of items 7 (controlled bite) and 16 (number of swallows), observers were relatively consistent in their assessment after a period of two weeks.
Table 3. Average intra-observer agreement (standard deviation in parentheses) for MOE items used to evaluate 20 children by student-raters (n=5). The lowest score of three bites was used to calculate the ICC.

<table>
<thead>
<tr>
<th>Intra-observer agreement</th>
<th>Average ICC (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Items</td>
<td>Bread</td>
</tr>
<tr>
<td>1. Lip closure</td>
<td>0.92 (0.06)</td>
</tr>
<tr>
<td>4. Tongue protrusion</td>
<td>0.98 (0.02)</td>
</tr>
<tr>
<td>5. Lateral tongue movement</td>
<td>0.91 (0.02)</td>
</tr>
<tr>
<td>6. Munching</td>
<td>0.95 (0.07)</td>
</tr>
<tr>
<td>7. Controlled bite</td>
<td>n.a.</td>
</tr>
<tr>
<td>9. Jaw movement</td>
<td>0.92 (0.07)</td>
</tr>
<tr>
<td>10. Chewing duration</td>
<td>0.89 (0.13)</td>
</tr>
<tr>
<td>13. Loss of food or saliva</td>
<td>0.88 (0.08)</td>
</tr>
<tr>
<td>14a. Gagging or disgust</td>
<td>0.96 (0.09)</td>
</tr>
<tr>
<td>14b. Coughing or aspiration</td>
<td>1.00 (0.00)</td>
</tr>
<tr>
<td>15a. Displacement of food with fingers</td>
<td>0.92 (0.05)</td>
</tr>
<tr>
<td>15b. Backwards displacement of food</td>
<td>0.92 (0.08)</td>
</tr>
<tr>
<td>16. Number of swallows</td>
<td>0.89 (0.03)</td>
</tr>
<tr>
<td>17. Fluency and coordination</td>
<td>0.95 (0.05)</td>
</tr>
<tr>
<td>n.a. = not applicable</td>
<td></td>
</tr>
</tbody>
</table>

Inter-observer agreement

The inter-observer agreement was calculated separately for the expert and student raters. The dichotomized scores were used to calculate the ICCs. Table 4 lists the ICC results for both groups. We could not calculate the ICC for four items (gagging, coughing or choking, using fingers to transport, and using head movement to transport) as all observers rated these items with the maximum score. ICCs for scoring biscuit were slightly lower than for scoring bread. The experts attained slightly higher levels of agreement than the students, especially for item 5 (lateral tongue movement), item 6 (munching), item 9 (jaw movement), item 14 (gagging), and item 16 (swallowing).
Table 4. Inter-observer agreement by consistency type and type of observer. The ICC (two-way random effects model with absolute agreement, single measurements) was calculated for each item.

<table>
<thead>
<tr>
<th>Inter-observer agreement</th>
<th>Bread ICC (95% CI)</th>
<th>Biscuit ICC (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Items</td>
<td>Experts n=2 Students n=5</td>
<td>Experts n=2 Students n=5</td>
</tr>
<tr>
<td>1. Lip closure</td>
<td>0.84 (0.41-0.90) 0.84 (0.70-0.93)</td>
<td>0.88 (0.68-0.96) 0.80 (0.61-0.91)</td>
</tr>
<tr>
<td>4. Tongue protrusion</td>
<td>0.88 (0.71-0.96) 0.89 (0.78-0.95)</td>
<td>0.74 (0.36-0.89) 0.81 (0.63-0.92)</td>
</tr>
<tr>
<td>5. Lateral tongue movement</td>
<td>0.90 (0.73-0.91) 0.79 (0.67-0.93)</td>
<td>0.83 (0.57-0.93) 0.56 (0.21-0.79)</td>
</tr>
<tr>
<td>6. Munching</td>
<td>0.83 (0.27-0.94) 0.68 (0.40-0.85)</td>
<td>0.59 (0.04-0.84) 0.35 (0.03-0.67)</td>
</tr>
<tr>
<td>7. Controlled bite *</td>
<td>n.a. n.a.</td>
<td>0.56 (0.15-0.83) 0.85 (0.72-0.93)</td>
</tr>
<tr>
<td>9. Jaw movement</td>
<td>0.62 (0.42-0.84) 0.64 (0.31-0.84)</td>
<td>0.83 (0.58-0.93) 0.37 (0.22-0.67)</td>
</tr>
<tr>
<td>10. Chewing duration</td>
<td>0.36 (-0.46-0.72) 0.65 (0.46-0.84)</td>
<td>0.68 (0.22-0.87) 0.64 (0.33-0.83)</td>
</tr>
<tr>
<td>13. Loss of food or saliva</td>
<td>0.81 (0.52-0.92) 0.92 (0.85-0.96)</td>
<td>0.74 (0.36-0.90) 0.84 (0.69-0.93)</td>
</tr>
<tr>
<td>14a. Gagging or disgust*</td>
<td>1.00 1.00</td>
<td>1.00 0.53 (0.10-0.91)</td>
</tr>
<tr>
<td>14b. Coughing or choking*</td>
<td>1.00 1.00</td>
<td>1.00 1.00</td>
</tr>
<tr>
<td>15a. Displacement of food with fingers *</td>
<td>1.00 0.93 (0.86-0.97)</td>
<td>1.00 0.89 (0.80-0.95)</td>
</tr>
<tr>
<td>15b. Backwards displacement of food *</td>
<td>1.00 0.74 (0.52-0.88)</td>
<td>1.00 0.80 (0.61-0.91)</td>
</tr>
<tr>
<td>16. Number of swallows</td>
<td>0.94 (0.86-0.98) 0.59 (0.25-0.81)</td>
<td>0.90 (0.75-0.96) 0.42 (0.21-0.72)</td>
</tr>
<tr>
<td>17. Fluency and coordination</td>
<td>0.88 (0.68-0.95) 0.76 (0.49-0.90)</td>
<td>0.72 (0.28-0.89) 0.64 (0.33-0.84)</td>
</tr>
</tbody>
</table>

* These items were not observed in the children included in this study.

DISCUSSION

We documented the development of an observational instrument called the MOE for assessing chewing skills in children. The focus of this study was to select and develop items and investigate whether these items could be reliably recognized on video recordings. Although the process of mastication is largely intra-oral and it remains difficult to detect small differences in mastication, we completed the first step in developing an instrument with acceptable levels of content validity and observer agreement.
Content validity
As found by Santos et al.\textsuperscript{18} and Reilly et al.\textsuperscript{21}, experienced SLTs did not unanimously agree on the relevance of items concerning mastication in children. Some SLTs preferred a minimum number of distinguishable items whereas others preferred many items to ensure that all sub-skills were considered. With the exception of jaw movement, there was consensus with respect to the description of most items after three Delphi rounds. Although jaw movements during mastication are widely described in EMG and kinematic research\textsuperscript{8,24,26,36}, defining and describing these movements appears to be a difficult task. The SOMA\textsuperscript{20} assesses jaw movements over six items, which may reflect the difficulties in defining jaw movements for clinical practice. We discussed the item jaw movement and opted for an item description based on the description used by Woda et al.\textsuperscript{26}

Consistency
As was seen in the excellent intra-observer agreement scores, observers were relatively consistent in their assessment of the items over two sessions. Similar results were reported by Santos et al.\textsuperscript{18} Excellent inter-observer agreement was found for items with the highest scores on relevance and clarity of item descriptions (e.g. lip closure, tongue protrusion, loss of food or saliva, gagging or disgust, coughing or choking, and support in the bolus transport). However, it must be noted that the last four items were not present in the children included in this study. We decided to keep these items in the final version of the MOE as we know from the literature and clinical experience that these items provide important information on chewing ability for children with CP.\textsuperscript{7} In a future study with a larger cohort of children with CP we will investigate whether these items are observable.

Unlike the SOMA studies\textsuperscript{21,22}, we found good agreement between the experts for items on tongue movements (items 4 and 5), which can be difficult to rate because these movements are not easily observed. Our results confirm that the number of swallows is a difficult item to rate. In our study a lower level of agreement was also scored on ‘munching’, ‘chewing duration’ and ‘fluency and coordination’.

We selected a four-point scale to encourage observers to decide whether an oral-motor behavior was appropriate or not. Although we specified the degree of (in) appropriateness for every item, we recoded the responses into dichotomous scores for analysis as observers disagreed about the degree of (in) appropriateness. The agreement could be improved by focusing on the degree differences during the rater training. Although the students achieved acceptable agreement results, their results are slightly lower than those of the experts and their variability was larger. Inexperienced observers (i.e. students) appear to have more difficulty recognizing some oral-motor movements than experienced SLTs. Extra attention for these specific items would be necessary for training. A good possibility would be to improve the training by using video recordings in conjunction with written material.
Overall, the agreement ratings for bread were slightly higher than for biscuit. Items were probably more difficult to evaluate as the mastication duration for biscuit is shorter because of the firmer consistency\textsuperscript{6,24} and this results in a shorter observation time for the rater.

It must be noted that although observers scored three bites we only used the lowest score for analysis as we wanted to concentrate on the performance and not on the capacity of the process of mastication. As in the clinical situation, it is more important to know if a child chokes on one of the attempts than to know if he is able to swallow the bolus sufficiently in some cases.

The MOE was used to evaluate mastication skills for healthy children and children with CP. Expert raters were able to observe all MOE items for both groups of children and reported that the variation of locomotion in mastication seemed to be smaller and the duration of mastication was longer for children with CP. Although not all mastication problems common for children with CP were observed in this study (e.g. choking, gagging, adaptation to bolus transport), this may be the result of a small cohort. Future studies should investigate the incidence of the marked problems when children with a higher GMFCS score are included in a study.

**Future research**

Future studies should investigate the reliability of the MOE using the criteria proposed by Terwee et al.\textsuperscript{32} These criteria refer to internal consistency, floor and ceiling effects and whether the tool is sensitive for developmental stages in healthy children. In the future, we would like to include a larger cohort of healthy children to investigate this. Moreover, we would like to test if this instrument is sensitive to change as result of an intervention. We intend to validate the MOE by using electromyography, kinematics and ultrasound measurements.

In this study, we identified which items are important and can be reliably scored for clinical assessment of mastication in healthy young children and children with CP. The MOE is a feasible instrument to use in clinical practice and future research will focus on further improvement of the validity and sampling of reference values.
ACKNOWLEDGEMENTS

The authors express their sincere thanks for the cooperation of the children and parents of the Sint Maartenskliniek and of the KION day-care centers in Nijmegen, who participated and consented to join this study. We would like to thank the participants of the panel who provided their time and knowledge. We are very grateful to Hanny Weersink for her job on the ratings of the children. We would also like to thank the students of the HAN University of Applied Sciences for collecting and scoring the video recordings. Our thanks also go to the staff at the Department of Child Rehabilitation Center at the Sint Maartenskliniek for their support and the use of their facilities.
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chapter 5

Validity and reliability of the Mastication Observation and Evaluation (MOE) instrument

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R Speyer
BE Groen
J van Limbeek
MWG Nijhuis-van der Sanden

Research in Developmental Disabilities 2014; 35: 1551–1561
ABSTRACT

The Mastication Observation and Evaluation (MOE) instrument was developed to allow objective assessment of a child’s mastication process. It contains 14 items and was developed over three Delphi rounds. The present study concerns the further development of the MOE using the COSMIN (Consensus-based Standard for the Selection of Measurement Instruments) and investigated the instrument’s internal consistency, inter-observer reliability, construct validity and floor and ceiling effects. Consumption of three bites of bread and biscuit was evaluated using the MOE. Data of 59 healthy children (6 - 48 months) and 38 children (bread) and 37 children (biscuit) with cerebral palsy (24 - 72 months) were used.

Four items were excluded before analysis due to zero variance. Principal Components Analysis showed one factor with eight items. Internal consistency was >0.70 (Chronbach’s alpha) for both food consistencies and for both groups of children. Inter-observer reliability varied from 0.51 to 0.98 (weighted Gwet’s agreement coefficient). The total MOE scores for both groups showed a normal distribution for the population. There were no floor or ceiling effects.

The revised MOE now contains eight items that (i) have a consistent concept for mastication and can be scored on a 4-point scale with sufficient reliability and (ii) are sensitive to stages of chewing development in young children. The removed items are retained as part of a criterion-referenced list within the MOE.
INTRODUCTION

Intake of solids is the last step in the development of food consumption in children. Learning to consume solid foods begins between 8 and 12 months. At the age of 12 months, children can manage most food structures and can move food from the center of the tongue to the side of the mouth, chew it and move it back to the center again when the food no longer requires chewing. They can also move the bolus to the back of the mouth for swallowing. Mastication coordination and strength improves up until 4 years of age and enables children to eat increasingly complex foods (e.g. beef and peanuts) or food with multiple consistencies (e.g. grapes). Not all children develop this process smoothly or quickly (e.g. children with neurological disorders, such as cerebral palsy) and these children may experience unpleasant and dangerous situations, such as gagging or choking. Oromotor feeding problems are an issue in approximately 90% of children with cerebral palsy (CP), but no data are available for mastication. A dysfunctional mastication system can result in an altered digestive process and in respiratory and dental health problems. It is also associated with an increase in mortality. Identifying (developmental) chewing difficulties is necessary to prevent such outcomes.

Clinical analysis of chewing can be assessed in a spontaneous mealtime context to indicate the proficiency of mastication (what the child does) and can be assessed in a clinical setting under optimal conditions (what the child can). Speech-language therapists (SLTs) require a structured observational instrument that (a) is child-friendly and easy to implement and (b) can be used to guide therapy goal-setting and/or indicate whether further assessment of the oral or oropharyngeal phase of the swallow is required.

Benfer et al. evaluated nine oral-pharyngeal dysphagia observation instruments and concluded that only two instruments, the Schedule for Oral Motor Assessment (SOMA) and Functional Feeding Assessment modified (FFAm), had suitably strong psychometric properties. The SOMA includes 22 items concerning chewing capacity to be scored on a dichotomous scale. Items in the SOMA do not have detailed descriptions of all items and does not include an item on lateral tongue movement. However, the FFAm, which is scored on a 5-point scale, evaluates a broad range of oral-motor skills and does not provide enough detail for a chewing evaluation. In addition to these drawbacks, there is no Dutch version of the SOMA and FFAm nor training possibilities for clinicians in the use of these scales. For these reasons, we developed the Mastication Observation and Evaluation (MOE) for the assessment of chewing in healthy infants and children with CP. In our original study, we developed the 14 MOE items over three Delphi rounds with 15 experts. Although we advocated removing the item 'chewing duration' because of low reliability, we decided to change the answer option for this item and we included this item for the present study to recheck its reliability.
The results from our first content validity study were of a level to warrant further development of the instrument. In the present study, we used a consensus-based approach for the selection of measurement instruments, called COSMIN.\textsuperscript{17} Only four measurement properties were relevant for the validation of the MOE instrument. The measurement properties that were not relevant or could not be assessed concerned the measurement error (not assessed due to the use of an ordinal scale), criterion validity (not assessed as there is no golden standard for assessing mastication) and responsiveness (we did not evaluate changes over time).

The goal of this study was to establish the internal consistency, inter-observer reliability and construct validity of the MOE and identify any floor and ceiling effects based on the chewing performance of a group of healthy children and a group of children with CP. As part of the analysis of the instrument’s construct validity, associations between MOE scores and age MOE scores and gross motor function of the children with CP were also investigated.

**METHODS**

**Participants**

In this study, 80 healthy children aged 6 - 48 months (healthy group) and 44 children with CP (CP group) aged 24 - 72 months with classification II-IV gross motor function according to the Gross Motor Function Classification Scale (GMFCS) took part in this study. Children with food allergies were excluded from the study.

Children in the healthy group were recruited from childcare centers in the Nijmegen and surrounding areas (Kinder Opvang Nijmegen (KION)). Children in the CP group were recruited from four rehabilitation centers and affiliated schools in the Netherlands. All parents received written information about the study and provided written informed consent. The Slotervaart Hospital’s Medical Ethics Committee and READE in Amsterdam approved the study (study number NL40472.048.12).

**Procedure**

The testing procedure was similar to that described by Remijn et al.\textsuperscript{16} While sitting in a suitable chair in a quiet environment, participants were offered a total of five pieces of wheat bread with chocolate spread (1.5 x 1.5 cm) and five pieces of a crunchy biscuit. A food item was presented to the child and the item was held in front of the child’s mouth. The child’s mouth had to be empty before another piece of food was offered.

Seven SLT students in their final study year performed the feeding sessions with the healthy children and the first author performed the sessions with the children with CP. All feeding sessions were recorded using a digital hard-disk camcorder (Sony DCR-SR90E). The camera was mounted on a tripod 1.5 m from the child and at an angle of 30 degrees.
to obtain a semi-profile view of the child’s face and neck. One SLT student not involved in the feeding sessions selected three of the five recorded bites per consistency (biscuit and bread) for assessment. Inclusion criteria for assessment were (i) that the SLT followed the instruction protocol when offering the food and (ii) there were three visible recorded bites from the moment of food intake until swallow. Exclusion criteria included recordings during which the child consistently talked, smiled or gazed. The recordings were coded and stored without child identifying information.

**Assessments**
The three recordings of a child’s attempt at biting and chewing each of the two consistencies were evaluated with the MOE. The MOE had 13 items for bread consistency and 14 items for biscuit to be scored on a 4-point ordinal scale. The items covered movement of lips, tongue, jaw and head and integrated behaviours, such as gagging, coughing, swallow coordination and number of swallows. The 4-point scale varied from reflecting ‘very inappropriate’ to ‘very appropriate’ movements and behaviours. For all items, responses reflected the degree of (in) appropriateness of an observation compared to mature mastication.

Seven SLT students involved in the feeding sessions received three 2-hour training sessions run by the first author. All MOE items including the answer options were explained and recordings of healthy children and children with CP (none of whom participated in this study) were used to illustrate each item. At the end of the training, all SLT students scored five recordings using the MOE (each recording contained three bites) and evaluations were compared to the first author’s evaluations. Each SLT student required an agreement level of 75% or more with the author to participate as an observer in this study. The differences in the interpretations of the five recordings were discussed in the last session. Students with an adequate agreement level received a set of recordings (randomised subset per observer) to score. All recordings were scored twice by different observers.

**Statistical analyses**
All statistical analyses were performed with the statistical analysis software SPSS (version 20) and AgreeStat. Descriptive statistics per MOE item were used to investigate the score distributions of the two groups. The prevalence of the maximum score (score 4) was calculated as a ratio for each item and for each consistency. We did not include items with no variance in score in the next step of the analysis.

We used Principal Component Analysis (PCA) with varimax rotation to identify clusters of items. Prior to the PCA we determined the Kaiser-Meyer-Olkin (KMO) measure of sampling (represented as the ratio of the squared correlation between variables to the squared partial correlation between variables). The KMO statistic summarizes the magnitude of the partial correlations relative to the original (zero-order) correlations. The partial
correlations are to be expected small in magnitude if the variables share common factor(s), and the KMO should be close to 1.0. A value of >0.60 indicates a relatively compact pattern of correlation and that factor analysis is appropriate for the data set. In the PCA, we used the criteria for eigenvalues to be greater than 1 time the mean eigenvalue and used the criterion that a loading of more than 0.40 on factors in this analysis was necessary.\textsuperscript{19,20} After extracting the factor(s), we examined the analysis of communalities as they indicate how much of the variance in each of the original variables is explained by the extracted factor. Items with a loading below 0.40 were removed.

In view of our interest in the capacity (optimal performance) of mastication, per observer we used the highest score per item out of three bites. The Cronbach’s alpha was computed for both groups and both consistencies using the highest score out of the three bites. As a measure of internal consistency, a value of 0.70 was considered to indicate that the scale was consistent.\textsuperscript{18}

Inter-observer reliability was calculated using the chance-corrected agreement coefficient (AC) statistic proposed by Gwet.\textsuperscript{18} The AC was designed for situations in which the kappa coefficient analysis might be paradoxically low when there are predominantly low or high scores in the set on which the observers agree. In a dataset with small variation, Gwet’s AC enables the unbiased calculation of rater agreement between two or among multiple observers.\textsuperscript{18} We used Gwet’s $AC^2$, the weighted version of the AC, which corrects for ordinal scaled data. According to Terwee et al.\textsuperscript{21,22}, $AC^2$ values above 0.70 indicate a positive level of agreement in a sample size of at least 50 persons.

We calculated the $AC^2$ including the 95% confidence interval for the remaining MOE items (i.e. after removal of items with no variation and items not selected in the PCA) for both groups of children and both consistencies using all scores per item of three bites. After this analysis, we determined the final list of items for the MOE.

For determining construct validity, we used regression analyses to determine the correlation between the total MOE scores by age of the children in the healthy group and children in the CP group, separately. Only data from the bread condition was appropriate for this analysis because of the standardised size of the bite. We hypothesized that the overall scores of the children with CP would be lower than those of the healthy children. We expected a correlation between age and MOE total score and decreasing variance in score with an increase in age for children in the healthy group. We expected no correlation between age and total MOE score for children in the CP group and a negative correlation between GMFCS and total MOE score. Finally, we calculated the score distributions by age and by group to identify any floor and/or ceiling effects.
RESULTS

Participants
Although almost all 80 healthy children were offered both bread and biscuit, data from all children for both conditions could not be included for analysis because the child was not used to eating biscuits \( n=13 \), the protocol for offering bread was not followed \( n = 18 \), the child displayed non-cooperative behaviour or did not achieve three good bites per consistency (bread condition \( n=3 \); biscuit condition \( n=6 \)). Data of 59 children available for statistical analysis of which 46 children had data for both food consistencies.

The data of 38 children with CP were included for bread and data of 37 children with CP for biscuit. Five children were not able to eat the biscuit and four children showed non-cooperative behaviour or had less than three good bites per consistency. Table 1 displays the group characteristics for both the healthy group and CP group.

Table 1. Descriptive data of the healthy children (healthy group) and children with cerebral palsy (CP group).

<table>
<thead>
<tr>
<th></th>
<th>Healthy group</th>
<th>CP group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Bread</td>
<td>Biscuit</td>
</tr>
<tr>
<td>( n )</td>
<td>59</td>
<td>59</td>
</tr>
<tr>
<td>Mean age in months (SD)</td>
<td>21.7 (12.8)</td>
<td>27.2 (11.6)</td>
</tr>
<tr>
<td>Age range in months</td>
<td>7 - 48</td>
<td>10 - 48</td>
</tr>
<tr>
<td>Gender Boys ((n))</td>
<td>33 (55.9%)</td>
<td>32 (54.2%)</td>
</tr>
<tr>
<td>Gender Girls ((n))</td>
<td>26 (44.1%)</td>
<td>27 (45.8%)</td>
</tr>
<tr>
<td>GMFCS* II ((n))</td>
<td>n.a.</td>
<td>n.a.</td>
</tr>
<tr>
<td>GMFCS* III ((n))</td>
<td></td>
<td></td>
</tr>
<tr>
<td>GMFCS* IV ((n))</td>
<td></td>
<td></td>
</tr>
<tr>
<td>GMFCS* V ((n))</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*GMFCS = Gross Motor Function Classification System
n.a. = not applicable

Variance in score
When considering the score distributions per item, we noted that for four items in more than 95% of the data no variation was present since nearly all scores were 4 (maximum score) for all three bites (i.e. gagging or disgust, coughing or aspiration, displacement of food with fingers and backwards displacement of food were not observed). The maximum performances of the oral behaviours are presented in Table 2. The performances in both bread and biscuit conditions were comparable.

Compared to the healthy group, the children with CP showed decreased performance on lip closure, tongue protrusion, lateral tongue movement, munching, controlled biting, jaw movement and chewing fluency and coordination.
Table 2. Prevalence of the maximum scores (score 4) as a ratio with total responses as denominator per item of the Mastication Observation and Evaluation instrument (MOE).

<table>
<thead>
<tr>
<th>Items</th>
<th>Healthy group</th>
<th>CP group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Bread</td>
<td>Biscuit</td>
</tr>
<tr>
<td>Lip closure</td>
<td>0.06</td>
<td>0.07</td>
</tr>
<tr>
<td>Tongue protrusion</td>
<td>0.63</td>
<td>0.86</td>
</tr>
<tr>
<td>Lateral tongue movement</td>
<td>0.24</td>
<td>0.37</td>
</tr>
<tr>
<td>Munching</td>
<td>0.28</td>
<td>0.70</td>
</tr>
<tr>
<td>Controlled bite</td>
<td>n.a.</td>
<td>0.79</td>
</tr>
<tr>
<td>Jaw movement</td>
<td>0.31</td>
<td>0.29</td>
</tr>
<tr>
<td>Chewing duration</td>
<td>0.27</td>
<td>0.42</td>
</tr>
<tr>
<td>Loss of food or saliva</td>
<td>0.86</td>
<td>0.98</td>
</tr>
<tr>
<td>Gagging or disgust*</td>
<td>0.96</td>
<td>0.98</td>
</tr>
<tr>
<td>Coughing or aspiration*</td>
<td>0.97</td>
<td>1.0</td>
</tr>
<tr>
<td>Displacement of food with fingers*</td>
<td>0.96</td>
<td>1.0</td>
</tr>
<tr>
<td>Backwards displacement of food*</td>
<td>0.95</td>
<td>0.96</td>
</tr>
<tr>
<td>Number of swallows</td>
<td>0.21</td>
<td>0.49</td>
</tr>
<tr>
<td>Fluency and coordination</td>
<td>0.26</td>
<td>0.25</td>
</tr>
</tbody>
</table>

* Items with less than 5% variation in the maximum score and therefore removed from the list
n.a. = not applicable

Principal Components Analysis

In total, four items were excluded from the analysis because no variance was present in the data. These were the items with the maximum scores for all bites in both groups of children. The KMO ranged from 0.627 to 0.757 over the four analyses. We did not exclude any items based on KMO scores. We completed the PCA with 9 items for the bread and 10 items for the biscuit conditions. The results of the PCA are listed in Table 3.

Only one factor was detected with an eigenvalue of more than 1 for both food types. This factor explained 34.1 - 41.0% of the variance in the items. Items in the factor that explained more than 40% were: ‘tongue protrusion’, ‘lateral tongue movement’, ‘munching’, ‘jaw movement’, ‘chewing duration’, ‘number of swallows’, and ‘fluency and coordination’. ‘Lip closure’ and ‘loss of food or saliva’ only loaded in one of the four analyses. The load of ‘loss of food or saliva’ on the factor was larger than that of ‘lip closure’. The item ‘controlled bite’ was only used for biscuit consistency and loaded below 0.40.
Table 3. Results of the Principal Components Analysis.

<table>
<thead>
<tr>
<th></th>
<th>Bread</th>
<th>Biscuit</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Healthy group</td>
<td>CP group</td>
</tr>
<tr>
<td></td>
<td>Healthy group</td>
<td>CP group</td>
</tr>
<tr>
<td>Kaiser-Meyer-Olkin measure of sampling (KMO)</td>
<td>0.76</td>
<td>0.67</td>
</tr>
<tr>
<td></td>
<td>0.63</td>
<td>0.71</td>
</tr>
<tr>
<td>Number of factors with eigenvalue &gt;1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>% of variance of the items</td>
<td>41.0%</td>
<td>39.3%</td>
</tr>
<tr>
<td></td>
<td>34.1%</td>
<td>40.9%</td>
</tr>
</tbody>
</table>

Loadings >0.40:
- Lip closure: 0.431
- Tongue protrusion: 0.725
- Lateral tongue movement: 0.783
- Munching: 0.785
- Controlled Bite: n.a.
- Jaw movement: 0.841
- Chewing duration: 0.561
- Loss of food or saliva: 0.528
- Number of swallows: 0.465
- Fluency and coordination: 0.737

n.a. = not applicable

Internal consistency
In the healthy group, Cronbach’s alpha of the remaining items was 0.61 for bread and 0.58 for biscuit conditions. In the CP group, Cronbach’s alpha was 0.69 for bread and 0.69 for biscuit conditions. After removal of the item ‘lip closure’, Cronbach’s alpha increased in all analyses (healthy group: 0.70 and 0.63 for bread and biscuit conditions, respectively; CP group: 0.72 and 0.73 for bread and biscuit conditions, respectively). Additional deletion of the item ‘controlled bite’, further improved Cronbach’s alpha in the biscuit condition for the healthy group to 0.71.

Inter-observer reliability
In the group of healthy children in the bread condition, two of the eight items had a Gwet’s AC² score just below 0.70 (lateral tongue movement and munching). The agreement of the other items ranged from 0.72 (jaw movement) to 0.97 (loss of food or saliva). The average AC score over all items was 0.77 (range 0.67 - 0.97). The reliability results for biscuit were comparable to those of bread but one item (lateral tongue movement) had an insufficient AC score. The average AC score over all items was 0.82 (range 0.68 - 0.98).

The inter-observer agreement for the group of children with CP was comparable with that of the healthy group. The average AC score for the bread condition was 0.73 (range 0.51 - 0.90) with a score below 0.7 for items ‘munching’ and ‘jaw movement’. In the biscuit
condition, the average AC score was 0.81 (range 0.77 - 0.98). The results of the Gwet’s AC<sup>2</sup> are listed in Table 4. When looking at the scores of two observers, approximately 50% of all differences in scores were attributed to responses of 3 or 4 on the scale, hence the degree of appropriateness of an oral-motor behaviour.

Table 4. Gwet’s AC<sup>2</sup> + (95% Confidential interval) by group and by food consistency (bread and biscuit). The AC<sup>2</sup> was calculated for all bites (n x 3 bites per consistency).

<table>
<thead>
<tr>
<th>Items</th>
<th>Healthy children</th>
<th>Children with CP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>bread</td>
<td>biscuit</td>
</tr>
<tr>
<td>Tongue protrusion</td>
<td>0.88 (0.85-0.92)</td>
<td>0.94 (0.92-0.97)</td>
</tr>
<tr>
<td>Lateral tongue movement</td>
<td>0.68* (0.62-0.75)</td>
<td>0.68* (0.62-0.75)</td>
</tr>
<tr>
<td>Munching</td>
<td>0.67* (0.61-0.74)</td>
<td>0.86 (0.82-0.89)</td>
</tr>
<tr>
<td>Jaw movement</td>
<td>0.72 (0.66-0.79)</td>
<td>0.71 (0.64-0.79)</td>
</tr>
<tr>
<td>Chewing duration</td>
<td>0.73 (0.67-0.79)</td>
<td>0.82 (0.78-0.86)</td>
</tr>
<tr>
<td>Loss of food or saliva</td>
<td>0.97 (0.95-0.98)</td>
<td>0.98 (0.97-0.99)</td>
</tr>
<tr>
<td>Number of swallows</td>
<td>0.79 (0.75-0.84)</td>
<td>0.79 (0.74-0.85)</td>
</tr>
<tr>
<td>Fluency and coordination</td>
<td>0.75 (0.68-0.81)</td>
<td>0.80 (0.76-0.85)</td>
</tr>
<tr>
<td>Average</td>
<td>0.77</td>
<td>0.82</td>
</tr>
</tbody>
</table>

* refers to a AC score below 0.7

The results of afore analyses are summarised in Table 5. The list of the accepted MOE-items including the answer options is presented in the appendix. The list has been forward and backward translated by two Dutch and two English SLTs, independent of each other. Discussion about the discrepancies in the translation brought consensus over the text.17
### Table 5. Summary of all results for all original MOE items.

<table>
<thead>
<tr>
<th>Items</th>
<th>Variance in score</th>
<th>Loading in factor</th>
<th>Cronbach’s alpha</th>
<th>Inter observer reliability</th>
<th>Decision</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lip closure</td>
<td>+</td>
<td>±</td>
<td>-</td>
<td>n.a.</td>
<td>-</td>
</tr>
<tr>
<td>Tongue protrusion</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Lateral tongue movement</td>
<td>+</td>
<td>+</td>
<td>±</td>
<td>+</td>
<td>±</td>
</tr>
<tr>
<td>Munching</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>±</td>
<td>+</td>
</tr>
<tr>
<td>Controlled bite</td>
<td>+</td>
<td>-</td>
<td>-</td>
<td>n.a.</td>
<td>-</td>
</tr>
<tr>
<td>Jaw movement</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>±</td>
<td>+</td>
</tr>
<tr>
<td>Chewing duration</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Loss of food or saliva</td>
<td>+</td>
<td>±</td>
<td>+</td>
<td>±</td>
<td>+</td>
</tr>
<tr>
<td>Gagging or disgust</td>
<td>-</td>
<td>n.a.</td>
<td>n.a.</td>
<td>n.a.</td>
<td>-</td>
</tr>
<tr>
<td>Coughing or aspiration</td>
<td>-</td>
<td>n.a.</td>
<td>n.a.</td>
<td>n.a.</td>
<td>-</td>
</tr>
<tr>
<td>Displacement of food with fingers</td>
<td>-</td>
<td>n.a.</td>
<td>n.a.</td>
<td>n.a.</td>
<td>-</td>
</tr>
<tr>
<td>Displacement with head movement</td>
<td>-</td>
<td>n.a.</td>
<td>n.a.</td>
<td>n.a.</td>
<td>-</td>
</tr>
<tr>
<td>Number of swallows</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Fluency and coordination</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
</tbody>
</table>

1. Variance in score:  
- = maximum score in both groups > 95%
2. Loading:  
+ = >0.40 in at least 2 out of 4 analyses of consistency and groups
± = >0.40 in 1 out of 4 analyses of consistency and groups
3. Cronbach’s alpha:  
- = Cronbach’s alpha >0.70 if item was deleted in 2 out of 4 analyses
4. Inter observer reliability:  
+ = score $AC_2 > 0.70$ in all analysis
± = score $AC_2 < 0.70$ in 2 or 3 out of 4 analyses
n.a. = not applicable

**Construct validity**

We calculated frequency statistics using the median and maximum score of the three bite attempts. The mean scores were 24.7 (SD 3.4) and 22.8 (SD 3.4) for the healthy and CP groups, respectively. The difference between the mean of the median and maximum performance score for the healthy group was 2.0 and for the children with CP was 1.7. The results are displayed in boxplots in Figure 1.
Figure 1. Mean scores for bread for the scores for the healthy children and children with CP. These results concern 8 items of the MOE.

\[
r^2 \text{(healthy group)} = 0.543 \quad (p<.001) \\
r^2 \text{(CP group)} = 0.053 \quad (p=.05)
\]

Figure 2. The estimated curve of the MOE total score using the median score of three bites for healthy children (left plot) and for children with cerebral palsy (CP) (right plot) in the bread condition. These results concern 8 items of the MOE.

Figure 2 displays the results of the regression analysis with the total MOE scores as dependent variable and age (in months) as independent variable per group. Healthy children aged 6 to 48 months showed a smooth curve towards maximum performance, but the scores showed a lot of variation, especially in children younger than 12 months.
The total MOE score and age showed a correlation coefficient (r) of 0.73 (p<.01). In the CP group, there was a weak positive relationship between the MOE total score and age (r= 0.23; p=.05) and a weak negative relationship between GMFSC and MOE total score (r=-0.32; p<.01).

Floor and ceiling effects
We detected no floor or ceiling effects as the results of the MOE total median scores were normally distributed for both groups.

DISCUSSION
In this study, we have established that the MOE, an instrument developed by Remijn and associates\textsuperscript{16} for the analysis of mastication, has sufficient psychometric properties in both a group of healthy children (aged 6 to 48 months) and a group of children with CP (aged 24 to 72 months). In addition to the eight developmental items, the MOE contains several criterion-referenced items. These items are essential in order for an SLT to provide advice and intervention on consumption of solid foods. The original items were thought to be relevant for mastication, but in the previous and this present study several items were not reliably scored, provided information of the whole feeding session and could not be scored per bite, had no variation or did not contribute to the internal consistency. The MOE requires criterion-referenced ‘red flag’ items, such as gagging or choking, because when a child gags or chokes extreme caution is needed for the decision to offer a further bite of bread or biscuit. MOE items covering the child’s use of head movements or fingers to move a bolus laterally or from anterior to posterior in the mouth are compensational maneuvers and provide overall information on chewing and swallowing behaviors even as side preference of chewing, head and trunk stability.

In our previous study, we described mastication as a difficult construct to measure by means of observation. Our current results on the internal consistency are moderate and the deletion of two items (‘lip closure’ and ‘controlled bite’) increased the internal consistency to a sufficient level. For this reason, these two items were removed from the instrument. Removal of these items was supported by the PCA results that indicated the presence of only one factor when these items were removed. It is interesting to note that ‘lip closure’ and ‘controlled bite’ were items of considerable discussion in the Delphi rounds during the developmental phase of the MOE. Lip closure during the mastication process prevents food or saliva loss\textsuperscript{23} and the degree of lip closure provides information on whether lateral tongue movements could be identified.\textsuperscript{14,24} A controlled bite provided information about the bite force and would not to be one to one related to the chewing ability.\textsuperscript{3} As the items ‘lip closure’ and ‘controlled bite’ are related to the intake of solid food, they were included in
the MOE criterion-referenced list.

MOE items were developed for scoring on a 4-point ordinal scale to optimize its sensitivity. Our findings support those of others that it is difficult to observe the oral-motor movements required in chewing and swallowing. To detect lateral tongue movements while the lips are closed demands the interpretation of other movements. Trained SLT students, however, were able to score oral-motor behavior with sufficient reliability. This resulted in sufficient levels of agreement for all items except ‘munching’ and ‘lateral tongue movement’. The agreement for ‘lateral tongue movement’ (0.68) was just below the agreement threshold for the healthy group (0.70). This group had slightly more lip closure than the children with CP which makes it harder to view tongue movements for the healthy group.

The observers achieved sufficient agreement for the item on ‘munching’ only for the healthy group but the levels of agreement were low for the CP group. Although we are unable to explain this large difference in score, we accepted ‘lateral tongue movement’ and ‘munching’ as MOE items because of the PCA and internal consistency results.

Overall, the results of the inter-observer agreement were almost equal for both groups. This result differs from a previous pilot study and suggests that observer training in analyzing oral-motor behaviour is more effective when the training material includes more recordings from both groups of children (healthy and CP group). The training could be improved by including examples of every answer option (32 options) in an effort to improve the inter-observer reliability and improve differentiation between scores 3 and 4. Moreover, by reducing the number of items, the observer will be able to focus on the remaining items.

Healthy children aged 6 to 48 months showed a smooth curve to the maximum performance score. The shape of this curve concurs with the development of mastication as described in the literature, that is, fast development until 12 months of age followed by a period of gradual increase. Although we anticipated the maximum score of the MOE would be reached by the age of 48 months, this was not shown in our data. As observed in the mean total MOE score and standard deviation, children at this age displayed significant variability in performance. In addition, the chewing capacity improves after 4 years of age because of teeth eruption.

The children under 12 months of age showed a relatively good performance in both chewing tasks. This observation corresponds with previous research in which young children were able to process easy chewable foods from the age of 8 months. Our hypothesis that there would be differences in performance between younger and older children was supported. We observed that the younger children seemed more eager to eat than the older children and we felt that the performance of the older children was negatively influenced by the camera, the presence of the SLT students or both the camera and students during the recorded sessions. The results of the children with CP aged from 24 to 72 months...
showed a weak but increasing upward trend in performance scores until 72 months of age. Unlike healthy children, children with CP continue to develop mastication skills through to an older age. Although we expected large differences in chewing performance between healthy children and children with CP, this was not supported by our data: both groups of children showed variable performance in chewing and we observed a correlation between age and mastication performance. Note, however, that the groups were not matched for age (the healthy group was approximately 20 months younger) which makes it difficult to compare the results.

Although we had expected lower scores on all items for the children with CP compared to the healthy group, the results indicated a predominately good chewing performance for the CP group. There are several reasons for this observation: the sessions took place in an optimal environment; the sessions were short and included only five bites (it would be informative to assess the last bites at meal time); we included children who regularly ate bread (parents may have been more willing to let their child participate if the child ate bread well) and although we had a protocol for the way in which food was offered, if the child was not able to take it in the front of the mouth, bread was sometimes placed between the molars. Chewing is a sequence of repetitive movements of the jaw, tongue, and lips and a score (1 to 4) reflects the observer’s overall interpretation of the repetitive movements of a single bite. It is, therefore, understandable that the scores lacked specificity. Using the median score across the three bites reflects the overall performance score per item and the maximum score reflects an impression of capacity (i.e. what the child can achieve in optimal conditions). We found a weak correlation between GMFCS and the MOE scores ($r=-0.32; p<.01$), whereas other researchers reported a significant relationship between chewing problems and GMFCS scores.\textsuperscript{26,27} This difference in results may reflect the variation between the studies in the age of the participants and inclusion criteria.

This study is not without its limitations. Eighteen recordings of sessions in which bread was offered to children in the healthy children could not be included because the SLT student deviated from the study protocol. We were also unable to standardize the amount a child bit off in the biscuit condition meaning that we could not compare results between and within the groups. We propose that future studies offer the biscuit in fixed sizes. Finally, assessment of the recordings could have been biased as the observers were not blind to child age or which group the child was from (healthy vs. CP group). This is unavoidable when a tool is based on observation of a participant.

**CONCLUSION**

The definitive version of the MOE includes eight items to objectively evaluate observed oral-motor behaviors of mastication. The items achieved a sufficient level of internal consistency and construct validity and can be scored on a 4-point ordinal scale with sufficient
levels of reliability. The tool is sensitive to developmental changes in young children and is without floor and ceiling effects. Although the MOE will be suitable for analyzing oral-motor movements to provide a baseline for intervention, we suggest therapists include additional measurements for detailed information on intra-oral processes, such as lateral tongue movement or munching during the mastication process.
ACKNOWLEDGEMENTS

The authors are indebted to all parents and children, the rehabilitation centers Sint Maartenskliniek, Groot Klimmendaal, Rijndam and de Hoogstraat Rehabilitation as well as the affiliated schools for their participation. We thank Loes Arts, Mandy Diepeveen, Yvonne Hendrick, Anne Koenen, Julia Peeters, Sanne de Rijk, Sandra van Tuil, Mariken de Wilde, the SLT students of the University of Applied Sciences of the HAN who took part in this study. In addition, we thank Petri Holtus for her support during this project.

There were no financial grants or conflicts of interest for this project.
REFERENCES


APPENDIX

Mastication Observation and Evaluation items

1. **Tongue protrusion**
The tongue does not protrude beyond the teeth during mastication unless it is used to actively remove food from the lips (= functional use). Note: the rating for tongue protrusion should only be assessed during chewing and not for the swallowing stage.

Does the tongue extend beyond the teeth during mastication?
1. Yes; the tongue extends frequently beyond the teeth.
2. Yes; the tongue extends a few times beyond the teeth.
3. Yes; the tongue extends once beyond the teeth.
4. No; the tongue never extends beyond the teeth.

2. **Lateral tongue movement**
The tongue collects food pieces during mastication and places the food between the molars for grinding. If the tongue is not visible but there is observable temporary bulging of one of the cheeks or asymmetric activity in the corner of the mouth, this is an indication of lateral food transport and should be scored as present.

Are lateral tongue movements present?
1. No; there is no lateral tongue movement.
2. Yes; there is once a tongue movement.
3. Yes; there is a regular lateral tongue movement.
4. Yes; there is a constant adequate lateral tongue movement.

3. **Squashing or sucking movement**
The tongue moves independently of the jaw during mastication. There is no observable squashing or sucking tongue movement.

Are squashing or sucking tongue movements present?
1. Yes; there are constantly squashing or sucking tongue movements.
2. Yes; there is once a squashing or sucking tongue movement.
3. Yes; there are occasional squashing or sucking tongue movements.
4. No; there are no squashing or sucking tongue movements.

4. **Jaw movement**
The mandible makes predominately vertical and slightly horizontal movements during chewing.

Does the mandible move in varied directions?
1. No; only in vertical direction.
2. Yes; there is once an adequate movement from the midline.
3. Yes; there are regular adequate movements from the midline.
4. Yes; there are constant variable movements from the midline.
5. **Chewing duration**

The chewing duration is the period of time between food entering the mouth and the swallow. The chewing duration depends on the size of the mouthful and the food consistency.

Is the chewing duration adequate for the size of the mouthful and the food texture?
1. No; there is no chewing or munching present.
2. No; chewing duration is much too short or much too long to be adequate.
3. Yes; chewing occurs but the duration is a little too short or a little too long.
4. Yes; there is an adequate chewing duration.

6. **Loss of food or saliva**

Food and/or saliva do not escape the mouth during mastication. Note: This applies to food that has already been in the mouth.

Is loss of food and/or saliva present?
1. Yes; there is a constant or a lot of loss of food and/or saliva.
2. Yes; there is a regular loss or small amount of food and/or saliva.
3. Yes; there is once a loss of a very small amount of food and/or saliva.
4. No; there is never a loss of food and/or saliva.

7. **Number of swallows**

One or two swallows are required to swallow a small bolus.

Is the bolus adequately swallowed?
1. No; the child doesn’t complete the swallow.
2. No; the bolus requires multiple swallows.
3. Yes; the bolus requires two swallows.
4. Yes; the bolus requires a single swallow.

8. **Fluency / coordination**

Mastication movements are rhythmic in coordinated movements.

Is chewing rhythmic and fluent?
1. No; chewing is never rhythmic and fluent.
2. No; chewing is sometimes rhythmic with fluent or coordinated movements.
3. Yes; chewing is mostly rhythmic with fluent and/or coordinated movements.
4. Yes; chewing is constantly rhythmic with fluent and coordinated movements.

*All MOE items have been translated using a forward and backward translation procedure. Translations were completed independently by two native Dutch speakers and two native English speakers. Any differences in translations were resolved during consensus.*
chapter 6

Ultrasound imaging for analyzing lateral tongue movements during mastication in adults with cerebral palsy compared to adults without oral motor disabilities

L Remijn
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BE Groen
CL de Korte
Chapter 6

ABSTRACT

This technical report describes an ultrasound technique to study tongue movements, particularly lateral tongue movements, during mastication. A method to analyze spatial and temporal tongue movements was developed and the feasibility of this method was evaluated. Biplane ultrasound images of tongue movements of four adults without oral-motor disabilities and two adults with oral-motor disabilities as result of cerebral palsy were acquired. Tongue movements were analyzed in the coronal and sagittal planes using B-mode and M-mode ultrasonography. The inter-rater and intra-rater agreement for manual tracing of tongue contours was good (ICC = 0.81 and 0.84, respectively). There were significant differences between two adult groups for movement frequency in the horizontal direction in both coronal and sagittal planes. In the coronal plane, differences were found for movement frequency and range of vertical movement. Data obtained from sagittal images, with exception of vertical frequency, indicated no differences between the groups.

The protocol developed in this study (using B-mode and M-mode) proved to be valid and reliable. By applying this protocol to individuals with and without oral-motor disabilities, we were able to demonstrate the clinical application of our protocol for evaluating differences in tongue movements during mastication.
INTRODUCTION

Effective mastication is characterized by efficiently fragmenting, crushing and mixing ingested food to form a bolus ready for swallowing. The tongue is vital for moving and positioning food between the teeth, for selecting food fragments for further mastication, for mixing saliva with the food and for posterior transportation of the bolus into the oropharynx. Neuromuscular disorders of the central nervous system (e.g. cerebral palsy) may have profound effects on the tongue muscles and thus on the coordination of mastication but the exact nature of the disordered movements is difficult to define and measure. For such individuals, clinical examination of tongue movements during mastication is required to develop an individual intervention plan.

Clinical examination of tongue positioning and tongue movements during mastication is difficult because of the tongue’s concealment within the oral cavity and the constant dimensional changes in tongue shape and tongue position. Ultrasound has been widely used for evaluating intra-oral structures, such as the tongue. Ultrasound is a safe, non-invasive procedure and is relatively easy to perform. Ultrasound has been used to analyze relatively rapid tongue movements associated with articulation, sucking and swallowing. The repetitive tongue movements made during mastication, however, are more complex than the movements of articulation or sucking/swallowing because the tongue contour changes as the consistency of the food changes and the bolus is formed. Submental transducer placement is generally used to obtain real-time images of the oral cavity. These images are displayed in two modes (B-mode and M-mode) to quantify differences in duration, range, and speed of tongue movements. B-mode images allow visualization of the changing tongue surface contours (i.e. tongue shape). M-mode images allow visualization of the tongue surface movements along the scan line over time (Figure 1). Blair et al. and Bressmann et al. completed a quantitative analysis of four coronal tongue images (B-mode and M-mode) taken during chewing and speech tasks. Both studies analyzed four anterior to posterior images. Unlike the standard B-mode transducer used in two studies of Blair et al. and Bressmann et al., Burton et al. used a matrix-array transducer to create three-dimensional tongue images to track tongue movements made during breast feeding.

The studies mentioned in the previous paragraph differ in several methodological aspects, namely the locations for measuring tongue movements, the measurement parameters and the parameter calculations. Since the tongue movements of mastication are complex, we used a semi-three-dimensional approach involving biplane imaging. Analysis of data from biplane imaging requires an updated protocol and analysis tool. The previous mentioned studies had limited data as they only included twelve measurement
points (four points on the sagittal view and three points on the coronal view). The primary aim of the present study was to develop a method using ultrasound to identify an efficient method to explore spatial and temporal tongue movements and to establish parameters to quantify lateral tongue movement during mastication [Part 1]. The secondary aim was to investigate differences in tongue movements during mastication between individuals with and without oral-motor disabilities [Part 2]. We hypothesized that individuals with oral-motor disabilities will demonstrate less frequent lateral tongue movements and a lower lateral movement rate and range than individuals without an oral-motor disability.

**METHODS**

**Part 1. Developing the method of data analysis**

**Ultrasound recording procedure**

We simultaneously recorded coronal and sagittal images to allow visualization of a movement in two planes. This is termed biplane imaging. We used an iE33 real-time 3D ultrasound scanner equipped with an X7-2 matrix-array transducer (Philips Ultrasound, Andover, MA, USA). Ultrasound images were stored as DICOM (Digital Imaging and Communications in Medicine) files for further data processing.

The transducer was covered with gel and positioned submentally and parallel to the participant’s neckline. The transducer was moved until the clearest tongue image was found while keeping the two orthogonal imaging planes in the coronal and sagittal directions. While a participant chewed, the researcher maintained contact between the transducer and the participant by using her thumb and index finger to apply light upward pressure to stabilize the transducer. This allowed the transducer to move with the jaw during mastication (similar to the protocol described by Stone.5

**Ultrasound image analysis**

A custom-made post-processing and analysis protocol was developed using MATLAB (release 14; The Mathworks Inc., Natick, MA, USA). Tongue movement was quantified by changes in tongue shape over time observed in both B-mode and M-mode images. In this way, we tracked the interface between the tongue and the oral cavity (tongue contour). Interactive selection of a single tongue contour on a B-mode image required the placement of a marker (clicks) up to eight times per frame. A full analysis would have required ±4000 (8 x 500 frames) clicks. Therefore, the motion component was analyzed by extracting eight M-modes images in which the contour was interactively selected by placing markers at distinct points in time. This resulted in a reduction of the number of clicks by approximately a factor 4.
A back-scan conversion procedure was implemented for both images (coronal and sagittal) because the sector data is displayed as scan-converted (pie shape) on the DICOM data. As displayed in Figure 1, 40 echo lines with a spacing of 1.5 degrees were determined while keeping the number of samples per echo line identical. The corresponding echo levels for the individual echo lines were obtained by applying a 2D cubic interpolation between the DICOM data and the back-scan grid (Figure 1).

![Original DICOM biplane sectors with marked sector calibrations (red dots: coronal plane; yellow dots: sagittal plane) and estimated M-lines positions for back scan conversion.](image)

The back-scanned converted data was then divided into eight zones of five image lines. From these eight zones, we created M-lines by taking the maximum echo level versus depth and plotting them over time. M-lines were used to interactively trace the tongue. Line interpolation was applied to M-mode coordinates with a distance smaller than 50 pixels (i.e., 0.76 s as frame rate was 38 Hz) and gaps larger than 50 pixels were automatically marked as missing data and were thus not interpolated. This procedure was repeated for all M-lines to create a full contour.

Based on the tracked M-mode contours (Figure 2), the B-mode tongue contours per frame were reconstructed (Figure 3; red dots) after transformation (scan-conversion) to the sector-shaped DICOM data coordinates using equations 1 and 2.

\[
X_{bmode} = Y_{mmode} \cdot \sin(Mline_{alpha} \cdot \pi/180)\\
Y_{bmode} = Y_{mmode} \cdot \cos(Mline_{alpha} \cdot \pi/180)
\]  

A polynomial fit (5th order for coronal contours and 4th order for sagittal contours) was applied through these B-mode points to obtain the tongue contour (Figure 3; blue lines).
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Ultrasound imaging for analyzing lateral tongue movement during mastication in adults

To trace the initial contours or to overrule interpolated M-mode contours, interactive B-mode tongue contour selection was also implemented. A polynomial fit through the B-mode contour was applied before B-mode points were added to the M-mode contours. The intersections of the fitted contour with the central zone lines were then estimated (Figure 3; red dots on dotted M-lines). Contour artifacts were caused by missing points in the M-mode and these missing points were corrected by manually adding points in the B-mode contour. Sometimes, a double line on the image occurred when the transducer was incorrectly positioned.

Figure 2. M-mode (polair) tongue movement. This image is one of the eight extracted zones. Every zone is depicted as the movement over time for a specific zone of the total view. Yellow stars = manually selected M-mode coordinates. Red line = interpolated line between yellow stars

Figure 3. Reconstructed coronal (left image) and sagittal (right image) B-mode contours of tongue movement. The yellow point marks the highest point of the tongue. The positions of the red points are derived from the manually tracked and interpolated M-mode contours.
Variables

The maximum tongue contour position (highest vertical point along the tongue contour) was chosen as the point to be tracked in time (Figure 3; yellow dot). All study parameters (described below) were calculated using the maximum tongue contour position as the reference point. Changes in tongue movement were recorded based on the relative X and Y positions, where X is defined relative to the center of the sector and Y relative to the averaged absolute Y position (Figure 4). Variables included frequency of vertical movements (Up-Down: UD<sub>freq</sub>) and frequency of horizontal movements (Left-Right: LR<sub>freq</sub>). Only up-down events in which the Y movement exceeded a ±5mm threshold were considered vertical movements to prevent the inclusion of accidental movements in the frequency count. A left-right event was included in the frequency count when the X max-point exceeded the reference X-point for > 6/frame rate (167 ms). (Figure 3a; red vertical dotted lines).

Up-Down frequency:

\[
UD_{freq} = \frac{\text{number of Up-Down events}}{\text{analyzed mastication time}}
\]  

[3]

Left-Right frequency:

\[
LR_{freq} = \frac{\text{number of Left-Right events}}{\text{analyzed mastication time}}
\]

[4]

Displacement and velocity parameters (Figure 4b and 4c) were derived as average values over time for one food trial and calculated according to equations 5 to 10.

Horizontal displacement:

\[
X_d = \Delta x = x_1 - x_2
\]

[5]

Vertical displacement:

\[
Y_d = \Delta y = y_1 - y_2
\]

[6]

Total displacement:

\[
R_d = \sqrt{\Delta x^2 + \Delta y^2}
\]

[7]

Horizontal velocity:

\[
X_v = \frac{\Delta x}{\Delta t}
\]

[8]

Vertical velocity:

\[
X_v = \frac{\Delta x}{\Delta t}
\]

[9]

Total velocity:

\[
R_v = \sqrt{X_v^2 + Y_v^2}
\]

[10]
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Figure 4. Results curves for the relative position of the tongue maximum (in centimeters) (4a), tongue displacement (in centimeters) (4b) and velocity of tongue movement (in centimeters/second) (4c) of one bite. Red= x-axis, blue= y-axis, black= resultant of both motions along x-axis and y-axis.

Part 2. Evaluating the method

Participants

Four participants without neurological and oral-motor disabilities (referred to as ‘controls’) [1 male and 3 females; mean age 35.6 years (SD 11.4; range 23-47 years] and two participants with congenital cerebral palsy (CP) [2 men; mean age 24.5 years; range 23-25 years; both with spastic CP, tetraplegic, Gross Motor Function Classification Scale III and moderate oral-motor feeding disabilities and moderate dysarthria] participated in the study. Participants in both groups had full permanent dentition. None of the participants reported intra-oral discomfort during the ultrasound session.

The participants in the control group were recruited from the Medical Ultrasound Imaging Center of the Radboud University Medical Center and the participants with CP were former clients of the Sint Maartenskliniek (participants had no therapeutic relationship with the researchers). In compliance with the World Medical Association Declaration of Helsinki:
Ethical principles for medical research involving human subjects (2008), participants received written information on the study and oral instructions regarding the ultrasound procedure before signing an informed consent form. The Ethics Committee at ‘Slotervaart Hospital and Reade Rehabilitation Center’ in Amsterdam (the Netherlands) declared that no formal approval of the detailed protocol was needed according to the Dutch Medical Research Involving Human Subjects Act (number U/14.142).

Data collection
Each participant completed three food trials. A 1.5 x 1.5 cm slice of wheat bread spread with pate was used at each trial. The first author (LR) held the transducer under the chin while the participant chewed and swallowed each food trial. The participant was asked to sit comfortably and to eat the piece of bread in their usual way. Ultrasound recording began when the food was in the participant’s mouth and imaging ceased as the participant swallowed. Imaging session duration was approximately 15 minutes per participant.

Two raters (author LR and student MS) independently traced the tongue contours per food trial. To ensure that both raters measured the same frames, we selected the frames between the start of chewing and the initiation of the swallow (Figure 5; yellow right stripe). For part 1, the intra-rater and inter-rater reliability were calculated over the manual click data from the first food trial. Data from three of the four controls was used to calculate intra-rater reliability and data from two participants with CP was used to calculate inter-rater reliability.

In part 2, we investigated patterns in tongue movements during chewing and pattern differences between the participant groups. Therefore, three food trials of three controls and one food trial of one control (two trials failed) and three food trials of two participants with CP, achieved by the first author (LR), were used. For the pattern in the parameters, the average value of the tongue movements of 10 food trials of controls and 6 food trials of CP-participants were used to analyze tongue displacement, velocity in three directions and movement frequency (LRfreq and UDfreq). The coronal and sagittal images were analyzed for all food trials.
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Figure 5. M-modes overview for data selection with marked area (between vertical yellow solid lines) for results estimation.

Statistical analysis
All analyses were performed using Statistical Package for the Social Sciences (SPSS, version 20.0). In part 1, the intraclass correlation coefficients (ICCs) were calculated to establish the inter-rater and intra-rater reliability of the tongue contour lines (consistency, two-way random and two-way single measurements). Consistency measures indicate whether there is a consistent pattern of differences between scores. Missing data from one or both raters were excluded from reliability analysis. An ICC value of 0.70 was considered acceptable.25 In part 2, descriptive statistics were used to analyze tongue displacement, velocity in three directions, and movement frequency (LRfreq and UDfreq). A non-parametric Mann-Whitney U test was performed to determine whether there were differences between the participant groups for each variable. This test was selected because of the small sample size. We used $p<.05$ as significance level for all statistical tests.
RESULTS

Part 1. Developing the method of data analysis

The developed method provided an appropriate manner to draw the tongue surface of M-mode slides. The B-mode was automatically interpolated and a manual check confirmed an accurate tongue surface contour line. Approximately 45 minutes were required to upload and process a participant’s coronal and sagittal images. The average length of the recordings from the start of chewing to the first swallow was 507.7 frames (range 407 - 645 frames) or 13.3 seconds (range 10.7 - 17.0 s).

The ICC for the inter-rater reliability of the tongue contour lines was 0.77 (range 0.29 - 0.93) for the coronal images and 0.86 (range 0.55 - 0.95) for the sagittal images. The average number of missing pixels was 10.7% (range 0 - 49.6%) for coronal images and 9.3% (range 0 - 30.3%) for sagittal images (Table 1).

Table 1. Inter-rater reliability of the traced tongue contours by two raters. The participants have no oral-motor disabilities (control group).

<table>
<thead>
<tr>
<th>Zones</th>
<th>Participant 1</th>
<th>Participant 2</th>
<th>Participant 3</th>
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<tbody>
<tr>
<td></td>
<td>Missing (%)</td>
<td>ICC</td>
<td>Missing (%)</td>
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<tr>
<td></td>
<td>Coronal</td>
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<tr>
<td>1</td>
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<td>31.2</td>
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<tr>
<td>2</td>
<td>33.2</td>
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<tr>
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<td>3.4</td>
<td>0.92</td>
<td>11.2</td>
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<td>0.90</td>
<td>3.1</td>
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<tr>
<td>7</td>
<td>0.0</td>
<td>0.93</td>
<td>0.0</td>
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<td>8</td>
<td>3.4</td>
<td>0.93</td>
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<tr>
<td></td>
<td>Sagittal</td>
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<td>1</td>
<td>12.7</td>
<td>0.91</td>
<td>3.1</td>
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<td>8</td>
<td>6.4</td>
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Intra-rater reliability was 0.87 (range 0.84 - 0.91) for rater 1 and 0.81 (range 0.78 - 0.87) for rater 2 (Table 2). The ICC scores for coronal images were generally stronger than the sagittal images. Although all eight zones were visualized, each zone did not always have a clear ultrasound image for all frames and this resulted in missing data points. Missing data point resulted in lower ICC values.

**Table 2.** Intra-rater reliability of the manual clicking of 3 food trials of controls and 2 food trials of participants with CP.

<table>
<thead>
<tr>
<th></th>
<th>Participant 1</th>
<th>Participant 2</th>
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</tr>
<tr>
<td>8</td>
<td>5.8</td>
<td>0.87</td>
<td>6.4</td>
<td>0.92</td>
</tr>
</tbody>
</table>
Part 2. Evaluating the method
Due to an error in transducer placement, two recordings of one control were unclear and were excluded from data analysis. Therefore, ten food trails were available for data analysis. The descriptive statistics for displacement per frame (cm) and velocity (cm/s) and for UDfreq and LRfreq are listed in Table 3 (coronal images) and Table 4 (sagittal images). For the control group, the standard errors for the sagittal images were relatively small (3.8% [R-velocity] to 14.7% [LRfreq]). For the participants with CP, the standard errors for sagittal images of most variables (except the Y-displacement and UD 95% range) were approximately twice the size of the control group’s standard errors. With the exception of Y-displacement and X-displacement, the standard errors obtained from coronal images were similar to results from the sagittal images. The variance was larger for the controls than for the participants with CP for measures of Y-displacement (76.1% and 44.4%, respectively) and X-displacement (52.9% and 11.4%, respectively). In the coronal view, Y-displacement (p=.019), LRfreq (p=.008) and UDfreq (p=.04) were significantly larger for the controls. The Y-velocity showed a trend for a group difference (p=.099), with a higher value for the control group. In the sagittal view, only the UDfreq was significantly higher for the controls (p=.019).

Table 3. Results of coronal view. Mean (SD), range and SEM of the control group and CP group. In the last column lists the p-value of the differences between both groups.

<table>
<thead>
<tr>
<th></th>
<th>Tongue movements of control group n=10</th>
<th>Tongue movements of CP group n=6</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (SD) range</td>
<td>SEM</td>
<td>Mean (SD) range</td>
</tr>
<tr>
<td>Frames analyzed</td>
<td>491.40 (79.94)</td>
<td>25.28</td>
<td>503 (63.25)</td>
</tr>
<tr>
<td></td>
<td>407 - 684</td>
<td>471 - 616</td>
<td></td>
</tr>
<tr>
<td>R-displacement (cm)</td>
<td>0.187 (0.025)</td>
<td>0.008</td>
<td>0.158 (0.036)</td>
</tr>
<tr>
<td></td>
<td>0.16 - 0.23</td>
<td>0.11 - 0.19</td>
<td></td>
</tr>
<tr>
<td>Y-displacement (cm)</td>
<td>0.071 (0.017)</td>
<td>0.054</td>
<td>0.054 (0.005)</td>
</tr>
<tr>
<td></td>
<td>0.04 - 0.11</td>
<td>0.05 - 0.06</td>
<td></td>
</tr>
<tr>
<td>X-displacement (cm)</td>
<td>0.151 (0.026)</td>
<td>0.008</td>
<td>0.132 (0.034)</td>
</tr>
<tr>
<td></td>
<td>0.12 - 0.20</td>
<td>0.09 - 0.16</td>
<td></td>
</tr>
<tr>
<td>R-velocity (cm/s)</td>
<td>7.50 (1.759)</td>
<td>0.556</td>
<td>6.131 (2.097)</td>
</tr>
<tr>
<td></td>
<td>5.79 - 10.94</td>
<td>3.36 - 8.44</td>
<td></td>
</tr>
<tr>
<td>Y-velocity (cm/s)</td>
<td>2.043 (0.623)</td>
<td>0.197</td>
<td>1.562 (0.325)</td>
</tr>
<tr>
<td></td>
<td>1.34 - 3.59</td>
<td>1.12 - 1.90</td>
<td></td>
</tr>
<tr>
<td>X-velocity (cm/s)</td>
<td>6.705 (1.744)</td>
<td>0.551</td>
<td>5.573 (2.052)</td>
</tr>
<tr>
<td></td>
<td>5.00 - 10.26</td>
<td>2.89 - 7.88</td>
<td></td>
</tr>
<tr>
<td>LR frequency (Hz)</td>
<td>1.240 (0.363)</td>
<td>0.115</td>
<td>0.580 (0.402)</td>
</tr>
<tr>
<td></td>
<td>0.70 - 1.80</td>
<td>0.10 - 1.00</td>
<td></td>
</tr>
<tr>
<td>UD frequency (Hz)</td>
<td>2.390 (0.120)</td>
<td>0.038</td>
<td>1.600 (0.616)</td>
</tr>
<tr>
<td></td>
<td>2.20 - 2.60</td>
<td>1.00 - 2.50</td>
<td></td>
</tr>
<tr>
<td>LR 95% range (cm)</td>
<td>2.083 (0.371)</td>
<td>0.117</td>
<td>2.250 (0.410)</td>
</tr>
<tr>
<td></td>
<td>1.54 - 2.72</td>
<td>1.88 - 2.93</td>
<td></td>
</tr>
<tr>
<td>UD 95% range (cm)</td>
<td>1.142 (0.259)</td>
<td>0.082</td>
<td>1.177 (0.165)</td>
</tr>
<tr>
<td></td>
<td>0.61 - 1.56</td>
<td>1.00 - 1.39</td>
<td></td>
</tr>
</tbody>
</table>
**Table 4.** Results of sagittal view. Mean (SD), range and SEM of the control group and CP group. In the last column lists the $p$-value of the differences between both groups.

<table>
<thead>
<tr>
<th></th>
<th>Mean (SD) range</th>
<th>SEM</th>
<th>Mean (SD) range</th>
<th>SEM</th>
<th>$p$-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>R-displacement (cm)</td>
<td>0.194 (0.025)</td>
<td>0.008</td>
<td>0.174 (0.027)</td>
<td>0.012</td>
<td>.254</td>
</tr>
<tr>
<td>Y-displacement (cm)</td>
<td>0.068 (0.015)</td>
<td>0.005</td>
<td>0.062 (0.011)</td>
<td>0.005</td>
<td>.513</td>
</tr>
<tr>
<td>X-displacement (cm)</td>
<td>0.164 (0.022)</td>
<td>0.007</td>
<td>0.146 (0.027)</td>
<td>0.012</td>
<td>.206</td>
</tr>
<tr>
<td>R-velocity (cm/s)</td>
<td>8.215 (0.999)</td>
<td>0.316</td>
<td>7.227 (1.746)</td>
<td>0.781</td>
<td>.371</td>
</tr>
<tr>
<td>Y-velocity (cm/s)</td>
<td>1.967 (0.379)</td>
<td>0.119</td>
<td>1.804 (0.438)</td>
<td>0.196</td>
<td>.859</td>
</tr>
<tr>
<td>X-velocity (cm/s)</td>
<td>7.500 (1.021)</td>
<td>0.323</td>
<td>6.547 (1.720)</td>
<td>0.769</td>
<td>.371</td>
</tr>
<tr>
<td>LR frequency (Hz)</td>
<td>0.91 (0.423)</td>
<td>0.134</td>
<td>0.62 (0.726)</td>
<td>0.324</td>
<td>.165</td>
</tr>
<tr>
<td>UD frequency (Hz)</td>
<td>2.18 (0.274)</td>
<td>0.087</td>
<td>1.540 (0.472)</td>
<td>0.211</td>
<td>.019</td>
</tr>
<tr>
<td>LR 95% range (cm)</td>
<td>2.29 (0.402)</td>
<td>0.127</td>
<td>2.676 (0.705)</td>
<td>0.315</td>
<td>.513</td>
</tr>
<tr>
<td>UD 95% range (cm)</td>
<td>1.117 (0.214)</td>
<td>0.067</td>
<td>1.33 (0.077)</td>
<td>0.034</td>
<td>.129</td>
</tr>
</tbody>
</table>

**DISCUSSION**

In this study, we used semi three-dimensional ultrasound imaging for quantifying tongue movement during mastication. In according to previous studies, we have proven that ultrasound is a non-invasive assessment and a suitable procedure for assessing the tongue movements. In Part I we developed a study protocol to ensure consistent ultrasound data collection and data analysis conditions. The protocol described the eight coronal and sagittal M-modes images, constructed out of the biplane data, to track the tongue contour. These M-mode contours were recoded to B-mode positions from which the tongue contour was derived. By determining the highest point of the tongue contour in time, the distance and velocity of tongue movements could be measured. Compared to Bressmann et al.\textsuperscript{14}, our measurement protocol was more detailed as we had 64 measurement points (8 zones x 8 points) across two measurement planes instead of 48 measurement points.

In our study, approximately 10 percent of pixels in 500 frames were classified as missing data. Data loss was likely caused by air in the oral cavity as result of chewing with an open mouth. In particular, one participant with CP consistently used an open mouth position during chewing and this resulted in poor image quality in some parts of the ultrasound
images. Parthasarathy et al.\textsuperscript{26} also reported data loss at tongue tip and tongue root during a speech task; the tongue tip was unclear due to air beneath it and the tongue root was obscured by the hyoid bone shadow. In our study, we noticed more data loss for coronal images than for sagittal images. For one participant, we noted an insufficient view in one of the most lateral zones in the coronal views. Our hypothesis is that these zones were not visible due to the presence of air when the bolus was at the opposite side in the mouth. In addition, the apex of the tongue is thinner and could provide a clearer image of the tongue contour during mastication than the middle part of the tongue in the coronal view and/or the hyoid bone could provide a shadow as mentioned by Parthasarathy et al.\textsuperscript{26} We do not believe the missing data had a large influence on the parameters because the traced M-mode images were interpolated to the B-mode and two parts of the image (contour decline and the highest point) were manually checked. The parameters were counted on the highest point on the traced line.

On several occasions, we observed double-line artifacts in the M-mode ultrasound images. Stone suggested these artifacts are caused by sound refraction when the transducer is held slightly off midline. Inconsistency in transducer positioning is a critical factor for outcome evaluation, and minimizing changes in transducer angles across sessions is crucial for research accuracy.\textsuperscript{5}

The detection of the highest tongue position, based on the tongue contour in the B-mode was difficult when the tongue was centrally grooved (i.e. with two high lateral tongue positions) or the tongue was relatively flat. We suggest a stricter use of the protocol for the ultrasound recordings and the use of a standardized control procedure for the M-mode and the B-mode to improve identification of the highest tongue position.

The second goal of our study was to investigate differences in tongue movements during mastication between controls and participants with CP. Although we had hypothesized a larger lateral tongue displacement for controls compared to the participants with CP, the difference was not significant (as seen on the coronal images). Y-displacement on the coronal images, however, differed significantly between the groups and there was less variation in the amount of tongue movement for controls compared to the participants with CP. The large variability in the data for the participants with CP is also due to the heterogenic nature of the group and the small sample size.

The frequency of both horizontal and vertical tongue movements on the coronal images significantly differed between the groups. Our results suggest that tongue movements of the participants with CP in our study were slower and had a smaller range of motion on the Y-axis than the control group. In contrast with the coronal images, data obtained from sagittal images, with exception of vertical frequency, indicated no differences in the food trials between the groups. Given our limited sample size, we are unable to generalize results beyond the subjects in our study. These results should be confirmed in a study with a larger sample size.
CONCLUSION

The results of this study indicate that ultrasound images of tongue movements are promising for the clinical analysis of mastication. The protocol developed in this study (using B-mode and M-mode) proved to be valid and reliable in individuals with and without oral-motor disabilities. Measurements of range of the tongue movement and frequency of tongue movement on the coronal B-mode images seemed to allow differentiation between controls and participants with CP. These results require confirmation in a study with a larger sample size. In a subsequent study, we will include a larger group of children with CP with a homogeneous classification with oral-motor disabilities and controls.
ACKNOWLEDGEMENTS

The authors thank Robin van Dungen, student of Fontys Hogeschool in Eindhoven, for his assistance with the Matlab analysis of the raw tongue contours and Mike Stoopendaal, student of HAN University of Applied Sciences in Nijmegen, for manual drawings of the B-mode en M-mode. The authors wish to thank Renee Clapham for her corrections of the English text.
REFERENCES


chapter 7

Reproducibility of 3D kinematics and surface electromyography measurement of mastication

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BE Groen
R Speyer
J van Limbeek
MWG Nijhuis-van der Sanden

Physiology & Behavior 2016; 155: 112–121
ABSTRACT

The aim of this study was to determine the measurement reproducibility for a procedure evaluating the mastication process and to estimate the smallest detectable differences of 3D kinematic and surface electromyography (sEMG) variables. Kinematics of mandible movements and sEMG activity of the masticatory muscles were obtained over two sessions with four conditions: two food textures (biscuit and bread) of two sizes (small and large). Twelve healthy adults (mean age 29.1 years) completed the study. The second to the fifth chewing cycle of 5 bites were used for analyses. The reproducibility per outcome variable was calculated with an intraclass correlation coefficient (ICC) and a Bland-Altman analysis was applied to determine the standard error of measurement relative error of measurement and smallest detectable differences of all variables.

The ICCs ranged from 0.71 to 0.98 for all outcome variables. The outcome variables consisted of four bite and fourteen chewing cycle variables. The relative standard error of measurement of the bite variables was up to 17.3% for ‘time-to-swallow’, ‘time-to-transport’ and ‘number of chewing cycles’, but ranged from 31.5% to 57.0% for ‘change of chewing side’. The relative standard error of measurement ranged from 4.1% to 24.7% for chewing cycle variables and was smaller for kinematic variables than sEMG variables.

In general, measurements obtained with 3D kinematics and sEMG are reproducible techniques to assess the mastication process. The duration of the chewing cycle and frequency of chewing were the best reproducible measurements. Change of chewing side could not be reproduced. The published measurement error and smallest detectable differences will aid the interpretation of the results of future clinical studies using the same study variables.
INTRODUCTION

Mastication, or chewing, is the ability to process solid and lumpy food textures for safe swallowing and is the first step in the digestive process. The rhythmic mandibular movements (opening and closing) alternate with tongue movements to position food between molars and to select food fragments for further crushing and/or mixing with saliva before the resulting bolus is transported into the oropharynx for deglutition.1-5

Good understanding of the chewing process is needed for diagnostic, intervention and evaluative purposes and to achieve this, an adequate assessment tool is essential.6,7 Observational assessments based on video recordings are commonly used in clinical practice because they are easy to conduct, are non-invasive and are inexpensive.8,9 The disadvantages of observational assessments include movements of other facial structures overshadowing visualization of the mandible10 and, in case of lip closure, the impossibility to verify the intra-oral activity of the tongue objectively.9 Detailed measurement of mandibular and tongue movements requires the inclusion of objective and validated measurement techniques.

Kinematic and/or surface electromyographic (sEMG) measurements are regularly used for research purposes but are not yet used in routine clinical practice. The 3D kinematics enables assessment of mandibular movements and offers insight into the stability of the chewing process.1,11-13 Muscle coordination and muscle force produced during mastication can be evaluated using sEMG.10,14-17 Kinematic and sEMG measurements are often simultaneously collected11,12,17-19, but the kinematic and sEMG outcome variables reported are diverse; differences in measurement protocol and systems result in a variety of outcome variables with related but distinct definitions.6 Although many studies have focused on evaluating interventions or comparing patient groups17,20, the reproducibility and/or sensitivity of the variables have not been thoroughly investigated. Knowing measurement reproducibility is important for interpreting study results.21 To date, reproducibility has been determined based on the number of chewing cycles using chewing gum22 or has focused on the sEMG signals of masseter and temporalis muscles during maximum clenching.16,22,23

Reproducibility has two components: relative and absolute reproducibility.24 The relative component is the degree to which individuals maintain their position in a sample over repeated measurements and can be reported with the intraclass correlation coefficient (ICC). The absolute component concerns the degree to which variables vary among subjects and is expressed in the units of the original measurement or as a proportion of the measurement values.24 Examples are Bland-Altman’s limits of agreements, standard error of measurement (SEM) and the smallest detectable difference (SDD).21 The SEM and SDD allow clinicians to consider a tool’s measurement error when determining whether an observed change is a real change.24
The purpose of this study is (1) to determine the reproducibility of several measurement variables using 3D kinematic and sEMG to evaluate the mastication process of healthy adults and (2) to estimate the SDD of the reproducible variables.

**METHODS**

**Participants**
Participants were recruited from within the Research Department of the Sint Maartenskliniek. Participants with known dental problems, mouth pain, loose teeth or dentures/implants or problems with the temporomandibular joint were excluded from the study. Twelve healthy adults participated in this study (six men and six women; mean age ± SD: 29.1 ± 9.9 years). In compliance with the World Medical Association Declaration of Helsinki (2008), written informed consent was obtained from participants prior to participation. The Ethics Committee at ‘Slotervaart Hospital and Reade Rehabilitation Center’ in Amsterdam (the Netherlands) decreed that formal approval of the detailed protocol was not required according to the Dutch Medical Research Involving Human Subject Act (number U/14.142).

**Experimental procedure**
The participants attended two experimental sessions (test-retest) held on the same day with at least two hours between sessions. During data collection, participants were seated in an upright position in a chair and instructed to keep their heads forwards and to keep head movements to a minimum. Each session started with a maximum voluntary clenching (MVC) measurement in which the participant was asked to bite as hard as possible for 5 s on a flexible chewing tube. Three MVC measurements were recorded with a 1-min interval. The participant then consumed five small (1.5 x 1.5 cm) and five large (1.5 x 3 cm) pieces of wheat bread with a chocolate spread and a crunchy biscuit (total of 20 pieces). Participants placed each piece of food in her/his mouth and were requested to eat as naturally as possible. Between trials participants could take a sip of water or take a short break.

The different textures in four conditions: bread or biscuit and small or large size were offered in randomized order in the first session and in reversed order in the second session (Figure 1). Data collection started when food entered the mouth and was stopped after swallowing the food. The 3D kinematic and sEMG data were recorded simultaneously. The first and last trial of each session consisted of a reference trial in which the participant was asked to stabilize the head for 5 s.
To allow measurement of the 3D movements of the mandible, custom-made Polyvinylchloride marker frames consisting of reflective markers of 8 mm in diameter were attached to a participant’s head, mandible and thyroid cartilage using hypoallergenic tape (Figure 2a). The head marker frame with three markers (weight: 5.6 g) was placed on the forehead in a way that the lower two markers were parallel with the eyes. The upper marker was placed in the middle on the forehead. The frame with two markers (weight: 1.8 g) was then placed on the thyroid cartilage. Finally, a marker frame with three markers (weight: 3.2 g) was placed on the midline of the mandible, just posterior to the mental symphysis.

The sEMG data was obtained from: (i) the right and left masseter muscle (respectively masseterR and masseterL), (ii) the right and left temporalis muscle (respectively temporalisR and temporalisL), and (iii) the digastric muscles. Prior to electrode placement, the skin was cleaned with soap and water to reduce electrode-skin impedance. The male participants had a clean-shaven face. The self-adhesive electrodes (Ag/AgCl, Conmed Neotrode electrodes) on the masseter muscle were placed on the crossing of the muscle’s main belly with the angulus oris - lower ear reference line. The electrodes on the temporalis muscle were placed directly superior to the reference line from auricle ear to the corner of the eye. Due to the small size of the digastric muscles and the placement of the mandible marker, unilateral recordings were obtained. The electrodes were placed, immediately posterior to the mandible markers on the right and left belly of the digastric muscles (at a distance of 1 cm from each other).

The kinematic and sEMG data were simultaneously collected with the 3D motion-capture system (Vicon MX 1.7.1, Oxford Metrics, UK). The system consisted of ten infrared high-speed cameras capturing the location of the eight light-reflecting markers (sample rate $f_s = 100$ Hz) in three dimensions.
Outcome variables

The 3D kinematics of the mandible was determined relative to movements of a participant’s head. A local head orthogonal coordinate system was defined based on a participant’s head and mandible markers (Figure 2a): The origin was defined by the upper head marker; the y-axis (vertical vector) was defined as the line from the upper head marker to the central mandible markers; the frontal plane was defined by the x-axis and the line from the lower head markers and the z-axis was perpendicular to the frontal plane. The mandible markers were transformed to the local head orthogonal coordinate system to remove translational and rotational components of head motion from the mandible movement. Thyroid movements were measured in the global (lab) coordinate system (Figure 2b).

Raw sEMG data was full-wave rectified and digitally filtered. A filter was applied (Second order Butterworth low pass filter, cut off = 10 Hz) to remove high frequency noise. Muscle activity amplitude during chewing was measured in mV and expressed as a percentage of the maximal muscle activity during the MVC test (%MVC). The maximum muscle activity was defined as the average muscle activity between the second to the fourth second during the MVC measurement. The presence of muscle chewing activity was defined by sEMG exceeding a threshold of 15% of the MVC. Figure 3 displays an example of the kinematics and sEMG raw data.

Figure 2. Marker and electrodes setting.

a. Set up of the marker frames and electrodes for recording of the 3D kinematics and sEMG. Solid lines represent the axes of the local head coordinate system.
b. Illustration of the 3D reconstruction of the markers in Vicon. Marker frames are placed on the forehead (Hup, HRdo and HLdo), the mandible (MaCent, MaR and MaL) and the thyroid cartilage (AdR and AdL).
Reproducibility of 3D kinematics and surface electromyography measurement of mastication

Figure 3. The data of one trial for one participant of vertical displacement of the MaCent marker and sEMG of the masticatory muscles. a) Vertical displacement of the MaCent marker (in mm). b) sEMG signals for the left (solid line) and right (dotted line) masseter muscle (in %MVC). c) sEMG signals for the left (solid line) and right (dotted line) temporalis muscles (in %MVC). d) sEMG signals for the digastricus muscles (in %MVC).

The first mandible opening included the period of food placement in the mouth until bolus positioning between the molars.\textsuperscript{15,26,27} Hence, the start of chewing was defined as the first mandible opening following the initial bite (first solid vertical line in Figure 4). A chewing cycle was defined as one up and down movement of the mandible.\textsuperscript{4}

Figure 4. Vertical mandible displacement (mm) of one bite measured by MaCent in time (s).

Custom written scripts in Matlab (The MathWorks, 2007) were used to compute outcome variables.
**Table 1. Description of the outcome variables.**

<table>
<thead>
<tr>
<th>Variable</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Bite variables:</strong></td>
<td></td>
</tr>
<tr>
<td>Time-to-swallow (s)</td>
<td>Time interval between start of chewing to the last mouth closure prior to swallowing indicated by the minimum vertical displacement of the mandible marker (MaCent).</td>
</tr>
<tr>
<td>Time-to-transport (s)</td>
<td>Time interval between start of chewing to the final chew, indicating that the rhythmic chewing cycle had advanced to bolus transportation.</td>
</tr>
<tr>
<td>Number of chewing cycles (n)</td>
<td>Number of up and down mandible movements</td>
</tr>
<tr>
<td>Change of chewing side (n)</td>
<td>Change from maximum left to maximum right chewing side or maximum right to maximum left chewing side</td>
</tr>
<tr>
<td><strong>Chewing cycle variables (Kinematic variables):</strong></td>
<td></td>
</tr>
<tr>
<td>Chewing cycle duration (ms)</td>
<td>Time interval of one chewing cycle</td>
</tr>
<tr>
<td>Chewing frequency (Hz)</td>
<td>Number of up and down mandible movements per second</td>
</tr>
<tr>
<td>Opening duration (ms)</td>
<td>Time interval between mandible closure and maximum mandible opening per chewing cycle.</td>
</tr>
<tr>
<td>Closing duration (ms)</td>
<td>Time interval between maximum mandible opening and mandible closure per chewing cycle.</td>
</tr>
<tr>
<td>Opening velocity (mm/ms)</td>
<td>Ratio of maximum mandible opening per chewing cycle (mm) to opening duration (ms).</td>
</tr>
<tr>
<td>Closing velocity (mm/ms)</td>
<td>Ratio of maximum mandible closing per chewing cycle (mm) to closure duration (ms).</td>
</tr>
<tr>
<td>Occlusion duration (ms)</td>
<td>Time interval between closure duration and opening duration</td>
</tr>
<tr>
<td>Δy vertical (mm)</td>
<td>Displacement on the y-axis</td>
</tr>
<tr>
<td>Δx horizontal (mm)</td>
<td>Displacement on the x-axis</td>
</tr>
<tr>
<td>Δz anterior (mm)</td>
<td>Displacement on the z-axis</td>
</tr>
<tr>
<td><strong>Chewing cycle variables sEMG variables:</strong></td>
<td></td>
</tr>
<tr>
<td>EMG masseter R/L (%MVC)</td>
<td>Value of masseter muscles right (MR) and left (ML) expressed as % of maximum voluntary clenching (MVC)</td>
</tr>
<tr>
<td>EMG temporalis R/L (%MVC)</td>
<td>Value of temporalis muscles right (TR) and left (TL) expressed as % of maximum voluntary clenching (MVC)</td>
</tr>
</tbody>
</table>

Four outcome variables per bite (*time-to-swallow, time-to-transport, number of chewing cycles, and change of chewing side*) were defined based on previous studies with sEMG and 3D kinematics on mastication in adults and children.\textsuperscript{11,12} The mean value of 5 complete bites per participant per condition was used for analyses.

An ANOVA analysis (*p*<.05) with post-hoc Bonferroni correction showed that the first cycle of the chewing process deviated from the subsequent four chewing cycles. This is probably due to the bolus being moved towards and between the molars. Similar to other studies\textsuperscript{11,12,15} we, therefore, excluded the first cycle for further analyses. Hence the
characteristics of the second to the fifth chewing cycle were used for analyses per bite. The analyses concerned ten kinematic variables: duration and frequency of the chewing cycle, duration and velocity of the opening and closing stage, duration of the occlusion stage, and the range of displacement of the mandible in horizontal [x], vertical [y], and posterior [z] direction. From the chewing cycles, four sEMG variables were obtained: the sEMG value of the masseterR, masseterL, temporalisR, and temporalisL muscles were expressed as relative values of the percentage of the MVC. Table 1 displays an overview of the chewing variables. In total, four variables per bite and fourteen variables per chewing cycle were used to describe the masticatory activity. All variables were determined for all four conditions separately.

Statistical analyses
Descriptive statistics as mean score and standard deviation (SD) were calculated for all variables based on the mean values of five bites per condition and per participant. To determine measurement reproducibility, the ICC (with 95% confidence interval (CI)) and t-test were calculated on all bite and chewing cycle variables (kinematic and sEMG data) for each condition. The reproducibility for each variable was determined using the ICC for average measurements based on a two-way random effects model and absolute agreement, with session and participants as random factors. A value of ICC > 0.70 was considered ‘good’. In addition, the two-tailed paired t-test was conducted to determine whether differences between session one and two were significant. A significant difference between the two sessions would indicate a systematic error.

Bland-Altman plots were constructed for the between-session difference against the mean of the two sessions for each variable for each variable per participant. The relationship between mean and between-session difference (mean\_diff) was calculated using Kendall’s Tau correlation to ascertain whether a variable was unequally distributed across its range of values. The limits of agreement were defined as the average difference ± 1.96 SD of the mean difference (SD\_diff). The absolute measurement error indicates the within-subject variability across repeated trials and may result from performance differences of non-specific sources of error. The SEM was computed as \(SD\_diff/2\). General guidelines for interpreting SEM are not available and had to be considered in regard to the clinical interpretation of the data. Therefore, we also express the SEM relative to the mean (%SEM), computed as \(SEM/Mean\_total\) (session 1 and session 2) x 100%. The SDD was derived from the SEM as \(SEM \times 1.96 /2\). We then calculated the z-score per variable for each condition to indicate the amount of SD relative to the mean and to enable results to be compared. We considered z-scores between -2 and +2 as acceptable. The level of significance was set to \(p<.05\) for all statistics analyses. All calculations were made using Statistical Package for Social Sciences (SPSS, version 23).
RESULTS

Relative reproducibility

The ICCs of the four bite variables ranged from 0.71 to 0.98 (Table 2a). For the bite variables ‘time-to-swallow’, ‘time-to-transport’, and ‘number of chewing cycles’ the ICCs were >0.90 for all conditions. The lowest ICC was for the variable ‘change of chewing side’ for the small size for biscuit and bread texture (0.71 [95% CI 0.12 to 0.92] and 0.72 [95% CI 0.13 to 0.92], respectively).

The ICCs of the chewing cycle variables ranged from 0.76 to 0.98 (Table 2b). The ICC values were similar for the different conditions. The lowest ICC value was for the variable ‘opening duration’ in the large size of bread texture (ICC = 0.76 [95% CI 0.19-0.92]). All other ICCs were >0.80.

Table 2a. Intraclass correlation coefficient (ICC) with 95% confidence interval (CI) between brackets for means for kinematic and sEMG variables for five bites based on two-way random absolute agreement for all conditions.

<table>
<thead>
<tr>
<th>Variables</th>
<th>Biscuit ICC (95% CI)</th>
<th>Bread ICC (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Time-to-swallow (s)</td>
<td>0.96 (0.81-0.99)</td>
<td>0.96 (0.86-0.99)</td>
</tr>
<tr>
<td>Time-to-transport (s)</td>
<td>0.95 (0.81-0.97)</td>
<td>0.94 (0.80-0.98)</td>
</tr>
<tr>
<td>Number of chewing cycles (n)</td>
<td>0.91 (0.61-0.98)</td>
<td>0.92 (0.73-0.98)</td>
</tr>
<tr>
<td>Change of chewing side (n)</td>
<td>0.77 (0.17-0.94)</td>
<td>0.71 (0.12-0.92)</td>
</tr>
</tbody>
</table>

Table 2b. Intraclass correlation coefficient (ICC) with 95% confidence interval (CI) between brackets for means for kinematic and sEMG variables per chewing cycle (2-5) based on two-way random absolute agreement for all conditions.

<table>
<thead>
<tr>
<th>Variables</th>
<th>Biscuit ICC (95% CI)</th>
<th>Bread ICC (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chewing cycle duration (ms)</td>
<td>0.91 (0.69-0.98)</td>
<td>0.94 (0.80-0.98)</td>
</tr>
<tr>
<td>Chewing frequency (Hz)</td>
<td>0.93 (0.74-0.98)</td>
<td>0.93 (0.77-0.98)</td>
</tr>
</tbody>
</table>
## Absolute reproducibility

The two-tailed $t$-test showed significant between-session differences for the bite variable ‘number of chewing cycles’ ($t=2.471, p=.031$) in the large size of the biscuit texture, for the cycle variables ‘opening velocity’ ($t=-4.379, p=.013$) in the large size of bread texture and for ‘closing velocity’ ($t=2.768, p=.018$) in the small size of biscuit texture. For the variable ‘vertical displacement’, significant differences were found in three of the four conditions ($t=-3.495, p=.005; t=-5.068, p=.009; t=-3.691, p=.004$). Table 3 displays the absolute reproducibility measurements for all variables and each condition. The number of ‘changes of chewing side’ was up to eight per bite. The values of the %SEM for the bites ranged from 8.2% for ‘time-to-swallow’ for the large size of biscuit texture (Table 3a) to 57.0% for ‘change of chewing side’ for the small size of bread texture (Table 3d). The values of the %SEM for the cycle variables ranged from 4.1% for ‘chewing cycle duration’ to 24.7% for ‘EMG masseterR’ for the large size of bread texture (Table 3a). The SDD and limits of agreement are shown in the last columns of Table 3a to 3d. Results of Kendall’s Tau showed a significant correlation between the sessions for the variables ‘closing velocity’ ($p=.028$), ‘vertical displacement’ ($p=.014$) and ‘EMG masseterR’ ($p=.028$) in one condition. The z-scores of variables in all conditions showed values from .003 for ‘opening duration’ (small size of bread texture) to 1.46 for ‘vertical displacement’ (large size of bread texture).

<table>
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<tr>
<th>Variable</th>
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<tr>
<td>Opening duration (ms)</td>
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</tr>
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<td>0.95 (0.83-0.99)</td>
</tr>
<tr>
<td></td>
<td>0.91 (0.69-0.98)</td>
</tr>
<tr>
<td></td>
<td>0.76 (0.17-0.93)</td>
</tr>
<tr>
<td>Opening velocity (mm/ms)</td>
<td>0.87 (0.59-0.96)</td>
</tr>
<tr>
<td></td>
<td>0.95 (0.82-0.99)</td>
</tr>
<tr>
<td></td>
<td>0.94 (0.09-0.97)</td>
</tr>
<tr>
<td></td>
<td>0.87 (0.54-0.97)</td>
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<tr>
<td>Occlusion duration (ms)</td>
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<tr>
<td></td>
<td>0.97 (0.91-0.99)</td>
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<tr>
<td></td>
<td>0.92 (0.75-0.98)</td>
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<tr>
<td></td>
<td>0.81 (0.34-0.95)</td>
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<tr>
<td>Closing duration (ms)</td>
<td>0.88 (0.56-0.96)</td>
</tr>
<tr>
<td></td>
<td>0.83 (0.38-0.95)</td>
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<tr>
<td></td>
<td>0.89 (0.64-0.97)</td>
</tr>
<tr>
<td></td>
<td>0.82 (0.34-0.95)</td>
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<tr>
<td>Closing velocity (mm/ms)</td>
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</tr>
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<td></td>
<td>0.85 (0.50-0.96)</td>
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<td>Δy vertical (mm)</td>
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<tr>
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<td>0.96 (0.57-0.98)</td>
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<tr>
<td></td>
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<td></td>
<td>0.94 (0.46-0.99)</td>
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<td>Δx horizontal (mm)</td>
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<td>0.96 (0.85-0.99)</td>
</tr>
<tr>
<td></td>
<td>0.93 (0.78-0.98)</td>
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<tr>
<td>Δz anterior (mm)</td>
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<tr>
<td></td>
<td>0.81 (0.03-0.95)</td>
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<td></td>
<td>0.86 (0.49-0.96)</td>
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<td>0.98 (0.91-0.99)</td>
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<td>0.81 (0.38-0.95)</td>
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<td>0.90 (0.65-0.97)</td>
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<td>EMG masseterL (%MVC)</td>
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<td>EMG temporalisL (%MVC)</td>
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<td>0.82 (0.40-0.95)</td>
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Table 3a. Descriptive statistics and limits of agreement (LoA) of large size of biscuit texture.

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<td>Upper LoA</td>
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<td>0.71</td>
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<td>1.07</td>
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<td>-1.88</td>
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<tr>
<td>Chewing cycle duration (ms)</td>
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<td>0.07</td>
<td>0.00</td>
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<td>0.03</td>
<td>5.70</td>
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<tr>
<td>Opening velocity (mm/ms)</td>
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<td>Closing duration (ms)</td>
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<tr>
<td>Closing velocity (mm/ms)</td>
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<tr>
<td>EMG TempR (%MVC)</td>
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<td>46.67</td>
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<td>57.02</td>
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* t test p < 0.05  
Kendall’s τa p <0.05  
Note: SEM= standard error of measurement; %SEM= relative standard error of measurement; SDD = smallest detectable difference; LoA = limits of agreement
Table 3b. Descriptive statistics and limits of agreement (LoA) of small size of biscuit texture.

<table>
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<th>Differences session 1-2</th>
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<td>Time-to-swell (s)</td>
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<td>Time-to-transport (s)</td>
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<td>Number chewing cycles (n)</td>
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<td>Occlusion duration (ms)</td>
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<td>Closing duration (ms)</td>
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<td>Closing velocity (mm/ms)</td>
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<td>Δz anterior (mm)</td>
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* t- test p < 0.05
Kendall’s tau p < 0.05
Note: SEM= standard error of measurement; %SEM= relative standard error of measurement; SDD = smallest detectable difference; LoA = limits of agreement.
### Table 3c. Descriptive statistics and limits of agreement (LoA) of large size of bread texture.

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<td>2.79</td>
</tr>
<tr>
<td>Number chewing cycles (n)</td>
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</tr>
<tr>
<td>Change of chewing side (n)</td>
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<td>1.93</td>
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<td>Closing velocity (mm/ms)</td>
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<td>Δy vertical (mm)</td>
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<td>Δx horizontal (mm)</td>
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<tr>
<td>Δz anterior (mm)</td>
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<tr>
<td>EMG ML (%MVC)</td>
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<td>6.53</td>
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* t-test p < 0.05  
Kendall’s tau p < 0.05  
Note: SEM= standard error of measurement; %SEM= relative standard error of measurement; SDD = smallest detectable difference; LoA = limits of agreement
Table 3d. Descriptive statistics and limits of agreement (LoA) of small size of bread texture.

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<th>Differences session 1-2</th>
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<td>Opening duration (ms)</td>
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<td>Δz anterior (mm)</td>
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<tr>
<td>EMG TempR (%MVC)</td>
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<td>31.95</td>
<td>3.43</td>
<td>15.60</td>
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<td>18.0</td>
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* t-test p <0.05
Kendall’s tau p <0.05
Note: SEM= standard error of measurement; %SEM= relative standard error of measurement; SDD = smallest detectable difference; LoA = limits of agreement
DISCUSSION

In this study, we determined the reproducibility and the SDD of 3D kinematic and sEMG variables of the mastication process in 12 healthy adults. We used two textures (biscuit and bread) and two sizes (small and large) and measured five bites and four chewing cycles per bite. With the exception of the variable ‘change of chewing side’, we found, in general, good measurement reproducibility for all kinematic and sEMG variables for the two textures and two sizes. The variables ‘chewing cycle duration’ and ‘chewing frequency’ had the most reproducible results.

Relative reproducibility

All variables for all conditions had good reproducibility (ICC > 0.70). This indicated larger variability between sessions than within subjects for all variables. Although all ICC scores were good, the 95% CI was large in some variables, particularly in the bread textures. The effects of these 95% CI might be reduced in a larger sample size. Our ICC results are in agreement with between-session analyses in other studies on sEMG of masseter and temporalis muscles (95% CI 0.58 - 0.93) involving healthy individuals and in a study with individuals with cerebral palsy ICC between 0.68 and 0.85 were reported. The lowest ICCs were found for the ‘change of chewing side’.

Absolute reproducibility

The %SEM for the bite variables showed a variation of up to 57% for ‘change of chewing side’ (small size of bread texture, Table 3d). The relatively large %SEM may be explained by only a few changes of chewing side during one bite. In healthy individuals, a preferred chewing side was established and about 40-80% chewed on both sides. A preference for chewing side is thought to be used in the first few chewing cycles because the food is then at its most difficult to chew. Our results support this idea in which the biscuit textures had fewer changes in the side of mastication than bread textures, because of the large measurement error in all conditions. ‘Change of chewing side’ seemed difficult to use for evaluating purposes. In addition, it must be noted that the number of changes of chewing side was much higher than expected and differed between zero up to eight changes of chewing side per bite. For the ‘number of chewing cycles’, the t-test indicated systematic differences between the two sessions of the large biscuit texture. The SDD of ‘number of chewing cycles’ is relatively small. In line with several other studies, we found that smaller sized food offerings required fewer chewing cycles for both biscuit texture (11.81 versus 8.20 cycles) and bread texture (11.18 versus 8.55 cycles). We did not find differences between biscuit and bread texture with the same size. These results are in contrast with Wilson et al. who reported a 3-cycle difference between solid and semisolid textures.
Reproducibility of 3D kinematics and surface electromyography measurement of mastication

‘Chewing cycle duration’ and ‘chewing frequency’ exhibited good to excellent between-session reproducibility as seen by the ICC. Moreover, no systematic differences were observed between the session and the %SEM was low in all conditions. The chewing cycle comprises three stages: closing, occlusion, and opening. The results of the chewing stages had good reproducibility and provided detailed information about the mastication process. The %SEM and SDD of the stages variables were slightly smaller in the large conditions than in the small conditions. These differences were larger for the bread textures than the biscuit textures.

The time spent in the various stages in the mandible displacement changed both from cycle to cycle and from texture to texture (biscuit and bread). The occlusion stage in the biscuit textures was slightly longer than in the bread textures (0.23 versus 0.20 s). The ‘opening duration’ and ‘opening velocity’ in biscuit textures were longer and faster than in the bread texture. In another study, these variables were even faster in silicone material than the biscuit texture in our study. Systematic differences between the two sessions were detected in ‘opening velocity’ and ‘closing velocity’, but only in one of the four conditions. The data of these two variables were not normally distributed in all conditions.

Similar to other studies, ‘chewing frequency’ was one of the most reliable variables. The chewing frequency of all conditions in our study (1.73-1.75 Hz) is in line with other studies investigating average adults chewing solids (1.5 Hz) and chewing gum (1.2-1.8 Hz).

Despite the significant systematic between-session differences for the variable ‘vertical displacement’ in three conditions, we decided not to remove this variable. Consequently, we should take into account the systematic differences in interpreting results of future studies. The %SEM of the ‘vertical displacement’ was 7.5%, indicating that the measurement error was relatively small and consequently the SDD was small. The ‘vertical displacement’ derived in this study, which ranged from 13.66 (SD 4.24) to 14.65 (SD 4.21) in all conditions, had a similar displacement as mentioned by other studies (16.87 [SD 2.77] and 12.6 [SD 3.1], respectively), but showed a larger variability. These differences in variability could be due to the textures used in the different studies. Chewing gum or artificial silicon-based materials keep their overall size and texture during mastication whereas natural products, such as biscuit and bread used in this study, change when mixed with saliva. This may result in differences in adaptation of the vertical displacement between subjects.

The %SEM for sEMG variables was larger than kinematic variables, indicating larger measurement error for sEMG variables. The relatively large %SEM for sEMG variables could be due to how we derived MVC used for the normalization of sEMG as %MVC and the differences in sEMG during MVC. In some participants, the EMG %MVC data exceeded the 100% MVC during chewing, indicating that clenching was not maximal when participants were asked to bite on a rubber tube with maximum force. Unfortunately, we did not standardize the placement of the rubber tube during MVC nor provide a training session
as was done by Giannasi et al. A more posterior placement of the rubber tube should result in a larger biting force due to the mechanical lever system of the mandible. It has been stated that as the mandible separation diverged from the optimum opening (17 mm of incisor opening), the strength of the maximum incising force decreased. Moreover, chewing on one side resulted in large between-session variability. Large variability in sEMG variables was also observed between subjects (SD was about 40% of the mean). These results are in line with previous studies indicating large variability between subjects.

The between-subject’s variability in sEMG variables could be attributed to the variability in the characteristics of sEMG as result of anatomical and physical factors, such as thickness of the muscle fibers or subcutaneous tissue layers, distribution and number of fibers in the motor unit, length of the muscle fibers and timing of the muscle contraction. In addition, results could change significantly between studies with changes in the detection points, the detection system applied and the estimation method used. Similar to Piancino et al., our sEMG variables were separately analyzed for mastication on the left and right sides. The t-tests did not indicate systematic differences for left and right masseter and temporalis muscles. We did, however, identify a large %SEM of sEMG variables of about 30% for all conditions. To improve estimating the relative value of the sEMG data, future research should focus on improving the MVC measurement method.

In this study, we used biscuit and bread samples as they are natural products often consumed in society and would be familiar to the participants. The diverse texture of the samples was selected to ensure different functional stimuli for the masticatory system.

**Limitation**
Identifying the swallow was difficult in some trials and resulted in a time-consuming process. We identified the swallow by the superior and anterior displacement of the thyroid cartilage. However, movement of the hyoid bone, which influences the movement of the thyroid cartilage, varies as a result of bolus location in the oropharyngeal tract. For detecting the swallowing onset, we used a combination of the sEMG data and the kinematics. To mark the end of the chewing process, we used the last closure of the mandible before the swallowing onset for the bite variables. Additional research is needed to improve the detection of swallowing onset.
CONCLUSION

Measurements using 3D kinematic and sEMG to assess mastication were reproducible. The variables ‘chewing cycle duration’ and ‘chewing frequency’ achieved the strongest results in terms of being reproducible whereas ‘change of chewing side’ was not identified as a reproducible variable. For determination of differences between subjects and treatment outcomes, large variability in data can be expected. SDD values can be used in future diagnostic studies to evaluate research with the same protocol as our study. The choice of the conditions used in future studies will depend on the research question(s) and the characteristics of the participant group(s).

ACKNOWLEDGMENTS

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REFERENCES


Can mastication in children with cerebral palsy be analyzed by clinical observation, ultrasound and kinematics?

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MWG Nijhuis-van der Sanden

**ABSTRACT**

The aim of this study was to explore the feasibility of the Mastication Observation and Evaluation (MOE) instrument, dynamic ultrasound and 3D kinematic measurements to describe mastication in children with spastic cerebral palsy and typically developing children. Masticatory movements during five trials of eating a biscuit were assessed in 8 children with cerebral palsy, spastic type (mean age 9;08 years;months) and 14 typically developing children (mean age 9;01 years;months). Differences between trials were tested (t-test) and the mastication of individual children with cerebral palsy was analyzed. MOE scores ranged from 17 to 31 (median 24) for the children with cerebral palsy and from 28 to 32 (median 31) for the typically developing children. There were an increased chewing cycle duration, a smaller left-right and up-down tongue displacement, and larger anterior mandible movements for the trials (n=40) of cerebral palsy children (p<.000 for all comparisons) compared to the trials of typically developing children (n=70). The MOE captures differences in mastication between individual children with cerebral palsy. The MOE items ‘jaw movement’ and ‘fluency and coordination’ showed the most similarity with the objective measurements. Objective measurements of dynamic ultrasound and 3D kinematics complemented data from the MOE instrument.
INTRODUCTION

Cerebral palsy (CP) is a non-progressive disease that occurs in the developing brain. The motor disorders are often accompanied by impairments in oral movements (e.g., reduced movements of the mandible and tongue, inappropriate closing of the lips), which can result in difficulties safely managing foods. Advances have been made in understanding the normal mastication process, yet literature on mastication in children with CP is scarce. Mastication concerns a combination of movement patterns with differences in duration, displacement, and velocity in tongue, mandible, and cheeks. In literature is described that children with spastic CP take more time to eat solid foods than typically developing (TD) children, and have a higher intensity and longer period of masseter and temporalis muscle activity during mastication.

The Mastication Observation and Evaluation (MOE) instrument was developed to structure clinical observation and evaluation of chewing in children. Additional objective measurements of the tongue movements and mandible movements may, however, be useful. While the MOE provides information on the observable characteristics of mastication, dynamic ultrasound measurements and 3D kinematics offer information on the movement patterns. Literature is scarce when looking at the use of ultrasound and kinematic measurements to describe the masticatory movements. Ultrasound measurements have been used in children in speech and swallowing studies to measure tongue movements, but mastication has not been measured yet. Recently, a pilot study on mastication with dynamic ultrasound measurements found that the tongue movements of the adults with spastic CP were slower and had a smaller range of motion on the Y-axis than those of healthy controls. To measure mandible movements, 3D kinematics have been performed in adults and TD children. However, these methods have not been used to describe mastication in children with CP.

The aim of this study was to explore the feasibility of the MOE instrument, dynamic ultrasound and 3D kinematic measurements to describe masticatory movements. We expected that it was feasible to distinguish differences in normal and disturbed patterns in mastication in children with spastic CP and in TD children using the three instruments with respect to different mastication mechanisms. This study focused on the feasibility of measurement techniques, in which mastication was the subject of research.

METHODS

Participants

Eight children with CP, spastic type (three boys, group mean age 9;08 years; months; range 7;03 - 11;05 years; months), and fourteen TD children (six boys, group mean age 9;01 years; months; range 5;05 - 11;10 years; months) took part in this study. They supplied 40 and 70
trials, respectively. According to the Gross Motor Function Classification System (GMFCS)\textsuperscript{20}, the eight children with CP included three with mild motor impairments (GMFCS I–II), two with moderate motor impairments (GMFCS III), and three with serious motor impairments (GMFCS IV–V). Four children with CP had a bilateral paresis and four had a unilateral paresis (two right-sided and two left-sided). Four children had intellectual abilities as average (IQ >85), two children had intellectual disabilities below average (70< IQ< 85), and two children had extremely low intellectual disabilities (IQ < 70)\textsuperscript{21}, based on information from patient files.

Children with CP were recruited from two rehabilitation centers in the Netherlands (Sint Maartenskliniek, Nijmegen and Groot Klimmendaal, Arnhem) and from a national organization for individuals with congenital physical impairments (BOSK). The TD group was recruited from the authors’ social network and was group-matched for age with the children in the CP group. Exclusion criteria included food intolerance for gluten, illness, loose teeth, less than two maxillary and two mandibular molars per side, not being familiar with eating biscuits, not being able to perform the task due to emotional, cognitive or other problems, or hypersensitivity to stimuli on the face.

The research protocol was approved by the Ethics Committee at the Slotervaart Hospital and Reade Rehabilitation Center in Amsterdam, The Netherlands (NL47397.048.14). Parents of all participants gave signed written informed consent.

\textit{Procedure}

Participants attended two consecutive sessions held on the same day: session A for dynamic ultrasound measurements and video recording and session B for 3D kinematic measurements. The order of sessions was randomized. During both sessions, each participant was seated in a chair in an upright position and instructed to keep their head forward and minimize head movements. Five trials were measured per session. In each trial, children consumed one piece (1.5 x 1.5 cm) of a crunchy biscuit (LU Véritable Petit Beurre\textsuperscript{®}). The participants were instructed to eat as naturally as possible. Data collection within a trial started just before the biscuit entered the mouth and stopped when the biscuit was swallowed.

\textit{Data collection and data analysis}

Similar to a previous study\textsuperscript{9}, each session B trial was recorded for subsequent observational assessment. Two trained speech-language therapists (SLTs) individually analyzed the recordings using the MOE and discussed results for each item to achieve consensus scores. The MOE consists of eight items: tongue protrusion, lateral tongue movement, munching, jaw movement, chewing duration, loss of food or saliva, number of swallows, and fluency and coordination. The items are scored on a four-point scale: scores 1 and 2 indicate
insufficiency and scores 3 and 4 indicate sufficiency. Score 4 is the best (matured) score and the maximum total MOE score is 32. This instrument is developed for SLTs to observe and analyze the variability in mastication as a basis for intervention. The instrument has an intra-observer and inter-observer reliability varying from 0.68 to 0.98 (weighted Gwet’s agreement coefficient) and an internal consistency of 0.71 and 0.73 (Cronbach’s alpha), which was tested with pieces of biscuit in TD children and children with CP, aged 6-72 months.9

Ultrasound was used to measure horizontal and vertical movements of the tongue. The transducer (C6-8 Pediatric Head, Philips Ultrasound, Andover, MA, USA) was placed under the floor of the participant’s mouth by the researcher (Figure 1a). Consecutive tongue movements were recorded while constant contact between skin and transducer was maintained.22 The highest point of the tongue surface on the ultrasound recordings was used as a reference point for measuring tongue movements. We determined left-right (LR) and up-down (UD) tongue displacement and frequency of tongue displacement in these two directions.16 Data was analyzed using MATLAB (The MathWorks, 2014). Tongue movements were quantified by changes in tongue shape over time observed. The inter-rater and intra-rater agreement was good as measured with intraclass correlation coefficient (ICC) for manual tracing of tongue contours (ICC >0.80).16

To allow measurement of the 3D movements of the mandible, two custom-made PVC marker frames consisting of reflective markers were attached to a participant’s head and mandible (Figure 1a). A 3D motion-capture system (Vicon MX 1.7.1, Oxford Metrics, UK; sample frequency 100 Hz) captured the positions of the markers (Figure 1b). Data was analyzed using MATLAB (The MathWorks, 2007). In a study of mastication in healthy adults, the ICC as indicator for reproducibility ranged from 0.83 to 0.98 and the relative measurement error was <15.1% for outcome variables.17 For chewing cycles 2-5 of each trial, mandible movements in three directions (horizontal, vertical, anterior) and cycle duration stages (opening, occlusion, closing) were determined. In addition, ‘time-to-swallow’ and ‘number of chewing cycles’ were calculated for each trial.17 Individual trials were used for analyses. The standard deviation of the cycle duration of five trials per participant was considered to be a measure of inter-trial variability.
Statistical analysis

Descriptive statistics were used to analyze group MOE outcomes (by median and range), and ultrasound and kinematic outcomes (by mean and 95% CI). Data was checked for normal distribution with Kolmogorov-Smirnov test. Independent $t$-tests were used to explore differences between groups. In addition, individual measurements of trials of children with CP were compared to the TD group; using the TD group as reference data, differences outside the 95% CI were considered to be significantly deviant.

Individual scores of children with CP were used to describe similarities and differences between MOE scores obtained by subjective observations and objective measurements.
Can mastication in children with cerebral palsy be analyzed by clinical observation, ultrasound and kinematics?


Statistical analyses were performed using Statistical Package for the Social Sciences (SPSS) version 23. Level of significance for multiple testing with Bonferroni correction was used, that is, $p<.05$ divided by the number of tests per ‘subgroup’ variable.

**RESULTS**

The median MOE score was 24 (range 17 - 31) for the CP group and 31 (range 28 - 32) for the TD group. Figure 2 shows the MOE profiles for the TD group and the individual children with CP. The TD group achieved maximum scores on the MOE items except for ‘number of swallows’, whereas large variations were observed in the profiles of the children with CP. For the children with CP, the MOE items ‘number of swallows’ and ‘fluency and coordination’ showed the lowest scores (scores 1 or 2) and none scored 1 or 2 on ‘loss of food or saliva’. In both groups, no child achieved the maximum 4 points on ‘number of swallows’.

![Mean profile typically development group; n=14](image)

![CP 1: boy; 10.03 yrs; GMFCS I Unilateral left; IQ > 80](image)

![CP 2: girl; 9.00 yrs; GMFCS I Bilateral Worster Drought syndrome IQ > 80](image)

![CP 3: girl; 9.04 yrs; GMFCS II; unilateral right; IQ > 80](image)

![CP 4: boy; 10.01 yrs; GMFCS III; unilateral left; IQ 60-80](image)

![CP 5: girl; 8.01 yrs; GMFCS III; unilateral right; IQ 60-80](image)
Figure 2. Profile of the MOE instrument of typically developing children \( (n=14) \) and individual profiles of children with spastic cerebral palsy (CP), based on the median score of five trials. The points on the outside of the plot correspond with MOE items: 1 = Tongue protrusion; 2 = Lateral tongue movement; 3 = Munching; 4 = Jaw movement; 5 = Chewing duration; 6 = Loss of food or saliva; 7 = Number of swallows; 8 = Fluency and coordination. The dark solid line in the plot corresponds with the scores per item. Score 4 indicates best (mature) performance and score 1 indicates poor performance.

GMFCS: Gross Motor Functioning Classification System. IQ: Intelligence Quotient.

Table 1 shows the results of ultrasound and kinematic measurements on the group level. The CP group showed significantly smaller LR and UD tongue displacement \( (p<.000) \) and a smaller frequency of tongue displacement in both directions \( (p<.012) \).

As shown in Figures 3, all individual children with CP had smaller LR tongue displacement and lower LR frequency compared to the TD group (below 95% CI). For the CP group, ‘anterior mandible movement’ was significantly larger \( (p<.000) \); ‘cycle duration’, ‘occlusal duration’, and ‘opening duration’ were significantly longer \( (p<.001) \); and ‘time-to-swallow’ and ‘number of chewing cycles’ were significantly larger \( (p<.000) \) than in the TD group (Table 1). The standard deviation (SD) of the cycle duration of five trials per participant was 0.14 s (95% CI 0.08-0.21 s) for the CP group and 0.06 s (95% CI 0.04-0.08 s) for the TD group, reflecting a significant larger SD for the CP group \( (t=-3.49; df= 20; p=.002) \). Figure 4 displays the mean scores derived from kinematic analysis of mandible movements (horizontal, vertical, and anterior direction) for the TD and CP groups.
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Table 1. Results of the ultrasound and kinematic measurements for both groups. The independent t-test was calculated over the sum of individual trials for all subjects per group. Cerebral palsy (CP) group: n=40 trials; typically developing (TD) group: n=70 trials.

<table>
<thead>
<tr>
<th></th>
<th>Group</th>
<th>Mean</th>
<th>95% CI</th>
<th>t-test</th>
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<tr>
<td><strong>Ultrasound measurement</strong></td>
<td></td>
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<tr>
<td>LR-displacement (mm)</td>
<td>CP</td>
<td>0.67</td>
<td>0.62-0.73</td>
<td>t=5.59; df=108; p&lt;.000*</td>
</tr>
<tr>
<td></td>
<td>TD</td>
<td>0.92</td>
<td>0.86-0.98</td>
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<tr>
<td>UD-displacement (mm)</td>
<td>CP</td>
<td>0.48</td>
<td>0.44-0.53</td>
<td>t=4.07; df=108; p&lt;.000*</td>
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<td></td>
<td>TD</td>
<td>0.59</td>
<td>0.56-0.62</td>
<td></td>
</tr>
<tr>
<td>LR-frequency (Hz)</td>
<td>CP</td>
<td>0.51</td>
<td>0.43-0.60</td>
<td>t=3.06; df=108; p&lt;.003*</td>
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<td>TD</td>
<td>0.81</td>
<td>0.67-0.95</td>
<td></td>
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<tr>
<td>UD-frequency (Hz)</td>
<td>CP</td>
<td>2.25</td>
<td>2.07-2.44</td>
<td>t=2.55; df=108; p&lt;.012*</td>
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<tr>
<td></td>
<td>TD</td>
<td>2.55</td>
<td>2.41-2.68</td>
<td></td>
</tr>
<tr>
<td><strong>Kinematic measurement; Mandible movement variables</strong></td>
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<tr>
<td>Horizontal mandible movement (mm)</td>
<td>CP</td>
<td>5.83</td>
<td>5.12-6.56</td>
<td>t=1.02; df=107; p=.309</td>
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<tr>
<td></td>
<td>TD</td>
<td>6.20</td>
<td>5.86-6.53</td>
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<tr>
<td>Vertical mandible movement (mm)</td>
<td>CP</td>
<td>12.77</td>
<td>11.63-14.75</td>
<td>t=2.42; df=107; p=.017</td>
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<td>14.28-15.64</td>
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<td>Anterior mandible movement (mm)</td>
<td>CP</td>
<td>10.31</td>
<td>9.11-11.44</td>
<td>t=6.94; df=107; p&lt;.000**</td>
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<tr>
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<td>TD</td>
<td>6.61</td>
<td>6.31-7.21</td>
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<tr>
<td><strong>Kinematic measurement; Cycle duration variables</strong></td>
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<tr>
<td>Chewing cycle duration (s)</td>
<td>CP</td>
<td>0.83</td>
<td>0.75-0.94</td>
<td>t=-4.54; df=107; p&lt;.000*</td>
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<tr>
<td></td>
<td>TD</td>
<td>0.64</td>
<td>0.60-0.68</td>
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<tr>
<td>Chewing cycle closing duration (s)</td>
<td>CP</td>
<td>0.27</td>
<td>0.23-0.33</td>
<td>t=-2.19; df=107; p=.030</td>
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<td>0.22-0.24</td>
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<td>Chewing cycle occlusal duration (s)</td>
<td>CP</td>
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<td>0.29-0.42</td>
<td>t=-4.43; df=107; p&lt;.000*</td>
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<td>TD</td>
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<td>0.21-0.26</td>
<td></td>
</tr>
<tr>
<td>Chewing cycle opening duration (s)</td>
<td>CP</td>
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<td>0.19-0.22</td>
<td>t=-3.29; df=107; p=.001*</td>
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<td>TD</td>
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<td>0.16-0.18</td>
<td></td>
</tr>
<tr>
<td><strong>Kinematic measurement; Total trial variables</strong></td>
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<tr>
<td>Time-to-swallow (s)</td>
<td>CP</td>
<td>15.58</td>
<td>13.56-18.14</td>
<td>t=-9.54; df=108; p&lt;.000***</td>
</tr>
<tr>
<td></td>
<td>TD</td>
<td>7.12</td>
<td>6.53-7.71</td>
<td></td>
</tr>
<tr>
<td>Number of chewing cycles (n)</td>
<td>CP</td>
<td>13.89</td>
<td>12.12-15.94</td>
<td>t=-6.50; df=108; p&lt;.000***</td>
</tr>
<tr>
<td></td>
<td>TD</td>
<td>8.91</td>
<td>8.36-9.47</td>
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</table>

*significant p<.0125; ** significant p<.0166; *** significant p<.025
Figure 3. Results for the typically developing (TD) group (n=14) and individual children with spastic cerebral palsy (CP) for ultrasound. Displacement of the tongue in left-right (LR) and up-down (UD) direction (respectively a and b). Frequency of the LR and UD tongue displacement (respectively c and d). Bold horizontal line = 95% CI.

The MOE results and objective measurements were compared for the trials of individual children with CP. Three of them (CP1, CP3, and CP6) scored 4 points on the MOE item ‘lateral tongue movement’; although these children had among the highest individual scores within the CP group for ‘LR tongue displacement’ (CP1: 0.78 mm; CP2: 0.84 mm; CP3: 0.72 mm), these values were lower than the TD group’s 95% CI. One child (CP2) scored 2 points on ‘lateral tongue movement’ and had a small LR tongue displacement, but not the smallest of the CP group, while the other child (CP7) who scored 2 points had a large LR tongue displacement. Hence, the scores for ‘lateral tongue movement’ showed a moderate similarity with the objective measurement.

Three children with CP (CP1, CP3, and CP6) scored 3 or 4 points on the MOE item ‘munching’ and had a UD tongue displacement within the 95% CI of the TD group. CP2 and CP8 scored 2 points on ‘munching’ and showed a smaller UD tongue displacement than the TD group (0.30 mm vs. 0.36 mm). Hence, only the high scores on ‘munching’ were in line with the UD displacement data.

Six children with CP scored 3 or 4 points on the MOE item ‘jaw movement’; for four of them, the horizontal mandible movements were within the TD group’s 95% CI. CP2 and CP7 scored 2 points on ‘jaw movement’ and had smaller horizontal mandible movements than
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In this study, we looked at the feasibility of three measurement instruments to explore differences in normal and disturbed patterns of mastication in children. As expected, differences in mastication patterns could be determined using these measurement tools. Mastication, measured by the MOE, showed higher scores in the TD children compared to the children with CP. Measures of tongue displacement (LR and UD) and displacement frequency in these directions were larger for the TD group, whereas the mandible movements in anterior direction were larger for the CP group. Chewing duration was twice as long in the CP group as in the TD group.

The TD group’s 95% CI. Hence, scores on MOE item ‘jaw movement’ were in line with the horizontal mandible movements.

Four children with CP (CP2, CP5, CP7, and CP8) scored 2 points on MOE item ‘fluency and coordination’ and had a variability in cycle durations above the TD group’s 95% CI (0.15 - 0.30 s). The other four children scored 3 points on ‘fluency and coordination’ and had variability in cycle durations that ranged from 0.05 s to 0.11 s. Two of these values were below the TD group’s 95% CI (CP3: 0.08 s; CP6: 0.05 s). Hence, the MOE item ‘fluency and coordination’ showed good similarity with variability in the chewing cycle duration.

**DISCUSSION**

In this study, we looked at the feasibility of three measurement instruments to explore differences in normal and disturbed patterns of mastication in children. As expected, differences in mastication patterns could be determined using these measurement tools. Mastication, measured by the MOE, showed higher scores in the TD children compared to the children with CP. Measures of tongue displacement (LR and UD) and displacement frequency in these directions were larger for the TD group, whereas the mandible movements in anterior direction were larger for the CP group. Chewing duration was twice as long in the CP group as in the TD group.

**Figure 4.** Results for the typically developing (TD) group (n=14) and individual children with spastic cerebral palsy (CP) for kinematic measurements. Mandible movement in horizontal (a), vertical (b) and anterior direction (c). Bold horizontal line = 95% CI.
The TD children achieved maximum scores for almost all MOE items (Figure 2). This indicates a ceiling effect and might be expected in view of the age of the children. Mature mastication can be expected at age 8 years and, in our study, the average age was 9.01 years. The only item for the TD group that did not attain the maximum score was ‘number of swallows’. The scale options specify that a score of 3 should be given when two swallows are required for a chewed food, which is a normal amount for an adult, and a score of 4 be given in case of a single swallow.

The MOE scores for the children with CP were generally in line with the GMFCS data (Figure 2). This relationship between GMFCS severity and feeding and swallowing problems has been reported elsewhere. Two children with CP (CP1 and CP3), who had almost identical MOE profiles as the controls, had also low levels of motor function impairments (GMFCS I and II, respectively). The one child (CP2) for whom MOE score and GMFCS was not in line, had been diagnosed with the Worster-Drought syndrome, a variant of CP. This lack of relationship was expected because that syndrome includes serious oral-motor problems and less gross motor problems resulting in limitations on biting, chewing and swallowing safely.

Children with CP had the most deviated scores on ‘number of swallows’ and ‘fluency and coordination’. ‘Loss of food or saliva’ was not scored as inappropriate, likely because (i) the inclusion criteria only allowed participants who could eat a biscuit, (ii) the offered food was bite-sized and (iii) eating occurred in an optimally controlled environmental (e.g., stable seat, quiet environment). Fluency and coordination problems in mastication were expected in children with CP and were indicated by the MOE score on this item and the considerable variability in chewing cycle duration. Mastication results from neural patterns which are generated in the cerebral cortex and controlled by the central pattern generator (CPG), which is damaged in CP. The CPG is involved in timing signals for a rhythmical alternation of mandible opening and closing and in the spatiotemporal activities of mandible, tongue and facial muscles.

Dynamic ultrasound measurements showed a generally smaller LR and UD tongue displacement for the CP group than for the TD group (Figure 3). These limited lateral tongue movement in children with CP are in accordance with those reported in the literature. Vertical mandible movements for the TD group were in line with those of Wilson & Green, but horizontal movements were almost 50% smaller in our study. These differences could be explained by the age range of the participants; Wilson & Green included children up to 30 months of age, whereas our participants were between 5.05 and
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11.10 years. Wilson and Green attributed larger horizontal mandible excursion as part of normal chewing development in which overshoot occurs before maturation. Our results showed no difference between the groups in horizontal mandible excursion. This is likely due to matured dentition in the participants of this study, which restricts lateral mandible excursion. Children with CP required prolonged time to perform the mastication task, despite the small size of the biscuit. Chewing duration measured as ‘time-to-swallow’ was more than twice as long for children in the CP group than those in the TD group. We found larger group differences in our study than Gisel et al. did; they reported average chewing durations for children aged 4 to 16 years of 12.6 s in children with CP and 8.1 s in TD children. The CP group had a longer chewing cycle duration than the TD group (0.83 s and 0.64 s, respectively). This was due to longer durations of the discrete stages of the chewing cycle, in particular, the duration of the occlusion stage (Table 1). The differences in our study in duration of chewing cycle, occlusion, and opening were also larger than the SDD of adults reported in a previous study.

Our study also found differences between the CP group and TD group for the number of chewing cycles required (CP: 13.98 cycles; TD: 8.91 cycles), whereas Gisel et al. found no significant differences. These differences can be explained by methodological differences in size and type of solid food and differences in the age of participants. The differences for ‘number of chewing cycles’ and ‘time-to-swallow’ suggest that the TD group had more effective chewing movements and required fewer cycles per trial for adequate bolus breakdown than the CP group. The long chewing duration of the children with CP has two implications for clinical practice. Children with CP need more time than TD children to (1) consume meals safely and (2) have sufficient quantitate food intake during regular lengths of mealtimes.

This study also described similarities and differences between MOE results and the objective measurements. The MOE items ‘jaw movement’ and ‘fluency and coordination’ showed the best similarity with the 3D kinematic measurements, while the MOE items ‘lateral tongue movement’ and ‘munching’ had results similar to those for dynamic ultrasound measurements for the best performance only. Unlike normal lateral tongue movements, deviated lateral tongue movements did not seem to be well observable. A large UD tongue displacement was supposed to be present in the case of munching, in which tongue movements are not separated from mandible movements and are thus related to a low score on the MOE item ‘munching’. Munching is not well measured by ultrasound as the position of the transducer does not detect the isolated movements between tongue and mandible. The difference between ultrasound measurement and MOE item scores for tongue movements may be explained by the type of tongue displacement measurement. Tongue displacement was measured from the highest point of the tongue contour, which may differ from the complete tongue movements. The difference could also be explained
by the difficulty in evaluating intraoral tongue movements by observation.

The strength of our study was the use of three instruments for describing masticatory movements in children with normal and disturbed patterns in mastication. This study proved that the different measurements for mastication are feasible to use in children with CP and TD children. The differences in the results between the measurements suggest that they considered other mechanisms of mastication. However, a limitation of our study was that we collected data in two sessions. Consequently, the comparison of the trials of the MOE and kinematics referred to different trials. Given the small sample size (40 and 70 trials, respectively), we have to be careful to generalize the results. We found differences between the measurements indicating that isolated parts of masticatory movements measured by using dynamic ultrasound or 3D kinematics are not captured in the same way as in clinical observations of mastication.

**CONCLUSION**

The MOE captures differences in mastication pattern between children with spastic CP and TD children, and among children with spastic CP. Not all MOE items are directly related to objective measurements, indicating that isolated parts of mastication measured using dynamic ultrasound or 3D kinematics are not captured in the same way as in clinical observations of mastication. Objective measurements of tongue movements by dynamic ultrasound, and of chewing duration by time measurement, could complement data from the MOE instrument for clinical purposes.
ACKNOWLEDGMENTS

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Competing interests
The authors have stated that they had no interests which might be perceived as posing a conflict or bias.
REFERENCES


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chapter 9

General discussion
GENERAL DISCUSSION

This thesis is dedicated to increasing the understanding of the mastication mechanism important and relevant for decision-making by speech-language therapists (SLTs) working with children with feeding and swallowing problems, particularly children with cerebral palsy (CP). This thesis focused on the development and validation of an observational assessment for mastication: the Mastication Observation and Evaluation (MOE) instrument. In addition, we assessed the feasibility of quantitative methods using ultrasound measurement, surface electromyography (sEMG), and 3D kinematics. Finally, in this chapter, the relevance for decision-making for subsequent interventions will be discussed.

Measurement in clinical health practice is important to provide insight into the relationship between the disease, consequences at the level of function, the actual performance at the level of activities and participation, and the influence of personal and environmental factors. For clinical purposes, we developed an observation instrument for mastication in children with CP. This proved to be a challenging goal, as previously predicted by Streiner and Norman: “the most common error committed by clinical researchers is to dismiss existing scales too lightly, and embark on the development of a new instrument with the unjustifiably optimistic and naïve expectation that they can do it better.” Keeping this in mind, we started with a search for available instruments and factors involved in mastication; consulting experts in the field.

Good measurement instruments should have sufficient psychometric characteristics. In clinical practice, data on the psychometric characteristics of tests are frequently missing, which makes it difficult to interpret outcome data. In this thesis, the intra-observer and inter-observer reliability, test-retest reliability and reproducibility of measurements in mastication were the main topics in five studies.

The successful development of the Mastication Observation and Evaluation (MOE) instrument was the result of patience, tenacity and persistence and the help of many SLT students (described in chapters 4 and 5). Next, we considered three quantitative measurement methods for analyzing parts of the mastication process: namely dynamic ultrasound measurement (chapter 6), surface electromyography (sEMG), and 3D kinematics (chapter 7). We focused on the development of the measurement protocols and on the reproducibility of the outcome variables as defined by Terwee et al. Finally, we explored the feasibility of the MOE instrument, dynamic ultrasound, and 3D kinematics measurements to describe masticatory movements in children with CP and typically developing children (chapter 8).

The following aims were addressed in this thesis:
1. to describe feeding and swallowing problems, including masticatory problems in children, and their impact on daily life in adolescence and young adulthood in individuals with CP;
2. to develop and validate an observation instrument for mastication;
3. to evaluate quantitative instruments for measuring mastication and to establish the contrasts with the observation instrument;
4. to test the feasibility of using different instruments to distinguish differences in mastication between children with CP and typically developing children.

This discussion section starts with an overview of the main findings of the seven studies conducted in this thesis and a consideration of some methodological issues. Subsequently, the results will be discussed within a broader perspective, focusing on the applicability of assessment instruments of mastication in children. Finally, the significance of the findings and implications for clinical practice will be described.

**MAIN FINDINGS**

1. Characteristics of children with feeding and swallowing problems and the impact on daily life in adolescence and young adulthood in individuals with CP

In a multiple case history study, we described a 4-6-week in-patient multidisciplinary intervention for children with feeding and swallowing problems, including a behavioral program based on theories of operant conditioning combined with oral-motor training, parental coaching and dietary support. It appeared that the intervention was effective in increasing the qualitative as well as the quantitative food intake in the children and decreasing the parental stress. Parents were unanimously positive about the in-patient intervention, possibly because of the high frequency of sessions and their active role in the intervention. The in-patient setting enabled us to observe characteristics of both children and parents in addition to the feeding sessions. By changing the parents’ perception and interpretation of their child’s behavior, parents gained insight into their child’s temperament and into coping strategies that could be used to resolve conflicts during feeding sessions. Therefore, it seemed that the primary goal for children with tube feeding should not be focused on eliminating the tube feeding as fast as possible, but on the capacity of parents to handle the refusal behavior of their child.

In this first exploratory study, we did not quantify the degree of improvement in oral-motor skills and the conclusions concerning the oral motor improvements were only based on observations. Moreover, high-quality evidence from past studies was missing to provide data on the effectiveness of any oral motor therapy for children with neurological impairments. However, while handling the heterogeneity of the group in this study by defining subgroups based on the feeding problem and considering the interaction between physical and behavioral problems, we got insight into the factors which should be included
in the diagnostic and intervention package. The combined behavioral and oral-motor program components are linked to the subgroup profile and the intervention provided insight for clinical decision-making.

In the second study (chapter 3), we explored the impact of feeding and swallowing problems on daily life in adolescents and young adults with CP. It appeared that levels of difficulty in managing food consistencies varied, but all participants experienced practical and emotional problems related to their eating and drinking. They mentioned coping strategies that included adapting or avoiding foods, and they reported feelings, such as shame, frustration, distress, concerns, and even fear for the future. It was obvious that they did not take the initiative to find help. However, health care providers were also not active in monitoring and evaluating the effect of growing up to adulthood with reduced abilities in eating and drinking. We concluded, therefore, that regular monitoring of eating and drinking in individuals with CP throughout the life course, is necessary. This would provide early detection of a decrease in functions and promote optimal interventions, since facial growth occurs during puberty and changes in anatomic proportions can result in a modified oral-motor pattern. Moreover, the social environment will constantly change and the assistance needs to be reduced as much as possible to increase autonomy. The previous advice packages, often given when children were young, need to be updated to guide these young adults towards increasing self-management skills by new insights achieved by research and/or technological developments, new materials, and situational differences.

2 The development and validation of an observation instrument for mastication

Based on literature and the opinions of experts, we developed and evaluated the MOE instrument in two studies (chapters 4 and 5). The MOE consists of eight items with four answer options each and provides insight into discrete oral movements (e.g., lateral tongue movement) and functional units of mastication (e.g., number of swallows). The MOE showed an almost maximum score for typically developing children from 6 years old and up, and is sensitive to developmental changes in young children aged 6-48 months (chapter 5). Moreover, mastication measured by the MOE instrument significantly differed between children with CP and typically developing children (chapter 8). Children with CP had distinctive MOE profiles, which can be used to tailor interventions on deficits in the chewing profiles as presented in the MOE overview (Figure 1).
Figure 1. Examples of individual mastication profiles. The left picture presents the average score of typically developing children. The middle and right pictures show the score of a child with CP (chapter 8). The points on the outside of the plot correspond to MOE items: 1 = Tongue protrusion; 2 = Lateral tongue movement; 3 = Munching; 4 = Jaw movement; 5 = Chewing duration; 6 = Loss of food or saliva; 7 = Number of swallows; 8 = Fluency and coordination. Higher item scores indicate a higher level of performance.

The observation and rating of a video recording by the MOE instrument is easy and takes experienced and trained raters 15 minutes to conduct. The items ‘tongue protrusion’, ‘munching’, and ‘jaw movement’ proved to be the most difficult to interpret, despite an acceptable inter-rater reliability. We suggested, if scores are low on these items, to use additional measurements to acquire detailed quantitative data about tongue and mandible movements, if necessary, for research purposes.

3 Evaluation of quantitative instruments for measuring mastication

Various quantitative assessment techniques are used to measure masticatory movements. Our studies concerned the pros and cons of dynamic ultrasound, sEMG and 3D kinematic measurement in addition to the MOE instrument for evaluating mastication.

Ultrasound
Tongue movements were traced using ultrasound images of healthy adults and adults with CP taken during a mastication task. The raw ultrasound recordings are useful to show temporal and spatial information about the tongue movements in both the sagittal and coronal planes. Since a uniform method of analyzing dynamic ultrasound images of the tongue is not yet available, we developed a customized post-processing and analysis protocol (chapter 6). Good intra-observer and inter-observer reliability for the analyzing method was established, however, the method was time-consuming. We found indications of differences in tongue movements between healthy adults and adults with CP. In the
coronal plane, we found that the movement frequency and the range of vertical tongue movements was lower in the adults with CP. In the sagittal plane, only the vertical movement frequency of the tongue was lower in adults with CP.

3D kinematics
We determined the test-retest reliability and the smallest detectable differences (SDDs) of the kinematic variables in healthy adults eating pieces of bread and biscuit (chapter 7). Good reproducible variables were: number of chewing cycles, chewing frequency, displacement of the jaw movements in horizontal, vertical, and anterior directions, frequency of the chewing movement, and chewing duration and velocity of the opening, occlusion, and closing stages. Next, we used the acquired SDDs to describe the clinical relevance of the outcome measures in masticatory movements of children with CP and controls (chapter 8).

Surface electromyography (sEMG)
In addition, we determined the masseter and temporalis muscle strength and activity during mastication. We detected larger SDDs for the sEMG measurements than for kinematic measurements in healthy adults in a test-retest setting, indicating limited reproducibility (chapter 7). We concluded that sEMG showed large intra-individual and inter-individual variations, due to intrinsic and extrinsic factors and the voluntary clenching task.

We did not use sEMG measurements in children with CP because of the variance in the anatomic and physiologic characteristics of their masticatory muscles caused by different ages in the sample group and the expected measurement error. Moreover, ultrasound data and kinematic measurements provide sufficient information about the masticatory movements of children with CP. We suggest that sEMG measurements of mastication are more relevant in a population with muscle strength problems (e.g., neuromuscular diseases). Therefore, the maximum voluntary clenching force measurement would need to be improved.

4 The feasibility of instruments to measure mastication and the differences in mastication between children with CP and typically developing children

The MOE instrument, ultrasound, and 3D kinematics proved to be feasible for assessing mastication in children with CP and typically developing children (chapter 8). All the measurement tools recorded differences between normal and disturbed mastication movements. The most obvious differences were: longer chewing durations, smaller lateral tongue movements, and larger anterior mandible movements. In conclusion, observation is a good first assessment tool for gaining insight into children’s mastication performance. Additional measurement of the chewing duration and number of chewing
cycles will provide important quantitative data about mastication. For research purposes, ultrasound measurement is convenient for analyzing displacement and frequency of horizontal and vertical tongue movements. 3D kinematics are useful for recording detailed mandible movements in three directions, duration and frequency of chewing cycles, total chewing duration, and numbers of chewing cycles. In general, the MOE instrument seems to be sufficient for analyzing the mastication performance in clinical practice. Additional measurements are only justified by doubts on the mastication capacity, the need for more detailed data, or findings on effects of intervention studies.

METHODOLOGICAL CONSIDERATIONS

Psychometrics
The intra-observer and inter-observer reliability of the MOE instrument were mainly assessed by SLT students. Several groups of students participated in the MOE analysis. For reliability purposes, all students were trained by LR (developer and trainer of the MOE instrument) and passed a test for inter-observer agreement on MOE ratings. Therefore, we can guarantee the quality of the ratings. To improve the intra-observer and inter-observer reliability of the MOE instrument and to implement the MOE in clinical practice, a standardized training of SLTs is necessary. Even though the responsiveness of the MOE has not yet been extensively studied, we expect that it will be applicable to the evaluation of mastication over time. Data on responsiveness need to be sampled in clinical practice to complete the review of the psychometric properties of the MOE.

Target population
Although children were the target group of our research, we also included adults to gain better insight into the feasibility of the assessments and to determine their reproducibility and intra-rater and inter-rater reliability. Since children are often asked to participate in studies and parents are reluctant to agree to participate in new studies with no direct therapeutic effect, only a few children were included. Moreover, one study (chapter 8) described the masticatory movements in a small number of participants; since the movements rather than the participants were the subject of the analyses, the use of a small group of participants is justified. However, we advise structured and systematic data sampling in clinical practice, to avoid unnecessary burden on the children and parents.

Selection and information bias
Since recruitment of participants took place through indirect advertisement without an intensive preselection, selection bias could not be prevented. Parents of children with CP had ethical motives for allowing their children to participate in our study and were sure that
their children could successfully perform the task. The children with CP who participated in the trial had a relatively good chewing ability, although some of them had severe gross motor impairments. The observed oral-motor abilities (speech and eating skills) were better in our target group than we expected based on our clinical experience. The generalizability of the results to clinical practice may be limited due to the large differences in clinical population regarding the gross and fine motor abilities, mostly classified conform the Gross Motor Function Classification (GMFCS)\(^1\), the oral-motor functions, and intellectual abilities. We suggest collecting more data in clinical practice to confirm and complete our results.

Although we tried to avoid information bias, the observations were not blinded and we could not prevent observers from obtaining unintentional information about the diagnosis.

**RESULTS IN A BROADER PERSPECTIVE**

*Intervention of feeding and swallowing problems*

The deficits in caloric and nutrient intake need to be prioritized in the analysis and intervention of children with mastication problems and feeding, and swallowing problems in general. Parents often have routines in daily dietary patterns and do not notice the deficits in food intake, especially in children who ‘only’ have problems in processing solid foods. Moreover, a total overview of the child and his or her environment in terms of abilities and disabilities is necessary and should be updated regularly for new life situations (e.g., school lunch, medical or conditional changes, or increasing independence during adolescence). We agree with Arvedson that a multidisciplinary approach to feeding and swallowing analyses is essential; one that considers both consistency and the nutritional intake.\(^6\) The intervention needs to be based on shared decision-making tailored to the child’s needs and environment, and guaranteeing the acceptability of possible necessary adaptations. Further, the intervention needs to be integrated into the child’s actual life situation to increase adherence.

*Are our findings applicable in clinical practice of mastication problems in children?*

Over the last decade, measurements have increasingly gained importance in the daily practice of SLTs, encouraging them to be more accountable and transparent in their choices of assessment and intervention goals.\(^7\) Observation of oral-motor behavior is one of the most frequently used measurement methods by SLTs. However, we should be aware that there is limited standardization of observations and most results are based on subjective judgment, hampering a reliable evaluation and the comparability of the results.\(^2\) Therefore, we also looked at other measurement instruments to assess mastication. Various aspects play a role in the applicability of assessments of mastication. To facilitate choices of assessments of mastication in children, we developed a criteria list (Table 1). Based on
the results of this thesis, we addressed the applicability and feasibility of the different assessment methods for mastication in children.

The following criteria were considered:
- **Purpose**: the assessment provides data on elements or stages of mastication.
- **Availability**: available on a large scale in clinical settings.
- **Measurement time**: amount needed to prepare and plan assessments.
- **Analysis time**: amount needed to analyze data.
- **Knowledge and training**: amount required for data analysis and interpretation (taking SLTs’ backgrounds into account).
- **Reliability**: the extent to which scores are the same for repeated measurement (test-retest, intra-observer and inter-observer reliability, measurement error).
- **Costs**: of the investments and consuming materials.

<table>
<thead>
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<th>Table 1. Applicability criteria for assessment of mastication.</th>
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<td><strong>Meaning of the ++ rating</strong></td>
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<td>Purpose</td>
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++ see column ‘meaning of the ++ rating’; + moderate; - opposite of ++

The masticatory movements of children with CP significantly differed from those of typically developing children, as indicated by all the above-mentioned assessments. The MOE instrument is, obviously, the easiest, cheapest, and least invasive assessment for analyzing mastication of various consistencies with increasing difficulty. To study mastication or to obtain specific quantitative data about the detailed movements of the tongue or jaw, additional assessments may be useful. The added value and purpose of additive assessments should be clearly defined beforehand to justify costs and increased burden, especially in children.

Ultrasound measurement is recommended for imaging tongue movements; it is non-invasive for the subject, easy to conduct, and provides good insight into tongue movements. A well-equipped hospital setting and knowledge of the analysis methods are necessary.
to transform the recordings into data suitable for analysis. 3D kinematics can be used to make detailed measurements of chewing cycles and mandible movement\textsuperscript{8,9}, but it requires placing markers on a child’s face and a well-equipped setting. Another simpler method of measuring chewing efficiency would be to add measurement of the number of chewing cycles and time necessary for oral processing to the MOE instrument.

The MOE describes masticatory capacity: the child’s performance on a mastication task during an assessment. Apart from that, it would be interesting to know more about the child’s daily performance. Additional assessments may be recommended to complete information about mastication performance, capacity, or to indicate the possible improvements. For example, the Karaduman Chewing Performance Scale describes mastication performance in five levels, ranging from normal chewing to a child who cannot bite or chew.\textsuperscript{10} This scale indicates the level of processing solid foods. The Test of Mastication and Swallowing Solids in children (TOMASS-c) can be used to determine the maximum chewing capacity.\textsuperscript{11} However, the instruction to “eat this as quickly as possible” suggests sufficient mastication and cognitive skills to prevent choking during the task and, therefore, is not suitable for every child. Despite sufficient results on motor aspects of mastication, some children could still have problems processing solids foods. In such cases, cognitive and behavioral issues need to be considered.

Figure 2 provides an overview of the recommended assessments of mastication in children, classified in different levels of complexity and extensiveness. The third level of assessments is only recommended in special cases or research purposes.
Figure 2. Measurement instruments for mastication. At the top, a broad inventory of nutritional status; in the middle, observational and qualitative data; at the bottom, quantitative data about mastication.

Clinical reasoning

Based on the integrated framework created by Schenkman et al.\textsuperscript{12} and the derived evidence statement for motor writing problems in children\textsuperscript{13}, we developed a preliminary systematic framework for clinical reasoning about children with mastication problems. (Figures 3a and 3b). Schenkman et al.’s integrated framework was based on the Hypothesis-Oriented Algorithm for Clinicians (HOAC), which described steps for assessment and decision-making about intervention.\textsuperscript{12,14} All health care professional should follow all of the steps as described.
Figure 3a. Overview of the Hypothesis-Oriented Algorithm for Clinicians in mastication problems in children.
Figure 3b. Overview of the Hypothesis-Oriented Algorithm for Clinicians in mastication problems in children.
The HOAC for mastication problems (Figures 3a and 3b) reflects on the decision-making process in the clinical practice of health professionals for mastication problems in children. Firstly, patient- and environment-identified problems can be diagnosed using interviews (step 1). It is important to consider both perspectives, as there may be discrepancies between the child and his or her environment. In an initial interview, barriers and facilitators in personal and environmental factors are detected and a preliminary hypothesis about the causes of the mastication problem is formulated. In the feeding performance (step 2), observations (using the MOE instrument) are made of live and home video feeding sessions. Various food consistencies and materials (tasks) can be used and personal and environmental issues can be observed to determine factors involved in the feeding problem. The results of steps 1 and 2 will lead to applied assessments covering task-specific features.

Next, all the data about body functions, activities, and participation, as well as personal and environmental factors can be summarized using the ICF classification and ordered into possibilities and disabilities. A hypothesis about the cause and type of the mastication problem can be formulated based on the obtained data, using multidisciplinary perspective and the circumstances influencing the performance (step 4). This analysis leads to an intervention goal to be formulated in terms of specific, measurable, achievable, relevant, time-bound and inspiring (SMARTI). The intervention (step 5) follows guiding principles: the issues that may result in success and are based on evidence and best practices. Finally, the results of the intervention must be systematically evaluated and adapted or changed if needed (step 6). The whole process should be repeated several times and needs to be adapted to new circumstances.

CLINICAL IMPLICATIONS

This thesis is intended to guide SLTs in the clinical decision-making process when choosing assessments of mastication for children with CP. We assume that this process not only has implications for the clinical practice involving children with CP with mastication problems, but also for pediatric feeding and swallowing problems in general. The recommendations are based on the HOAC framework of clinical reasoning in mastication and are related to the main findings in this thesis. The numbers of the paragraphs refer to the steps in the HOAC scheme for mastication (Figures 3a and 3b).

1 Request for help

Multidisciplinary approach

Problems with mastication have an impact on an individual’s eating and drinking, nutrition, health, communication, social relationships, and quality of life. All these aspects should be reflected in the assessment of mastication and involve multiple health care professionals.
A team of health care professionals involved in feeding and swallowing problems should consist of a pediatrician specializing in gastroenterology, a dietician for the qualitative and quantitative feeding analyses and intervention, a speech-language therapist for oral-motor analyses and intervention, a psychologist for diagnosis and intervention of the behavioral aspects and parent-child interaction, and, eventually, a physical or occupational therapist for gross and fine motor tasks. The diagnosis of mastication problems in children with CP, or larger feeding and swallowing problems, needs to be established from a broad perspective since these problems are not only caused by various underlying medical diagnoses but also by other influencing factors. This variety in performance and underlying causes of such problems requires a multidisciplinary approach towards assessments, analyses, and interventions.

2 Feeding performance

Mastication Observation and Evaluation instrument

The MOE instrument provides insight into disrupted oral-motor functions in the mastication process and is validated for pieces of biscuit and bread. Mastication should be considered with various food consistencies and contexts. We suggest using the MOE instrument to observe different tasks in order to compare the masticatory capacity (what the child can do in an optimal situation) and the performance (what the child does in daily practice). Information about the gap between capacity and performance provides insight into therapy perspectives and prognoses. The MOE profiles could also be an aid in determining intervention goals (Figure 1).

3 Diagnostic assessment

A single outcome may not provide a complete overview of mastication. However, we do not advocate using all quantitative assessments for mastication problems in children. The MOE instrument, enhanced with qualitative data about various conditions and quantitative data derived from measuring chewing frequency and chewing duration, provides good insight into the management of solid foods. In addition to the motor aspects of mastication, sensory issues and personal and environmental factors also need to be considered to complete the performance assessment of mastication. Instrumental assessments (e.g., dynamic ultrasound, 3D kinematics) are only indicated to study specific research questions about mastication. Ultrasound measurement is the simplest assessment to use with children, however, the analysis method is time-consuming.

Safety first

Safely processing solid foods depends on both the quality of mastication and the complete swallowing process, including the pharyngeal and esophageal phases. Video-fluoroscopy
Chapter 9

is most appropriate for detecting dysfunction in the pharyngeal or esophageal stages of swallowing and could exclude or confirm pathology in these stages. However, due to the risk of radiation, the use of video-fluoroscopy in children is generally restricted. Therefore, SLTs should be alert for signs and signals of (silent) aspiration via observation, such as a hoarse voice, disrupted breathing, coughing, or respiratory problems. In case of serious signs, they should consult a pediatrician. Foods should be adapted to be safely managed, this however could conflict with the intervention goal of eating a wide variety of foods.

4 Analyses

Shared multidisciplinary analyses with indications for the best intervention goal should be made together with parents or caregivers, leading to tailored care for the child and parents. Depending on the results of analyses, the most appropriate professional should conduct the intervention. This results in an individually adapted (or client-centered) intervention. The intervention plan should be formulated using SMARTI goals, to facilitate the evaluation of the intervention. Moreover, health care professionals should be inventive in using less invasive adaptations for feeding and swallowing problems, as adolescents have drawn our attention to feelings of shame about their adapted tools for eating and drinking.

5 Intervention

Intervention for feeding and swallowing problems

In line with Arvedson, we support taking a holistic approach towards feeding with the primary goal of having every child receive adequate nutrition and hydration without health complications and without stress for the child and caregivers. This statement results in a serious dilemma: an adaptation of food consistency could be necessary to improve caloric intake but may hinder the child from gaining experience with more challenging foods, such as solids or finger foods. We assume that as the focus on the amount of solid food intake decreases, the quality of the feeding skills could increase.

Intervention in mastication problems

Interventions for mastication problems can focus on rehabilitation (e.g., improving tongue movements) or on compensation (e.g., a small amount of chewable food). There is still limited knowledge about successful interventions for mastication problems related to children with CP. Recent insights about motor learning, also for oral- motor tasks, recommend intensive training using tasks in a real daily-life context. Therefore, we suggest training with small amounts of chewable foods during mealtimes. We advocate starting with one type of food within a given time, and giving social rewards after every effort. This training session should be repeated a few times a day while gradually increasing
either the amount or the type of food, depending on the child’s performance. Moreover, we should understand the individual need for accommodating at a slow pace.

6 Evaluation

*When growing older*

Periodic reassessment is necessary to determine the effect of the intervention, to facilitate the identification of mastication problems, to indicate additional intervention, to reduce symptoms, and to increase health, safety, and comfort during mealtimes. Eating and drinking problems may still be present in adolescents and adults with CP, even in those who are less seriously affected. Moreover, there is evidence of a decrease in ability from youth to adolescence.\textsuperscript{18,19} Therefore, we promote a lifelong attention to eating and drinking activities.\textsuperscript{18} In addition, we need to look at new insights based on research or technological developments, especially in chronic diseases, such as CP. In line with the modern perspective on health, we also need to consider individual preferences and self-management.\textsuperscript{20} A client-centered approach to feeding and swallowing problems is also needed when children with CP grow older.

**In general**

Guidelines and multidisciplinary education for feeding and swallowing problems in children are increasingly well-structured and based on theoretical concepts, evidence, and best practices.\textsuperscript{21} These should be implemented into various settings of clinical practice and considered for children with specific diagnoses, such as CP. Due to time constraints and high workloads, the screening and assessment of feeding and swallowing problems are at risk. Therefore, SLTs need to be effective and up to date in their use of interventions. Moreover, they should be encouraged to participate in research projects to enrich data on this topic and to publish their findings in (inter)national journals, thus allowing other clinicians and researchers to access their findings.

The social environment of individuals with CP and health care professionals should be aware of the impact of relatively minor oral motor problems that result in restricted participation in mealtimes with their social environment.

**FUTURE PERSPECTIVE**

Although we performed several studies on mastication mechanisms and assessment methods, many questions remain unanswered. Concerning the MOE instrument, we suggest generalizing its applicability for evaluation purposes, providing more psychometric evidence, evaluating its usability for other target groups, and determining the optimal training for its use. The MOE instrument has already been successfully used in pilot studies.
of children with a repaired cleft palate\textsuperscript{22} and premature children.\textsuperscript{23} Further research into the use of the MOE instrument should confirm its applicability to other groups. We recommend conducting future studies using the MOE instrument as the outcome measure with various solid foods, other than pieces of bread and biscuit. Moreover, we are considering adding an easy to conduct chewing time and frequency measurement to the MOE, but the inter-rater and intra-rater reliability of the measurements still needs to be determined.

Apart from isolated mastication performance, mastication efficiency is also important. In research with oral oncology patients, two-colored wax tablets (a mixing ability test) was used to assess the mastication efficiency and seemed promising for use with children.\textsuperscript{24} Therefore, we should determine the applicability of this mixing ability test. Moreover, we could compare these results with the previous data on bread and biscuit, as we know that children with CP show overall limited variability in movement or adapt less well to a motor task.\textsuperscript{16}

In this thesis, we used ultrasound measurements to distinguish tongue movements in normal mastication and disturbed mastication patterns. We recommend refining this analysis method to improve the assessment of relative flat tongue movements in the data and to improve the automatic data analysis. Moreover, it would be useful to determine whether dynamic ultrasound measurements could be used as visual feedback in mastication interventions because the lateral tongue movements are relatively easy to distinguish, even for persons lacking specific training.

A preliminary design for decision-making in mastication problems is presented in this chapter. This scheme still needs to be completed using evidence from available and future studies before being used as a guideline for health care professionals in this field.

Finally, feeding and swallowing problems in children, mastication included, are an interesting and relevant topic that awaits more research and a transition of study results into clinical implications. Moreover, including adolescents with CP in further research would also be important, because they still experience many problems in processing different food consistencies (chapter 3).
REFERENCES


Summary
SUMMARY

The aims of this PhD thesis are: (1) to describe feeding and swallowing problems, including masticatory problems in children, and their impact on daily life in adolescence and young adulthood in individuals with cerebral palsy (CP); (2) to develop and validate an observation instrument for mastication; (3) to evaluate quantitative instruments for measuring mastication and to establish the contrasts with the observation instrument; and, finally, (4) to test the feasibility of using different instruments to distinguish differences in mastication between children with CP and typically developing children. This PhD thesis aims to contribute to the understanding of the mechanism of mastication in children with CP, to improve clinical reasoning of speech-language therapists (SLT), and to provide tailored interventions for optimizing participation in social life, specifically in mealtimes.

Chapter 1 describes the introduction, the purposes, and outline of this thesis. The definition of feeding and swallowing disorders, used in this thesis, is derived from the American Speech-Language-Hearing Association (ASHA): “difficulties gathering food and getting ready to suck, chew, or swallow it”. This includes “difficulty with any step of the feeding process from accepting foods and liquids into the mouth to the entry of food into the stomach and intestines”, resulting in “developmentally atypical eating and drinking behaviors, such as not accepting age-appropriate liquids or foods, being unable to use age-appropriate feeding devices and utensils, or being unable to self-feed”.1 A description of a case from our clinical practice illustrates the complexity of feeding and swallowing problems. The interaction in this case between body functions, related impairments in activities, and the influence of the parents and child factors on the learning process of eating and drinking is presented using the framework of the World Health Organization’s (WHO) International Classification of Functioning, Disability and Health, Child and Youth version (ICF-CY).2 Furthermore, this chapter describes: (i) the different stages in the mastication process, the anatomical structures and sensory-motor processes involved in normal development of mastication as a basis for understanding the pathology on mastication; (ii) characteristics of children with CP; and (iii) observational and quantitative measurements tools of mastication; ultrasound, surface electromyography, and 3D kinematics.

Chapter 2 describes a retrospective case study of 29 children (aged 1;00 - 5;07 years;months) with a variety of medical issues with feeding and swallowing problems. The children exhibited various degrees of physical disabilities and they often had behavioral problems, a variety of differences in food intake, varying durations of feeding and swallowing problems, and differences in context variables related to the children and their families. The characteristics of these children were described using the ICF-CY classification, and the increase in qualitative and quantitative oral intake was used as an outcome measure to
establish the effect of a 4 to 6-week multidisciplinary in-patient intervention. Based on their food intake, the children were classified into three groups: (i) tube feeding; (ii) selective food refusal by texture; and (iii) unpredictable food refusal. In addition to the characteristics present in all groups (e.g., problems with taste and sensory issues, ingestion, and prolonged and selective eating activities), every subgroup also displayed some specific characteristics. In the ‘tube feeding’ group, the prevalence of problems in energy and drive functions was obvious due to metabolic dysfunctions, medical diagnosis, or a far too low caloric intake. The ‘selective food refusal by texture’ group had relatively more problems with ingestion and control of voluntary movements due to neurologic dysfunctions. The ‘unpredictable food refusal’ group showed a prominent prevalence of problems in the middle ears or tonsils, or in sleeping, and handling stress. The environmental factor of individual attitudes of immediate family members was also noticed as a barrier to eating in this group.

Of the 29 children in the study, 26 made progress in their qualitative and quantitative food intake. Only children with very complicated medical issues, such as metabolic illness \(n=3\), could not improve their oral intake within the 4 to 6-weeks intervention due to the complexity of their underlying issues. Parents were positive about the multidisciplinary intervention and appreciated the in-patient program. Throughout the intervention, we considered parental opinions and beliefs which contributed to the final positive outcomes. Parents gained insight into their child’s temperament and learned how to cope with conflicts during mealtimes and to avoid the focus on improving oral food intake as quickly as possible. Moreover, the intervention elements could be transferred to the home situation through parental training.

In conclusion, an intervention’s success is not only dependent on the type or presence of a chronic illness or disorder, but also on each child’s personal factors, such as character and coping style, and the environmental factors, such as parental educational style. Therefore, an intervention for feeding and swallowing problems with behavioral, oral-motor and dietary components needs to be individually adapted to each child and family.

Chapter 3 continues with exploring the experiences of adolescents and young adults with CP about their eating and drinking in daily life, using semi-structured interviews. Adolescents and young adults need to become increasingly independent from parents and caregivers, and this may be complicated by their disabilities. Moreover, physical growth in puberty influences the motor coordination during activities, such as eating and drinking.

The young people with CP in our study, aged 15 - 23 years, exhibited varying degrees of disabilities related to managing all food textures, and they face practical and emotional problems related to eating and drinking activities. Four main themes were extracted from the interviews: (a) feelings (e.g., shame, frustration, fear, and distress); (b) coping strategies (e.g., adaptation or food avoidance); (c) the influence of the social and physical context (e.g., the accessibility of restaurants or assistance); and (d) concerns about the future.
Some participants had relatively minor oral-motor problems but mentioned more limitations in participating in their social environment related to eating and drinking than did severely impaired individuals with CP. Severely impaired individuals seemed to have fewer problems with accepting help from others or dealing with restrictions than did mildly affected individuals. One striking finding was that all but one of the participants had not recently received either monitoring or intervention for eating and drinking skills.

We conclude that regular multidisciplinary rehabilitation programs with regard to eating and drinking ability are needed for purposes of evaluation, advice, and intervention in order to increase the participation of young people with CP. Such programs should draw on the latest insights and they should involve socially acceptable and age-appropriate food adaptation. Moreover, young people with CP need to be trained in self-management so that they are able to ask for tailored assistance with their environment.

Chapter 4 describes the first phase in the development of the Mastication Observation and Evaluation (MOE) instrument. Adequate insight into mastication ability allows us to understand how children process solid foods. This study reported on item selection and item definition, content validity, and intra- and inter-observer reliability of the MOE instrument. Items were retrieved from the literature and discussed by 15 experts in three Delphi rounds. This process resulted into 14 items for which more than 75% of the experts reached consensus. The items were scored on a four-point ordinal scale in which score 1 was the worst performance and score 4 was the best, most mature performance. The four-point scale enabled the respondent to express the extent to which an oral-motor behavior is achieved.

To test the intra- and inter-observer reliability, two experts and five students SLT evaluated video recordings of 20 children (10 children with CP, aged 29 - 65 months, and 10 typically developing children, aged 11 - 42 months) eating pieces of bread and a biscuit. All items showed good to excellent intra-observer agreement (ICC 0.73 - 1.0). The inter-observer agreement was fair to excellent for all items, with the exception of 'chewing duration' and 'number of swallows'.

Chapter 5 describes the development of the final version of the MOE instrument using the Consensus-based Standard for the Selection of Measurement Instruments (COSMIN) framework\(^3\), and determines the instrument’s internal consistency, inter-observer reliability, construct validity and floor and ceiling effects. Data from three bites of two food textures (bread and biscuit) taken by 59 typically developing children and 38 children with CP, aged 6-48 months, and scored on the 14 item version of the MOE, were used for this study.

Four items from the MOE were excluded due to zero variance in the scoring. One item was removed because it was only useful in the case of biting off a piece of food. The
Principal Components Analysis (PCA) showed one factor with eight items, having a weight of >40%. One item loaded below 0.40 in the PCA and was therefore eliminated. The internal consistency of the MOE instrument as determined by Cronbach’s alpha was 0.71 for the two food textures and for both groups of children. The inter-observer reliability calculated using the weighted Gwet’s agreement coefficient, varied from 0.51 to 0.98. Agreement on item ‘lateral tongue movement’ and ‘munching’ was lower than on the other items. The total MOE scores for both groups showed normal data distributions. There were no floor or ceiling effects.

The final version of the MOE contains eight items with four answer options for discrete bites. The MOE instrument: (a) has sufficient internal consistency, (b) can be scored on a 4-point scale with sufficient reliability, (c) is sensitive to changes as a result of the development of mastication in young typically developing children and (d) has maximum item performance scores that are usually reached by the age of 48 months.

However, the reliability of scores on intra-oral movements by observation is not as strong as for the other items. Therefore, training about interpreting of the items and the answer options is highly recommended to optimize the reliability.

Chapter 6 shows how the ultrasound technique is used to visualize dynamic tongue movements. This technical report describes the method used to analyze the coronal and sagittal tongue movements with ultrasound measurement. The tongue curves were manually drawn in the one-dimension mode (M-mode) and automatically extrapolated to the real-time Brightness or B-mode. The highest point per frame was considered to be the most active point and used as reference point for the tongue position. The distance of the highest point between two consecutive frames was the base for calculations resulting in the displacement and velocity parameters. Good intra-observer and inter-observer reliability for the manually drawing was established with average ICC scores of 0.84 and 0.81, respectively. Significant differences between trials of adults with CP and controls were identified. In the coronal plane, we found differences for movement frequency and range of vertical tongue movements. Data obtained from sagittal images, apart from vertical frequency, revealed no differences between the groups.

A disadvantage of the ultrasound technique is the difficulty of interpreting some frames due to faint lines caused by air in the oral cavity, poor contact between the transducer and the skin, or problems in detecting the tongue body contour. Moreover, detection of the highest tongue position based on the tongue contour in the B-mode is difficult when the tongue is centrally grooved (i.e., with two high lateral tongue positions) or relatively flat. We are still working on improving the analysis method for the flat tongue position.
Chapter 7 investigates two other quantitative techniques for evaluating mastication in twelve healthy adults. 3D kinematics of mandible movements and surface electromyographic (sEMG) activity of the masticatory muscles were obtained during two sessions consisting of four conditions: two food textures (biscuit and bread) in two sizes (small and large). 3D kinematics was used to determine the following outcome measures in mastication: amplitude of the jaw movements in horizontal; vertical, and anterior directions; frequency of chewing cycles; and chewing cycle duration and velocity of the opening, occlusion, and closing stages. Although these variables were frequently used to evaluate interventions or to make group comparisons, only limited information is available about their reliability and validity. The measurement reproducibility was good to excellent; ICC ranged from 0.71 to 0.98 for all kinematic and sEMG outcome variables. The standard error of measurement, relative error of measurement, and smallest detectable differences of all variables were calculated with a Bland-Altman analysis. The relative standard error of measurement of the bite variables was up to 17.3% for ‘time-to-swallow’, ‘time-to-transport’ and ‘number of chewing cycles,’ but ranged from 31.5% to 57.0% for ‘change of chewing side.’ The relative standard error of measurement ranged from 4.1% to 24.7% for chewing cycle variables and was smaller for kinematic variables than sEMG variables.

In summary, measurements obtained with 3D kinematics and sEMG are reproducible techniques for assessing the mastication process. ‘Chewing cycle duration’ and ‘chewing frequency’ are the best reproducible variables. The published measurement errors and smallest detectable differences may aid in interpreting the results of future clinical studies that use these variables.

Chapter 8 is dedicated to exploring the feasibility of the MOE instrument, ultrasound, and 3D kinematics through a study conducted with eight children with spastic CP (mean age 9;08 years;months) and 14 typically developing children (mean age 9;01 years;months). We assessed masticatory movements over five trials with a piece of biscuit. Moreover, we compared the clinical observations from the MOE measurements with the quantitative measurements of ultrasound and kinematic measurements.

The MOE scores ranged from 17 to 31 (median 24) for the children with CP and 28 to 32 (median 31) for the typically developing group. Differences between individual children as determined with the MOE were visualized in mastication profiles. The total MOE score decreased with declining gross motor function in children with CP. When ultrasound and 3D kinematics were used to assess the masticatory movements of children with CP in comparison to those of typically developing children, the results showed:

1. a longer chewing cycle duration (0.84 s and 0.64 s, respectively) with a prolonged opening and occlusion duration of the chewing cycle;
2. a longer time-to-swallow (15.6 s and 8.1 s, respectively) and more chewing cycles (13.8 times and 10.4 times, respectively);
3. a larger anterior mandible movement (10.2 mm and 6.8 mm, respectively), but similar horizontal and vertical mandible movements;
4. a lower frequency of tongue movement in the horizontal direction (0.51 Hz and 0.81 Hz, respectively) and the vertical direction (2.25 Hz and 2.55 Hz, respectively).

In addition, the MOE items ‘jaw movement’ and ‘fluency and coordination’ had the best relationship with the 3D kinematic outcome measures. The best performances on the MOE items ‘lateral tongue movement’ and ‘munching’ aligned with the results of the ultrasound measurements. We conclude that the quantitative measurements complement the data of the MOE instrument on tongue and jaw movements.

Chapter 9 provides a comprehensive outline of the main findings of all studies and consequently, the clinical implications for daily practice of SLTs involved in children with mastication problems. A schematic overview of assessments for mastication is presented including a broad inventory of the nutritional status, observational data, qualitative data and quantitative data of the food intake. A preliminary systematic framework for clinical reasoning with regard to mastication problems in children is proposed. Based on this current framework, we provide recommendations for intervention about mastication problems:

- a multidisciplinary approach with the larger perspective on feeding and swallowing;
- observation of mastication including both capacity and performance;
- carefully consideration of the use of quantitative measurement tools;
- attention to the efficiency of mastication in relation to nutritional intake;
- shared decision-making in interventions for mastication problems;
- use of SMARTI (specific, measurable, achievable, relevant, time-bound, and inspiring) formulated intervention goals;
- a lifelong attention to mastication issues, especially in less affected children and young adults with CP.

We suggest for future research:

- to establish the usability of the MOE instrument with various solid foods, other than pieces of bread and biscuit, and for other clinical groups;
- to determine the applicability of the mixing ability test in children with CP to mastication efficiency;
- to improve the analysis method of ultrasound measurements and to establish its usability for visual feedback on tongue movements;
- to improve the framework with evidence for decision-making in mastication problems for health care professionals.
REFERENCES


Appendices

Samenvatting in het Nederlands
Dankwoord
Curriculum vitae
List of publications
List of abbreviations
SAMENVATTING

In mijn werk als logopedist op de Sint Maartenskliniek heb ik me jarenlang bezig gehouden met de diagnostiek en behandeling van kinderen met eet- en slikproblemen ten gevolge van onderliggende medische problematiek. Al in mijn beginjaren als logopedist stelde ik vast dat deze, veelal complexe, problematiek in interdisciplinair perspectief behandeld moest worden met een belangrijke rol voor ouders of verzorgers. Een deel van de kinderen met eet- en slikproblemen had ernstige moeite met het verwerken van vast voedsel. Dit had enerzijds te maken met een gebrek aan motorische en sensomotorische vaardigheid, maar ook de sociale context speelde hierin een rol. De zoektocht naar een passende interventie voor de kauwproblemen is de aanleiding voor dit proefschrift geweest.

De doelstellingen van dit proefschrift zijn: (1) het beschrijven van eet- en slikproblemen, inclusief kauwproblemen bij kinderen en de impact hiervan op het dagelijks leven van adolescenten en jong volwassenen met cerebrale parese (CP); (2) het ontwikkelen en validatieschakel van een observatie instrument voor kauwen; (3) het meten van het kauwen met kwantitatieve meetmethoden, zoals echografie, oppervlakte-elektromyografie (EMG) en 3D kinematica (bewegingsleer), om overeenkomsten en verschillen met het observatie instrument vast te stellen; en tot slot (4) het testen van de gevoeligheid van de verschillende instrumenten om verschillen in kauwbewegingen van kinderen met CP en zich normaal ontwikkelende kinderen vast te stellen. Dit proefschrift is bedoeld om een bijdrage te leveren aan het begrijpen van het kauwmechanisme van kinderen met CP en om een handvat te geven voor het klinische redeneren van logopedisten om zo een op maat gemaakte interventie te ontwerpen die bijdraagt aan de participatie in het sociaal leven.

Hoofdstuk 1 beschrijft de introductie, het doel en kader van dit proefschrift. De definitie van eet- en slikstoornissen die in dit proefschrift gehanteerd wordt, is afgeleid van de Amerikaanse vereniging voor spraak, taal en gehoor, genaamd ASHA, en luidt: “problemen in het verzamelen van voedsel en het gereed maken voor zuigen, kauwen en slikken”. Hierbij horen “problemen in elke fase van het voedingsproces: van het accepteren van voedsel of vloeistof in de mond tot de passage in het maag-darmkanaal”. De problemen leiden tot een atypische ontwikkeling van eet- en drinkgedrag en zijn zichtbaar in het niet accepteren van leeftijdsadequate voeding, het niet kunnen hanteren van leeftijdsadequaat eet- en drinkgerei, of het niet in staat zijn om zelfstandig te eten. Een beschrijving van een casus uit de dagelijkse praktijk illustreert de complexiteit van deze eet- en slikproblemen. De interactie tussen lichaamsfuncties en de hieraan gerelateerde beperkingen in activiteiten en de invloed van ouder- en kind factoren op het leerproces van het eten en drinken wordt geïllustreerd aan de hand van het raamwerk van de Wereld Gezondheidsorganisatie (WHO); de kind en jeugd versie van de internationale classificatie van menselijk functioneren (ICF-CY). Verder beschrijft dit hoofdstuk; (i) de verschillende fases van het slikproces, de
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anatomische structuren en sensorisch motorische processen van de normale ontwikkeling van het kauwen als basis om de pathologie van het kauwen te begrijpen, (ii) kenmerken van kinderen met CP; en (iii) de observationele en kwantitatieve methodes om het kauwen in kaart te brengen, waaronder: achografie, EMG en 3D kinematica.

Hoofdstuk 2 beschrijft een retrospectieve dossierstudie van eet- en slikproblemen bij 29 kinderen (leeftijd 1;00-5;07 jaar) met uiteenlopende medische diagnoses. De kinderen hadden verschillende fysieke problemen en vaak ook gedragsproblemen en hadden een verschillende voedselintake en uiteenlopende duur van de eet- en slikproblemen. Tevens waren er verschillen in de contextvariabelen van de kinderen en de gezinnen. De karakteristieken van deze kinderen werden beschreven aan de hand van de ICF-CY-classificatie en de verbetering in kwalitatieve en kwantitatieve orale voedselinname werd gebruikt als uitkomstmaat om het effect van de 4 tot 6 week durende interdisciplinaire klinische interventie te evalueren. De kinderen werden verdeeld in drie groepen, gebaseerd op de wijze van voedselname; (i) sondevoeding, (ii) selectieve voedselweigering van textuur en (iii) onvoorspelbare voedselweigering. In aanvulling op de kenmerken die in elke groep voorkwamen (o.a. problemen met smaak en sensorische informatieverwerking, spijsvertering, en langdurende en selectieve etactiviteiten), had elke subgroep specifieke karakteristieken. In de groep ‘sondevoeding’ was er een opvallende hoge prevalentie van problemen in energiehuishouding, ten gevolge van metabole ziekten of andere medische diagnose of door veel te lage calorische voedselinname. De groep ‘selectieve voedselweigering van textuur’ had relatief meer problemen met de spijsvertering en controle van willekeurige bewegingen ten gevolge van neurologische aandoeningen. De groep ‘onvoorspelbaar voedselweigering’ liet een opvallende prevalentie zien van problemen in het middenoor of de tonsillen, slaapproblemen en het omgaan met stress. De omgevingsfactor betreffende de attitude van de gezinsleden werd in deze groep opvallend vaak opgemerkt als beperkende factor voor het eetgedrag.

Van de 29 kinderen in deze studie boekten 26 kinderen vooruitgang in zowel de kwalitatieve als kwantitatieve voedselname. Alleen bij de kinderen met zeer gecompliceerde medische kenmerken, zoals metabole ziekten (n=3), verbeterde de orale voedselintake in de 4 tot 6 weken niet vanwege de onderliggende complexiteit van hun ziekte. Ouders waren positief over de multidisciplinaire interventie en waren er blij mee later de aanpak thuis door ouders toegepast worden.

Concluderend kunnen we vaststellen dat het succes van een interventie niet alleen afhankelijk is van de aard en kenmerken van de ziekte of stoornis, maar ook
afhankelijk is van persoonlijke factoren, zoals karakter en aanpassingsvermogen, en van de omgevingsfactoren, zoals opvoedingsstijl van ouders. Een interventie voor eet- en slikproblemen met gedrags-, oraal motorische en diëtetische elementen moet aangepast worden aan het individuele kind en zijn familie.

Hoofdstuk 3 vervolgt met het verkennen van de ervaringen met het eten en drinken in het dagelijks leven van adolescenten en jongvolwassenen met CP door middel van semigestructureerde interviews. Adolescenten en jongvolwassenen worden steeds meer onafhankelijk van ouders en verzorgers en dit kan worden bemoeilijkt door hun beperkingen. Bovendien beïnvloedt de lichamelijke groei in de puberteit de motorische coördinatie van activiteiten, zoals het eten en drinken.

De jongeren met CP (15-23 jaar) van onze studie, hadden uiteenlopende beperkingen met de verwerking van verschillende voedseltexturen en werden geconfronteerd met praktische en emotionele problemen tijdens eet- en drinkactiviteiten. Uit de interviews werden behalve de praktische problemen vier hoofdthema’s vastgesteld: (a) gevoelens (bijv. schaamte, frustratie, angst en verdriet); (b) coping strategieën (bijv. aanpassing of vermijden van voedsel); (c) de invloed van de sociale en fysische context (bijv. de toegankelijkheid van restaurants of hulp); en (d) zorgen over de toekomst.

Enkele deelnemers hadden relatief beperkte oraal-motorische problemen, maar vermeldden meer beperkingen in de deelname aan hun sociale omgeving dan de meer ernstig beperkte deelnemers met CP. Ernstig beperkte personen leken minder problemen te hebben met de acceptatie van hulp van anderen of het omgaan met de beperkingen, dan de minder ernstig beperkte personen. Een opvallende bevinding was dat alle geïnterviewden, met uitzondering van één persoon, niet recent een controle of een interventie voor de eet- en drinkvaardigheden had gehad.

We concluderen dat multidisciplinaire revalidatieprogramma’s met betrekking tot de eet- en drinkvaardigheid voor de evaluatie, het advies en de interventie regelmatig moeten plaatsvinden om de participatie van jongeren met CP te optimaliseren. Dergelijke programma’s moeten gericht zijn op de nieuwste inzichten, moeten sociaal acceptabel zijn en leeftijdsadequate voeding bevatten. Bovendien moeten jongeren met CP in zelfmanagement worden getraind zodat zij in staat zijn om gerichte hulp aan hun omgeving te vragen.

Hoofdstuk 4 beschrijft de eerste fase in de ontwikkeling van het Kauw Observatie en Evaluatie (KOE) instrument. Voldoende inzicht in het kauwproces is nodig om te kunnen begrijpen hoe de kinderen vast voedsel verwerken. Deze studie betreft de itemselectie en itemdefinitie, inhoudsvaliditeit en intra- en interbeoordelaarsbetrouwbaarheid van het KOE-instrument. De items werden samengesteld uit de literatuur en besproken met 15
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deskundigen in drie Delphirondes. Dit proces resulteerde in 14 items waarvoor minimaal 75% van de experts consensus bereikt werd. De items werden gescoord op een ordinale vier-puntenschaal waarbij score 1 de slechtste prestatie was en score 4 de beste en meest ontwikkelde prestatie. De vier-puntenschaal geeft de beoordelaars de mogelijkheid om de mate weer te geven waarin het oraal motorisch gedrag is bereikt.

Om de intra- en interbeoordelaarsbetrouwbaarheid te bepalen hebben deskundigen en vijf studenten logopedie (SLT) video-opnamen van 10 kinderen met CP, leeftijd 29-65 maanden, en 10 kinderen met normale ontwikkeling, leeftijd 11-42 maanden, geëvalueerd tijdens het eten van stukjes brood en een koek. Alle items gaven een goede tot uitstekende intra-beoordelaarsbetrouwbaarheid (ICC 0.73-1.0). De interbeoordelaarsbetrouwbaarheid was redelijk tot uitstekend voor alle items behalve voor de items ‘kauwduur’ en ‘aantal slikbewegingen’.

Hoofdstuk 5 beschrijft de ontwikkeling van de definitieve versie van het KOE-instrument met behulp van het raamwerk van op Consensus gebaseerde Standaard voor de selectie van Meet Instrumenten (COSMIN) en stelt de interne consistentie, interbeoordelaarsbetrouwbaarheid, construct validiteit en vloer- en plafond effecten van het instrument vast. Gegevens van drie happen van twee voedsel texturen (brood en koek) van 59 kinderen met normale ontwikkeling, in de leeftijd van 6-48 maanden, en 38 kinderen met CP, in de leeftijd 24-72 maanden, werden gescoord op de 14-item versie van de KOE. Vier items uit de KOE werden verwijderd als gevolg van het ontbreken van variantie in de scores. Eén item werd verwijderd omdat het alleen bruikbaar was bij het afbijten van voedsel. De factoranalyse toonde één factor met acht items die voor > 40% een verklarende waarde hadden. Eén item had een lagere verklarende waarde in de factoranalyse en werd daarom verwijderd uit de lijst. De interne consistentie van het KOE-instrument zoals bepaald met de Cronbach’s alpha was 0.71 voor de twee voedseltexturen en voor beide groepen kinderen. De interbeoordelaarsbetrouwbaarheid berekend met de gewogen Gwet’s coëfficiënt, varieerde van 0.51 tot 0.98. De overeenstemming voor de items ‘laterale tongbeweging’ en ‘munching’ was lager dan voor de andere items. De totale KOE-scores toonden voor beide groepen een normale verdeling van de resultaten. Er waren geen vloer- of plafond effecten.

De definitieve versie van de KOE bevat acht items met vier antwoordopties per hap. Het KOE-instrument: (a) heeft voldoende interne consistentie, (b) kan worden gescoord op een 4-punts schaal met voldoende betrouwbaarheid, (c) is gevoelig voor de veranderingen op basis van ontwikkeling van het kauwen van jonge kinderen met normale ontwikkeling, en (d) heeft maximale prestatiescores die doorgaans worden bereikt op de leeftijd van 48 maanden.
De betrouwbaarheid van de scores op de intra-orale bewegingen met observatie is echter niet zo sterk als bij de overige items. Voor het optimaliseren van de betrouwbaarheid wordt daarom training voor de interpretatie van de items en de antwoordopties aanbevolen.

Hoofdstuk 6 toont op welke wijze de echografie techniek kan worden gebruikt voor het visualiseren van de dynamische tongbewegingen. Dit technische rapport beschrijft de methode die wordt gebruikt voor het analyseren van de tongbewegingen in het coronale en sagittaal vlak met echografische metingen. De tongcurves werden handmatig ingetekend in de eendimensionale modus (M-modus) en automatisch geëxtrapoleerd naar de real-time of B-modus. Het hoogste punt per frame werd beschouwd als het meest actieve punt en werd gebruikt als referentiepunt voor de tongpositie. De afstand van het hoogste punt tussen twee opeenvolgende frames was de basis voor berekeningen van de verplaatsing en snelheid parameters. Er was een goede intra- en interpersoonlijke betrouwbaarheid voor het handmatig tekenen met gemiddelde ICCs van respectievelijk 0,84 en 0,81. Er werden aanzienlijke verschillen gevonden tussen de metingen van de happen van de volwassenen met CP en de controle proefpersonen. In het coronale vlak vonden we verschillen voor de frequentie van tongbewegingen en het aantal verticale tongbewegingen. Gegevens die werden verkregen uit de sagittale beelden, afgezien van de verticale frequentie, vertoonden geen verschillen tussen de groepen.

Een nadeel van de echografie is de moeilijkheid van het interpreteren van sommige frames als gevolg van de vage contouurlinjen veroorzaakt door lucht in de mondholte, slecht contact tussen de transducer en de huid of problemen bij het vaststellen van de tongcontour. Bovendien is het vaststellen van de hoogste tongpositie op basis van de tongcontour in de B-modus moeilijk wanneer de tong een centrale groef heeft (d.w.z. twee hoge laterale tongposities) of relatief vlak is. We zijn nog bezig met het verbeteren van de analysemethode in het geval van de platte tongpositie.

Hoofdstuk 7 onderzoekt twee andere kwantitatieve technieken, voor de beoordeling van de kauwbewegingen van 12 gezonde volwassenen. Bewegingen van de onderkaak, gemeten met 3D kinematica, en de activiteit van de kauwspieren, gemeten met EMG, werden gemeten tijdens twee sessies bestaande uit vier condities: twee voedseltexturen (koek en brood) en twee maten (klein en groot). Uit de 3D kinematica metingen werden de volgende uitkomstmatten voor het meten van het kauwen vastgesteld: amplitude van de bewegingen van de kaak in horizontale, verticale en voor-achterwaartse richtingen, frequentie van de kauwcycli, de duur van de kauwcyclus en de snelheid van de openings-, occlusie- en sluitingsfase. Hoewel deze variabelen vaak gebruikt worden voor de evaluatie van interventies of de vergelijking van groepen, is informatie over de betrouwbaarheid en validiteit van deze variabelen slechts beperkt beschikbaar. De reproduceerbaarheid
van de metingen was goed tot uitstekend; ICCs varieerden van 0,71 tot 0,98 voor alle kinematica en EMG variabelen. De standaard meetfout, de relatieve meetfout en de kleinste waarneembare verschillen van alle variabelen werden berekend met een Bland-Altman analyse. De relatieve standaard meetfout van de variabelen per hap was tot 17,3% voor ‘tijd tot slikken’, ‘tijd tot transport’ en ‘aantal kauwcycli,’ maar varieerde van 31,5% tot 57,0% voor ‘wijziging van kauwzijde’. De relatieve standaardafwijking van de metingen varieerden van 4,1% tot 24,7% voor de variabelen van de kauwcyclus en was kleiner voor de kinematica variabelen dan voor de EMG variabelen.

Samenvattend zijn 3D kinematica en EMG reproduceerbare technieken voor de beoordeling van het kauwen. ‘Duur van de kauwcyclus’ en ‘kauwfrequentie’ zijn de beste reproduceerbare variabelen. De gepubliceerde meetfouten en de kleinste waarneembare verschillen kunnen als norm gebruikt worden bij het interpreteren van resultaten van toekomstige klinische studies met deze variabelen.

Hoofdstuk 8 is gewijd aan het verkennen van de toepasbaarheid van het KOE-instrument, echografie en 3D kinematica in een studie uitgevoerd met acht kinderen met een spastische CP (gemiddelde leeftijd 9;08 jaar) en 14 kinderen met normale ontwikkeling (gemiddelde leeftijd 9;01 jaar). We beoordeelden de kauwbewegingen van vijf happen van een koekje. Bovendien vergeleken we de KOE-metingen met de kwantitatieve metingen van echografie en 3D-kinematica.

De KOE-scores varieerden van 17 tot 31 (mediaan 24) bij de kinderen met CP en van 8 tot 32 (mediaan 31) bij de kinderen met normale ontwikkeling. De verschillen tussen de individuele kinderen, bepaald met de KOE, zijn gevisualiseerd in kauwprofielen. Bij kinderen met CP daalde de totaalscore van de KOE met het afnemen van het grof motorisch functioneren. Vervolgens zijn de metingen van echografie en 3D kinematica gebruikt om de verschillen in kauwbewegingen vast te stellen tussen kinderen met CP en kinderen met normale ontwikkeling. De resultaten toonden:
1. een langere duur van de kauwcyclus (respectievelijk 0,84 s en 0,64 s) met een langere openingsduur en de duur van de occlusie van de kauwcyclus;
2. een langere ‘tijd tot slikken’ (respectievelijk 15,6 s en 8,1 s) en meer kauwcycli (respectievelijk 13,8 keer en 10,4 keer);
3. een grotere voor-achterwaartse beweging van de onderkaak (respectievelijk 10,2 mm en 6,8 mm), maar een vergelijkbare bewegingsuitslag van de onderkaak op het horizontale en verticale vlak;
4. een lagere frequentie van tongbeweging in de horizontale richting (respectievelijk 0,51 Hz en 0,81 Hz) en de verticale richting (respectievelijk 2,25 Hz en 2,55 Hz).
Daarnaast vertoonden de KOE-items ‘kaakbeweging’ en ‘vloeienheid en coördinatie’ de beste overeenstemming met de kwantitatieve 3D kinematica resultaten. De beste
prestaties op de KOE-items ‘laterale tongbeweging’ en ‘munching’ kwamen overeen met de resultaten van de echografie metingen. We concluderen dat de kwantitatieve metingen een goede aanvulling zijn op de gegevens van het KOE-instrument voor de bewegingen van de tong en kaak.

Hoofdstuk 9 bevat een overzicht van de voornaamste bevindingen van alle studies en de implicaties voor de dagelijkse praktijk van logopedisten die betrokken zijn bij kinderen met kauwproblemen. Een schematisch overzicht van evaluaties voor het kauwen wordt gepresenteerd met inbegrip van een brede inventarisatie van de voedingstoestand, observationele gegevens, kwalitatieve en kwantitatieve gegevens van de voedselintake. Er wordt een concept voor een systematisch kader voor het klinisch redeneren met betrekking tot kauwproblemen bij kinderen voorgesteld. Op basis van het dit kader worden aanbevelingen gedaan voor interventie van kauwproblemen:

• een multidisciplinaire aanpak met het breder perspectief op voeding en slikken;
• observatie van het kauwen, inclusief wat het kind kan en wat het doet in het dagelijks leven;
• zorgvuldige aandacht voor het gebruik van kwantitatieve meetprogramma’s;
• aandacht voor de efficiëntie van het kauwen met betrekking tot voedingsinname;
• gedeelde besluitvorming met betrekking tot de interventies voor kauwen;
• gebruik van SMARTI (specifiek, meetbaar, haalbaar, relevant, tijdgebonden en inspirerend) geformuleerde doelen van de interventie;
• levenslange aandacht voor het kauwen, ook bij minder ernstig aangedane kinderen en jonge volwassenen met CP.

Suggesties voor toekomstig onderzoek:

• de bruikbaarheid van het KOE-instrument vaststellen voor verschillende vaste voedingsmiddelen, anders dan stukjes brood en koek, en voor andere klinische groepen;
• de bruikbaarheid van de mixing-ability-test vaststellen voor de efficiëntie van het kauwen bij kinderen met CP;
• het optimaliseren van de analysemethoden van de echografie en de bruikbaarheid van echografie als visuele feedback op de tongbewegingen te onderzoeken;
• het systematisch kader voor professionals ten aanzien van besluitvorming bij kauwproblemen te verbeteren door verder wetenschappelijk onderzoek.
DANKWOORD

De inspiratie voor dit onderzoek is voortgekomen uit mijn werk als logopedist, waarin ik frequent te maken had met de gerichte hulpvraag van kinderen en hun ouders om ‘gewoon’ te kunnen eten. In dit proefschrift heb ik me gericht op de kinderen met een cerebrale paresis. Tim, Nathalie, Thijs; ik heb jullie als peuters gekend en recent hebben jullie een rol gespeeld binnen mijn onderzoek. Jullie hebben een belangrijke bijdrage geleverd aan het ontwikkelen van mijn kennis en vaardigheden op het gebied van eten en drinken. Kinderen met verschillende ziektebeelden hebben me aangezet om me steviger in de kenmerken van het kauwen vast te bijten; Mike, Maud, Tim, Justin en vele anderen; jullie waren mijn inspiratiebron voor dit werk. Het werken aan een proefschrift is het bedrijven van topsport op een cognitief en pragmatisch niveau. Maar daarnaast is er passie nodig om de klus te volbrengen. Mijn passie voor kinderen met beperkingen en het verminderen van de dagelijkse problemen op het gebied van eten en drinken heeft ervoor gezorgd dat het de inspanning waard was.

Het schrijven van een proefschrift vraagt veel tijd en doorzetten en het was een langdurig en intensief traject. Ik wil degenen die samen met mij deze weg bewandeld hebben, hartelijk bedanken. Jullie zijn van onschatbare waarde geweest gedurende dit proces. Het traject dat startte met de stellige uitspraak van Jacques van Limbeek, toenmalig directeur van Research, Development & Education bij de Sint Maartenskliniek, dat mijn masterthesis in 2007 met het onderwerp “een meetinstrument voor kauwproblemen” een aanloop was voor een promotie. Daar had ik toen sterke twijfels over, maar niettemin kreeg een promotietraject langzaam vorm in 2011. De Hogeschool van Arnhem en Nijmegen (HAN) en de directie van het Instituut Paramedische Studies hebben mij optimaal gefaciliteerd om dit traject uit te kunnen voeren, waarvoor ik zeer dankbaar ben.

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duo in de uitvoering van de metingen; een combinatie van jouw structuur, precisie en rust en mijn zoeken naar pragmatisch oplossingen in onvoorziene situaties. Ik waardeer de tijd, de thee en de rust die je altijd nam voor onze gesprekken en om coherente kennis te maken aan te brengen. Renée Speyer, wie had in 1989, toen we als jonge logopedisten startten op de Sint Maartenskliniek, kunnen vermoeden dat jij vele jaren later mijn copromotor zou zijn. Toen jij je toeleggen op de wetenschappelijke kant van het vakgebied, scheidden onze wegen. Jaren later trof ik je weer op de HAN; de eerste keer bij toeval, later structureel als collega. Jij sleepte me door de dalen heen en kon vele gedachten relativeren. Je was altijd snel met je reactie en positief gestemd over de uitslag van analyses of de voortgang van een artikel.

Petri Holtus, jij hebt gedurende mijn hele carrière een bijzondere rol gespeeld. Jij verleende onder alle omstandigheden support en je voorzag me van waardevolle adviezen op professioneel, maar ook persoonlijke gebied. Jij bent sensitief voor mogelijkheden en kansen, waardoor je de initiator en stimulator was van vele projecten. Zonder jou was ik niet gekomen waar ik nu ben. Een rol als paranimf is een blijk van mijn waardering voor jouw bijdrage.

Uiteraard ben ik veel dank verschuldigd aan de mensen die me ondersteund hebben in het verwerven of verwerken van de data. Maike Koch en Jorine Vermaire, jullie hebben veel werk verzet met de metingen in het looplab van de Sint Maartenskliniek. Jullie zijn nu afgestudeerd als bewegingswetenschappers en ik gun jullie nog vele van deze leuke projecten. Ook de vele studenten van de opleiding Logopedie die mij ondersteund hebben met hun afstudeerproject ben ik erkentelijk; het is een genot om met jonge mensen zoals jullie te werken. Jullie hebben me geholpen met de analyses van talloze kauwfilmpjes, het intekenen van echografiebeelden en het verwerken van interviews. Door het stellen van kritische vragen hielden jullie me scherp en brachten jullie andere perspectieven in. Renée Clapham, collega promovenda, dank voor de zorgvuldige correcties van de tekstuele onzekerheden in de artikelen.

Er zijn meerdere mensen die ik gedurende deze weg heb ontmoet en wiens weg onze wegen nooit gekruist zouden hebben. Zij hebben me geholpen om dit proces beter te maken dan ik zou durven hopen; Caroline Speksnijder, Gert Weijers, Chris de Korte. Dank voor jullie inspirerende wijze van kennisdelen en de geboden faciliteiten. Frans van Grunsven, jij was een kritisch proefpersoon met de gave om mee te denken over wetenschappelijk onderzoek.

Collega logopedisten van de Sint Maartenskliniek; het werken met jullie was een waar genoegen. Met jullie heb ik een geweldige periode gehad waarin we elkaar stimuleerden om de revalidanten de beste logopedische interventies te geven op basis van deskundigheid,
maar ook dankzij de goede teamgeest en plezier. Dankzij jullie kreeg ik de mogelijkheid om me toe te leggen op de eetproblematiek van kinderen. Peter Jongerius, Dorine van Bentum-Schouwink, Esmeralda Spanjaards, Nicole Struis, Martine van Atteveld, Babette Hermans, Jordy Sohier en Tessa Broekhuijsen; met jullie een eetteam vormen en steeds weer uitdagende eet-problemen te analyseren en kinderen begeleiden tot optimaal eetgedrag was bijzonder. We waren een goed eetteam en we kregen te maken met veranderende speerpunten van de Sint Maartenskliniek! Eigenlijk waren we de basis van de revalidatie; zonder eten is er namelijk helemaal geen motoriek mogelijk.

Het team van de opleiding Logopedie van de HAN; jullie hebben moeten dealen met een leidinggevende die meerdere taken en rollen had. Dat kan alleen als je werkt met goede professionals en dat zijn jullie. Vier jaar achtereen een waardering als ‘Topopleiding’ bewijst dat. Colleagues logopedisten van de Radboudumc; het is altijd prettig om met jullie te delen in onderwijs en onderzoek en jullie bij internationale congressen te ontmoeten. In het bijzonder wil ik Lenie van den Engel-Hoek bedanken voor de inspirerende bijdrage tijdens de laatste begeleidingsgesprekken.

Ik kan geen dankwoord schrijven zonder de mensen in mijn naaste omgeving te noemen. De aanmoedigingen en belangstelling gedurende dit traject hebben veel voor mij betekend. Hierdoor kreeg ik telkens weer de energie om door te gaan. Regelmatig heb ik sociale activiteiten moet missen of moeten inkorten. Ik dank iedereen die mij desondanks nog in zijn vriendenlijst heeft opgenomen. Ik hoop deze tijd nu goed te maken.

Lilian Beijer; ik kan me niet voorstellen hoe ik dit onmogelijke avontuur zonder jouw vriendschap en briljante adviezen had kunnen doorstaan. Ik ben heel blij dat jij mijn paranimf wil zijn, ook al is dat voor een associate lector geen gebruikelijke rol.


Lieve Lauren, Nathan en Rogier; het is geweldig om jullie te zien opgroeien en te zien ontwikkelen tot jongvolwassenen. Een struggle-for-life waarin je steeds keuzes moet maken, waarvan je nu de consequenties nog niet kan overzien. Jullie zijn goed op weg en papa en ik hebben vertrouwen dat het jullie ook zal lukken om je doel te bereiken. Ik ben super trots op jullie.

Tot slot, JeanPaul, je hebt met jouw stabilité en rust gezorgd dat we het gezin draaiende konden houden. Jij was de bron van constante support en waardevolle adviezen. Zonder jou had ik dit proefschrift nooit kunnen voltooien.


In 2012 volgde een overstap naar de Hogeschool van Arnhem en Nijmegen (HAN) als hoofd van de opleiding Logopedie. Een functie waarin de koppeling tussen werkveld, onderwijs en onderzoek als puzzelstukjes in elkaar paste. Vanaf april 2017 heeft zij een nieuwe uitdaging als programmacoördinator van de master Logopediewetenschap aan de Universiteit Utrecht, gecombineerd met onderwijs- en onderzoeksaanjagen binnen het Instituut Paramedische Studies van de HAN.

Lianne is getrouwd met JeanPaul Zondag en zij hebben 3 kinderen; Lauren (19), Nathan (18) en Rogier (16).
LIST OF PUBLICATIONS

Remijn L, van den Engel-Hoek L, de Swart BJM, Nijhuis-van der Sanden MWG. Experiences with eating and drinking among adolescents and young adults with cerebral palsy: A qualitative study. [Submitted].

da Costa SP, Remijn L, Weenen H, Vereijken C, van der Schans C. Exposure to texture of foods for 8-month-old infants: does the size of the pieces matter? [Accepted in J Text Stud].


Remijn L. De kauwfunctie bij kinderen. Logopedie & Foniatrie 2009 (9).
# LIST OF ABBREVIATIONS

<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Full Form</th>
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<tbody>
<tr>
<td>AC</td>
<td>Agreement coefficient</td>
</tr>
<tr>
<td>ASHA</td>
<td>American Speech-Language-Hearing Association</td>
</tr>
<tr>
<td>COSMIN</td>
<td>COncensus-based Standards for Measurement INstrumnts of health</td>
</tr>
<tr>
<td>CP</td>
<td>Cerebral palsy</td>
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<tr>
<td>DDS</td>
<td>Dysphagia Disorder Survey</td>
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<tr>
<td>DICOM</td>
<td>Digital Imaging and Communications in Medicine</td>
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<tr>
<td>ICC</td>
<td>Intraclass correlation coefficient</td>
</tr>
<tr>
<td>ICF-CY</td>
<td>International Classification of Functioning, Disability and Health, Child and Youth</td>
</tr>
<tr>
<td>FFAm</td>
<td>Functional Feeding Assessment, modified version</td>
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<tr>
<td>GMFCS</td>
<td>Gross Motor Function Classification System</td>
</tr>
<tr>
<td>KION</td>
<td>Kinder Opvang Nijmegen</td>
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<tr>
<td>KMO</td>
<td>Kaiser-Meyer-Olkin</td>
</tr>
<tr>
<td>LR</td>
<td>Left-Right</td>
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<tr>
<td>MOE</td>
<td>Mastication Observation and Evaluation</td>
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<tr>
<td>MVC</td>
<td>Maximum voluntary clenching</td>
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<tr>
<td>n.a.</td>
<td>not applicable</td>
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<tr>
<td>OMAS</td>
<td>Oral Motor Assessment Scale</td>
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<tr>
<td>PCA</td>
<td>Principal Component Analysis</td>
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<tr>
<td>SD</td>
<td>Standard deviation</td>
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<tr>
<td>SDD</td>
<td>Smallest detectable differences</td>
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<tr>
<td>SEM</td>
<td>Standard error of measurement</td>
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<tr>
<td>sEMG</td>
<td>surface Electromyography</td>
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<tr>
<td>SLT</td>
<td>Speech-language therapist</td>
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<tr>
<td>SPSS</td>
<td>Statistical Package for the Social Sciences</td>
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<tr>
<td>SOMA</td>
<td>Schedule for Oral Motor Assessment</td>
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<tr>
<td>TD</td>
<td>Typically developing</td>
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<tr>
<td>UD</td>
<td>Up-Down</td>
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<tr>
<td>WHO</td>
<td>World Health Organization</td>
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