OROFACIAL FUNCTION IN CHILDREN WITH SPEECH SOUND DISORDERS

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Stockholm 2021
Orofacial function in children with speech sound disorders
THESIS FOR DOCTORAL DEGREE (Ph.D.)

By

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which, by due permission from Karolinska Institutet, will be publicly defended in lecture hall Månen, Alfred Nobels Allé 8, Karolinska Institutet, Campus Flemingsberg, Stockholm, Sweden

November 12th, 2021, at 1 p.m.

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In memory of Rut Eriksson and Gunilla Eriksson
ABSTRACT

Speech sound disorder (SSD) is one of the most common neurodevelopmental disorders in children and can have different aetiologies and outcome. Speech difficulties often co-exist with other disorders, such as motor difficulties and orofacial dysfunction. These co-existent difficulties may have the same biological background. It is important to assess and describe orofacial function in children with SSD, as it may be relevant in differential diagnostics of speech disorders. Orofacial dysfunction can lead to eating difficulties, saliva leakage, reduced oral clearance, reduced mimic, deviations in speech production, voice and resonance and malocclusion. The overall aim of this project was to investigate and describe orofacial function, speech characteristics, occlusion, and other co-existing symptoms in children with SSD persisting after the age of six years.

This PhD project consisted of four prospective cross-sectional studies. The participants included 61 children with SSD aged 6.0-16.7 years (mean age 8.5), 14 girls and 47 boys, and 44 children with typical speech development (TSD) aged 6.0-12.2 years (mean age, 8.8), 19 girls and 25 boys. In Study I, orofacial function was assessed with NOT-S together with phonetic transcription of consonant and vowel production and perceptual ratings of nasality in the participants with SSD. Parents also completed the Intelligibility in Context Scale (ICS) and a questionnaire including anamnestic questions. In Study II, a kinematic assessment of lip and jaw movement was made with a 3D motion analysis and the results were compared for children with SSD and children with TSD. In Study III, the prevalence, type, and severity of malocclusions in children with SSD and TSD were assessed using the IOTN-DHC index. In Study IV, orofacial function in the SSD group and TSD group, respectively, was further assessed by using a bite force meter, the two-coloured chewing gum test, a bite block for jaw stability and oral stereognosis. The results of the two groups were compared and related to malocclusions in the SSD group.

The results showed that all participants had impaired consonant production to a varying degree. Many participants also had impaired vowel production. Half of the participants were found to have deviant nasality. Children with SSD had worse performance on all orofacial function assessments than children with TSD, especially regarding assessments involving jaw stability and sensory function. In addition, children with SSD had a higher prevalence of malocclusions and displayed more functional than structural malocclusions compared the TSD group. The malocclusions were also rated as more severe. In children with SSD, those with poorer orofacial function were at greater risk of malocclusion. General motor difficulties and other neurodevelopmental disorders were reported in children with SSD.

The findings from this thesis suggest that children with persistent SSD are at risk of orofacial dysfunction, malocclusions, general motor difficulties and other neurodevelopmental disorders, and should therefore be screened for co-occurring disorders. Children with SSD and poor orofacial function are at greater risk of malocclusion. Clinicians working with children with SSD need to have knowledge and awareness of this co-occurrence and a multi-professional approach is necessary to ensure appropriate care. An assessment of orofacial function is important when describing the characteristics of children with SSD, as it adds valuable information in differential diagnostics and in future genetic testing.
SAMMANFATTNING

Talstörningar är relativt vanligt hos barn och kan ha flera olika orsaker. Det är också vanligt att svårigheter med talet förekommer samtidigt med andra utvecklingsneurologiska tillstånd såsom ADHD, autism och motoriska svårigheter. Detta kan bero på att de samexisterande svårigheterna har samma biologiska grund. Nedsatt oralmotorisk förmåga kan påverka ansiktsmimik, röst och talklang, åtförmåga, salivkontroll, förmågan till självrengöring i munhålan och bettutveckling. Det är viktigt att bedöma orofacial funktion hos barn med talstörning eftersom det kan vara viktigt vid differentialdiagnostik av talstörningar. Det övergripande syftet med detta projekt var att utforska/undersöka och beskriva orofacial funktion, talkkarakteristika, bettutveckling och andra samtidigt existerande symptom hos barn med talstörning som kvarstår efter sex års ålder.


En stor andel barn med kvarstående talstörning i det här avhandlingsprojektet uppvisade negativ påverkan på orofacial funktion och bettutveckling. Barn med talstörning och orofacial dysfunktion hade också en ökad risk för bettavvikelser. Föräldrarna rapporterade en hög grad av generella motoriska svårigheter och neuropsychiatriska tillstånd. Det är därför viktigt att man är uppmärksam på samesterande svårigheter i omhändertagandet av barn med talstörning och ett tvärprofessionellt arbetsätt är att föredra.


III. Mogren, Å., Havner, C., Westerlund, A., Sjögreen, L., Barr Agholme, M., McAllister, A. Malocclusion in children with speech sound disorders. (Submitted)

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<th>Abbreviation</th>
<th>Description</th>
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<tr>
<td>ADHD</td>
<td>Attention Deficit Hyperactivity Disorder</td>
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<td>AI</td>
<td>Articulation Impairment</td>
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<td>ASD</td>
<td>Autism Spectrum Disorder</td>
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<td>AOB</td>
<td>Anterior Open Bite</td>
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<td>CAS</td>
<td>Childhood Apraxia of Speech</td>
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<td>DD</td>
<td>Developmental Dysarthria</td>
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<td>DCD</td>
<td>Developmental Co-ordination Disorder</td>
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<td>DLD</td>
<td>Developmental Language Disorder</td>
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<td>ESSENCE</td>
<td>Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations</td>
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<tr>
<td>ICS</td>
<td>Intelligibility in Context Scale</td>
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<td>ID</td>
<td>Intellectual Disability</td>
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<td>IOTN-DHC</td>
<td>Index of Orthodontic Treatment Need, Dental Health Component</td>
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<td>MME</td>
<td>Mimic Muscle Evaluation</td>
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<td>NDD</td>
<td>Neurodevelopmental Disorders</td>
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<tr>
<td>NOT-S</td>
<td>Nordic Orofacial Test-Screening</td>
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<td>NNS</td>
<td>Non-Nutritive Sucking</td>
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<td>PCC</td>
<td>Percentage Consonants Correct</td>
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<td>PVC</td>
<td>Percentage Vowels Correct</td>
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<tr>
<td>PROMPT</td>
<td>Prompts for Restructuring Oral Muscular Phonetic Targets</td>
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<tr>
<td>ROM</td>
<td>Range of Motion</td>
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<tr>
<td>SDCS</td>
<td>Speech Disorders Classification System</td>
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<td>SLP</td>
<td>Speech-Language Pathologist</td>
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<td>SMD</td>
<td>Speech Motor Delay</td>
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<td>SSD</td>
<td>Speech Sound Disorder</td>
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<tr>
<td>SVANTE</td>
<td>Swedish Articulation and Nasality Test</td>
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<td>TD</td>
<td>Typical Development</td>
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<td>TSD</td>
<td>Typical Speech Development</td>
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THESIS AT A GLANCE

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<th>Methods</th>
<th>Results</th>
<th>Conclusions</th>
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<tr>
<td>Prospective cross-sectional studies</td>
<td>SSD varied according to PCC (8–95%) and PVC (55–100%) measurements. Percentages of co-occurring disorders included: 51% nasality deviations, 90% intelligibility issues, and 87% orofacial difficulties. The most affected orofacial domains were “Chewing and swallowing” (41%), “Masticatory muscles and jaw function” (38%) and “Sensory function” (38%). The majority (64%) had co-existing dysfunctions relating to general motor and neurodevelopmental disorders.</td>
<td>Children with persistent SSD are at risk of orofacial dysfunction, general motor difficulties and other neurodevelopmental disorders and should therefore be screened for co-occurring disorders.</td>
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**I Prevalence of orofacial function**

Clinical assessment of 61 children with SSD and parental questionnaire. The severity of SSD was estimated using Percentage Consonants Correct (PCC), Percentage Vowels Correct (PVC), and assessments of nasality based on the Swedish Articulation and Nasality Test (SVANTE). Orofacial function was screened using the Nordic Orofacial Test-Screening (NOT-S). Parents completed the Intelligibility in Context Scale (ICS) and a questionnaire including questions about heredity, medical and neurodevelopmental conditions and speech development.

**II Speech motor analysis**

Instrumental assessment. 51 children with SSD and 42 children with TSD. Range of motion (ROM) in lips and jaw in the vowels [a, ʊ, ɪ] produced in a syllable repetition task and median values in resting position were measured with a system for 3D motion analysis. The analysis was based on the coordinates for the mouth corners and the chin centre.

There were significant differences between the groups regarding lateral movement in both the lips and jaw. Children with TSD generally had smaller and more symmetrical lip and jaw movements in all three dimensions compared with children with SSD. There were no significant differences between the groups in the resting position.

Children with SSD showed more asymmetrical and more variable movement patterns of the lips and jaw during vowel production compared with children with TSD in a simple syllable repetition task. The differences were more pronounced in the lateral direction in both the lips and jaw.
### Methods

<table>
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<th>III Prevalence of malocclusion</th>
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<tr>
<td>Clinical assessment and parental questionnaire. 61 children with SSD and 44 children with TSD. Extra-oral and intra-oral examinations were performed by an orthodontist and an SLP. The severity of malocclusion was scored using the IOTN-DHC Index. Questionnaire on oral habits was used.</td>
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### Results

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<td>There were differences between the SSD and TSD groups with regards to the prevalence, type, and severity of malocclusions; 61% of the children in the SSD group had a malocclusion, as compared to 29% in the TSD group. In addition, the malocclusions in the SSD group were rated as more severe. Functional posterior crossbite and habitual lateral and/or anterior shift appeared more frequently in the SSD group. Class III malocclusion, anterior open bite and scissors bite were found only in the SSD group.</td>
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### Conclusions

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<tr>
<th>III Prevalence of malocclusion</th>
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<tr>
<td>Children with SSD had a higher prevalence of and more severe malocclusions than children with TSD. Hyperactivity in mentalis was more common in children with SSD and specifically related to AOB. Oral habits or earlier NNS behaviour were not related to malocclusion in this study, except for in children with a Class II malocclusion for whom it was somewhat more common to have an ongoing oral habit</td>
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### Methods

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<tr>
<th>IV Relationship between orofacial function and malocclusion</th>
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<tr>
<td>Clinical assessments. 61 children with SSD and 44 children with TSD. Assessments of orofacial function included bite force, jaw stability, chewing efficiency and intraoral sensory function. Possible malocclusions were also assessed.</td>
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### Results

<table>
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<tr>
<th>IV Relationship between orofacial function and malocclusion</th>
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<tr>
<td>Children with SSD differed from the control group regarding orofacial dysfunction score (NOT-S), bite force, jaw stability, chewing efficiency and intraoral sensory function. The strongest relationship between orofacial function and malocclusion was for the NOT-S total score. Bite force and jaw stability also strongly predicted the risk of having a malocclusion.</td>
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### Conclusions

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<th>IV Relationship between orofacial function and malocclusion</th>
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<tr>
<td>Children with SSD suffered from orofacial dysfunction more often than a control group with children with TSD. In children with SSD, those with poorer orofacial function were also at greater risk of malocclusion.</td>
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1 INTRODUCTION

1.1 GENERAL MOTIVATION

Over the years, I have met many children with motor speech disorders and orofacial dysfunction in my work as a speech-language pathologist (SLP) at a multi-professional orofacial resource centre for children with rare diseases. These symptoms are very common in this population and are often of great concern for the children themselves and their families (Johnson et al., 2016; Klingberg, Hallberg, & Oskarsdóttir, 2010). However, symptoms of motor speech disorders and orofacial dysfunction are not only restricted to known genetic conditions but are also common in children assessed by SLPs for their speech difficulties in the regular health care service. All participants in this thesis are children that have an unknown cause of their speech sound disorders (SSD) and orofacial dysfunction. Even though all the participants had a long-term contact with an SLP and had persistent SSD, very few had undergone an assessment of orofacial function. The motivation for this thesis arose from the clinical observation that many children who were referred to the clinic had co-existing orofacial dysfunctions and malocclusions in addition to their speech disorder. These co-existing symptoms are documented in the literature but rarely described in detail.

In the developmental psychology literature motor difficulties have been described as “the Cinderella syndrome” (Rosenbaum, 2005). Motor difficulties are a commonly co-existing symptom in many genetic conditions and neurodevelopmental disorders (NDD) (Gillberg, 2010), but the scientific literature is sparse in this field despite the symptoms being visually reachable and accessible compared with cognitive functions.

The need for oral motor assessments and the lack of the same are reported in other scientific studies (Braden, Leventer, Jansen, Scheffer, & Morgan, 2019; Kent, 2015; McCauley & Strand, 2008; Murray, McCabe, Heard, & Ballard, 2015). Murray et al. (2015) strongly emphasised the need for an oral motor assessment since it “… is a practical, inexpensive screen to check for any overt structural deficits or functional impairments related to muscle strength and tone.” (p.53) Murray et al. (2015) also referred to an unpublished review study on children with childhood apraxia of speech (CAS) by McCauley et al. (2012), where they found that only 52% of the studies reviewed included oral motor assessments. A review study of speech and language in children with bilateral perisylvian polymicrogyria (BPP) by Braden et al. (2019) also reports the same pattern. Only 47% of the reviewed studies reported any formal or structured assessment of oral motor function despite oral motor deficits being regarded as a core symptom in this diagnosis.

The notion that speech difficulties and oral motor difficulties overlap and co-exist is not controversial but whether and how those oral motor difficulties should be addressed in intervention is another question. This thesis does not try to answer that question.
2 BACKGROUND

2.1 SPEECH SOUND DISORDERS

Speech and language disorders are some of the most common neurodevelopmental disorders (NDDs) (Bishop, 2010). Despite this, they are not as thoroughly researched as other NDDs, such as autism and attention deficit hyperactivity disorder (ADHD) (Bishop, 2010). Speech errors are some of the most common symptoms in children with speech and language disorders of different aetiology (McLeod & Harrison, 2009). The relationship between motor control, speech articulation and intelligibility is complex (Namasivayam et al., 2013).

SSD is used as an umbrella term for speech sound difficulties of both known and unknown origin (International Expert Panel on Multilingual Children’s Speech, 2012). Children with SSD may have difficulty with articulation, phonology, or motor speech performance, including childhood apraxia of speech (CAS). SSD has been defined as “…a significant delay in the acquisition of articulate speech sounds” (Lewis et al. 2006, p.1294). Limbrick et al. (2013) defined it somewhat differently, stating that SSD is used as a generic term for a diverse population and refers to “…problems with speech sound production, perception, and/or phonological representation, which may make speech difficult to understand” (p.296).

When subtle motor control difficulties and phonological delays co-occur, it is difficult to make a differential diagnosis between the different SSDs (Namasivayam, 2013). Primary difficulties with production may also affect the speaker’s perceptual ability (Byun, 2012).

Already when trying to define SSD it is obvious that children with SSD form a heterogeneous group, and there is an ongoing debate about terminology and classification that is relevant in both a clinical and research setting (Namasivayam, Coleman, O’Dwyer, & van Lieshout, 2019; Waring & Knight, 2013). Problems with definitions are not unique to SSD. Several other neurodevelopmental disorders have the same problem. A recent large consensus work involving researchers and SLPs from the English-speaking world has suggested the umbrella term “Developmental Language Disorder” (DLD) instead of the previously used “Specific language impairment” (SLI) (Bishop, Snowling, Thompson, & Greenhalgh, 2016). DLD also includes children with co-morbidity with other disorders and SSD is suggested as one subgroup. In the CATALISE project (Bishop et al., 2016), the term SSD covers speech disorders with a motor or physical origin as well as expressive phonological problems. They also state that it can be difficult to differentiate between phonological disorders and other types of speech production problems.

Different attempts have been made to create a classification system for SSD (Dodd, 2005; Fox, Dodd, & Howard, 2002; Shriberg, 2010; Stackhouse, 1997), but no agreement has been reached as to which system provides the best model. One of the reasons why it has been difficult to design an agreed-upon classification system is that the theoretical views on SSD differ. Shriberg’s “Speech Disorders Classification System” (SDCS) has an aetiological approach (Shriberg et al., 2010), Dodd’s Differential Diagnosis system (Dodd, 2005) has a descriptive linguistic approach and Stackhouse and Wells’ Psycholinguistic Framework
(1997) is primarily a psycholinguistic processing approach. Depending on the classification system the disorder will be described differently. Shriberg’s SDCS divides the speech disorders into subgroups. The three main types are Speech delay, Motor speech disorders and Speech errors. The three typological subgroups are then divided into eight aetiology subgroups. The SDCS approach has been met with some criticism due to the problem of defining the subgroups and because the nature and severity of the speech difficulties in the different subgroups are not well described. The Motor speech disorder subgroup includes apraxia of speech, dysarthria and Speech Motor Delay (SMD) (earlier described as motor speech disorder–not otherwise specified (MSD-NOS)) (Shriberg, Campbell, Mabie, & McGlothlin, 2019; Shriberg et al., 2010). The SMD term is described as an umbrella term for a speech disorder that shares features with CAS and dysarthria regarding voice, prosody, rate and consonant production, but the difficulties are not as specific as in those subgroups.

Shriberg’s SDCS is the only classification system that describes this subgroup of children with a clear motor speech disorder that is not typical CAS or developmental dysarthria (DD). In Shriberg et al. (2019) and in Vick et al. (2014), this subgroup is described in greater detail. Shriberg et al. (2019) state that: “Specifically, the ten most frequent signs of early SMD included age-inappropriate motor behaviours in subdomains of Speech (Vowels, Consonants, and both Vowels and Consonants), Prosody (Rate, Stress), and Voice (Laryngeal Quality).” (p.745). The intervention study by Namasivayam et al. (Namasivayam, Huynh, Granata, Law, & van Lieshout, 2020), which studied a Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) intervention for children with SSD, state that: “Clinically, these children may present with decreased jaw stability (e.g., lateral jaw sliding), limited control of the degree of jaw height (jaw grading) for mid-vowels (e.g., [ɛ], [ɔ], [ɛ], and [ɔ]), excessive jaw movement range, decreased lip rounding and retraction, and occasionally overly retracted lips.” (p.1). In the study by Vick et al. (2014), non-word repetition and chewing were studied with a kinematic analysis, and it was concluded that children with SMD have greater variability in speech movements compared with children with SSD but not SMD. They also use this analysis to distinguish between the groups. The findings from the chewing assessment are not further described. Orofacial function is not assessed or described in either the study by Shriberg et al. (2019) or by Vick et al. (2014). In the study by Namasivayam et al. (2020), the participants with SMD were assessed with the Verbal Motor Production Assessment for Children (VMPAC) test. Interestingly, their result on the VMPAC improves after intervention with PROMPT. In Shriberg et al. (2019), this subgroup is also described as having co-existing general delays in motor development, in line with the criteria for developmental coordination disorder (DCD), and having difficulties with gross, fine and oral motor tasks.

In the Differential Diagnosis system, speech disorders are divided into three groups: Phonetic, Phonemic and Motor planning, programming and execution (Dodd, 2005). Three types of SSD are described in all the classification systems, even if the authors use different terminology. They are: 1) articulation-based speech disorder with substitution and distortion of speech sounds, 2) a motor planning/programming subgroup with CAS included, and 3) a
phonological subgroup characterised by linguistic simplification processes. CAS is a subgroup in SSD that is described by many researchers (Murray et al., 2015; Shriberg, Lohmeier, Strand, & Jakieliski, 2012). Over the years there has been an ongoing discussion about how to define CAS. American Speech-Language-Hearing Association (ASHA) published a technical report in 2007 where CAS is characterised by three primary features: 1) inconsistent errors on consonants and vowels in repeated production of syllables or words, 2) lengthened and disrupted coarticulatory transitions between sound and syllables, and 3) inappropriate prosody (ASHA, 2007). The problem of differential diagnostics is illustrated by the debate and the development of different feature lists (Iuzzini- Seigel, Murray, 2017; Murray et al., 2015; Shriberg, Potter, & Strand, 2011). However, several problems are linked to these lists as symptoms change with age, interventions, and severity. CAS is also known to occur in many genetic disorders, e.g., Down’s syndrome (Rupela, Velleman, & Andrianopoulos, 2016), 16p11.2 deletion (Fedorenko et al., 2016), and galactosaemia (Shriberg et al., 2011), and NDDs, language disorders and motor difficulties (Iuzzini-Seigel, Hogan, & Green, 2017). The other motor speech disorder described is dysarthria. Dysarthria is most often caused by neurological and/or neuromuscular impairment or associated with a congenital disorder, such as cerebral palsy. The problems with execution of movement result in difficulties controlling and coordinating the speed, range, strength and duration of speech movements. The affected muscles are weak and/or slow, independently of the task (speech, oral motor, chewing, eating). This results in deviations in phonation, loudness and pitch, and resonance, and often generally slurred speech (Pennington, 2016). In this thesis, the classification system by Shriberg (SDCS) is used for the theoretical background of the diagnostics of SSD.

2.1.1 Speech characteristics in SSD

Children with SSD exhibit different speech difficulties, dependent on their age and the characteristics of the disorder. They present with age-inappropriate speech sound deletions and/or substitutions (Namasivayam et al., 2020). One way to describe speech errors is the SODA analysis, where the errors are described as consisting of Substitutions, Omissions, Distortions and Additions (Dodd, 2013). The speech process errors can be categorised into syllable error patterns (final consonant deletion, weak syllable deletion, reduplication, consonant cluster reduction, assimilation, epenthesis, metathesis) or substitution error processes (gliding of liquids, stopping of fricatives, velar fronting and coronal (dental) backing, voicing and devoicing of consonants) (Dodd, 2013). Most of these speech process errors are found in typical speech development as well.

Even if speech difficulties, like backing and velar fronting, may sound the same, they don’t necessarily have the same origin. Research by Cleland et al. (Cleland & Scobbie, 2021) show that motor speech difficulties may be more common in children who traditionally have been described as having “Phonological issues”. In a study of children with persistent SSD, 71% had undifferentiated lingual gestures,” reflecting a speech motor constraint involving either
delayed or deviant control or functionally independent regions of the tongue” (Gibbon, 1999, p. 393).

Atypical speech is not only characterised by deviant consonant production. Deviations in voice, resonance, prosody, fluency, tempo and vowels may also be present. Resonance and voice deviations are often described as features of dysarthria but the difficulties with timing and planning in children with CAS or SMD may also result in difficulties with voice and resonance (Shriberg, 2010; Shriberg, 2019). Reduced coordination between breathing and phonation may be one reason for voice deviations in children with motor speech disorder (Potter, 2011). Both voice disorders and resonance disturbances are reported in children with different genetic diagnoses such as Dravet syndrome (Turner et al., 2017), Down’s syndrome (Rupela et al., 2016), galactosaemia (Shriberg et al., 2011; Potter, 2011), Koolen deVries syndrome (Morgan et al., 2018), and Prader-Willis syndrome (Akefeldt, Akefeldt, & Gillberg, 1997). Voice disorders have been suggested to be related to oral motor deficits in children with severe speech disorders (Amorosa, von Benda, & Wagner, 1990). A study of 38 children with SSD and oral motor deficits could not confirm this relationship (McAllister, 2003). However, the results showed that voice quality also improved significantly after oral motor treatment. Resonance deviations have been reported in children with high-functioning autism (Shriberg et al., 2001). The boys in the study had no severe resonance differences but they were severe enough to be perceived as deviant speech. Deviant voice and resonance may influence intelligibility and result in speech that is perceived as “different” by peers (Nyberg & Havstam, 2016). Speech requires coordination of several muscles and neural subsystems and this inter-articulator coordination is necessary to be able to produce intelligible speech without sound distortions and with the dynamic and timing required for typical voice and resonance (Smith & Zelaznik, 2004). Co-existing language difficulties may further influence intelligibility.

Difficulties with prosody is described both in CAS (Murray et al., 2015) and DD (Patel, Hustad, Connaghan, & Furr, 2012). It is also described in adolescents and adults with high-functioning autism (Shriberg et al., 2001). Exaggerated prosody and monotone intonation are described in children and adolescents with Williams syndrome (Rossi & Giacheti, 2017). Prosody is important for intelligibility and monotonic speech may reduce intelligibility (Klopfenstein, 2009). Correct prosody may also reinforce intelligibility in speech disorders where articulation is affected in other ways (Klopfenstein, 2009).

To conclude, not only speech sounds are affected in children with SSD. The underlying motor speech difficulties may affect several aspects of speech production, such as voice, resonance, prosody and speech rate.

### 2.1.2 Persistent SSD

Persistent SSD is suggested to be strongly correlated primarily with impaired motor skills rather than phonological linguistic processing (Flipsen, 2002, 2015; Johnson, Beitchman, & Brownlie, 2010; Lewis et al., 2015). In a longitudinal study, Wren and co-workers (Wren,
Miller, Peters, Emond, & Roulstone, 2016) found that early motor skill deficits, such as weak sucking at four weeks of age and a history of suspected motor coordination difficulties, were strongly correlated with persistent SSD at eight years of age. The most common persistent speech difficulties were distortion errors that affected intelligibility or the listener’s speech perception; however, to a minor extent. Nevertheless, such speech difficulties may have a negative impact on social life and future employment opportunities for the individual (Flipsen, 2015). In a research article on older children with speech difficulties related to cleft lip and palate, Nyberg & Havstam (2016) showed that peers note and react even to minor articulatory difficulties. There is no consensus about when a speech disorder is regarded as persistent. Wren et al. (2016) include children in the term “persistent SSD” from eight years of age. They also exclude children with the most common distortions from the definition. The DSM-5 manual proposes that the most frequently misarticulated sounds, the so-called “late eight” (l,r,s,z,th,ch,dzh, and zh) should have been learned before eight years of age (American Psychiatric Association, 2013). Flipsen (2015) divides persistent SSD into two categories: residual speech errors and persistent speech errors. He argues that residual speech errors are distortion errors produced around the end of the developmental period by children who have had earlier omission and substitutions errors, while persistent speech errors are abnormal production of speech sounds that were present from the beginning. Flipsen (2015) states that residual speech errors are more common than persistent speech errors and that most of them (75%) will resolve by the end of high school. In this thesis, speech disorder is defined as persistent after six years of age, as all Swedish speech sounds are expected to be mastered at this age (Blumenthal & Lundeborg Hammarström, 2014).

There is also an ongoing debate about the negative consequences of waiting too long to provide interventions for speech disorders (McGill, McLeod, Crowe, Wang, & Hopf, 2021). Using a “wait-and-watch” approach for young children with SSD may not be beneficial, as this will increase the risk of automating incorrect motor programmes for speech. Incorrect speech motor patterns can result in speech sound distortion, incorrect speech movements and persistent SSD (Cleland & Scobbie, 2021; Cleland, Scobbie, Heyde, Roxburgh, & Wrench, 2017; Grigos & Kolenda, 2010; Kabakoff, Harel, Tiede, Whalen, & McAllister, 2021; Klein, McAllister Byun, Davidson, & Grigos, 2013). Well-established speech motor patterns may be difficult to alter.

2.2 SPEECH DEVELOPMENT AND THEORIES OF EMBODIED COGNITION

Motor development and speech and language development occur in a complex and multi-faceted interaction (Iverson, 2010). It is reported that as children develop and mature, the duration of the movements decreases in the lips and tongues while the speech rate increases. Children have also been described as having have less precision of articulatory movements than adults. Increased maturation results in more stable movements and reduced variation during articulation (Grigos, 2009). In Swedish, all consonants are established by the age of six years, including /t/ and /s/ sounds (Blumenthal & Lundeborg Hammarström, 2014), like English-speaking five-year-olds (Dodd, Holm, Hua, & Crosbie, 2003). The development of
speech sounds follows the oral motor development (Lundeborg, Nordin, Zeipel-Stjerna, & McAllister, 2015). Less motorically challenging speech sounds are developed earlier (Lohmander, Lundeborg, & Persson, 2017). The lips and jaws are suggested to be more important in early speech development, as 40% of the consonants are produced with the lips and jaw as the primary articulators (Stoel-Gammon, 1985). Most speech motor functions relevant for speech have reached an adult-like pattern at 14 years of age (Smith & Zelaznik, 2004).

Theories of Embodied Cognition could be one way to understand why oral motor development affects speech and language development (Adams, 2016). Several studies have shown a considerable overlap between motor difficulties and speech and language difficulties (Hill, 2001, Nip, Green, & Marx, 2011). The theory behind this interplay between motor and cognitive/linguistic processes is that they share the same neural network (Adams, 2016; Alcock, 2006; Hill, 2001). Studies on the relationship between language development and oral motor development in typically developing children indicate a strong relationship between language and motor skills (Alcock, 2006; Alcock & Connor, 2021). Two studies on motor performance and expressive language development in children with typical development (TD) by Alcock (2006) and Alcock & Connor (2021) have shown that children with poorer oral motor performance also have less developed expressive vocabulary at the age of 21 months and 3-4 years. Green et al. (Green, Moore, Higashikawa, & Steeve, 2000; Green, Moore, & Reilly, 2002) have shown that important changes in the lip and jaw musculature take place at the age of two years, at the same time as an extensive increase in expressive vocabulary occurs.

The interplay between motor function and cognition is undeniably important and movement/motor function plays an important role in speech and language development. This interaction theory has been further developed as a part of the embedded cognition/enactivism theory. The oral motor development influences the learning of speech sounds and there is also a phonological representation in the motor cortex (Adams, 2016). Impaired oral motor function could result in restricted speech sound development, which may lead to a limited vocabulary (Nip et al., 2011). Mimic musculature is also important, both for how the child is interpreted by their surroundings and for how the child itself interprets and understands expressed emotions and possibly words related to emotions (vocabulary again).

According to theories and research on mirror neurons (De Stefani & De Marco, 2019), areas in the brain are activated when we see someone else performing an action even if we don’t perform the action ourselves. We also quite automatically respond to a smile from another person with a smile. This interaction is likely influenced if the child is restricted in their own mimic musculature (which was the case in many of the children in this study; several children had difficulties raising the corners of their mouth to a smile). Motor development is important, both for emotional and cognitive development (De Stefani & De Marco, 2019), and presumably more important in early than in later speech and language development (Iverson, 2010; Nip, Green, & Marx, 2009). This underlines the importance of addressing
motor and oral motor development in children who are referred for speech and language difficulties. Alcock & Connor (2021) argue that motor skills—both oral motor and limb motor skills—need to be included in studies examining language development in order to understand the underpinnings of language development, and they conclude that “Speech and language development across the typically developing range is closely associated with both manual and oral motor skills.” (p.1957).

Studies of how children with SSD perform on detailed oral motor assessments could add knowledge to this growing field of research. Embodied theory suggests the need for some prerequisites to be met in order to develop certain skills. If they are lacking, the individual child’s development will be affected in different ways; for instance, the development of compensatory strategies.

2.3 AETIOLOGY

The genetics behind congenital speech and language disorders is a growing field of research and recent findings indicate that there could be a shared genetic foundation for several neurodevelopmental brain disorders, such as speech and language disorders, CAS, reduced cognitive function and deficits in motor development (Eising et al., 2018).

It is likely that the genes that put the child at risk of communication disorders also affect motor development (Bishop, 2002). This association seems to be strongest when speech production is affected. The heredity for speech and language disorders is well known and well documented. The underpinning genetics are suggested to be complex and involve multiple loci (Chen et al., 2017). Alteration of the FOXP2 gene is known to cause speech and language disorders, including CAS and dysarthria (Newbury & Monaco, 2010). Several other gene mutations and diseases have been identified to result in specific impairment of speech and language (Morgan et al., 2017). Gillberg (Gillberg, 2010) has estimated that approximately 1% of the population is affected by a genetic condition, in ESSENCE terminology called “behavioural phenotype syndromes”. Many of these syndromes are associated with a wide palette of neurodevelopmental disorders. The underlying genetic cause is often missed (Gillberg, 2010).

Subtle abnormalities in motor neural circuitry have been suggested to be affected in children with persistent speech disorders (Redle et al., 2015), as well as aberrations in the corpus callosum (Luders et al., 2017). Connectivity anomalies in specific brain regions involved in speech/language function have been seen in children with CAS (significant alterations of inter- and intra-hemispheric connections of bilateral brain regions) (Fiori et al., 2016). In their article on the aetiology of CAS, Morgan and Webster (Morgan & Webster, 2018) suggest that altered connectivity of the left corticobulbar tract may be a neural marker of developmental speech disorders. The study by Eising et al. (2108) identified molecular pathways that are involved in the regulation of gene expression during early brain development that may be critical for the acquisition of fluent spoken language.
Morgan & Webster (2018) showed that oromotor disorders and/or oromotor praxis were present in all the genetic conditions they present as examples of a genetic cause of CAS (FOXP2-only, FOXP2-plus, GRIN2A, SETBP1, 2p15p.16.1 microdeletion, 12p13.33 microdeletion, 17q21.31 microdeletion, 16p11.2 deletion). Intellectual disability (ID), language deficits and oromotor disorders were present in almost all those conditions and show that there is an overlap of neurodevelopmental symptoms in conditions associated with CAS. Liegeois et al. (Liegeois et al., 2019) found the same overlap of SSD/CAS and orofacial dysfunction in a study of a family with a genetically unidentified inherited SSD, where 12/13 of the family members had CAS and oral motor impairment. Barnes et al. (2006) found atypical oral structures and oral motor function in boys with Fragile X and Down’s syndrome. The oral motor and speech motor difficulties in the Down’s syndrome population are well documented (Kumin, 2006) and CAS is probably more common in this population (Kumin, 2006) than previously thought.

Bilateral perisylvian syndrome (also called Bilateral Perisylvian Polymicrogyria (BPP) and Worster-Drought syndrome) is a neurological condition where orofacial dysfunction and motor speech disorders are very common (Braden et al., 2019). It is characterised by malformations in the perisylvian region (sylvian fissures). In some cases, these malformations are not visible on magnetic resonance imaging (MRI) (Clark, Chong, Cox, & Neville, 2010), but the severity of the disorder is not related to how large the malformation is. This condition is probably underdiagnosed, and the diagnosis is often made quite late in childhood, despite the difficulties with oral motor function and speech. The orofacial dysfunction and speech disorder may be very severe while other cognitive and gross motor functions are often less affected. This may be one reason for the late diagnosis in many cases. Both heritable and de novo genetic causes of BPP have been described (Mirzaa et al., 2015; Stutterd & Leventer, 2014).

2.3.1 Coexistent difficulties

The complex genetic/polygenetic cause underlying NDD is probably the reason for the high incidence of coexistent symptoms in children with SSD. The definition “coexistent” or “co-occurrence” describe the phenomenon better than the term “co-morbidity”, as this term implies that one condition is regarded as the primary condition and suggests individual aetiologies (Brimo et al., 2021). Several studies indicate a co-occurrence of different NDDs in line with suggestions within the ESSENCE concept (Brimo et al., 2021; Gillberg, 2010; Gillberg & Billstedt, 2000; Kaplan, Dewey, Crawford, & Wilson, 2001; Lundström et al., 2015). The ESSENCE concept offers a model describing the interaction between different neurodevelopmental disorders. Gillberg (2010) claims that specific disorders, such as language disorders, ADHD and DCD, should not be seen as separate conditions but as a combination of symptoms that largely overlap. In a study following children identified with language problems through child health screening at the age of 2.5 years in Sweden, 72% had neuropsychiatric or learning disorders at the age of seven years (Miniscalco, Nygren, Hagberg, Kadesjo, & Gillberg, 2006).
DCD is regarded as one of the most common neurodevelopmental disorders and probably highly underdiagnosed (Gillberg, 2018). In a study by Miniscalco et al. (2006), a third of the children with early detected language disorders met the criteria for DCD at seven years of age. Brimo et al. (2021) also found that DCD was more common in children with dyslexia than in children without dyslexia. In ADHD, around 50% are considered to also fulfil the criteria for DCD (Athanasiadou et al., 2020). DCD is characterised by a delay in the development of gross and fine motor skills, poor motor planning and coordination, resulting in difficulties to acquire everyday skills (Leonard & Hill, 2015). The prevalence of DCD varies in different countries according to diagnostic practice, but is estimated to occur in a severe form in 5% of children (Kadesjö & Gillberg, 1999).

Hypermobility of joints is another symptom that may coexist with motor difficulties (Adib, Davies, Grahame, Woo, & Murray, 2005; Kirby & Davies, 2007) and NDD (Adib et al., 2005). (Baeza-Velasco, Grahame, & Bravo, 2017). It is also a common symptom in many genetic diagnoses, such as Down’s syndrome, Williams syndrome, Ehlers-Danlos syndrome (EDS), Marfan syndrome and Fragile X, and is more common in females than in males (Adib et al., 2005). There are some genes that are known to cause severe connective tissue disorders, such as vascular, classic, and kyphoscoliotic forms of EDS (Castori et al., 2012), but more mild and more common hypermobility is thought to have a complex genetic background, like many NDDs. Connective tissue disorders may cause voice disorders, speech difficulties and orofacial dysfunction, such as low muscle tone and hypermobility of oral structures (Celletti et al., 2015; Rimmer, Giddings, Cavalli, & Hartley, 2008).

There are several studies that have documented gross and fine motor difficulties in children with speech and language impairments (Hill, 2001; Visscher, Houwen, Scherder, Moolenaar, & Hartman, 2007). Redle et al. (Redle et al., 2015) reported that children with SSD exhibited poorer oral and fine motor skills compared with typically developing children. They used functional MRI (fMRI) to examine the brain networks and one of their findings was that children with persistent speech disorders displayed overactivation in the cerebellum during motor tasks (Redle et al., 2015). This was assumed to be related to a subtle abnormality in the motor neural circuitry which could affect fine motor praxis.

Few studies have investigated orofacial function in children with SSD, but it is likely that the same functions that control gross and especially fine motor skills also influence oral motor function. Oral motor difficulties could be a symptom of a coexistent motor disorder.

2.4 OROFACIAL FUNCTIONS AND SENSORY-MOTOR FUNCTION

2.4.1 Definition

Chewing, sucking and swallowing, saliva control, breathing, sensory function, facial expression, and speech, are all vital orofacial functions. Using the term “orofacial functions” is a way to state that the condition not only refers to specific muscle movements, but also to the different activities that these muscles are involved in. The underlying theoretical background in this thesis is that if oral motor function (strength, tone, mobility, motor
planning/co-ordination of the muscles) and/or intraoral and extraoral sensory function (tactile and proprioceptive feedback) are affected in a negative way for some reason, this will affect functions such as chewing, swallowing, moving the articulators for speech, etc. This could relate to the International Classification of Functions, Disability and Health (ICF) components (WHO, 2001) regarding oral sensory-motor function as “Body structures” and orofacial functions as “Body functions”.

Orofacial dysfunction is common in syndromes and rare diseases, where as many as every other child has problems with speech, eating and saliva control (Sjogreen, Mogren, Andersson-Norinder, & Bratel, 2015). In children with CAS, oral motor difficulties were one of the most common coexisting symptoms according to parental reports (Teverovsky, Bickel, & Feldman, 2009). Orofacial dysfunctions may have a great impact on quality of life (Klingberg et al., 2010, Johnson et al., 2016). Deviant or delayed general development may be associated with orofacial dysfunctions (Bergendal, Bakke, McAllister, Sjogreen, & Asten, 2014).

The mouth and face are richly innervated and the orofacial muscles are used in a variety of functions. Even if the same muscles are used for different functions, such as chewing, swallowing and speech, they are not controlled in the same way. It is unique for the orofacial muscles that they can be used in such a heterogenic way (Kent, 2015). There is an ongoing discussion about how this overlap of functions should be interpreted. Some argue that oral motor difficulties and language and speech difficulties are to be interpreted as symptoms of the same underlying disorder (Kent, 2015). Others claim that speech is specific to the domain of linguistic expression and that those functions cannot be compared with other motor activities (Ziegler & Ackermann, 2013). Typically developing children have good oral-motor control before the age of four (Martinez & Puelles, 2011), even if the development continues and is refined, especially for speech, throughout childhood.

The sensory part of motor disorders is crucial (Patel, Jankovic, & Hallett, 2014) and it is not possible to separate those functions from each other as they interact and influence each other; for example, the importance of peripheral sensory feedback in the execution and planning of voluntary movement is well described (Nijs et al., 2012). Thus, the term sensory motor/sensorimotor is a more correct description (Patel et al., 2014). Well-balanced sensory motor function in the mimic muscles, lips, jaw, and tongue is important for eating, drinking, swallowing and managing saliva control (Martinez & Puelles, 2011). Sensory feedback is an essential factor in regulating mastication (Peyron, Lassauzy, & Woda, 2002) and in speech development (Crary, Fucci, & Bond, 1981). Sensory function includes both tactile and proprioceptive feedback. There are superficial and deep mechanoreceptors in the oral tissues that react to different stimuli. The superficial mechanoreceptors identify more tactile input (two-point discrimination), and the deep mechanoreceptors have more proprioceptive qualities (oral stereognosis) (Sivapathasundharam et al., 1995). Oral stereognosis is the process where sensory information is perceived through mucosal receptors, the tongue, and receptors in the gums, lips, and temporomandibular joints, and then interpreted (Park, 2017).
This skill is decreased in the elderly population and declines markedly after a stroke (Park, 2017). It is also reported that children with mouth breathing have difficulties with oral stereognosis (Norström, 2003). Proprioception is both unconscious and conscious perception of different positions of body structures and the movements of those structures in space (Strand, 2020). Children with CAS are thought to have impaired proprioception skills (Strand, 2020).

2.4.2 Jaw function

The jaw is suggested to be the most prominent articulator in early speech production. In early speech development, the opening and closure of the jaw produce bilabial consonants and syllable gestures. Control of the jaw develops earlier than control of the lips and tongue (Green et al., 2000). According to Green et al. (2000), jaw movements are adult-like already at twelve months of age, but lip and tongue movements develop much later. Coordination between lip and jaw movements appears to be adult-like at six years of age (Green et al., 2000), but coordination between the jaw and tongue develops later (Cheng, Murdoch, Goozee, & Scott, 2007). Jaw control is the foundation for the development of movement in the lips and tongue (Kent, 1999). Control of the jaw is essential for complex speech. The jaw muscles and the temporomandibular joint are coordinated in a functional synergy, and controlled, graded movements are important for both speech and chewing (Hebert, 2013). Being able to use regulated force is an important factor in both chewing and speech and immature motor control is often characterised by the inability to use adequate force. This may be one reason why children master stops before fricatives in early speech development (Green & Nip, 2010). Grigos and Kolenda (Grigos & Kolenda, 2010) showed that children with CAS have different jaw movements than typically developing children and that improved jaw stability resulted in improved precision and reduced variability in consonant production.

2.4.3 Methods to assess orofacial functions and sensory-motor function

In a narrative review of non-speech oral movements and oral motor disorders, Kent (2015) states that it is important to examine oral motor skills as a part of an overall assessment of children with delayed language development, as their difficulties are rarely limited to language only, and such an assessment may reveal important information about neurology and motor function. He also argues for a holistic assessment and underlines the importance of separating motor skill learning from strength and endurance, both in assessment and treatment, as those skills require different performance. Murray et al. (Murray, Iuzzini-Seigel, Maas, Terband, & Ballard, 2021) state that an oral motor assessment is important in the diagnostic procedure of CAS. They recommend a complete oral motor assessment, including diadochokinetic (DDK) tasks and polysyllabic single-word production for a reliable diagnosis of CAS.

Green and Nip (2010) state that the development and function of speech motor control has been sparsely studied compared with other motor functions and one of the reasons for this is
the lack of proper methods. McCauley and Strand (McCauley & Strand, 2008) conclude that “The field’s limited understanding of motor speech disorders in children is demonstrated most powerfully by a lack of agreement on core characteristics that can help guide test construction and validation and lead to the development of a test that can serve as a gold standard.” (p.82). They found that tests in this area were “inadequately developed from a psychometric perspective” (McCauley & Strand, 2008, p.88). The VMPAC (Hayden & Square, 1999) was the only test in the review by McCauley and Strand that met the criteria for adequate norms. This test is not officially distributed in Swedish. A translation into Swedish was used in a master thesis (Björelius Hort, 2009) but the translation is not available from the publisher. Many international studies on oral motor performance have used the Robbins & Klee Oral Speech Motor Protocol (1987). It is a standardised assessment based on direct observation of orofacial structures and function during both speech and non-speech tasks. The test includes normative information for children between the ages of two and six years. TD children are expected to master all items in the test by the age of six. The original study included 90 children with TD and the author states that additional evaluation of the protocol is needed to determine if the test is sensitive enough to distinguish TD children from children with orofacial dysfunction (Robbins & Klee, 1987). However, no further studies have investigated the reliability, sensitivity and validity of the test. An Italian study of 191 TD children (Granocchio et al., 2021) presented normative data for Italian children.

There are four tests for assessing sensory-motor function and orofacial functions in Sweden: Stockholms oralmotoriska bedömningsprotokoll (STORM – the Stockholm oral motor assessment protocol) for children and adults, as yet without normative values or validity testing (Henningsson et al., 2007, Hartstein, 2020), the ORIS – with normative values for children up to seven years of age (Holmberg & Bergström, 2008), the Dysarthria test with normative values for adults (Hartelius, 2015), and the Nordic orofacial test (NOT-S) with normative values for children and adults (Bakke, Bergendal, McAllister, Sjogreen, & Asten, 2007; McAllister & Lundeborg, 2013). The NOT-S is a validated screening test developed to assess orofacial functions (Bakke et al., 2007). It is regarded as a comprehensive test that covers several orofacial functions. It consists of a structured interview and a clinical examination. The NOT-S has been translated into many languages and is widely used in scientific studies. It has also been used for several different patient groups to describe the orofacial dysfunction profile (Bergendal et al., 2014; Edvinsson & Lundqvist, 2016).

Assessments of sensory-motor function are often made during observations, using a structural test protocol. Measuring orofacial muscle strength can be a way to acquire a quantitative measure of muscle function. Measures of bite force, lip force and tongue force can provide important information on muscular status, and such strength measurements are used in several studies of orofacial function in different populations (Clark & Solomon, 2012; Hagg, Olgarsson, & Anniko, 2008; Potter, Nievergelt, & VanDam, 2019; Sjogreen, Tulinius, Kiliaridis, & Lohmander, 2010).
Assessments of intraoral sensory function have been performed using several different methods, both clinically and in research, and so far, there is no consensus about which method is the most reliable and valid (Boliek et al., 2007). Two-point discrimination tasks and oral stereognosis are two of the most widely used methods (Jacobs, Bou Serhal, & van Steenberghe, 1998; Jacobs et al., 2002; Kumin et al., 1984). Two-point discrimination is regarded as a reliable method and has been used for neurological examinations for a long time. One limitation is that it is difficult to control for applied force when using the tool on the lips and tongue (Jacobs et al., 2002). Assessment of intraoral stereognosis is also known to be a clinically applicable method (Park, 2017).

Kinematic measurement methods are one way to study orofacial movements. Visual motion analysis programmes in 2D or 3D have been used to study lip and jaw movements during speech (Grigos, 2009; Grigos & Kolenda, 2010; Terband, 2013; Ward, Strauss, & Leitão, 2013). By using measurement points in the face (natural landmarks or reflectors that provide information on position), it has been possible to calculate the duration, displacement and velocity of lip and jaw movements (Grigos, Saxman, & Gordon, 2005; Sjogreen, Lohmander, & Kiliaridis, 2011). In most visual motion analysis programmes, only movements of visible articulators could be analysed (Green & Nip, 2010), so there are more studies of the coordination of the lips and the jaw than of the tongue. Electropalatography (EPG) (Gibbon, 1999) and ultrasound have been used in studies of tongue movements (Sugden, Lloyd, Lam, & Cleland, 2019). It is difficult to use kinematic measurement methods in research in young children as those methods often require full participation.

To summarise, there is consensus about the importance of an orofacial functional assessment for children with motor speech disorders but there is no consensus on which tests and methods to use. Also, there is still a lack of validated and reliable methods to assess orofacial functions in children with SSD.

2.5 MALOCCLUSION

Occlusal development is affected by both genetic and environmental factors. Growth pattern, muscle function, breathing patterns, oral habits and early tooth extractions are known to influence occlusal development (Linder-Aronson, 1970; Ovsenik, Farcnik, Korpar, & Verdenik, 2007). At approximately 16 months, the occlusal contact between the first molars, which provides the prerequisites for stable jaw movements, is established (Widmer, 1992). Malocclusion has been defined as “…not a disease but rather a variation from accepted societal norms that can lead to functional difficulties or concerns about dento-facial appearance...” (Dimberg, 2015, p.16). The prevalence of malocclusion varies with age and across different populations. In Sweden, approximately 58% of seven-year-old children exhibit malocclusions (Dimberg, Lennartsson, Arrrup, & Bondemark, 2015). Malocclusions are common in individuals with neurodevelopmental disorders (de Castilho et al., 2018; Fontaine-Sylvestre, Roy, Rizkallah, Dabbagh, & Ferraz Dos Santos, 2017; Miamoto et al., 2010; Vellappally et al., 2014).
2.5.1 Relationship between orofacial dysfunction and malocclusion

Orofacial dysfunction and malocclusion often coexist. A relationship between oral dysfunction, open mouth posture, oral habits and tongue protrusion was found in individuals with open bite and posterior crossbite (Dimberg, Lennartsson, Soderfeldt, & Bondemark, 2013; Grabowski, Kundt, & Stahl, 2007). Also, individuals with rare diseases and orofacial function had a higher prevalence of malocclusions than individuals with rare diseases without orofacial dysfunction (Sjogreen, Andersson-Norinder, & Bratel, 2015).

Malocclusion can affect the person’s orofacial function. Good occlusal contacts are important to prepare the bolus (Fontijn-Tekamp et al., 2000) and malocclusion can cause decreased chewing efficiency if occlusal contacts are reduced (Magalhães, Pereira, Marques, & Gameiro, 2010). Some speech sounds may be influenced by different types of malocclusion, especially structural deviations in the anterior part of the oral cavity, but often to a minor degree (Jensen, 1968; Laine, 1987; Subtelny, Mestre, & Subtelny, 1964). An anterior open bite can result in interdental production of dental fricatives (e.g., /s/), and the articulation of labio-dental fricatives (/f/, /v/) can be affected by Class III occlusion (Profitt, 2013). However, speech is a complex cognitive and motor activity with specific requirements on precision and neurological control (Moore & Ruark, 1996). Koskela et al. (Koskela et al., 2020) have reported that children with severe malocclusions have speech difficulties more often than control subjects. However, they concluded that this might reflect a shared genetic aetiology rather than a causal relationship.

There are several reports that indicate that orofacial dysfunction can affect occlusal development (Behlfelt, 1990; Dimberg et al., 2013; Kiliaridis, Johansson, Haraldson, Omar, & Carlsson, 1995; Kiliaridis & Katsaros, 1998; Linder-Aronson, 1970; Sjogreen, Andersson-Norinder, et al., 2015). There are several studies showing that orofacial dysfunction and reduced oral muscular strength can influence facial growth in a negative way (Kiliaridis et al., 1995; Kiliaridis & Katsaros, 1998). Posterior crossbite can also develop due to a low resting position of the tongue (Ovsenik, 2009). Folletti and colleagues (Foletti, Antonarakis, Galant, Courvoisier, & Scolozzi, 2018) saw an association between atypical swallowing patterns and relapse after orthognathic surgery.

2.6 RATIONALE FOR THE THESIS

Several areas are included in this thesis where there are knowledge gaps in the literature. A rationale for this thesis was to add knowledge to the existing literature on orofacial functions and malocclusion in children with persisting SSD while using a multi-professional approach, and as far as possible using validated, reliable, and objective methods to assess orofacial functions.
3 RESEARCH AIMS

3.1 GENERAL AIM
The overall aim of this project was to investigate orofacial functions, speech characteristics, occlusion, and co-existing symptoms in children with SSD of unknown origin persisting after the age of six years.

3.2 SPECIFIC AIMS
The thesis includes four studies, with the following specific aims:

I. To investigate speech, orofacial functions and neurodevelopmental symptoms in children with SSD.

II. To compare movement patterns of the lips and jaw during vowel production in children with TSD and children with SSD.

III. To investigate the occurrence, type, and severity of malocclusions in children with SSD and compare these findings to children with TSD.

V. To investigate differences in orofacial functions between children with SSD and children with TSD and explore possible associations between orofacial functions and malocclusion.
4 MATERIALS AND METHODS

4.1 PARTICIPANTS

The children with SSD were all consecutive patients who met the inclusion criteria and were referred to Mun-H-Center, a national orofacial resource centre, for a speech and oral motor examination in 2014-2016. The inclusion criteria were SSD persisting after the age of six. This age was selected as Swedish-speaking children are expected to manage all Swedish speech sounds at the age of six (Blumenthal & Lundeboerg Hammarström, 2014). The exclusion criteria were moderate to severe ID, cerebral palsy and/or severe autism spectrum disorder (ASD). Sixty-two patients met the inclusion criteria and were offered to participate in the study, one adolescent declined to participate. In total, 61 children with SSD were included, aged 6.0-16.7 years (mean age 8.5), 14 girls and 47 boys.

Referrals came from speech-language pathologists (SLPs) (n = 47), physicians (n = 7), school health services (n = 6) and self-referral (n = 1). The reasons for referral were requests for an oral motor and speech motor examination and/or a second opinion, as the child had not improved their speech as expected from previous speech-language interventions. All participants were children who had SSD of unknown origin where speech difficulties had not resolved at the age of six years, despite long-term contact with an SLP. The median age for the first SLP visit was 4.0 years (2.0-7.3 years). Thirteen per cent of the parents were unable to remember how old their child was at his/her first appointment with an SLP. Five participants were raised in bilingual homes but had Swedish as their first language and two children were adopted internationally at 2.6 and three years of age, respectively. Three sibling pairs were included. All participants but one followed the regular curriculum for compulsory schooling.

There was a wide range of descriptions of the children’s speech difficulties in the referral; 29 children came with a diagnosis of CAS or with suspected CAS, 25 had a diagnosis of phonological impairment stated in the referral, 18 had a diagnosis of DLD even if their language ability in some cases was described as age adequate, four did not have a diagnosis of DLD but were described as having delayed speech and language development, 15 were described as having oral motor difficulties, seven as having unclear speech, three had a diagnosis of reading and writing disorders, and two children had a fluency disorder in addition to SSD. It was specifically stated for eight children that they had no language difficulties. Several children had more than one of the above diagnoses.

A control group of children with TSD was included in three of the studies in this thesis (II, III, IV). The TSD group consisted of 44 children, aged 6.0-12.2 years (mean age, 8.8), 19 girls and 25 boys, recruited from the Public Dental Health service and through personal contacts. The inclusion criteria were typical speech development and no known neurodevelopmental disorder. All the children in the TSD group were screened with the Nordic Orofacial Test – Screening (NOT-S) to confirm that no oral motor or speech sound disorders were present. Two children in the TSD group were bilingual but had Swedish as their first language.
Table 1. Distribution of age and sex in the SSD and TSD groups.

<table>
<thead>
<tr>
<th>Age groups (year: months)</th>
<th>SSD Girls</th>
<th>SSD Boys</th>
<th>SSD Total</th>
<th>TSD Girls</th>
<th>TSD Boys</th>
<th>TSD Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>6–6:11</td>
<td>3</td>
<td>19</td>
<td>22</td>
<td>3</td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td>7–7:11</td>
<td>5</td>
<td>9</td>
<td>14</td>
<td>4</td>
<td>3</td>
<td>7</td>
</tr>
<tr>
<td>8–8:11</td>
<td>1</td>
<td>4</td>
<td>5</td>
<td>4</td>
<td>7</td>
<td>11</td>
</tr>
<tr>
<td>9–9:11</td>
<td>1</td>
<td>5</td>
<td>6</td>
<td>5</td>
<td>5</td>
<td>10</td>
</tr>
<tr>
<td>10–10:11</td>
<td>0</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>6</td>
<td>8</td>
</tr>
<tr>
<td>11–11:11</td>
<td>3</td>
<td>2</td>
<td>5</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>12–16:7</td>
<td>1</td>
<td>6</td>
<td>7</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>14</td>
<td>47</td>
<td>61</td>
<td>19</td>
<td>25</td>
<td>44</td>
</tr>
</tbody>
</table>

### 4.2 PROCEDURE

#### 4.2.1 Data collection

The data used in all four studies were collected during one, or in some cases two, visits to the clinic at Mun-H-Center, Gothenburg, Sweden. The same SLP (the author) performed all the assessments. The dental examinations were mainly performed by the same orthodontist. All speech assessments were audio-recorded (Tascam HD-P2; Tascam, USA) using a stereo microphone (SONY ECM-MS957; Sony, Japan), and videotaped (Canon Legria HF S11; Canon, Japan) with an external microphone (Canon DM-100; Canon, Japan). All assessments of orofacial function were videotaped. All recorded data were coded and saved on a locked and secured, password-protected hard drive used specifically for this purpose. No personal data were saved digitally. As the participants were consecutive patients, all personal data and photos were saved in the digital patient record system used in the public dental health service, Region Västra Götaland, Sweden.

The entire assessment procedure took about two hours. The child was given breaks when needed. The dental examination was performed in a dental chair with full dental equipment. The other assessments were carried out in a separate room with the child seated in a stable sitting position during the whole assessment. The chair used was an ergonomic work chair for children with manual handbrakes, adjustable backrest, neck support, armrest, and foot plate (Mercado Medic).
4.3 MATERIAL

Table 2. Overview over material and analyses used in the different studies included in the thesis project “Orofacial function in children with SSD”.

<table>
<thead>
<tr>
<th></th>
<th>Study I</th>
<th>Study II</th>
<th>Study III</th>
<th>Study IV</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Speech</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PCC</td>
<td>X</td>
<td>X</td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>PVC</td>
<td>X</td>
<td>X</td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>Consonant inventory</td>
<td>X</td>
<td></td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>Nasality deviations</td>
<td>X</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Intelligibility (ICS)</td>
<td>X</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Speech disorder diagnosis</td>
<td></td>
<td></td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td><strong>Orofacial function</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>NOT-S</td>
<td>X</td>
<td>X</td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>Bite force</td>
<td></td>
<td></td>
<td></td>
<td>X</td>
</tr>
<tr>
<td>Jaw instability (bite blocks)</td>
<td></td>
<td></td>
<td></td>
<td>X</td>
</tr>
<tr>
<td>Chewing efficiency</td>
<td></td>
<td></td>
<td></td>
<td>X</td>
</tr>
<tr>
<td>Oral stereognosis</td>
<td></td>
<td></td>
<td></td>
<td>X</td>
</tr>
<tr>
<td>3D video analysis</td>
<td></td>
<td></td>
<td></td>
<td>X</td>
</tr>
<tr>
<td><strong>Malocclusion</strong></td>
<td></td>
<td>X</td>
<td>X</td>
<td></td>
</tr>
<tr>
<td><strong>Oral characteristics</strong></td>
<td></td>
<td>X</td>
<td></td>
<td>X</td>
</tr>
<tr>
<td>Background data (co-existing symptoms, oral habits)</td>
<td>X</td>
<td>X</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Control group of children with TSD</td>
<td>X</td>
<td>X</td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>Prospective cross-sectional Study</td>
<td>X</td>
<td>X</td>
<td>X</td>
<td>X</td>
</tr>
</tbody>
</table>

1 Result only used as background data and not for statistical analysis

4.3.1 Speech assessment and analysis

For speech assessment, the Swedish articulation and nasality test (SVANTE) (Lohmander et al., 2005; Lohmander et al., 2017) was used. It includes picture naming of single words for assessment of consonants and repetition of sentences for assessment of nasality. The ICS questionnaire was used for assessment of intelligibility.
**Consonant and vowel production**

SVANTE is a validated test for the assessment of speech production, including consonant proficiency and consonant errors (Lohmander et al., 2017). It consists of 86 single words (48 monosyllabic and 38 disyllabic) elicited via pictures. It also includes sentence repetition to assess connected speech. All Swedish consonants are included, and most occur in three possible realisations in the initial, medial, and final position. The consonants /ɕ/ and /h/ only exist in initial positions and /ŋ/ only exists in medial and final positions according to Swedish phonotax. An analysis of the speech material was made in accordance with test instructions (Lohmander et al., 2017), but assessment of vowels is not included in the original use of the test.

The outcome measurements were consonant proficiency (Percentage Consonants Correct (PCC)), Percentage Vowels Correct (PVC), and consonant inventory. A narrow phonetic transcription of single words was made from the audio recordings. The consonants and vowels were then scored as correct or incorrect, according to instructions in Shriberg et al. (1997). When scoring PCC, all distortions were rated as incorrect, but all Swedish allophones of /r/ were rated as correct. Typically developing five and seven-year-old Swedish children have a mean PCC on the SVANTE test of 95.8 (± 4.8) and 97.8% (± 3.3), respectively (Lohmander et al., 2017). At 19 years of age, the mean PCC is almost 100% (99.4 ± 1.5). For the consonant inventory, a 90% threshold was used in accordance with Blumenthal & Lundeborg Hammarström (2014). A consonant phoneme is regarded as established if it is correctly produced in 90% or more possible realisations. SVANTE is originally not designed for assessing vowels, but it includes all Swedish vowels, except for /ʏ/. Most vowels are realised several times but /ɪ/, /ʏː/, /ʊ/, /œː/, and /œ/ only exist once. Typically developing children have a 100% PVC from the age of four years (Blumenthal & Lundeborg Hammarström, 2014).

PCC is a widely used method to measure articulation competence. It was first described by Shriberg & Kwiatowski in 1982, but since then, several adjustments have been made and alternatives developed, both by the developer and other researchers. In the original version, it was used with spontaneous speech samples (Shriberg & Kwiatkowski, 1982) but it has also been used with naming tests (Klinto, Salameh, Svensson, & Lohmander, 2011). When spontaneous speech is used, there is a risk that the child only uses the speech sound that he/she masters. By using a naming test, it is possible to get a more detailed picture of the child’s performance. Consonants are scored as correct or not correct based on phonetic transcription. Shriberg et al. (1997) argue that narrow phonetic transcription should be used as this gives a more detailed description of the speech errors and a more reliable assessment of substitutions or distortions. Alternatives to the original way to assess PCC, where all distortions, deletions and substitutions are counted as incorrect, are the PCC-A (Adjusted) and the PCC-R (Revised). In PCC-A, common clinical distortions in the age group are scored as correct, and in PCC-R all distortions are scored as correct. The reason for using different PCC scores is that the original PCC counts distortions equal to more phonologically based difficulties such as omissions and substitutions. As a complement to PCC scores, PVC and PPC may also be used (Shriberg et al., 1997). PVC assesses the percentage of vowels produced in a correct way and in PPC, all speech sounds are counted, both consonants and
vowels. In PPC, only omission and substitution are scored as incorrect. All variants of the PCC measure are binary. Shriberg & Kwiatowski (1982) used the PCC score to classify the severity of the speech disorder. They categorised the PCC scores into four sectors: mild = 85-100% correct, mild-moderate = 65-85%, moderate-severe = 50-65%, and severe = less than 50%. How these cut-offs were established is not described.

Another way to measure consonant proficiency is to calculate the number of established consonants and this is often used together with a metric measurement like PCC. For a consonant inventory, two different thresholds can be used. In Lohmander et al. (2017), a consonant phoneme is estimated to be established in children at three and five years of age if correctly produced in 50% or more of possible realisations. In Blumenthal & Lundeborg, Hammarström (2014), a 90% accuracy criterion was used. The 90% accuracy criterion seems to be more widely used. In this thesis, the 90% criterion was used as the children were six years of age and older.

*Nasality*

Recordings of sentence repetitions in SVANTE were used to assess nasality perceptually. Nasality variables were rated on a four-point ordinal scale (no, mild, moderate, or severe deviance), according to Lohmander et al. (2017). The parameters assessed were hypernasality, hyponasality, audible nasal air leakage and reduced pressure on consonants. The parameter mixed/varying nasality was added to the original scale. The term deviant nasality is used throughout this thesis. Hyper- and hyponasality are regarded as resonance disorders but audible nasal air leakage and reduced pressure on consonants are nasality variables (Lohmander et al., 2017).

*Intelligibility*

Assessments of articulation are not sufficient to assess intelligibility (Ertmer, 2010). A child can sometimes produce more correct speech in single words than in connected speech. PCC was developed to determine the severity of an SSD but does not give the full picture of a child’s intelligibility. Intelligibility can be rated from spontaneous speech samples. However, children that are highly unintelligible often refrain from talking in a test situation. The ICS is a questionnaire used to measure the functional intelligibility of children with SSD (McLeod, Harrison, & J. McCormack, 2012). It consists of seven items where the parents rate the degree of the child’s intelligibility in different contexts and in everyday life on a five-point scale. The ICS questionnaire has been translated into Swedish (Lagerberg, 2014). Normative data for Swedish children have recently been published by Lagerberg and colleagues (2021). Swedish children between three and nine years of age had a mean score of 4.73 on the ICS questionnaire (Lagerberg et al., 2021). German-speaking children with SSD, between three and six years of age, had a mean score of 3.97 in a study by Neumann and colleagues (2017). Lagerberg et al. (2021) also investigated validity, comparing the results from the ICS questionnaire with transcription-based evaluations of spontaneous speech. They found weak correspondence between the ICS and the transcription-based evaluations and conclude that the ICS should be used in combination with other methods when assessing intelligibility.
4.3.2 Differential diagnostics of SSD

It was not an aim of this thesis to differentiate between disorders within SSD and the process and results of differential diagnostics of SSD are not fully described in any of the included articles; however, the participants’ different SSD diagnoses are presented as background information in Study II, III and IV.

Nevertheless, as the participants were also consecutive patients at the clinic, this was part of the clinical assessment and of importance for advice on further interventions. A clinical diagnosis was made at the visit but to determine in a more structured way what diagnosis was present, the operationalised 12 CAS features list by Iuzzini-Seigel & Murray (2017) was used. This list has also been used for Swedish children with CAS in a thesis by Malmenholt (2020). For the other diagnoses, the detailed description of the features of different SSDs, according to Shriberg’s classification system SDCS by Namasivayam et al. (Namasivayam et al., 2019), was used. The criteria for obtaining a diagnosis of SMD are not clearly specified in the literature. SMD is described as a “disorder of execution: a delay in the development of neuromotor precision-stability of speech motor control” (Shriberg, Campbell, et al., 2019). In this thesis, a child was diagnosed with an SMD if they did not fulfil the criteria for CAS but had obvious signs of motor speech disorders such as nasality (without having a cleft), voice deficits and consonant distortions and vowel errors. Also, in the SMD group there were several participants that had inconsistency (n = 7) but did not get a diagnosis of CAS as they did not fulfil the criteria of five features from the list and had no other specific CAS features, such as stress errors and groping.

Twenty-five (41%) children were eventually assessed as having CAS, 23 (38%) as having SMD, nine (15%) had SMD/sCAS, three (5%) AI, and one (2%) DD. In 15 (52%) of 29 participants with a diagnosis of CAS or suspected CAS in the referral, the diagnosis was confirmed (CAS or sCAS).

Some children did not fulfil all the proposed criteria of CAS (five features from the operational list + inconsistency), but still had a motor speech disorder that included several CAS features. They were diagnosed with sCAS. The group sCAS consists of children that showed obvious motor speech features, like the SMD group, but did not fulfil the criteria of CAS. Participants were placed in this group if they had less than five features from the list but several features of CAS, like inconsistency (all but two had this feature), groping and stress errors.
Table 3. Features list by Iuzzini-Seigel & Murray (2017) used in differential diagnostics for the different motor speech disorders. Data on voice deviation and PCC is added. Calculation of percentages was not used for AI and DD as the subgroups contained few participants.

<table>
<thead>
<tr>
<th>Feature</th>
<th>All N=61</th>
<th>CAS N=25</th>
<th>SMD N=23</th>
<th>sCAS N=9</th>
<th>AI N=3</th>
<th>DD N=1</th>
</tr>
</thead>
<tbody>
<tr>
<td>Consonant distortion</td>
<td>60 (98%)</td>
<td>25 (100%)</td>
<td>22 (96%)</td>
<td>9 (100%)</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>Vowel error</td>
<td>51 (84%)</td>
<td>25 (100%)</td>
<td>17 (74%)</td>
<td>8 (89%)</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Stress error</td>
<td>31 (51%)</td>
<td>23 (92%)</td>
<td>6 (26%)</td>
<td>2 (22%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Syllable segregation</td>
<td>17 (28%)</td>
<td>12 (48%)</td>
<td>2 (9%)</td>
<td>2 (22%)</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Groping</td>
<td>23 (38%)</td>
<td>17 (68%)</td>
<td>3 (13%)</td>
<td>3 (33%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Intrusive schwa</td>
<td>15 (25%)</td>
<td>9 (36%)</td>
<td>5 (22%)</td>
<td>1 (9%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Voicing error</td>
<td>36 (59%)</td>
<td>21 (84%)</td>
<td>9 (39%)</td>
<td>5 (55%)</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Slow rate</td>
<td>5 (8%)</td>
<td>4 (16%)</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Increased difficulties</td>
<td>1 (2%)</td>
<td>0</td>
<td>1 (4%)</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>with multisyllabic words</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
4.3.3 Assessment of orofacial functions and sensory-motor function

‘NOT-S
The NOT-S screening test is divided into twelve domains (six domains in the interview part and six in the examination part) addressing “Sensory function”, “Breathing”, “Habits”, “Chewing and swallowing”, “Drooling” and “Dryness of the mouth” in the structured interview, and “Face at rest”, “Nose breathing”, “Facial expression”, “Masticatory muscles and jaw function”, “Oral motor function” and “Speech” in the clinical examination. Each domain includes one to five items depending on complexity. The scoring is based on “yes” or “no” and the criteria are well defined in the manual. One or more positive answers in a domain generate a “dysfunction score”. The maximum NOT-S score is twelve, one score for each domain. Typically developing children (> 5 years) have a mean score of < 2 (McAllister & Lundeborg, 2013). In the present study, the interview part was scored based on interviews with the parents of participants aged < 12 years and with participants > 12 supported by parents.

Bite force
Maximum voluntary bite force could be an indicator of the functional state of the masticatory system (Koc, Dogan & Bek, 2010). Measurements of bite force was performed with an

<table>
<thead>
<tr>
<th></th>
<th>All N=61</th>
<th>CAS N=25</th>
<th>SMD N=23</th>
<th>sCAS N=9</th>
<th>AI N=3</th>
<th>DD N=1</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nasal resonance</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>31 (51%)</td>
<td>14 (56%)</td>
<td>12 (52%)</td>
<td>3 (33%)</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Difficulties achieving initial articulatory configurations</td>
<td>31 (51%)</td>
<td>19 (76%)</td>
<td>4 (17%)</td>
<td>7 (78%)</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Numbers of features Mean ± (min-max)</td>
<td>4.9 ±2.0 (1-9)</td>
<td>6.8 ±1.2 (5-9)</td>
<td>3.5 ±0.9 (2-6)</td>
<td>4.4 ±1.0 (3-6)</td>
<td>1.7 ±1.1 (1-3)</td>
<td>6</td>
</tr>
<tr>
<td>Inconsistency</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>37 (61%)</td>
<td>25 (100%)</td>
<td>6 (26%)</td>
<td>7 (78%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Other</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Voice deviation (hoarsness, pitch, weak voice)</td>
<td>24 (39%)</td>
<td>9 (36%)</td>
<td>9 (39%)</td>
<td>4 (44%)</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>PCC mean %</td>
<td>66</td>
<td>52.9</td>
<td>78.3</td>
<td>66.9</td>
<td>88.7</td>
<td>57</td>
</tr>
</tbody>
</table>
occlusal force meter (Occlusal Force-Metre GM 10, Nagano Keiki Co., Japan). The biting element was placed on the first molar and the participant was instructed to bite as hard as possible. The measurement was repeated three times on each side. To calculate a mean value for each participant the highest value on each side was added up and then divided by two. In typically developing adults, normal maximum bite force is reported to be between 300-600 N (Antonarakis, Kjellberg, & Kiliaridis, 2013; Owais, Shaweesh, & Abu Alhaija, 2013; Sonnesen & Bakke, 2005). Owais and colleagues (2013) found a mean maximum bite force of 433 kN in typically developing ten-year-old children in the late mixed dentition stage.

**Jaw stability**

Jaw grading bite blocks (Rosenfeld-Johnson, 2005) were used for assessing jaw stability. Six bite blocks of different size were used. The assessment started with the smallest bite block (no. 2). Participants were asked to hold the bite block between the molar teeth during 15 seconds on each side. The examiner attached the bite block to a gauge meter and pulled the bite block gently, carefully monitoring the pull to be exactly 1kN on the gauge meter while measuring and counting the seconds out loud. The assessment was finished when the participant was unable to retain the bite block. If the participant managed to hold the bite block for 15 seconds, the examination was repeated in the same way but with the next size of the bite block. The minimum value on this task was size 2 and the maximum was 6.

**Chewing efficiency**

To assess chewing efficiency, a two-coloured chewing gum test developed by Schimmel et al. (Schimmel, Christou, Herrmann, & Muller, 2007) and described in Halazonetis et al. (Halazonetis, Schimmel, Antonarakis, & Christou, 2013) was used. By using a digital image processing program (Viewgum), the analysis quantifies how well the colours have been mixed, resulting in a measure of chewing efficiency. A two-coloured chewing gum specifically developed for this purpose was used (Firmenich, Switzerland). The participants were asked to chew 30 times on the chewing gum and then spit it out in a cup. The chewing strikes were counted by the examiner. Each side of the flattened gum was scanned in a flatbed scanner (resolution 600dpi) and analysed by the Viewgum software (ViewGum©software, dHAL Software, Greece, [www.dhal.com](http://www.dhal.com)). Chewing efficiency was measured in SD Hue (the standard deviation of the variance of Hue), which is a measure of how well the two colours in the chewing gum have been mixed. A low SD Hue value indicates greater masticatory performance. This method has never been used in a young population before. In healthy adults without malocclusion, a SD Hue mean value of 0.05 was found after chewing 30 times (Halazonetis et al., 2013). Another chewing gum was used in their study, which means that the values from Halazonetis et al. (2013) are not completely comparable with the values from this study.

**Oral stereognosis**

When testing intraoral stereognosis, figures in different shapes and sizes are used. However, there is no standardised procedure (Boliek et al., 2007; Park, 2017). Oral stereognosis tests the ability to recognise different shapes in the mouth. In this study, a set of four different shapes in two different size was used (star, triangle, circle, half-circle) (Byteme AB). The assessment was performed as described in the STockholm ORalMotoriska bedömningsprotokoll (STORM) (Henningsson, McAllister, Hartstein, & Raud Westberg, 2007). Participants were asked to open their mouth and close their eyes. The figure was
placed on the tongue by the examiner. The participant was then asked to close the mouth and “feel the shape of the figure”. After ten seconds, the figure was removed and the participant was asked to point to pictures of the figures to determine which picture matched the figure in the mouth. The maximum score was eight when all figures were correctly identified.

3D video recordings
In Study IV, the range of motion (ROM) in the lips and jaw was measured using the SmartEye® Pro – MME software (Mimic Muscle Evaluation) (SmartEye AB, Gothenburg). MME is an add-on that tracks lip and jaw movements, and it has been shown in earlier studies to be a reliable instrument (Antonarakis & Kiliaridis, 2019; Schimmel et al., 2010; Sjogreen & Kiliaridis, 2012; Sjogreen et al., 2011). The SmartEye® Pro system was originally a head and gaze tracking system able to measure head pose and gaze direction in full 3D. Infra-red (IR) diodes are used to illuminate the face and minimise the effects of varying light conditions (Sjögren et al., 2011).

Two calibrated video cameras (Sony XC-HR50) with IR lighting were placed on a fixed metal bar at a distance of 25 cm. The participant was seated approximately 80 cm in front of the cameras. A PC computer with SmartEye Pro 5.7 – MME software was used. The cameras shot 60 frames/second with a resolution of 640 x 480 pixels. The system compensates for head movements, provided that the face is captured by the cameras. Three repetitions of the syllables [mama], [mimi] and [momo] were recorded. One recording was also made of the face at rest for 30 seconds. After the assessment was finished, an individual profile was created where specific landmarks were plotted manually on ten snapshots extracted from the landmark profile recording of each child. The following poses were captured: head upright, head turned slightly to the left and to the right, open mouth smile and lip pucker. The landmarks were needed for the automatic tracking of the lips and jaw in 3D. The results of the tracking were transferred and saved in log files. The log files were exported to an MS Access application where extreme values (values due to momentary loss of tracking) were deleted and replaced with the closest accepted data. A visual review of the video recordings in tracking mode was made to ensure that the tracking procedure was performed correctly.

The mouth width value was calculated from values of the left mouth width and right mouth width. Lip movement asymmetry was calculated by subtracting the movement of the left mouth corner from the movement of the right mouth corner. ROM was expressed in mm and was defined as the maximum displacement of the mouth corners and the jaw (chin centre) during vowel production.
Figure 1. Different methods for assessing oral sensory-motor function and orofacial functions, including bite force, jaw stability, chewing efficiency, oral stereognosis, and lip and jaw movements in 3D.

4.3.4 Malocclusions and oral characteristics

Extraoral and intraoral examinations were performed by an orthodontist. Assessments of tonsils, tongue-tie and mentalis were performed in consensus agreement with the Speech-Language Pathologist (the author).

**IOTN Index**

The scores on the IOTN-DHC (Index of Orthodontic Treatment Need, Dental Health Component) Index (Brook & Shaw, 1989), malocclusion (yes/no), and type of malocclusion were assessed on intraoral and extraoral photographs by two orthodontists. The IOTN-DHC (hereinafter referred to as IOTN) was used to describe the orthodontic treatment need in the groups and to describe the severity of the malocclusions according to the index priority. The malocclusions were scored from 1-5. As no x-ray photographs were taken, hypodontia, hyperdontia and ectopic eruption or impacted teeth could not be examined or included in the assessment. Participants with IOTN grades 1 (almost perfection), 2 (minor irregularities) or 3d (moderate space anomalies) were not considered to have a malocclusion. The exception to this was 2c, which describes minor functional anterior or posterior crossbites and bilateral
crossbites without shift. Participants with IOTN grades 2c, 3abcef, 4 and 5 were considered to have a malocclusion. The reason for excluding moderate space anomalies was to minimise the number of false-positive malocclusions in young individuals, in whom crowding in the transition to permanent dentition is to some extent considered a normal finding in occlusal development.

4.3.5 Questionnaires

Background information
A questionnaire (MHC questionnaire + additional questions) was sent to the families prior to the first visit to collect information on medication, general disabilities (hearing, vision, epilepsy, presence of neurodevelopmental condition, general motor difficulties), a family history of speech and language disorders and delayed speech and language development, present and previous oral habits, and orthodontic treatment. The questionnaire used included other questions that were not used in the project. The NOT-S and ICS also includes questions about orofacial function and communication that were used instead.

4.4 RELIABILITY

4.4.1 Study I

The percentage of point-by-point comparison was used to assess interrater and intrarater agreement for PCC, PVC, nasality and NOT-S assessments. Twenty-three percent of the recordings from the speech assessments and 31% of the recordings of nasality and NOT-S assessments were randomly selected and reassessed. Inter- and intrarater agreement varied between good and excellent. Three SLPs were included in the interrater assessments, one for PCC and PVC, one for nasality and one for the NOT-S assessments. Their assessments were then compared with those of the main assessor.

4.4.2 Study II

All individual Smarteye profiles were constructed by the same examiner. Individual profiles from 19 (20%) randomly selected participants (nine from the TSD group and ten from the SSD group) were reconstructed to assess intraexaminer reliability. Based on the new profiles, new trackings were made and compared with the original results. A single measurement, absolute agreement, two-way mixed effects model was used for measurement of the Intraclass Correlation Coefficient (ICC). The overall ICC was 0.85 with a 95% confidence interval (CI) of 0.83 – 0.87. The ICC for the TSD group was 0.88 (CI = 0.85 – 0.91) and for the SSD group 0.82 (CI = 0.78 – 0.85). Based on the ICC results, it was concluded that the intraexaminer reliability for the study was good.

4.4.3 Study III

A set of intraoral and extraoral photographs was acquired during the clinical examination and used to assess the inter- and intrarater reliability of the dental assessment. The interrater agreement between two orthodontists was assessed for all participants. The intrarater agreement was calculated by re-assessing 20% randomly selected participants. The levels of interrater and intrarater agreement on the IOTN and the type of malocclusions were calculated using Cohen’s kappa coefficient and were estimated to be good (κ = 0.706)
(Altman, 1991). The interrater agreement about the type of malocclusion was slightly lower but still judged to be good, with 83% point-by-point agreement. The intrarater reliability of the IOTN Index was estimated to be very good ($\kappa = 0.901$). The intrarater agreement on the type of malocclusion was also very good with 95% point-by-point agreement.

4.4.4 Study IV

Interrater and intrarater reliability testing was performed for the NOT-S assessment (the same as in Study I) and malocclusions (the same as in Study III). For other assessments, reliability could not be estimated because they were performed in a clinical setting and dependent on live assessments.

4.5 STATISTICAL ANALYSIS

The data in Study I, II, III were analysed using the Statistical Package for the Social Sciences (SPSS statistics 22). The level of significance was set at $p < 0.05$ throughout.

In Study IV, all analyses were carried out in R (R Core Team, 2013) and no significance testing was used.

Descriptive statistics were used in all four studies for the background variables, age, and sex of the participants, and to characterise PCC, PVC, NOT-S, orofacial function measurements and dental characteristics.

4.5.1 Study I

Non-parametric tests were used for comparisons due to non-normal distributions, Spearman’s rho was used for correlation analysis (age, PCC, PVC, number of established consonants, ICS), and the Mann-Whitney U test was used for analyses of the significance of two independent samples. A linear regression analysis, using the enter option, was conducted to examine the predictive ability of the independent variables, age, and NOT-S, on the dependent variables, PCC and PVC, respectively.

4.5.2 Study II

Parametric tests were used for comparisons, since continuous data were used and based on relatively large participant groups. The independent samples t test was used to analyse differences in ROM of the lips and jaw between children with TSD and SSD in three different syllables, and Pearson’s $r$ to analyse correlations between age and ROM.

4.5.3 Study III

For nominal data, Pearson’s Chi-squared test was used for between-group comparisons, and when the data were ordinal, the Mann-Whitney $U$ test was used. The Pearson correlation coefficient (Pearson’s $r$) was used for the correlation analysis of age with IOTN.

4.5.4 Study IV

In this study we focused the inferential statistics on confidence intervals (CI) rather than statistical significance testing, in line with modern recommendations (Wasserstein et al., 2019). Where relevant, groups were compared using relative risk (RR), which describes how
much greater the risk of a particular outcome is in one group compared with the other. Separate logistic regression analyses were used to describe the relationship between orofacial function and malocclusion. This analysis focused only on the children with SSD as most children with TSD had typical orofacial function and no malocclusion.

4.6 ETHICAL CONSIDERATIONS

All four studies were approved by the regional ethical review board in Gothenburg (reg. no. 363-14). All the participants received both oral and written information about the study. The children received a simplified version of the information including pictorial support. All the children were involved in the discussion about their participation, but younger children were involved to a lesser degree than adolescents. The parents provided informed consent to their child’s participation before any assessments took place. All assessments that were included in the study, except for the 3D video analysis, were also a part of the clinical assessment that should have taken place even without participation in the study. All participants were consecutive patients referred for a consultation at the clinic and their decision to participate in the study or not did not influence the care they were given. The only difference between a regular visit to the clinic and an assessment in the study was that the visits were somewhat longer and somewhat more extensive. This was challenging for some children who had difficulties with concentration and attention. For this reason, the assessments had to be divided into two occasions in a few cases. The pictorial support that all children received before the visit was helpful to many children. We found that the children were well prepared for the assessment and curious about several of the assessment methods. One reason for not dividing the assessment into several occasions for all participants was that some families were travelling quite far to visit the clinic and we wanted to reduce long distance travel for the children.

A child should have the same right to cancel their participation in a research study as adults. It is important to respect the child’s will and needs, even when the purpose is data collection for a research project. We did not push children to continue the assessment if they showed that they did not want to perform a specific assessment or could not participate any longer in the session. Some missing data in the project are related to this.

Another ethical consideration was the fact that the extensive assessment could reveal difficulties that the parents were not fully aware of before. To communicate this in an understandable and pragmatic way was very important. In some cases, a result of the assessment was a referral to an ear, nose, and throat (ENT) specialist or a discussion about whether it would be beneficial to initiate a genetic investigation of the child. This information may trigger worries and questions in the families that had to be met in the clinical setting.

Some children experienced the assessments as very difficult, and it was important not to let the assessment and participation in the project become a negative experience for the child. The assessor was attentive to this and tried to include the child as an active participant as far as possible, explaining what and why we were assessing. As the participants were all school children it was important to make sure that they understood and were included in the conversation and the decisions.
The benefits of participating in the study were considered to outweigh the possible disadvantages. The extensive assessment was important for clinical decisions and further interventions. Many parents expressed that they received confirmation—sometimes for the first time—that validated their experience of their child’s struggle with eating, chewing and sensory issues, like toothbrushing, and that all the difficulties their child had were related and connected. The assessment offered an explanation model that was valued by many of the parents and older participants.
5 RESULTS

5.1 STUDY I

All participants had speech difficulties to a varying degree. Impaired consonant production assessed by PCC varied from 8 to 95 (median 71, mean 66 (SD 22.1)). Vowel production assessed by PVC varied from 55-100 (median 95, mean 91 (SD 10.1)). No single consonant was fully established in any participant. Thirty-one participants (51%) were found to have deviant nasality, according to a perceptual evaluation. According to the ICS questionnaire, filled out by the parents, intelligibility was affected in 90% (53/59) of the children. The mean ICS score was 3.72 (SD 0.60). The majority (87%) displayed difficulties with orofacial functions in more domains than expected for their age (total NOT-S score ≥ 2. The most affected domains apart from “Speech” were “Chewing and swallowing” (41%), “Masticatory muscles and jaw function” (38%), “Sensory function” (38%), and “Face at rest” (36%). There were significant correlations between orofacial function and consonant and vowel production. NOT-S examination scores and age together explained 25% of the variability in PCC and 21.8% of the variability in PVC, age alone 13.7% for PCC and 6.7% for PVC. In all, the parents of 39 (64%) participants reported one or more co-existing symptoms. A total of 34 (56%) participants reported motor difficulties and/ or confirmed or suspected hypermobility in joints.

Figure 2. Distribution of NOT-S scores in different domains in children with SSD (n = 61) and children with TSD (n = 44). The results are also compared with 116 TD children, aged six to eight years, from McAllister & Lundeborg (2013).
5.2 STUDY II

Most participants in the TSD group (80-86%) had an asymmetry < 2 mm. Children with SSD had a larger lateral ROM and more asymmetrical movements in both the lips and jaw than children with TSD. The ROM in the lips and jaw was generally small in all dimensions for children with TSD and the variation between individuals was also small. Lateral lip movements were ≤ 3 mm in all but one of the TSD participants. There was a significant difference between the groups on several of the studied variables and the individual variation was larger. Differences were especially pronounced in the lateral direction of both the lips and the jaw. No differences were found in the resting position. Younger children had somewhat larger moments both in the TSD and the SSD group.

![Figure 3. Example of lateral jaw movement during the syllable [mim] in one participant with typical speech development (left) and one with speech sound disorder (right). The figure shows that the movement in the jaw is larger in the child with SSD and of longer duration.](image)

5.3 STUDY III

The children with SSD were more likely to have more and more severe malocclusions than children with TSD. There were differences between the SSD and TSD groups with regard to the prevalence, type, and severity of malocclusions; 61% of the children in the SSD group had a malocclusion, as compared to 29% in the TSD group. In addition, the malocclusions in the SSD group were rated as more severe. Functional posterior crossbite and habitual lateral and/or anterior shift were more frequent in the SSD group. Class III malocclusion, anterior open bite and scissors bite were only found in the SSD group. None of the participants in either the SSD group or the TSD group used a pacifier or engaged in thumb sucking at the time of the assessment, although 19 participants (31%) in the SSD group reported a current oral habit, such as nail biting, biting the lips or teeth grinding during daytime. In the TSD group, nail biting was the only reported habit for eight participants (18%). There were no differences in earlier non-nutritive sucking habits (NNS) between the children in the SSD group and the children in the TSD group. Hyperactivity of the mentalis was significantly more common in the children with SSD and more children in the SSD group had a tongue-tie than in the TSD group. There was no significant difference between the groups regarding...
enlarged tonsils. Enlarged tonsils were somewhat more common in the children with SSD and TSD with posterior crossbite (both skeletal and functional). More children with anterior open bite had enlarged tonsils and hyperactivity of the mentalis.

<table>
<thead>
<tr>
<th>Angle Class I</th>
<th>Angle Class II (postnormal)</th>
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<tbody>
<tr>
<td>SSD: 69% TSD: 80%</td>
<td>SSD: 25% TSD: 18%</td>
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<tr>
<th>Angle Class III (prenormal)</th>
<th>Deep bite</th>
</tr>
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<tbody>
<tr>
<td>SSD: 15% TSD: 0</td>
<td>SSD: 23% TSD: 4%</td>
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<tr>
<th>Anterior open bite</th>
<th>Scissors’ bite</th>
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<tr>
<td>SSD: 12% TSD: 0</td>
<td>SSD: 3% TSD: 0</td>
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<tr>
<th>Skeletal posterior crossbite</th>
<th>Functional posterior crossbite</th>
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<tr>
<td>SSD: 0 TSD: 4%</td>
<td>SSD: 21% TSD: 0</td>
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**Figure 4.** Different types of malocclusions and prevalence of the different types in the SSD group (N = 61) and the TSD group (N = 44).
5.4 STUDY IV

Children with SSD had worse results on all measurements of orofacial function and had a greater risk of malocclusion than children with TSD. The differences between the SSD and the TSD group regarding bite force, jaw stability and intraoral sensory function were greater than the difference in chewing efficiency. Some children with SSD had orofacial functions at similar levels as their peers with TSD but most children with SSD had poorer scores on all assessments of orofacial function than any of the children with TSD. Children with SSD and orofacial dysfunction also had a greater risk of having a malocclusion. The strongest relationship between orofacial function and malocclusion was for the NOT-S total score. There was an increased risk of having an open bite for individuals with a positive response on “Face at rest”. Other noteworthy relationships were between “Nose breathing” and Class III, and between “Breathing”, “Chewing and swallowing”, and “Masticatory and jaw function” and having a Class II malocclusion.

Figure 5. Example of chewing efficiency measured in SDHue with a two-colour chewing gum in one participant with SSD with low chewing efficiency (left) and one participant with TSD and high chewing efficiency (right).
6 DISCUSSION

In this thesis, the aim was to conduct a comprehensive investigation of orofacial function, speech characteristics, occlusion and co-existing symptoms in children with SSD of unknown origin persisting after the age of six years. Different methods were used to assess orofacial function and the results were related to both speech characteristics and anatomical characteristics. The results showed that most of the included children with SSD had poor performance on the included tests of orofacial function. Orofacial dysfunction had a relationship both with speech impairment and malocclusion, which confirms the importance of assessing orofacial function in children with SSD.

6.1 SPEECH CHARACTERISTICS, INTELLIGIBILITY AND DIFFERENTIAL DIAGNOSTICS

Speech production and intelligibility were affected in all participants in the study group but to varying degrees, according to PCC, PVC, consonant inventory, and ICS results (Study I). All participants had impaired consonant production and vowel production was affected in most of the participants. Many participants also exhibited deviant nasality. All those speech difficulties resulted in impaired intelligibility according to the ICS results. Participants with orofacial dysfunction (NOT-S ≥ 2) had lower PVC. Participants with reported motor difficulties displayed deviant nasality to a higher degree. The high percentage of participants with vowel distortions, difficulties with motoric challenging speech sounds, and deviations in nasality confirms the presence of underlying motor speech disorders (Shriberg, Campbell, et al., 2019; Shriberg, Strand, Jakielski, & Mabie, 2019).

It is noteworthy that all participants were assessed as having motor speech involvement while most of them were presented with a phonology impairment diagnosis or DLD on referral. Even the child with an obvious DD came with a diagnosis of phonological impairment. All participants were children who had not improved as expected, despite receiving traditional speech language therapy, and had persistent SSD, in many cases severe.

Differential diagnostics in SSD is regarded as difficult (Waring & Knight, 2013) and this is also confirmed in the present study as some children did not fulfill the proposed criteria for CAS (five features from the operational list + inconsistency), but still had a motor speech disorder that included several CAS features. They were divided into the subgroups SMD/sCAS.

The discrepancy noted between the speech diagnosis in the referral and the diagnosis given after the extensive assessment and speech analysis shows a need for more knowledge of CAS and motor speech disorders among Swedish SLPs. This is in accordance with results from a survey study by Malmenholt and colleagues (2017), where Swedish SLPs reported a need for further education about CAS. Until recently, there has not been a specific test for CAS in Swedish but since 2016, there is a Swedish version of the DEMS that can be used in the clinic as a supplement to phonology, language and oral motor examinations (Rex, McAllister & Hansson, 2016; Rex, Hansson, Strand, & McAllister, 2021; Rex, Sand, Strand, Hansson, & McAllister, 2021). The speech feature of increased difficulties with multisyllabic words was unfortunately difficult to assess based on the speech assessment used in the present study, as
it did not contain enough multisyllabic words. Ideally, more challenging speech material
could have been added. It is not possible to rule out that if other speech material had been
used, with more focus on repetition of more complex words and sentences, a more specific
description of the inconsistency feature would have been gained, which would have made the
determination of a diagnosis in the sCAS group easier. CAS features can also change with
age and maturation (Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004). The children in the
CAS group had the lowest mean age. The children with sCAS and AI were older, even if
there were younger children in the sCAS group as well.

The differences between the diagnostic groups were naturally most distinctive regarding
inconsistency as this feature is mandatory, together with having five other features from the
CAS checklist for a CAS diagnosis (Iuzzini-Seigel, Murray, 2017). But it is also clear that the
CAS group presents with stress errors to a higher degree, which strengthens the diagnosis of
CAS, as this is one of the core criteria. Groping and voicing errors were also far more
common in the CAS group. Difficulties achieving initial articulatory configurations were
present both in the CAS group and in the sCAS group. It is possible that participants in the
sCAS group were children with milder CAS as they also had a higher PCC value. The SMD
group had nasality and voice deviations as two of the most common features.

6.2 SSD AND OROFACIAL FUNCTIONS

Orofacial function was affected in several domains in this group of children with persistent
SSD, compared with a control group with children with TSD (Study I, II, IV), especially
functions related to jaw stability and intraoral sensory function. “Chewing and swallowing”,
“Masticatory muscles and jaw function”, “Sensory function” and “Face at rest” were the most
affected domains, according to a screening test of orofacial function (NOT-S) (Study I).
Chewing efficiency, jaw stability, bite force, and intraoral sensory function were also
decreased compared with a control group of children with TSD (Study IV).

The literature is quite consistent; it is common in children with both SSD and DLD to have
co-occurring motor issues. However, this is not completely implemented in the clinical work
or the research into this population. Few of the children participating in this thesis had
undertaken an oral motor assessment prior to being included in the study or met a
physiotherapist for general motor difficulties. In a recent survey study of Swedish speech-
language pathologists’ practice regarding assessments of SSD by Wikse Barrow and
colleagues (2021) this is confirmed. Oral-motor function was the least frequent assessed
function by Swedish SLPs working with children with SSD. In a review on the literature on
speech and language in children with BPP, the authors mention a lack of results based on
formal or structured assessments of oral motor function even though oral motor deficits are
core symptoms in the diagnosis (Braden et al., 2019).

One of the aims of this project was to use reliable tests and objective methods to assess
orofacial function, as far as possible. Some of the methods lack normative values for children
and with some of the methods it was difficult to assess reliability. However, several of the
results were surprising. Children with SSD were expected to have poorer performance on
tests of orofacial function than TD children, but not as many as was the case. Nor were children with SSD expected to perform so poorly on bite force and chewing efficiency (Study IV). Thus, the results from this thesis reveal that oral motor difficulties and the influence on orofacial function may be much more severe than expected based on other methods and assessments reported in earlier studies.

Several studies confirm the need for oral motor assessments in the clinical work and suggest that this should be carried out as a part of assessments of children with SSD. However, it is relatively rarely motivated why this should be done and what further assessments should be used. Kent (2015) argues that coexisting oral motor difficulties are common in children with speech and language disorders and that an oral motor (orofacial) assessment could provide insight into other functional impairments and add comprehensive information on other affected functions.

Using non-speech oral motor exercises (NSOMEs) as an intervention for children with SSD has been questioned (Lof, 2008, Forrest & Iuzzini-Seigle, 2008, Lass & Pannbacker, 2008). Consequently, this may have affected the use of oral motor and sensory assessments in clinical work and in research. This should be evaluated against reports from SLPs finding NSOMEs useful for certain clinical populations and therapy targets (e.g., for phonetic placement) (Lof, 2008; Rumbach, Rose, & Cheah, 2019). However, Kent (2015) and McCauley & Strand (McCauley, Strand, Lof, Schooling, & Frymark, 2009) state that there is no scientific evidence to support or refute interventions including NSOMEs. In a review of orofacial myofunctional therapy and myofunctional devices, Shortland and colleagues (2008) state that there is limited evidence for those interventions, but that the strongest evidence is in the area of swallowing and mastication. They also emphasise that the use of orofacial myofunctional therapy in the management of oral hygiene is a field that may be worth exploring further. Kent (2015) also states that there may be a closer relationship between the use of NSOMEs and eating difficulties (swallowing and chewing) and it is therefore important to assess orofacial functions to get information on whether other functional impairments are present to recommend the proper interventions.

The results from this thesis indicate that SSD, orofacial dysfunction and motor difficulties often co-occur (study I). There were also several participants (15%) in the SSD group with a confirmed NDD, but none in the TSD group. Several parents and referrers reported that there were more children who were waiting for a neuropsychiatric assessment and that the parents and/or teachers thought that the child had ADHD and/or ASD (study I). This means the number of participants with NDDs is probably higher than presented in this thesis. However, the children with a confirmed neurodevelopmental disorder did not differ from those without regarding speech impairment.

It is problematic that it is included in the definition of SSD in diagnostic manuals, such as the DSM 5, that there should not be any other difficulties present to fulfil the criteria for the diagnosis. The definition of SSD in the DSM 5 says: “persistent difficulty with speech sound production that interferes with speech intelligibility or prevents verbal communication that
cannot be explained in terms of sensory problems, motor difficulties or other physical conditions” (American Psychiatric Association, 2013). This implies that coexisting difficulties are not expected, despite the growing amount of evidence that shows the opposite. The previously used term SLI has been widely criticised for using the word “specific”, which implies that children should only have difficulties with language. As this is rarely the case, this definition does not reflect the clinical reality and there is a risk that children are excluded from service and from research (Ebbels, 2014).

Hypermobility of joints was also surprisingly common in the participants in this thesis (Study I), as well as some rare malocclusions for this age group, such as scissors bite and Class III malocclusions (Study III). Those symptoms might be associated with an underlying genetic condition. Most of the participants had a known heredity for speech and language difficulties and three pairs of siblings participated. This is in agreement with the theories of a genetic cause behind SSD (Eising et al., 2018). For several of the participants with SSD in this thesis, a further investigation of genetic conditions and neuropsychiatric and/or neurological assessment was initiated.

6.2.1 Movement patterns of the lips and jaw during vowel production

Differences were seen in both lip and jaw movements in the children with SSD compared to the children with TSD in the three very simple syllable tasks used (Study II). With the observed difficulties during a simple syllable task, it is likely that difficulties are frequent in more demanding speech movements. The larger lateral jaw movements indicate jaw instability and could also result in compensatory fixing patterns such as retracted lips or clenched articulation (fixed jaw), and may also lead to functional malocclusions, such as functional crossbite. A fixing pattern may evolve as a compensatory strategy to stabilise the jaw and allow the lips and tongue to move more freely (Ward et al., 2013).

The lip movement asymmetry found in this study is also of relevance, both for coarticulation (Grigos, Moss, & Lu, 2015; Moss & Grigos, 2012) and for occlusal development. Asymmetry of the orofacial muscles can lead to dental malocclusion (Ovsenik, 2007). This is a reason why it is important to consider jaw instability and orofacial muscle movements in speech motor interventions.

6.3 THE OCCURRENCE, TYPE AND SEVERITY OF MALOCCLUSIONS

Children with SSD in this thesis exhibited differences concerning occurrence, as well as the types and severity of malocclusion (Study III). There was a more than twofold risk of malocclusion among the children with SSD than the children with TSD (Study IV). It is not possible to identify the cause of those differences from the results of those studies, but a shared genetic background for SSD/NDD and for the morphological traits is a possible explanation. There are other factors that are known to influence occlusal development, such as oral habits and orofacial dysfunctions (Grabowski, 2007; Ovsenik, 2007). The parents of the children with SSD reported somewhat more ongoing oral habits. However, this difference
is not likely to be the only explanation for the difference between the groups. Children with a functional posterior crossbite; that is, a malocclusion related to oral habits in the literature (Ovsenik, 2007), did not have a higher occurrence of oral habits than children with other malocclusions, nor had they used a pacifier for a longer time than other children. However, there was a relationship between Class II malocclusion and oral habits. Participants with a Class II malocclusion were more prone to have an ongoing oral habit, which is in line with previous studies (Baeshen, 2021; Grippaudo et al., 2016).

One explanation for the differences between the groups regarding malocclusion could be related to oral motor difficulties, and especially the observed jaw instability in children with SSD. The jaw instability related to malocclusion was described as a pattern with habitual lateral and/or anterior shift of the mandible during speech and rest.

Other oral characteristics that could influence occlusal development, such as tongue tie, hyperactivity in the mentalis and enlarged tonsils were also assessed (Study III). Hyperactivity in the mentalis was much more common in children with SSD and was related to an anterior open bite, as well as to enlarged tonsils, which is in line with the earlier described relationship between open bite and open mouth posture (Grabowski et al., 2007), and between activation of the mentalis in anterior open bite (Pichaya Pintavirooj, 2014). Tongue tie was somewhat more common in children with SSD but was not related to any of the malocclusions in this study.

6.4 RELATIONSHIP BETWEEN SSD, OROFACIAL FUNCTIONS AND MALOCCLUSIONS

In this thesis, a relationship was identified both between speech production and orofacial function and between malocclusion and orofacial function (Study I and Study IV). There was a relationship between the results on the orofacial function screening test (NOT-S) and consonant and vowel production. This is contrary to the results in a study by Torres et al. (2020) that aimed to determine which factors best explain the severity of speech impairment in a group of pre-school children with DLD and SSD. Torres and colleagues (2020) found no relationship between oral motor function and phonological process errors. They use a non-standardised test to assess oral motor function without normative values and without interrater and intrarater reliability. The differences in results can depend on the participants’ characteristics both regarding speech production and other background data. Even if speech symptoms may sound the same, they do not necessarily have the same origin. It is like coughing that could either depend on a cold or on lung cancer. The contradictory results may also depend on the different tests used for the purpose and emphasise the need to use reliable and valid tests.

The high incidence of deviant nasality among the participants in the SSD group could also be related to orofacial dysfunction (Study I). Low orofacial muscle tone may result in hypernasality, and abnormal resonance has been described as a motor-based articulatory deficit (Lewis et al., 2015). It is also noteworthy that all children in this study were assessed
as having a motor speech disorder, even if three of the children only had AI. It is likely that children with motor speech disorders have more oral motor difficulties than children without motor speech issues (Potter et al., 2019). On the other hand, there is a growing body of evidence that motor speech issues may be more common than suggested, especially in children with persistent SSD (Cleland & Scobbie, 2021; Flipsen, 2015; Gibbon, 1999; Lewis et al., 2015; Namasivayam et al., 2019). Consequently, it is of importance that assessments of children with persistent SSD include both assessments of motor speech symptoms and orofacial function. There is strong overlap and interaction between difficulties with phonology, language and motor speech planning and execution, and children with persistent SSD rarely exhibit difficulties in only one domain (Wren et al., 2016). Based on the results in the present thesis it could be beneficial for children with SSD if assessments of motor speech and orofacial function were implemented in the clinical SLP setting.

Orofacial dysfunction was predictive of malocclusion in children with SSD (Study IV). The total score on NOT-S was the strongest predictor of having a malocclusion but reduced bite force and jaw stability also strongly predicted the risk of malocclusion. Bite force and jaw stability could both be indicators of reduced oral muscular strength and hypotonic musculature. These results are in line with earlier studies by Kiliaridis et al. (1995;1998) where they showed that reduced oral muscular strength can influence facial growth. Anterior open bite was one of the malocclusions with the strongest relationship with orofacial dysfunctions, such as open mouth at rest and reduced strength and stability in the jaw musculature, and with hyperactivity in the mentalis. The hyperactivity in the mentalis could be related to jaw instability and oral motor difficulties. It could be a sign of a compensatory fixing pattern used to stabilise the jaw, recruiting an additional muscle unit to keep the jaw in position and to achieve lip closure.

A third of the children with SSD had difficulties with oral stereognosis compared with the control group and even more participants in the SSD group had a dysfunction score on NOT-S in the “sensory” domain (Study I and Study IV). Intraoral sensory function is considered important for both speech sound development (Crary et al., 1981) and chewing (Peyron et al., 2002). The results from Study I and IV support those earlier findings.

Altogether, orofacial function seems to influence both speech development and occlusal development and those relationships should be further studied to fully understand the interaction.

6.5 METHODOLOGICAL CONSIDERATIONS

A strength of this thesis is that several different and specific methods were used to assess orofacial function and that reliable, objective and quantitative methods were used as far as possible. Comparisons with the control group of children with TSD also strengthen the results. The participants with SSD are a clinically representative group and the multiprofessional approach used offers a more holistic view of the participants’ function. There are, however, some methodological considerations in the studies.
The high incidence of motor speech disorders in this sample is not equivalent to the numbers reported in other studies (Shriberg, 2019). This probably indicates a bias towards children with more severe SSD and a selection prior to our assessment. The children were all referred for a speech motor and oral motor assessment, which means that someone suspected a speech motor disorder and possibly also oral motor difficulties. However, this thesis did not aim to study or establish the prevalence of speech motor disorders, nor is it a thesis on differential diagnostics of SSD. The results from this thesis are a description of orofacial dysfunction and how it may manifest itself. On the other hand, the prevalence of DD was lower than in the Shriberg study (2019). Those differences in prevalence numbers could be related to differences in health care systems and the recruitment of participants as consecutive patients. The children in Shriberg (2019) were also younger than the participants in this study but that does not explain the differences.

If comparisons should have been done between different subgroups, an equal number of participants in each group would have been preferable. Most participants belonged either to the CAS or the SMD group and only three had AI and one had DD. To explore how orofacial dysfunction differs between different subgroups, another recruitment procedure would have been used.

The differential diagnostic procedure provided some problematic issues. As earlier described, there is no consensus in the literature on how to differentiate the different SSDs from each other. In this thesis, the checklist proposed by Iuzzini-Seigle & Murray (2017) was used for the diagnosis of CAS. The checklist states that a feature must only be present on one occasion, which could result in some false positive results. Inconsistency is given special importance in this checklist since it is the only mandatory speech feature for CAS. A simple speech material is used in this thesis. It could not be ruled out that more participants could have exhibited inconsistency if more complex words, multi-syllable words and sentences had been used. Perhaps the participants with SMD and sCAS should have been treated as one group but there are some distinct differences between the groups that justifies keeping the groups separate. The participants in the SMD group had a higher PCC mean and were less inconsistent than the sCAS group. The SMD group also had nasality deviations and voice deviations as a more specific feature of the group characteristics. The assessment of the differential speech disorder diagnosis is not perfect but perhaps “as good as it gets”, based on the included speech assessments. If the aim of this thesis had been differential diagnostics of SSD, other tests should have been added and reliability testing should have been performed.

The methods for assessing orofacial function had some limitations. The kinematic recording system in Study II, Smarteye MME, lack audio recording possibility in the program, which limited the analysis of vowel production. If audio recordings had been synchronised with the program, an acoustic analysis could have complemented the movement analysis, but this was technically difficult to solve within the frame of this thesis project. Even if Smarteye MME does not require attached markers or that participants are completely still, it still requires more attention and ability to follow instructions than other assessments in this thesis. This was also the assessment method with the greatest data loss and it was participants with the
most severe difficulties that were excluded, due to insufficient video recordings. This reflects an obvious limitation of the method, as participants with the most severe disorders are also of the greatest interest and important to study. The system could only capture lip and jaw movements but information on tongue movement would add valuable information on speech motor performance. This would require other assessment methods like ultrasound or electropalatography.

For the assessments of chewing efficiency, bite force, jaw stability and intraoral sensory function no normative values exist for the studied age groups. Reliability could not be estimated for any of those assessments, as they were performed in a clinical setting and dependent on live assessment. This reflects a methodological limitation but, on the other hand, the control group provides values for TSD children and can be used as comparisons and the methods supplies quantified values. For all assessments that lack reliable normative values the results in the control group are used for comparison.
CONCLUSIONS AND CLINICAL IMPLICATIONS

Several potentially important findings have evolved from this project on orofacial functions, speech characteristics, occlusion and other co-existing symptoms, in a group of children with SSD persisting after the age of six years. The results add knowledge to the existing literature on orofacial function in children with SSD. This is in line with the aim and rationale for this thesis. The main conclusions are that children with SSD persisting after the age of six years:

- often have motor speech involvement affecting consonant and vowel production as well as nasality;
- are at risk of orofacial dysfunction;
- may have jaw instability, low chewing efficiency, and intraoral sensory deficits;
- may have general motor difficulties (including hypermobility of joints) and other neurodevelopmental disorders;
- have more and more severe malocclusions than children with TSD and children with a combination of SSD and poor orofacial function are at greater risk of malocclusion.

Orofacial functions in children with SSD are rarely assessed and described in scientific research. The results from this thesis add to the growing body of evidence that children with SSD often have coexisting oral motor difficulties. Speech disorders are one symptom among other symptoms that likely arise from the same (often unknown) biological cause. It is not a causal but a coexistent relationship and the symptoms can most likely interact and influence each other. Difficulties with jaw stability and movement, deviations in sensory function and open mouth posture were some of the most common oral motor difficulties.

The jaw instability found in this thesis may represent a commonly overlooked difficulty in children with motor speech disorders that could influence speech movements, as limited jaw grading control could affect several speech sounds. In some intervention studies, increased jaw stability has been reported to lead to improved consonant production (Grigos & Kolenda, 2010; Namasivayam et al., 2013; Terband, 2013). The results from this thesis strengthen the need for intervention that aims to improve jaw stability. Identifying jaw instability and addressing this in intervention may be important for children with SSD. There are several therapeutic concepts, such as PROMPT (Dale & Hayden, 2013), developed to improve accuracy and stability of speech production using tactile-kinaesthetic proprioceptive input (Namasivayam et al., 2020). Namasivym et al. (2020) found significant improvement of speech motor skills in children with SMD after a ten-week PROMT intervention. The PROMPT focuses on integration of jaw, lip, and tongue movements and improvements in timing and co-ordination of movements (Ward et al., 2013). In the DTTC treatment method developed by Strand (2019), one of the core elements is the attention to proprioception. Children with CAS may have decreased intraoral proprioception and the results from this thesis confirm the need to assess sensory function and address this in the treatment of affected children. The participants with SSD in this thesis had poor performance on intraoral stereognosis and the parents also reported a high proportion of sensory-related issues (NOTS).
Several studies have shown that there is a difference in tongue movements measured with ultrasound and EPG in children with SSD compared with TSD children (Cleland & Scobbie, 2021; Gibbon, 1999; Kabakoff et al., 2021). If we assume that jaw stability and control is a prerequisite for tongue control (Kent, 1999), it is a somewhat expected finding that the children with SSD show limited jaw stability and control. As well as assessing tongue mobility and control, jaw stability and control should also be a part of the SLP assessment. Difficulties with jaw stability and control could probably also lead to malocclusions and chewing difficulties, as the results from this study indicate. To conclude, when orofacial functions are affected in children with SSD, a sensory-motor perspective should be included in the intervention strategies. There is a growing body of evidence for interventions based on principles of motor learning for children with motor speech disorders (Maas, Gildersleeve-Neumann, Jakielski, & Stoeckel, 2014) (Maas, Gildersleeve-Neumann, Jakielski, & Stoeckel, 2014, McAllister et al., 2018). These principles may at least, in part, explain the lack of progress following mainly language-based interventions experienced by many of the participants in the present studies.

Speech disorders rarely exist in isolation. The results from this thesis are in line with the ESSENCE concept (Gillberg, 2010). All professionals working with children with speech and language disorders need to be aware of the increased risk of having multiple difficulties. As differential diagnosis between different SSDs is difficult to perform, the most important distinction to make is the one between a motor speech disorder and a more language-based disorder as this will influence the choice of intervention. It is important that SLPs have sufficient knowledge to assess and diagnose children with SSD and are aware that SSDs often co-occur with other difficulties. It is often necessary to offer a multiprofessional approach when working with children with SSD and orofacial dysfunction to ensure that appropriate interventions are provided.

The results from this thesis also indicate that children with impaired orofacial functions have a higher risk of developing malocclusion. A causal relationship cannot be confirmed from the results in this thesis but the children with more severely impaired orofacial functions were also more prone to have a malocclusion. The difficulties with oral motor function in children with persistent SDD could also result in impaired ability for oral self-clearance, which implies a need for more frequent dental care, to monitor both occlusal development and caries prophylaxis.

The results show that clinicians need to pay attention to children with coexisting oral motor difficulties, which may have a negative impact on language development (Adams, 2016). Coexisting oral motor difficulties may also influence occlusal development and chewing efficiency negatively.
8 FUTURE PERSPECTIVE

The assessment methods in this thesis should be used on a wider and less biased group of children with SSD to confirm or refute the strength of the results. There is a need for population-based studies of orofacial function in children with SSD to gain information on the prevalence of orofacial dysfunction in children with SSD. It would also be beneficial to have more evenly distributed groups of different SSDs to compare symptoms between different types of speech disorders. In this thesis, children with AI performed better on several assessments, but since there were only three AI children, no conclusion can be drawn on such a small sample. A more in-depth analysis of the relationship between speech production and performance on different oral and sensory motor and orofacial function assessments would also be beneficial.

Developmental trajectories in a longitudinal study on orofacial function and malocclusion in children with SSD would also be most enlightening. It would be beneficial to follow how speech characteristics, orofacial functions and malocclusions interact and develop over time.

A study on quality of life in this group of children could offer knowledge on how orofacial dysfunctions influence quality of life, depending on age and the severity of the disorder. This would provide an opportunity to gain insight into the participants’ own perspective on their difficulties and what they think is the most important regarding interventions. Such studies could be made using different interview methods.

The methods used in this thesis could be used to evaluate results from interventions, both regarding motor speech and oral motor function. Oral motor interventions often have methodological difficulties due to heterogenic groups but also related to proper assessments. Some of the methods used in this thesis would be suitable for this purpose, such as Smarteye MME and the two-coloured chewing gum test, which provide objective quantitative data of a function. Bite force meter measurements also yield objective data, but muscle strength can increase without the function increasing (Sjogreen et al., 2010).
9 ACKNOWLEDGEMENTS

This work could not have been done without support and contribution from a lot of amazing people, both in my professional and personal life. I am deeply and sincerely grateful for this.

I really would like to thank all the participating children and their families. Without your patience with me and with the sometimes exhausting assessments this work could never have been done.

I have been blessed with the best combination of supervisors. We have had so many fruitful and interesting discussions over the years and also so much fun. You are the best of mentors both in science and life in general.

My main supervisor, Anita McAllister – you have been my supervisor and mentor since around 1997 when I started at the SLP program at KI. Life and science are never boring with you around. Your curiosity, knowledge and holistic view is a prerequisite for the work of this thesis. I’ve learned so much from you and most important of all, you are brilliant in keeping the fire alive.

My co-supervisor and former colleague Lotta Sjögreen - I don’t think it is too much to say that you made me to what I am in a professional way. You took me on board on a new professional journey 16 years ago, which still feels like a fantastic adventure. I am so thankful for the opportunity to have had you as a mentor over the years, always helpful, always inspiring, always full of knowledge. This thesis is created out of many of our discussions and experiences from your and our work at MHC.

My co-supervisor Monica Barr Agholme who has been a brave dentist among SLPs during this process. Your knowledge, engagement for patients, positivism and your caring personality have been of most importance. You have generously opened your home for “writing retreats” and you have patiently listened to us SLPs discussing speech disorders for hours.

I also want to express my gratitude to:

Ulf Lekholm, Marianne Bergius, Lotta Sundelin and Folk tandvården Västra Götalandsregionen who gave me the opportunity to start this PhD education. I could not have done this without your support.

My colleague and co-writer Christina Havner who helped me with the data collection and the writing of study III and IV but most of all been the best guide to the world of orthodontics. I’m looking forward to all our future adventures in life and in research.

Professor Anette Lohmander and colleagues at the Division of Speech Language Pathology at KI who has created a creative environment for PhD students in Speech Language Pathology.

My fellow PhD students Susanne Rex and Ann Malmenholt for support and friendship during this education. I really appreciate our discussions and exchange of frustration and joy over the
years. Susanne, we started this journey at the same time, and it has been so great to have your companion and support during publishing processes, courses and conferences. Also, a big thank you for your hospitality and generosity.

*All former and present PhD students* at division of speech language pathology for discussions at seminars and sharing of experiences. A special thanks to *Anna Persson* for answering all my questions regarding the application and preparations the last months.

*Natalie Davet* – aside from being one of my closest friends you have also been my PhD colleague but in a different subject. Our writing retreats and lunch breaks discussion have been so important and inspiring. Working side by side with you in deep concentration getting energy and inspiration by you is some of my best memories from those years. We have shared the ups and downs in the academic life and in life in general and I hope we will never stop doing that.

*AnnaCi Algers* – we started at the SLP program at KI in 1997 and you were a motivation to continue that education, I’m so thankful for that. Since then we have been walking side by side on the winding roads of speech and language and life. We will always wear the same socks.

*Anna Karin Larsson* - you have been the most supportive SLP and PhD-friend, I am so thankful for everything we have shared over the years, articles, conferences, lunches, and laughter. I’m also thankful for your help with re-assessments for reliability testing. You have been more important than you maybe know.

*Emilia Carlsson* - I so much appreciated supervising students with you with data from this project and I’m looking forward to completing the article we are working on.

*Anna Westerlund* for your contribution to study III and IV and your patience with the multivariate analysis that I hope we will have the possibility to use in future studies.

*Magnus Hakeberg* for friendly and fast statistical advices.

*Utbildningskliniken för barn at Odontologen* for helping with recruitment of the control group.

*Anders Sand* - you were a true hero when helping us with the statistics in study IV.

*Ann Nordberg* for your friendly support and for helping with re-assessments for reliability testing.

*Anna Petersson and Agneta Wittlock* for all help with administration.

Former SLP students *Josefine Andersson, Linn Hallgren, Kajsia Nilsson and Therese Nyström*, for helping with clinical assessment of the children with typical speech development
My friends in the IADH community that has offered a foundation for multidisciplinary work and research that was important especially when I started working in this field of “logodentistry”. A special thanks to Johanna Norderyd who is such an excellent role model regarding this.

Sara Rosenfeld Johnson who was the one person that first opened my eyes to the importance of jaw stability.

Ingrid Henriksson for your ears and support as my mentor.

Hannes Högberg for creating the illustration on the front page and for being the sweetest cousin to my children.

All my dear colleagues at Mun-H-Center. A special thanks to Inga Svensson for helping with the pictures and tables in the thesis and a very special thanks to my SLP colleagues Lisa Bengtsson, Helmine Brattfors, Agneta Rubensson and Torunn Liljegren for running the clinical work and everything else when I needed to finish this project. Also, thanks to Anna Ödman and Lena Romeling for help with administration and other important things. I am blessed with the best colleagues and I’m truly looking forward to seeing you all more in the future.

To travel between Gothenburg and Stockholm during those years was not always easy. Without the possibility to borrow a place to stay from my friends it would have been almost impossible. An extremely great thanks to Natalie and Håkan and to Julia and Peter for many nights of good sleep! Julia, we have been so close to each other for more than 35 years now, we have shared so much and I cannot think of who I would have been without you. I’m looking forward to sharing all the phases in life with you, from childhood and teenager to getting old together.

Work is important to me but without all my wonderful friends life would be meaningless, you offer love and compassion, important talks, reflections and laughter, exercise, parties, travels, music and art – yes, everything that makes life worth living. I would like to write an essay to you all but there is not enough space for that. Just promise me that we will continue!

A special thanks also to “Fredagsgruppen” and coach Ola for focusing on the physical health during this last intense period of this project. And Johanna, I’m so thankful for all the shared paces and shared thoughts over the years.

Hans Hansson and Annika Nordström close friends to my mother who were the first persons I knew that had a PhD, you’ve been important in many ways.

And at last, without my family I could not have done anything. Thanks to my father Ingemar and sister Tove for your support and for teaching me to argue and letting me practice over and over and to little Rut, who has brought so much sunshine into the world since she arrived in the middle of a pandemic and the end of this thesis project.
Words are simply not enough to express the love and gratitude I feel for my husband Martin and my children Sigrid and Gustaf. You have been so patient with me during those years. Sigrid and Gustaf – you were young children when I started this education and now you are almost grown up. I hope that being close to me in this process has brought you some perspective that you can have use for in the future and an urge for learning. Martin, the love of my life – the workload of household work that you have made those years is a love gift that I appreciate more than you can understand. You are the warmest and most supportive person anyone can imagine and being parents and creating home and garden together with you is the foundation for everything in my life.

I have dedicated this thesis to my grandmother Rut Eriksson, who was the most intelligent, brilliant, and humorous person I will ever know. Due to economic reasons and a different society she had to quit school after 6 years, but she never stopped reading. My mother Gunilla Eriksson was the first person in her family to study at the university. I still miss her every day. I wish that they both could have been able to follow my PhD education but their unconditional love and support for me still carries me through life.

This research was financially supported by Health and Medical Care Executive Board of the VästraGötaland Region Aina Börjessons foundation for Speech Language Pathology research, Jerringfonden, Majblommans riksförbund, Märtha and Gustaf Ågren’s Foundation, Petter Silfverskiölds minnesfond,
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