DEVELOPMENTAL COURSES OF CHILDHOOD DYSARTHRIA:

FUNCTION- AND COMMUNICATION-RELATED ANALYSES AGAINST THE BACKGROUND OF TYPICAL DEVELOPMENT

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Speech and language development in children are most complex processes. Children do not only have to learn thousands of words and their meanings, grammatical rules, and syntax, they also need to acquire the motor abilities to translate these units into speech. Yet, speech itself is a highly complex motor function as it requires the precise coordination of more than 50 pairs of muscles, the constant integration of sensory information, and relies on the support of linguistic and cognitive processing (Ziegler & Vogel, 2010). Like all voluntary motor skills, speech movements need to be acquired through constant practice. From the first cry upon birth to the full maturation of all movement patterns of speech, it is a prolonged process. In children with "typical" speech development (i.e., without impairments), the development and perfection of this coordinative high-level performance extends beyond the first decade of life (Kent & Vorperian, 1995).

Speech represents the most important and most natural means of communication used in the majority of everyday situations to interact with others. Impairments of the processes involved in speech, which may occur, for example, as a result of neurological diseases, can disturb the complex sensorimotor interaction and lead to characteristic disorders, such as dysarthria.

Traditionally, however, in the discipline of speech-language therapy, processes of the acquisition of speech and language, on the one hand, and neurologically induced disorders of speech motor skills, on the other, have been considered two independent research domains, that is, *speech-language development* and *neurophonetics*. Developmental language disorders in children and acquired neurogenic communication disorders in adults constitute two key elements of speech-language therapy. Despite theoretically and clinically relevant interfaces, insights of both fields have hardly found their way into the other discipline so far.

Within the field of speech-language development research, the development of specific speech motor skills has received little attention compared to other domains (e.g., phonology, vocabulary, grammar, or language comprehension). In German textbooks on

language development, phonetic-motor processes are not considered (Klann- Delius, 2016; Szagun, 2019) and only few empirical studies address the topic (e.g., Boliek et al., 2009). Consequently, the understanding of unimpaired speech motor development is still very limited. In neurophonetics, on the other hand, which is concerned with the neural basis of speech and the analysis of neurological speech disorders (e.g., dysarthrias), traditionally the major focus is on disorders acquired in adulthood. Thus, for the most part, children are currently not included in standard textbooks on dysarthria (Duffy, 2013; Ziegler & Vogel, 2010).

However, dysarthria can also occur early in life. *Childhood dysarthria* constitutes a speech motor problem that is caused by a congenital neurological disorder or a neurological condition occurring at an early age (i.e., before completion of speech motor development) (Murdoch 2011). The resulting communication disorders entail far-reaching consequences for affected children (e.g., Fauconnier et al., 2009). The abovementioned separation of speech-language development and neurophonetics, however, is reflected in the assessment and characterization of childhood dysarthria. In previous studies, concepts and methods established in the assessment of late acquired dysarthria were applied to children without question (e.g., Cahill et al., 2002). To answer central research issues of speech acquisition and to investigate the consequences of neurological disorders in childhood, however, concepts of speech development theory and neurophonetics need to be integrated. This dissertation is focused on childhood dysarthria and its assessment, employing neurophonetic approaches, but also considering developmental aspects.

In the following, basic information on childhood dysarthria will be presented, which are also highly relevant for clinical assessment.

1.1 Etiologies and prevalence

Dysarthria constitutes the most common neurologic communication disorder (Mei et al., 2014; Ziegler & Vogel, 2010). It may occur in the context of a variety of neurological conditions at any age (Morgan & Liégeois, 2010; Ziegler & Vogel, 2010). Some of the etiologies that are associated with a particularly high prevalence of dysarthria are characterized by an early onset. Accordingly, dysarthria often occurs in children. The most prominent example is cerebral palsy (CP), which is one of the most common

neurodevelopmental disorders of early childhood, with a prevalence of 2.4 per 1000 children (Cans, 2000; Odding et al., 2006).

Children affected by CP acquire brain damage before, during, or immediately after birth. Early brain damage represents an obligatory feature of the diagnosis of CP, although to date no upper age limit has been defined. CP is mainly characterized by gross motor dysfunction, which may include spastic paresis, hyperkinesia (especially athetosis), or ataxia (Rosenbaum et al., 2007). Associated impairments are most frequently communication disorders (i.e., dysarthria), but also cognitive impairments (which can range from mild learning difficulties to severe intellectual disabilities), sensory disorders (e.g., visual impairments), disturbances of perception and behavior, as well as epilepsy (e.g., Beckung & Hagberg, 2002; Novak et al., 2012; Rosenbaum et al., 2007). The prevalence for cerebral palsy is approximately 2.4 per 1,000 live births (Hirtz et al., 2007). Referring to the most recent population-based data, Mei et al. (2014) report that 90% of children with cerebral palsy present with symptoms of speech motor dysfunction. Thus, cerebral palsy represents the most common cause of childhood dysarthria (Lepage et al., 1998; Yorkston et al., 1999). Because of a remarkably high proportion of severe forms, it is also known that dysarthria in CP is of particular clinical relevance. Multiple studies have shown that the proportion of severe speech impairments in individuals with CP is considerably higher than, for example, in individuals who experience dysarthria due to Parkinson's disease or stroke (Bogousslavsky et al., 1988; Mei et al., 2014; Müller et al., 2001; Schölderle et al., 2016).

Congenital malformations of the brain or certain genetic syndromes (e.g., Down syndrome, Worster-Drought syndrome, Möbius syndrome) or neuromuscular disorders (e.g., Duchenne muscular dystrophy, spinal muscular atrophy) represent other causes of childhood dysarthria (Kooi-van Es et al., 2020; Murdoch, 2011; Yorkston et al., 1999). In addition, there are a variety of age-independent etiologies that may cause brain damage at an early age, such as traumatic brain injury, cerebrovascular events, or brain tumors (Cahill et al., 2003; Morgan & Liégeois, 2010; Murdoch, 2011; Ozimek et al., 2004; Yorkston et al., 1999). These are however less common.

Based on prevalence data regarding the underlying diseases as well as corresponding data on the frequency of dysarthria in these conditions, it is reasonable to assume that in Germany more than 50,000 children and adolescents have dysarthria (Schölderle et al., 2018). Yet, standard textbooks on dysarthria do not include childhood

dysarthria (Duffy, 2013; Ziegler & Vogel, 2010), and there are hardly any specific materials for the assessment of the disorder or its treatment (Ackermann et al., 2018; Morgan & Vogel, 2008; Pennington et al., 2016). Overall, both in research and clinical practice, childhood dysarthria has been significantly underrepresented compared to dysarthria acquired in adulthood. This is also reflected in the fact that studies conducted to describe the speech symptoms of childhood dysarthria so far are limited in number and scope.

1.2 Symptoms of childhood dysarthria

A review of the literature reveals that symptoms of childhood dysarthria have been described in all speech subsystems, i.e., respiration, voice, articulation and resonance, and prosody (e.g., Allison et al., 2017; Allison & Hustad, 2018a, 2018b; Blumberg, 1955; Cahill et al., 2002; Chen et al., 2012; Cornwell et al., 2003; Gibbon & Wood, 2003; Hong et al., 2011; Murdoch & Hudson-Tennent, 1994; Nip, 2013; Nordberg et al., 2014; Ozimek et al., 2004; Richter et al., 2005; van Mourik et al., 1998; Workinger & Kent, 1991). Most of the symptoms associated with dysarthria acquired in adulthood may also occur in children, though not necessarily in the same syndrome constellations (Schölderle et al., 2021). However, there are also certain features of adult dysarthria that have not yet been reported in children (e.g., pauses or iterations). These features can but need not necessarily be symptoms of childhood dysarthria, because they also occur in typically developing children of the same age.

It is important to point out that only a limited number of studies examined children's speech disorders across multiple speech subsystems (Allison & Hustad, 2018a, 2018b; Cahill et al., 2002; Cornwell et al., 2003; Nordberg et al., 2014; Workinger & Kent, 1991). In addition, only few studies have conducted auditory-perceptual analyses, though this approach represents the gold standard for the assessment of dysarthria (e.g., Cahill et al., 2002; Murdoch & Hudson-Tennent, 1994; Nordberg et al., 2014; Workinger & Kent, 1991). Furthermore, several studies exclusively employed acoustic methods or instrumental procedures (e.g., electropalatography) and thereby failed to examine whether the detected dysfunctions also manifest as audible symptoms (Chen et al., 2012; Gibbon & Wood, 2003; Hong et al., 2011; Nordberg et al., 2011). Such studies can only provide limited information about the clinical picture of childhood dysarthria.

There are a number of challenges to a systematic and comprehensive assessment of childhood dysarthria and many aspects have been neglected in research so far. These will be outlined in the following.

1.3 Challenges and unresolved issues in the description and assessment of childhood dysarthria

1.3.1 Assessment in the context of multiple disabilities

Childhood dysarthria hardly ever manifests as an isolated symptom, but usually merges into the picture of multiple disability. As mentioned before, several concomitant symptoms may occur, including sensory disturbances in form of visual or hearing impairment, motor impairment of the trunk and limbs, cognitive impairment, but also disturbances of perception and behavior (Beckung & Hagberg, 2002; Novak et al., 2012; Rosenbaum et al., 2007). In addition, children with dysarthria may also show impaired language development (e.g., regarding vocabulary or syntax; Pennington, 2008) or other types of speech disorders (e.g., phonological disorders). Thus, standardized assessment is not a trivial endeavor in children with dysarthria.

So far, no specific materials exist for the assessment of childhood dysarthria in Germany. But beyond that, also in the English-speaking countries there is a "lack of standard tools for eliciting connected speech" in children with dysarthria, as Morgan et al. already pointed out in 2014 (Morgan et al., 2014, p. 335). In the Anglo-American community, the *Park Play Scene* is the only available material specifically designed to elicit speech samples from children with dysarthria (Patel & Connaghan, 2014). Yet, the assessment based on this picture description task focuses on the transcription and analysis of single words and disregards a more comprehensive auditory-perceptual analysis of continuous speech. To the author´s knowledge, the Park Play Scene materials have not been used in published research so far.

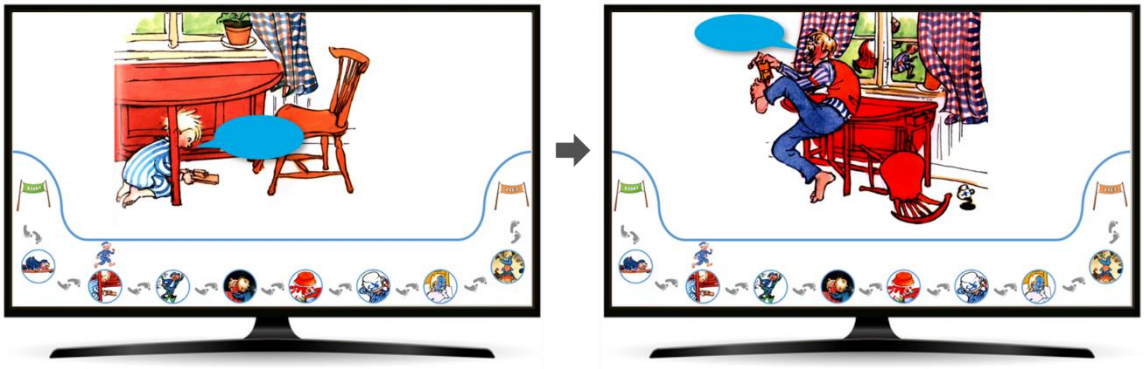
Hence, all studies conducted so far obtained speech samples from the children using materials that were not specifically designed for children with dysarthria. For example, some studies used the *Test of Children's Speech Plus* (TOCS+: Hodge & Daniels, 2007), which was originally developed to assess intelligibility in children, to collect standard speech samples for the assessment of auditory-perceptual characteristics related

to speech subsystems (e.g., Allison & Hustad, 2018a). But also concepts and methods were borrowed from assessments for dysarthria acquired in adulthood, such as the *Frenchay Dysarthria Assessment* (FDA: Enderby & Palmer, 2012; see Cahill et al., 2002, 2003, 2005; Cornwell et al., 2003). Yet, for example, the FDA protocol requires words and sentences to be read, or complex long sentences to be repeated without further support. These tasks require the individual to be able to read (i.e., to have certain cognitive abilities and intact vision) and to be able to memorize and repeat multi-word utterances (i.e., to have good working memory capacities). These abilities, of course, are limited in very young children or children with multiple disabilities. Therefore, as a prerequisite for the present dissertation, child-appropriate materials were developed, taking into account the specific requirements of the target group (BoDyS-KiD; Haas et al., 2015, 2020).

BoDyS-KiD materials: The assessment tool *Bogenhausen Dysarthria Scales - Childhood Dysarthria* (BoDyS-KiD; [German: *Bogenhausener Dysarthrieskalen – Kindliche Dysarthrien*]) was developed as part of the author´s master thesis (Haas, 2015) and is designed for the assessment of children from 3 to 9 years of age. The key principles of the *Bogenhausen Dysarthria Scales*, which had originally been designed for adults with dysarthria (BoDyS; Ziegler et al., 2018), were retained to the best extent possible. This means that, for instance, multi-word utterances should be collected to diagnose dysarthria in a comprehensive manner. Such speech samples are necessary to be able to make meaningful judgements of all relevant domains of continuous speech, especially fluency and prosodic modulation. Because different examination modalities demand different cognitive-linguistic as well as speech motor skills, the adult version involves the recording of speech samples in four modalities, namely spontaneous speech, sentence repetition, passage reading, and narration of a picture story. For example, an individual may produce longer phrases in a spontaneous speech context than in repeating sentences, which places greater demands on speech breathing. To obtain an overall picture of dysarthria which is as comprehensive and valid as possible, speech samples should therefore be collected in multiple modalities. Following this model, the BoDyS-KiD materials aim to collect repeated sentences and spontaneous speech samples, as these modalities can be adapted specifically for children with multiple disabilities of the respective age (Haas et al., 2020).

To visually support the elicitation of the speech samples, child-appropriate images from the illustrated book of "Michel from Lönneberga" by Astrid Lindgren (Lindgren et al., 2005; © Illustration: The Astrid Lindgren Company/Bildmakarna Berg/Björn Berg)

were employed. This means that various funny events that Michel experiences have been put together to form a coherent frame story. The original drawings were arranged in a way that two illustrations depict one funny event – one illustration displays one of Michel's actions, and the following illustration reveals the consequences of this action (see Figure 1). In its sequence of illustrations, the frame story was implemented as a computer game by means of a PowerPoint presentation and intended to engage the children in speaking within a setting similar to situations in which children are read to. A "narrator" first describes a short sequence of events for each picture. Following this, the child is asked to repeat a sentence that is essential to the story. To draw the child's attention to the stimulus sentence in question, a speech bubble is displayed as a trigger. The frame story as well as the repetition sentences were recorded by a professional speaker and embedded in the presentation ensuring a standardized presentation. After repeating a sentence, the next illustration appears, and the frame story proceeds. The progress in the game (displayed at the bottom of the screen) is directly related to the repetition of the target sentences, which enhances motivation. This interactive procedure adds a playful element to the repetition of the sentences, and the formal test character of the examination recedes into the background (Haas et al., 2020).



„Ich versteck´ die Mausefalle unter dem Tisch.“
[“I am hiding the mousetrap under the table”]

„Was ist denn da an meinem Zeh?“
[“What's this thing on my toe?”]

Figure 1. BoDyS-KiD materials: Two screens depicting the sequence of a funny event. © The Astrid Lindgren Company / Bildmakarna Berg / Björn Berg.

Children who have difficulties following the frame story or repeating sentences due to multiple disabilities are provided with the highest level of support. For example, each of the sentences is related to the picture at hand and is meaningful and plausible in relation to the action or situation depicted. Furthermore, multiple repetitions of the auditory target are possible and permitted. In addition, the repetition sentences are phrased as direct speech

by one of the characters involved. This allows the child to put him/herself in the position of the character.

The repetition task includes 12 sentences that were constructed systematically, taking into account phonetic and linguistic parameters. Sentences vary in length (6- to 12-syllables), intonation patterns (declarative, imperative, and interrogative), word length (1- to 4-syllable words), complexity of syllables (number of consonant clusters), as well as manner and place of articulation to elicit diverse and thus valid speech material. Two versions of the computer game were developed for children of different ages and cognitive abilities. Both versions differ in the number of repetition sentences (12 vs. 9 sentences) and the length and syntactic complexity of the frame story. The stimulus sentences were adapted to the children's level of linguistic development. Following the example of the adult version of the BoDyS, the 12 (or 9) sentences were also designed to form three groups of four (or three) sentences each, to obtain not only one overall judgment but several and more differentiated judgments in the subsequent auditory-perceptual analyses (see Ziegler et al., 2017; Ziegler et al., 2018).

Furthermore, the game also offers several opportunities to ask children specific open questions that are related to the content of the story (e.g., "What do you like to play with your friends?"). Thereby, as a standard, it allows for the recording of three sequences of spontaneous speech. Beyond that, the story provides other opportunities for spontaneous speech, so that children can also tell stories trough their own initiative.

In summary, the BoDyS-KiD materials allows for the elicitation of standardized speech samples from children with dysarthria in a child-appropriate and motivating way. It provides the basis for comprehensive auditory-perceptual analyses of all speech subsystems. Further information on the materials as well as a more detailed outline of the process of development can be obtained from Haas (2015), Haas et al. (2020), Schölderle et al. (2020), or online (https://lernmodule-sprachtherapie.de/).

1.3.2 Childhood dysarthria in the context of typical speech development

Due to anatomical-physiological characteristics (Lieberman et al., 2001; Vorperian et al., 2009) as well as the neuronal immaturity of the infant brain, speech in children differs markedly from adult speech throughout the first decade of life (Boliek et al., 2009; Hoit et

al., 1990; Murdoch, 2011; Smith, 2006; Vorperian et al., 2005). For instance, a reduced lung volume and immature coordination of the abdominal and costal breathing muscles may result in both shorter exhalation phases and audible inspirations when speaking (Boliek et al., 2009; Hoit et al., 1990). A slow speech rate may be related to cognitive–linguistic development (Martins et al., 2007). This represents a major challenge for the assessment of childhood dysarthria, as the above-mentioned physiological features of childhood speech coincide with common symptom categories of dysarthria. Thus, the identification of dysarthria symptoms inevitably requires normative data. Without norms, a valid distinction between salient, yet age-appropriate speech characteristics and atypical speech characteristics cannot be achieved. To date, normative data covering all relevant speech subsystems are not available for German-speaking children.

1. The first aim of the present dissertation was to investigate auditory-perceptual characteristics related to all speech subsystems (i.e., respiration, voice, articulation and resonance, prosody) in typically developing children aged three to nine years. These data then allow for a differentiation between age-appropriate ("typical") speech characteristics and specific symptoms of childhood dysarthria.

1.3.3 Developmental courses of childhood dysarthria

To date, there are mainly cross-sectional investigations of speech characteristics in childhood dysarthria and only little is known about the developmental courses (Hustad et al., 2019). Longitudinal studies of childhood dysarthria are however particularly challenging because the course of a child´s speech impairment interacts with the dynamics of typical speech development. Due to the lack of age-norms, earlier studies have not been able to relate their observations in dysarthric children to typical development. This is true for a comparison at a specific time point (i.e., in cross-sectional studies or for each point in time in a longitudinal study), but also for a comparison of the developmental dynamics of typically developing children and children with dysarthria. Normative data, though, are essential to classify a child's development in the context of typical development.

Such an approach would allow for specific analyses of the developmental courses, i.e., whether a child manages to keep up with the typical speech development or develops differently in comparison to children of the same age. Detailed knowledge of such trends in speech motor development of children with dysarthria can help to identify needs and potentials for speech-language intervention and are thus highly relevant for clinical treatment planning.

Most previous studies have carried out acoustic analyses of individual speech characteristics in children with dysarthria. In contrast, auditory-perceptual analyses have been performed only rarely, although they are the gold standard in clinical practice.

2. The second objective of this dissertation was to describe developmental courses of childhood dysarthria against the background of typical speech motor development.

1.3.4 Influence of childhood dysarthria on communicative abilities

Dysarthria is usually associated with considerable communicative restrictions (Bax et al., 2006; Mei et al., 2014). This means, for example, that the children's intelligibility is often reduced (e.g., Hustad et al., 2012) and the affected children consequently experience difficulties in communicating with others. Naturally, these difficulties are encountered in all areas of everyday life. Among other factors, intelligibility proved to be a decisive criterion for whether children interact with others at all (Pennington, 1999). Dysarthria acquired at an early age can thus constitute a significant impairment to a person's participation in everyday life and may ultimately severely affect the individual´s quality of life (Dickinson et al., 2007; Fauconnier et al., 2009; Mei et al., 2015; Young et al., 2010). These limitations are of major significance in children with neurologic conditions, as by definition, childhood dysarthria occurs in infancy and constitutes a lifelong disability.

Consequently, in addition to an assessment of the auditory-perceptual correlates of motor speech dysfunction, a comprehensive assessment of childhood dysarthria should also consider how the individual is able to participate in life situations, for example, through communication. One approach that accounts for both aspects is the model of the International Classification of Functioning, Disability and Health (see Figure 2: ICF;

WHO, 2001). This holistic view of a health condition, e.g., childhood dysarthria, may provide speech-language pathologists with a coherent structure to organize a diagnostic process that takes all relevant perspectives into account.

Figure 2 maps the multiple effects of dysarthria from the perspective of an affected child onto the ICF model. Environmental factors and personal factors are part of the ICF model as well but are not depicted in Figure 2, as they will not be part of present dissertation – despite their clinical relevance.

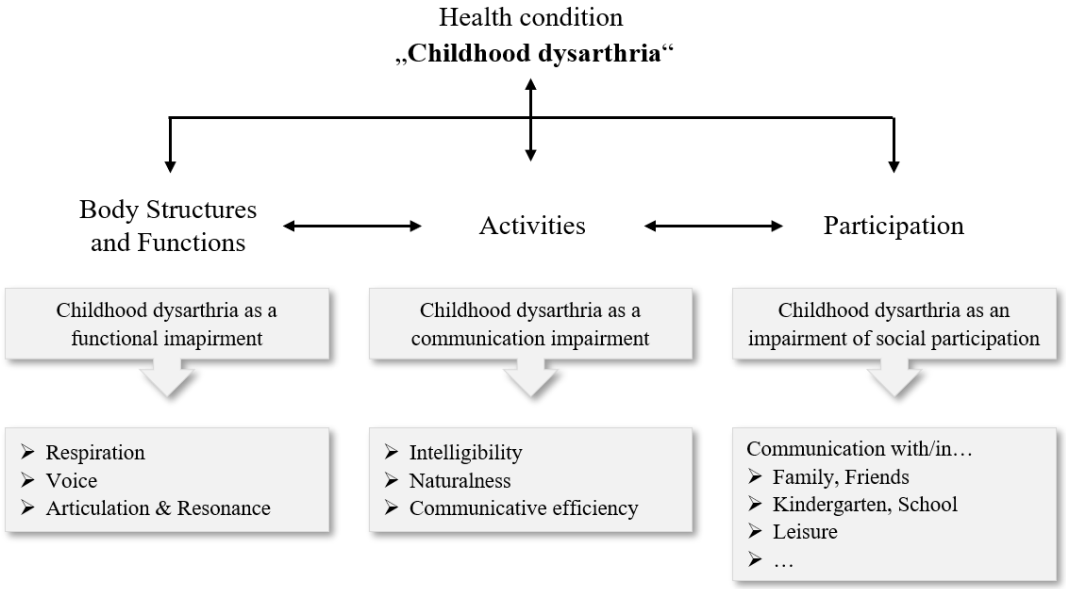


Figure 2. Adapted illustration of the interactions between the components of the International Classification of Functioning, Disability and Health (ICF; WHO, 2001) with the example of "childhood dysarthria" as the health condition. Environmental factors and personal factors are not included.

The primary goal of speech-language assessment is the comprehensive and detailed identification of a child's impairments and the resulting need for treatment. The main objective of treatment is then to improve the child's everyday communication.

To date, however, normative data of different speech features in typically developing children have not been established. Such data can provide a reference measure for the assessment of communication abilities in childhood dysarthria. Moreover, there is a lack of studies that systematically cover a wide age range and consider different parameters to describe communication skills. Hence, earlier studies on communication abilities in childhood dysarthria remained on a descriptive level (e.g., percent of intelligible words within a group of assessed children). In addition, communication-related parameters of neither typically developing children nor children with dysarthria have previously been

related to auditory-perceptual characteristics corresponding to speech subsystems. Moreover, there is a lack of longitudinal analyses which could provide insights into the relationship between the development of auditory-perceptual characteristics and changes of communication abilities.

3. The third aim of the dissertation was to examine the children's communicative abilities through intelligibility, rate, and communication efficiency. Again, communication was investigated against the background of typical development across a longer period of time.

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2 Outline of the dissertation and statement of own contribution

Four studies will be presented in this dissertation. Figure 3 illustrates their focusses and groups of subjects.

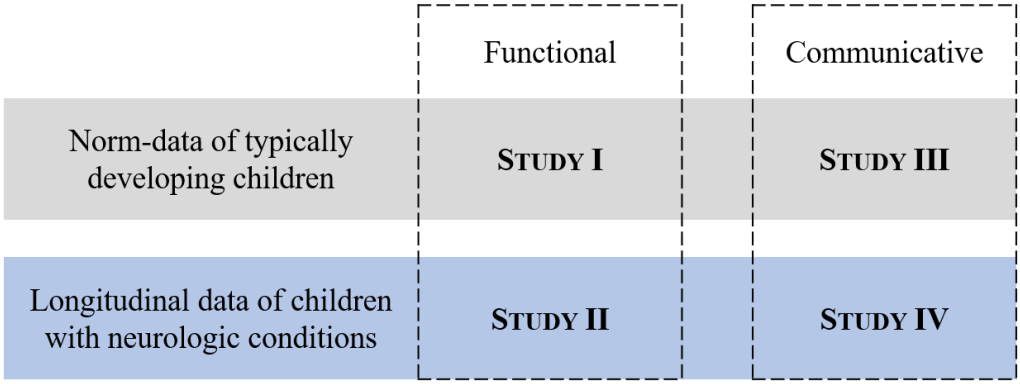

Figure 3. Schematic illustration of the four studies.

In studies I and II, the focus was on a comprehensive characterization of auditory-perceptual characteristics related to speech subsystems. Throughout this dissertation, the term "speech functions" will be used to describe the auditory correlates of motor speech functions. More precisely, speech functions will be understood as the deviations of these auditory correlates from an intuitive adult norm, which will be determined by experts through systematic auditory-perceptual judgements. Studies III and IV concentrated on communication abilities. In this dissertation, the analyses of communicative abilities will be confined to the level of verbal interaction. For this purpose, the analyses include intelligibility as well as articulation and speech rate, which then allow for a combined measure of communicative efficiency. Normative data were established for both auditory-perceptual characteristics related to speech subsystems and communicative parameters, as only typically developing children were included in studies I and III (see Figure 3). The normative data were then applied to children with neurological conditions in studies II and IV. Speech samples for all studies were obtained through the BoDyS-KiD materials.

To provide a basis for a valid assessment of childhood dysarthria, study I[1] was dedicated to the collection of comprehensive neurophonetic normative data for auditory-perceptual speech characteristics of kindergarten and elementary school aged children. These norms then formed the basis for study II[2], in which children with neurologic conditions were examined longitudinally. Thus, for the first time, it became possible to differentiate between speech characteristics that are part of the typical development and those that are symptoms of dysarthria.

As depicted in Figure 3, study III[3] presents normative data on communication-related parameters. In addition, to be able to bring both auditory-perceptual characteristics related to speech subsystems and communication parameters together, these were linked to the data collected in study I. In study IV[4], the normative data obtained in study III were applied to children with neurologic conditions. These were presented again in longitudinal section and, in addition, were also related to the data on auditory-perceptual characteristics of study II.

The present dissertation project was carried out in close collaboration with the DFG project headed by Dr. Theresa Schölderle and Prof. Dr. Wolfram Ziegler "Speaking in typical development and after early brain damage: A development-based neurophonetic investigation of childhood dysarthria". For this reason, the authors are almost the same for all articles included. To break down the contributions of the individual authors to the respective articles, a CRediT author statement is provided below for each study.

[1] Schölderle, T.*, Haas, E.*, & Ziegler, W. (2020). Age Norms for Auditory-Perceptual Neurophonetic Parameters: A Prerequisite for the Assessment of Childhood Dysarthria. *Journal of Speech, Language, and Hearing Research, 63*(4), 1071–1082. https://doi.org/10.1044/2020_JSLHR-19-00114
*shared first authorship

[2] Haas, E., Ziegler, W., & Schölderle, T. (2021). Developmental Courses in Childhood Dysarthria: Longitudinal Analyses of Auditory-Perceptual Parameters. *Journal of Speech, Language, and Hearing Research, 64*(5), 1421–1435. https://doi.org/10.1044/2020_JSLHR-20-00492

[3] Schölderle, T., Haas, E., Baumeister, S., & Ziegler, W. (2021). Intelligibility, Articulation Rate, Fluency, and Communicative Efficiency in Typically Developing Children. *Journal of Speech, Language, and Hearing Research, 64*(7), 2575–2585. https://doi.org/10.1044/2021_JSLHR-20-00640

[4] Haas, E., Ziegler, W., & Schölderle, T. (2022). Intelligibility, Speech Rate, and Communication Efficiency in Children with Neurological Conditions: A Longitudinal Study of Childhood Dysarthria. *American Journal of Speech-Language Pathology, 31*(4), 1817-1835. https://doi.org/10.1044/2022_AJSLP-21-00354

Study I: Age Norms for Auditory-Perceptual Neurophonetic Parameters: A Prerequisite for the Assessment of Childhood Dysarthria

CRediT author statement:

Theresa Schölderle and Elisabet Haas shared first authorship.

Theresa Schölderle:	Funding acquisition, Conceptualization, Methodology, Investigation, Data Curation, Writing – Original Draft, Project administration
Elisabet Haas:	Funding acquisition, Conceptualization, Investigation, Data Curation, Writing – Original Draft
Wolfram Ziegler:	Funding acquisition, Conceptualization, Formal analysis, Writing – Review & Editing, Supervision

Study II: Developmental Courses in Childhood Dysarthria: Longitudinal Analyses of Auditory-Perceptual Parameters

CRediT author statement:

Elisabet Haas:	Funding acquisition, Conceptualization, Methodology, Investigation, Data Curation, Writing – Original Draft, Project administration
Wolfram Ziegler:	Funding acquisition, Conceptualization, Formal analysis, Writing – Review & Editing
Theresa Schölderle:	Funding acquisition, Conceptualization, Writing – Review & Editing, Supervision

Study III: Intelligibility, Articulation Rate, and Communicative Efficiency in Typically Developing Children

CRediT author statement:

Theresa Schölderle:	Funding acquisition, Conceptualization, Methodology, Investigation, Data Curation, Writing – Original Draft, Project administration
Elisabet Haas:	Funding acquisition, Conceptualization, Methodology, Investigation, Data Curation, Writing – Review & Editing
Stefanie Baumeister:	Investigation, Data Curation
Wolfram Ziegler:	Funding acquisition, Conceptualization, Formal analysis, Writing – Review & Editing, Supervision

Study IV: Intelligibility, Speech Rate, and Communication Efficiency in Children with Neurological Conditions: A Longitudinal Study of Childhood Dysarthria

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Elisabet Haas:	Funding acquisition, Conceptualization, Methodology, Investigation, Data Curation, Writing – Original Draft, Project administration
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Theresa Schölderle:	Funding acquisition, Conceptualization, Methodology, Data Curation, Writing – Review & Editing, Supervision

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3 STUDY I:

Age Norms for Auditory-Perceptual Neurophonetic Parameters: A Prerequisite for the Assessment of Childhood Dysarthria

Theresa Schölderle*, Elisabet Haas*, & Wolfram Ziegler

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Schölderle, T.*, Haas, E.*, & Ziegler, W. (2020). Age Norms for Auditory-Perceptual Neurophonetic Parameters: A Prerequisite for the Assessment of Childhood Dysarthria. *Journal of Speech, Language, and Hearing Research, 63*(4), 1071–1082. https://doi.org/10.1044/2020_JSLHR-19-00114

(*shared first authorship)

4 STUDY II:

Developmental Courses in Childhood Dysarthria: Longitudinal Analyses of Auditory-Perceptual Parameters

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5 STUDY III:

Intelligibility, Articulation Rate, Fluency, and Communicative Efficiency in Typically Developing Children

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Schölderle, T., Haas, E., Baumeister, S., & Ziegler, W. (2021). Intelligibility, Articulation Rate, Fluency, and Communicative Efficiency in Typically Developing Children. *Journal of Speech, Language, and Hearing Research*, *64*(7), 2575–2585. https://doi.org/10.1044/2021_JSLHR-20-00640

6 STUDY IV:

Intelligibility, Speech Rate, and Communication Efficiency in Children with Neurological Conditions: A Longitudinal Study of Childhood Dysarthria

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7 Conclusion

In this dissertation, for the first time, comprehensive norm data of typically developing children for speech motor as well as communicative parameters were collected. These data constitute a highly relevant prerequisite for the description and clinical assessment of childhood dysarthria. The norm data were then applied to children with neurological conditions - in the first international longitudinal study on childhood dysarthria that contains normalized functional and communication parameters. The speech samples underlying all analyses were elicited using material specifically developed for use in children with neurologic disorders, i.e., the BoDyS-KiD materials.

Overall, this work contributes to a thorough theoretical understanding of childhood dysarthria as well as of typical speech motor development and provides a basis for a specific comprehensive and valid clinical assessment. Two central components of assessment were linked in the present thesis, namely auditory-perceptual characteristics related to speech subsystems and communicative parameters. Such extensive analyses of the interplay of both components had not been addressed before despite their high theoretical and clinical relevance. The findings presented in this thesis may ultimately provide a research-based guideline for clinicians supporting them in decision making in speech-language intervention. Therefore, this dissertation may contribute to an improvement in the health care of children with neurological conditions.

Future studies can now build upon the findings of this work and address further research questions in the field of childhood dysarthria.

Deutsche Zusammenfassung

Kindliche Sprachentwicklungsstörungen sowie neurogene Kommunikationsstörungen im Erwachsenenalter stellen zwei zentrale Säulen der Sprachtherapie dar. Erkenntnisse aus beiden Bereichen haben bislang jedoch kaum Eingang in die jeweils andere Disziplin gefunden, trotz theoretisch sowie klinisch relevanter Schnittstellen, wie beispielsweise den kindlichen Dysarthrien. Kindliche Dysarthrien treten in Folge einer neurologischen Erkrankung auf, die im frühen Kindesalter (d.h. vor Abschluss der sprechmotorischen Entwicklung) erworben wird und fügen sich zumeist in das Bild einer Mehrfachbehinderung.

Neurologische Erkrankungen im Kindesalter sind keine Seltenheit. Dabei sind einige Ätiologien, die besonders häufig Dysarthrien verursachen, durch einen frühen Krankheitsbeginn gekennzeichnet, wie beispielsweise die Cerebralparese oder genetische Syndrome. Aber auch andere, altersunabhängige Ätiologien können bereits im Kindesalter zu einer Dysarthrie führen, wie z.B. zerebrovaskuläre Ereignisse. Schätzungen zufolge leben mehr als 50.000 Kinder und Jugendliche in Deutschland mit einer Dysarthrie.

Trotz der großen Gruppe betroffener Kinder sind die kindlichen Dysarthrien sowohl in der Forschung als auch in der klinischen Praxis im Vergleich zu den im Erwachsenenalter erworbenen Dysarthrien bisher deutlich unterrepräsentiert. So wurden bislang z. B. kaum spezifische Ansätze zur Diagnostik der Sprechstörung entwickelt, weshalb in früheren Studien Testmaterialien und Auswertungskriterien übertragen wurden, die für spät erworbene Dysarthrien etabliert sind. Auf diese Weise werden jedoch der Entwicklungsaspekt sowie mögliche Begleiterscheinungen, die im Rahmen einer Mehrfachbehinderung auftreten können, vernachlässigt. Als vorbereitende Arbeit für die vorliegende Dissertation wurde deshalb zunächst der Untersuchungsansatz *Bogenhausener Dysarthrieskalen – Kindliche Dysarthrien* (BoDyS-KiD) entwickelt, der die standardisierte Aufnahme von Sprechproben bei Kindern mit neurologischen Erkrankungen auf kindgerechte und motivierende Art und Weise erlaubt.

Des Weiteren sind auch die bisher durchgeführten Studien zur Beschreibung der Symptome kindlicher Dysarthrien in Anzahl und Umfang begrenzt. Zusammenfassend zeigen sie jedoch, dass in allen Funktionskreisen des Sprechens (d.h. Atmung, Stimme, Artikulation und Resonanz sowie Prosodie) Symptome beschrieben wurden. Allerdings analysierte nur eine sehr begrenzte Anzahl von Studien die Sprechstörungen der Kinder über mehrere Funktionskreise hinweg. Darüber hinaus wurden nur selten auditive Analyseverfahren angewendet, die jedoch in der klinischen Diagnostik den Goldstandard darstellen. Zuletzt muss darauf hingewiesen werden, dass bislang in keiner Studie eine systematische Altersnormierung der Sprechmerkmale vorgenommen wurde.

Kindliches Sprechen unterscheidet sich jedoch aufgrund anatomisch-physiologischer Besonderheiten sowie der neuronalen Unreife des kindlichen Gehirns über die gesamte erste Lebensdekade hinweg deutlich von erwachsenem Sprechen. Diese Tatsache stellt ein erhebliches diagnostisches Problem dar, da viele physiologische Merkmale kindlichen Sprechens auditiv mit Phänomenen übereinstimmen, die bei einer Hirnschädigung im Erwachsenenalter als Symptom einer Dysarthrie interpretiert werden. Eine eindeutige Differenzierung physiologischer und pathologischer Merkmale kann daher ausschließlich auf Basis altersspezifischer Normdaten erfolgen. In diesem Zusammenhang können Längsschnittuntersuchungen, in denen ein Normvergleich stattfindet, besonders relevante Erkenntnisse liefern. So kann beispielsweise dokumentiert werden, inwieweit ein Kind mit der Entwicklungsdynamik typisch entwickelter Kinder im selben Alter Schritt halten kann, oder ob das Ausmaß beobachteter Veränderungen von typischen Entwicklungsprozessen abweicht.

Die Symptome dysarthrischer Sprechstörungen sind zudem meist mit erheblichen kommunikativen Einschränkungen verbunden und beeinträchtigen die Teilhabe eines Menschen. Jedoch liegen bislang auch hinsichtlich kommunikativer Parameter keine umfangreichen Normdaten typisch entwickelter Kinder vor. Die Anwendung einer Altersnormierung ist jedoch für eine valide Diagnostik unabdingbar, die wiederum die Grundlage für therapeutische Entscheidungen (z.B. zur Behandlungsbedürftigkeit einer Störung oder der Definition von Therapieschwerpunkten) darstellt.

Die vorliegende kumulative Dissertation umfasst vier Artikel, die unmittelbar auf den dargestellten Forschungslücken aufbauen und im Folgenden skizziert werden. Es wird erstmals ein funktions- und kommunikationsorientierter Ansatz zur Diagnostik kindlicher

Dysarthrien vorgestellt, der den physiologischen Entwicklungsprozess systematisch berücksichtigt. Abbildung 1 visualisiert die Zusammenhänge der Studien.

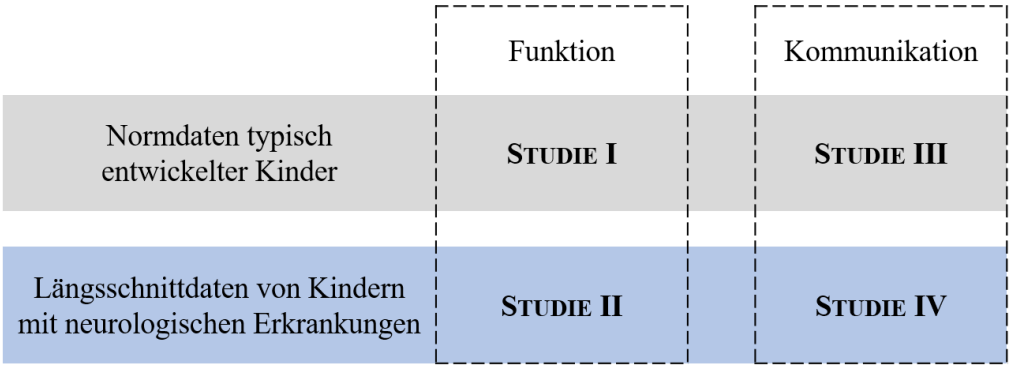

Abbildung 1. Schematische Darstellung der vier Studien.

Studie I hatte zum Ziel, über eine große Altersgruppe hinweg umfassende neurophonetische Normdaten für etablierte Symptomkategorien einer Dysarthrie zu erheben. Dazu wurden in einer Querschnittsstudie 144 typisch entwickelte Kinder zwischen 3 und 9 Jahren untersucht. Anhand des BoDyS-KiD Materials wurden Nachsprechsätze sowie Spontansprachproben aufgenommen. Die auditive Beurteilung erfolgte entlang der Analysemethoden der *Bogenhausener Dysarthrieskalen* (BoDyS), einem etablierten Diagnostikverfahren für Erwachsene, auf dem der BoDyS-KiD Ansatz unmittelbar aufbaut. Anhand von 9 Skalen (*Atmung*, *Stimmlage*, *Stimmqualität*, *Stimmstabilität*, *Artikulation*, *Resonanz*, *Artikulationstempo*, *Redefluss*, *Modulation*) werden dabei alle klinisch relevanten Funktionen des Sprechens systematisch abgeprüft.

Die Ergebnisse dieser Studie legten dar, dass die typisch entwickelten Kinder verschiedene Sprechmerkmale zeigten, die mit Symptomkategorien einer Dysarthrie überlappen. So traten beispielsweise häufig eine behauchte Stimmqualität, eine reduzierte Artikulation oder auch ein verlangsamtes Artikulationstempo auf. Darüber hinaus konnten bedeutsame Fortschritte zwischen den 3- und 9-jährigen Kindern beobachtet werden. Diese waren für die einzelnen BoDyS Skalen jedoch unterschiedlich stark ausgeprägt. So waren beispielsweise für die *Atmung* und die *Stimmqualität* auch bei den 9-jährigen Kindern noch erhebliche Abweichungen von der Erwachsenennorm erkennbar, während etwa im Bereich der *Modulation* bereits bei den 3-jährigen Kindern nur selten Abweichungen auftraten.

Zusammenfassend wurde in dieser Studie aufgezeigt, dass in dem untersuchten Alter teils noch erhebliche Abweichungen von der Erwachsenennorm bestehen. Die systematisch erhobenen Altersnormen für alle Funktionskreise des Sprechens erlauben nun

die Unterscheidung zwischen altersadäquaten physiologischen Sprechmerkmalen und Symptomen einer kindlichen Dysarthrie.

An dieser Stelle setze **Studie II** an, deren Ziel es war, Entwicklungsverläufe kindlicher Dysarthrien vor dem Hintergrund der typischen sprechmotorischen Entwicklung zu beschreiben. Im Rahmen einer Längsschnittstudie wurden 14 Kinder mit neurologischen Erkrankungen im Alter von 5 bis 8 Jahren über 18 Monate hinweg zu drei Zeitpunkten untersucht. Diese Gruppe wurde durch 14 typisch entwickelte, nach Alter und Geschlecht gematchte Kinder ergänzt. Die Sprechproben (Nachsprechsätze und Spontansprache) wurden anhand des BoDyS-KiD Materials erhoben und, wie für Studie I beschrieben, entlang der BoDyS Auswertungskriterien auditiv beurteilt.

Anhand der in Studie I erhobenen Normdaten erfolgte eine Altersnormierung für alle BoDyS Skalen, sowie für den BoDyS-KiD Gesamtschweregrad (= Mittelwert über die 9 BoDyS Skalen). Außerdem wurden die Daten der 14 typisch entwickelten Kinder herangezogen, um zu beurteilen, ob die Gradienten der Entwicklungsverläufe der Gruppe von Kindern mit neurologischen Erkrankungen die der typischen Entwicklung übertreffen. Dabei zeigte sich, dass die Störungsprofile der Kinder mit neurologischen Erkrankungen sehr heterogen waren und hinsichtlich des Schweregrades stark variierten. Auf Gruppenebene waren jedoch über den Beobachtungszeitraum hinweg relativ konstante Entwicklungsverläufe erkennbar. Außerdem wurde ersichtlich, dass die Entwicklung der Kinder mit neurologischen Erkrankungen hinsichtlich aller neun BoDyS Skalen größtenteils parallel zur Entwicklung der typisch entwickelten Kinder verlief.

Diese Ergebnisse ließen den Rückschluss zu, dass die meisten Kinder mit neurologischen Erkrankungen in der Lage waren, die Entwicklungsdynamik auszunutzen und sich parallel zur Norm weiterzuentwickeln.

In den Studien III und IV standen die kommunikativen Fähigkeiten im Vordergrund. Der Fokus von **Studie III** lag zunächst auf der Beschreibung der Entwicklungstrajektorien typisch entwickelter Kinder hinsichtlich vier kommunikationsorientierter Parameter: *Verständlichkeit, Artikulationstempo, Redefluss* und *kommunikative Effizienz*. Studie III baute unmittelbar auf Studie I auf und nutzte die dort erhobenen Sprechproben sowie die auditiven Daten der 144 typisch entwickelten Kinder. Um die Verständlichkeit zu beurteilen, wurde ein Hörexperiment durchgeführt, bei dem Laienhörer die aufgenommenen Nachsprechsätze orthografisch transkribierten. Ergänzend wurden

Artikulationstempo und Redefluss durch akustische Analysen bestimmt. Die kommunikative Effizienz wurde ermittelt, indem der Anteil korrekt transkribierter Silben mit der Sprechrate multipliziert wurde.

Für die Verständlichkeit zeigten sich steile Entwicklungstrajektorien. Der Großteil der Kinder war bereits im Alter von fünf Jahren nahezu uneingeschränkt verständlich. Dem gegenüber entwickelte sich das Artikulationstempo deutlich langsamer und zeigte eine hohe Variabilität – auch bei den älteren Kindern. Redeflussstörungen traten wiederum nur bei den jüngsten Kindern auf. Per Definition vereinte die kommunikative Effizienz Charakteristika der Entwicklungsverläufe von Verständlichkeit und Artikulationstempo. In einem weiteren Schritt wurden die vier kommunikationsorientierten Parameter mit den in Studie I auditiv erhobenen Funktionsparametern in Beziehung gesetzt. Dabei trat am deutlichsten ein signifikanter Zusammenhang zwischen der Verständlichkeit und der Artikulation zutage. Aber auch zwischen der kommunikativen Effizienz, der Artikulation und den Tempomaßen konnten Zusammenhänge gefunden werden.

Studie IV hatte daraufhin zum Ziel, kommunikative Parameter (*Verständlichkeit*, *Sprechtempo* und *kommunikative Effizienz*) bei Kindern mit neurologischen Grunderkrankungen zu untersuchen und deren Entwicklungsverläufe vor dem Hintergrund der typischen Entwicklung zu beschreiben. Darüber hinaus sollten auch für diese Stichprobe die Zusammenhänge zwischen den Entwicklungsverläufen auf kommunikativer und funktioneller Ebene untersucht werden.

Die Probanden entsprachen den in Studie II vorgestellten Kindern (14 Kinder mit neurologischen Grunderkrankungen, 14 typisch entwickelte Kinder). Die auditiven Ratings wurden ebenfalls aus Studie II übernommen. Das Vorgehen zur Beurteilung von Verständlichkeit, Sprechtempo und kommunikativer Effizienz war identisch mit Studie III. Die Altersnormierung erfolgte entlang der in Studie III ermittelten Normdaten.

Auf Gruppenebene zeigten die Kinder mit neurologischen Erkrankungen auffällige Roh- sowie Normwerte für alle der drei Kommunikationsparameter. Außerdem wurde im Vergleich zu den typisch entwickelten Kindern eine höhere Variabilität hinsichtlich der Entwicklungsverläufe offensichtlich, die jedoch für die Verständlichkeit deutlich ausgeprägter war als für das Sprechtempo und die kommunikative Effizienz. Zuletzt wurde auch ein starker Zusammenhang zwischen der Entwicklung der Verständlichkeit und der Artikulation gefunden.

Die vorliegende Dissertation trägt somit zu einem grundlegend besseren Verständnis der physiologischen sprechmotorischen Entwicklung sowie den Entwicklungsverläufen kindlicher Dysarthrien bei. Die adressierten Fragestellungen sind von unmittelbarer klinischer Relevanz und können Grundlage für eine spezifische, umfassende und valide klinische Diagnostik darstellen.

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