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Maassen, Ben A. M.

Published in:
Routledge Handbook of Communication Disorders

Publication date:
2015

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DEVELOPMENTAL MODELS OF CHILDHOOD APRAXIA OF SPEECH

Ben Maassen

During the last decade, research suggested that developmental communication disorders have multi-factorial origins at different levels of aggregation. Moreover, the influence of these levels of aggregation on the resulting behavioral output is interactive rather than uni-directional. In this chapter, a developmental, perceptual-motor model of childhood apraxia of speech (CAS) is described that uses this multi-level, multi-factorial concept as its foundation. In the first section, global outlines of the interactive multi-level model are described. This is followed by a discussion of each of the four levels as they relate to CAS, including the interactions between the levels. Since relatively little is known about the genetic and neurological background of CAS, the focus in this chapter is on the perceptual-motor level, including to some extent higher cognitive functions, such as lexical retrieval. In addition, the behavioral level is described, being the most directly observable and accessible level for clinical management. In the final section, implications for clinical practice and future research directions are discussed.

Multi-level models of developmental disorders

A model describing the different levels of causation of developmental disorders is presented in Figure 11.1 (adapted from Bishop and Snowling, 2004). Four levels of aggregation are distinguished, with some of the boxes labeled according to known underlying deficits in CAS. The etiological level describes the genetic constitution of the individual in combination with relevant environmental factors including biological factors, such as ante- and perinatal conditions. The Bishop and Snowling (2004) model also incorporates the influence a child's behavior has on his/her own environment. An example would be the communicative activity level of an infant. More active infants tend to elicit more communicative response from the care-taker as compared to more passive infants. Hence, during development, the etiological factors unfold into a neurological architecture combined with functionality, i.e., the neurobiological level. The model makes clear that the brain does not develop according to a genetic blueprint, but is continuously adapting to biological and behavioral (environmental) circumstances.

Cognitive functions form the third level of aggregation. The raison d'être of this intermediate level between neurobiology and behavior is that there is no one-to-one relationship between brain functions and behavior. Cognitive functions indeed constitute an intermediate level that operates at a more abstract level than the behaviors that result from it. For example, a deficit in

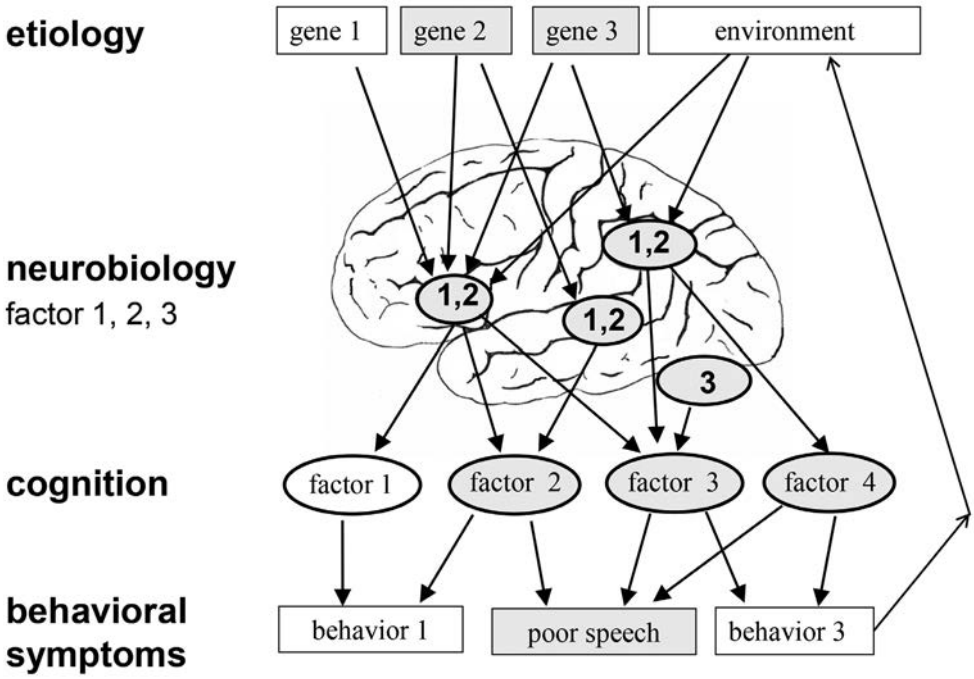

Figure 11.1 Levels of causation for Childhood Apraxia of Speech (CAS) (Modified after Bishop and Snowling, 2004, p. 859). Numbered gray cells indicate factors associated with CAS.

Note: Genes 2 and 3 refer to associated genes, such as FoxP2 (Graham and Fisher, 2013). Neurobiology factors 1 and 2 refer to distributed factors in speech areas, such as reduced neurite outgrowth (Graham and Fisher, 2013). Factor 3 depicts cerebellar deficits. Cognition factors 2, 3 and 4 refer to known factors, such as poor sequential motor programming, coordination, and auditory processing.

word and syllable retrieval could have an effect on sentence construction – such as when the speaker experiences difficulty in rapid retrieval of the correct word form for sentence continuation, which could result in speech dysfluencies. In this case, the speaker has difficulty with initiating word articulation. The fourth level is behavioral. It is directly observable and constitutes – in the case of CAS – the speech characteristics and clinical symptoms.

A series of theoretical papers summarizing the main arguments for the notion that developmental disorders are quite unlike acquired disorders in adults has been published. An exhaustive discussion is outside the scope of this chapter, but the key concepts and arguments are briefly presented here. Bishop (1997) argued that dissociation, or even better, double dissociation, which is the ideal neuropsychological evidence in the study of acquired disorders in adults, does not apply to developmental disorders. The reason is that a dysfunction in a developing child (e.g., poor auditory speech perception) will affect the acquisition of other functions and skills that partly depend on this function being intact (e.g., phonological development). Thus, in developmental disorders, it is more likely to find associations between functions than dissociations. From a slightly different perspective, Karmiloff-Smith (2006; Karmiloff-Smith *et al.*, 2003) comes to the same conclusion, arguing that in both normal and disordered development, cognitive modules are the outcome of development rather than its starting point. The progression to the adult system is a gradual and continuous process made up of interactions between emerging modules, resulting in associations among functions.

In an attempt to specify the developmental process for CAS, Maassen (2002) observed that infant speech development starts from random babbling and sensomotoric learning, which forms the basis for more abstract phonological acquisition. Starting from the assumption that the core deficit of CAS is a reduced sensomotoric learning capacity, one can predict not only poor articulation, but also poor auditory and somatosensory representation, and, from there, effects on the psycholinguistic domain because of the impact on phonological and higher-level processes. This contrasts with adults with apraxia of speech (AOS), who already have acquired stable top-down processes. Because the developmental trajectory sketched above is probabilistic and multi-factorial rather than deterministic, Karmiloff-Smith (2006) characterizes the process of childhood disorders as a tortuous route from genes to behavior.

Genetics

The indirect relationship (or tortuous route) between the underlying deficit at the neurological and cognitive level and speech symptoms, seriously complicates the search for heritability and genetic factors involved in CAS. Because the diagnostic speech symptoms of CAS change with age, and are influenced by multiple factors, family members cannot be directly compared at the behavioral level. Even within twins, the commonality of symptoms changes during speech development. The difficulty lies in the changing speech symptoms (Maassen 2002), which seriously complicates the specification of the phenotype. Therefore, Stein *et al.* (2011) have argued that studies comparing siblings, or parent-offspring pairs need to take into account that the expression of a particular underlying, genetically determined trait changes with age and is influenced by environmental factors.

Nevertheless, recent progress in genetic linkage methodology has revealed underlying genetic deficits in speech and language disorders. Since its discovery in 1998 (Fisher *et al.*, 1998), the *FOXP2* gene probably has been the most frequently studied gene in relation to speech and language functions. Its major function has been shown to be regulating the expression of other genes. That is, *FOXP2* encodes a transcription factor that controls the expression of a series of other genes regulating, among other things, language and motor functions. One of these genes regulated by *FOXP2* is *CNTNAP2*, which has been associated with specific language impairment (SLI). Specific parts of *CNTNAP2* also have been associated with phonological short-term memory, and thus with performance on non-word repetition tasks (Newbury *et al.*, 2010).

Whether directly, or in its role as transcription factor, variants of *FOXP2* have been shown to affect the development of the motor cortex, striatum and cerebellum. These structures are involved in neural circuits that facilitate language acquisition, and more specifically sequential speech motor learning. Interestingly, mutations of *FOXP2* lead to difficulties in sequential articulatory movements, like in AOS, and songbird orthologues of *FoxP2* are involved in the vocal learning of the courtship song. Knockdown of FoxP2 expression during song development results in inaccurate and incomplete imitation of tutor songs (Vernes and Fisher, 2009).

Four single cases of individuals with *FOXP2* disruptions have been summarized by Shriberg *et al.* (2012), showing relationships among speech, prosody, voice, cognition, language, and other findings with CAS. Other CAS related genes are *FOXP1*, *FOXG1*, *ELP4*, and *RAI1*. These authors note an important difference in CAS as compared to acquired AOS in adults, in that in CAS, cognitive and sensorimotor development may be affected in all brain regions and circuits in which gene expression is disrupted, as compared to more localized impairments in adults.

More detailed discussion is outside the scope of this chapter (the reader is referred to Section I of this volume) but it is important to realize that there are no direct genotype-phenotype links

in speech motor disorders. Rather, overlapping and interacting pathways from genes via neurological structure and function to cognition and behavior together result in a disorder with a complex phenotype, such as CAS.

Neurobiology

Direct data on neurobiological determinants of CAS is extremely scarce. No systematic brain imaging studies of young children with CAS are available for obvious ethical reasons, so the scarce studies with MRI, fMRI, or EEG are conducted with children with a medical condition requiring brain imaging. The ASHA Technical Report (American Speech-Language-Hearing Association (ASHA), 2007) notes that CAS can occur in different clinical contexts, the first of which comprises known neurological etiologies, such as intrauterine stroke, infections, and trauma. The second clinical context consists of complex neurobehavioral disorders (e.g., genetic, metabolic). Both contexts are promising sources of information on neural mechanisms underlying CAS. Thus, from a series of heterogeneous studies, it can be determined that associations exist between CAS on the one hand and benign rolandic epilepsy (5 patients), fragile-X syndrome (4 out of 10 patients), galactosemia (14 out of 24 patients), Down syndrome, Rett syndrome, autism, Coffin-Sirus syndrome, and increased theta-activity on clinical EEG in parietal-temporal cortical regions (Shriberg, 2010). In these cases, the underlying neurological impairment is known. The contribution of this clinical knowledge to our understanding of underlying mechanisms in CAS is limited, however, because these studies lack detail on the speech and prosody characteristics of the diagnosed children, such that the specific diagnosis CAS cannot be confirmed (ASHA, 2007).

In addition, CAS, which is not associated with any known neurological or complex neurobehavioral disorder, can occur as an *idiopathic neurogenic speech sound disorder* and can be accommodated by Shriberg's (2010) neurodevelopmental framework. This approach, in which core speech and non-speech features are based on diagnostic characteristics as sequelae of a known neurological or neurodevelopmental condition, may have research promise for resolving existing diagnostic controversies for CAS. Speech data from known neurological syndromes (CAS, possibly also AOS) should yield pathognomonic signs and markers, such that the diagnostic circularity – the problem that finding specific diagnostic markers requires clearly diagnosed patients (based on which markers?) – (see further discussion below) – can be breached. Once diagnostic markers are available, further research can be conducted to find the genetic and neural substrates by comparing validated cases of CAS with controls; then idiopathic motor speech disorders can be identified on the basis of established characteristics. A diagnostic category of special interest is dysarthria. There are several subtypes of dysarthria, and also the recently introduced diagnostic category, childhood motor speech disorder – not otherwise specified, which is a cover term for dysarthria and CAS, shows much overlap in symptomatology with CAS.

From a methodological point of view, Weismer and Kim (2010) argued to approach the diagnostic classification of dysarthrias and AOS from a taxonomic perspective, such that not only differential diagnostic, but also commonalities between disorders, are assessed to identify 'core' phenomena of these disorders. An example would be slow speech rate, which is characteristic of almost all speech disorders, and therefore needs to be accounted for rather than discarded because it does not contribute to differential diagnosis.

Cognitive and perceptual-motor processes

More than a decade ago, McNeil and colleagues (2004) made the following comment on the status quo of clinical research in AOS in adults:

> it is not a lack of theory or the inability to select the correct theory from the known alternatives that limits understanding of AOS [..]. It is, likewise, not the lack of neurologic or anatomic instantiation that limits AOS understanding. The most important impediment to theoretical and clinical advancement in AOS is, however, the lack of a comprehensive and clear definition that leads to an agreed-upon set of criteria for subject selection.
>
> *(p. 389).*

This comment still applies to AOS.

The comprehensive ASHA Technical Report (2007) on CAS concludes that: "… there presently is no one validated list of diagnostic features of CAS that differentiates this disorder from other types of childhood speech sound disorders, including those apparently due to phonological level deficits or neuromuscular disorder (dysarthria)" (p. 5). Thus, for both AOS and CAS, there is no consensus with respect to diagnostic criteria. However, as McNeil *et al.* (2004) above and the ASHA Technical Report (2007) conclude, there is consensus with respect to the underlying deficit at the cognitive level, thanks to a series of studies focusing on underlying mechanisms of CAS, by means of measuring kinematic parameters. Thus, as with AOS, in the last decade researchers have agreed on the processes or proximal causes underlying CAS. For CAS, the core impairment lies in planning and/or programming spatiotemporal parameters of movement sequences, resulting in imprecise and inconsistent speech movements (ASHA, 2007, pp. 3–4). Despite this clear definition of the underlying cause of CAS, the major obstacle for clinical management and research is that there is high variability in resulting speech symptoms, particularly if developmental aspects are also taken into account. This variability seriously complicates diagnostic classification.

It is clear that studying only the phonological output (percentage of consonants and vowels correct; number of substitutions – see next paragraph) is insufficient to collect the required data to diagnose the above mentioned underlying deficits. The study by Bahr (2005) was one of the earliest that focused on the movement of articulators rather than phonemic accuracy. Through comparisons of children with phonological disorder (PD) and CAS, it appeared that both groups evidenced significantly poorer phonemic accuracy than the typically developing children did, but these groups could not be distinguished based on their speech error profile. However, each child group with speech sound disorders (SSD) showed a specific pattern of gesture coordination difficulties. Both groups evidenced difficulty with gestures involving the tongue blade, but children with PD specifically showed poor voicing, and the children with CAS specifically had difficulty with correct coordination of the velum and the lips with the other articulators. An explanation for the coordination difficulty demonstrated by children with CAS might be that the independence of the velum and lips from the tongue is demanding from a gesture coordination point of view, and thus vulnerable to an underlying coordination deficit.

Also, Grigos and Kolenda (2010) studied kinematic parameters in addition to phoneme accuracy. They followed a 3-year-old boy with CAS for 8 months during the acquisition of /p/, /b/, and /m/. Transcription analyses showed that consonant and vowel errors decreased across sessions. More importantly, the child's kinematic jaw movement parameters became more similar to controls overall. However, only closing velocity and stability reached a level of accuracy comparable to controls, whereas opening velocities showed a much more erratic

pattern over time. Similarly, Terband *et al.* (2011) found higher variability of tongue tip movements trajectories in five children with CAS as compared to controls. Different patterns of coordination between CAS and controls were interpreted as an indication of underlying coordination difficulties.

Such coordination difficulties had been found by Green *et al.* (2002), who measured midsagittal displacements of the lips and jaw in typically developing 1-, 2- and 6-year-olds, as well as adults, while producing utterances 'baba', 'papa', and 'mama'. Their results showed that movement patterns of the jaw matured earlier than those of the lips. Further support for the specific development of lip movement coordination is found in a study by Grigos *et al.* (2005), who studied the development of voicing contrast in /p/ versus /b/. They found that variability of lip and jaw movement of 19-month-old children decreased as they began to acquire the voiceless phoneme /p/ in addition to the earlier acquired voiced /b/.

To summarize, the kinematic studies show that "the precision and consistency of movements underlying speech are impaired" (ASHA, 2007, p.3) in children with CAS, more so, or perhaps more specifically than in children with SSD. Thus, there seems to be substantial agreement on the basic underlying deficit. Less clear however, is what the sequelae are, both upstream and downstream in the speech production process. Upstream, phonological abilities and lexical storage are involved. Shriberg *et al.* (2012) consider auditory-perceptual encoding processes as proximal causes of CAS. These processes form the input of phonemic and lexical representations, memory processes that store and retrieve these representations, and transcoding processes for the planning and programming of motor gestures. Downstream effects result in speech sound errors that at the perceptual level are classified as substitutions or distortions. For instance, extreme voicing errors could be considered as a substitution. Such distortions and extreme distortions are similar to those found in dysarthria, and therefore contribute to the confusion about differential diagnosis.

Behavior: speech symptoms

The most accessible, but also the most variable, is the behavioral level, i.e., actual speech production. In the vast majority of cases, clinical intervention starts at this level. A child is referred to a speech-language pathologist (SLP) because parents, other caregivers, or teachers express concern about the observable delays or suspected deviances in speech as compared to their peers. In addition, the diagnostic process takes speech characteristics and symptoms as a starting point. One of the classical issues is whether CAS should be considered as a discrete diagnostic entity that occurs separately from other speech impairments, or if it consists of a symptom complex and should be considered as a syndrome (Guyette and Diedrich 1981, McCabe *et al.*, 1998).

Approaching differential diagnosis from a multi-level perspective, I personally have never really understood the distinction between a symptom complex and a syndrome. At a purely descriptive level, however, the line of reasoning is that, in order to show that CAS is a discrete diagnostic entity, one searches for a diagnostic marker that is present in all cases with CAS and not with other speech difficulties. Alternatively, CAS as a syndrome presumes a complex of co-occurring underlying deficits that in combination underlie the symptom complex. What is never expressed explicitly, but remains implicit, is the assumption that single and specific causes underlie each single speech symptom, and that no interactions between causes and/or symptoms occur. Although the multi-level model sketched above has – I hope – convincingly shown that the same symptom can refer to different underlying deficits, and the same deficit can result in different symptoms, among other factors due to developmental interactions.

This is why studies relying on clinical judgments alone have shown a wide variety of symptoms and much overlap between different SSDs. For instance, McCabe and colleagues (1998) compiled an inventory of features characteristic of CAS (labeled developmental apraxia of speech (DAS) at the time) from a search of the literature, and came up with 30 different characteristics. To test these characteristics, a retrospective file audit was conducted of 50 pediatric clients, who had been referred to the Communication Disorders Treatment and Research Clinic at The University of Sydney for articulatory or phonological impairment of an unknown origin. Nine children were diagnosed with CAS. It turned out that the order of features was highly similar for the nine children with CAS as compared to the total group of 50 children. The top five characteristics were: decreased performance with increased speech complexity; decreased expressive language level for age; multiple articulation errors; slow development of speech skills; and idiosyncratic sound substitutions. In another study, Forrest (2003) asked 75 SLPs to write down their top three characteristics used to diagnose CAS. This list showed little overlap with that of McCabe *et al.* The top five characteristics were: inconsistent productions; general-oral motor difficulties; groping; unable to imitate sounds; and increased errors with increased utterance length. These studies indicate that subjective ratings tend to yield variable results; clearer definition and quantification of diagnostic speech characteristics seem to be needed.

A more focused approach is the search for a diagnostic marker. Although this search has not yielded yet "… pathognomonic symptoms or necessary and sufficient conditions…" (Guyette and Diedrich 1981) to diagnose CAS, it has contributed much to the identification of core symptoms. In a series of papers, Shriberg *et al.* (1997a, b, c) studied different clinical populations of children with suspected CAS as compared to children with speech delay (SD) and adults with acquired AOS. The aim was to identify speech errors that differentiated CAS from SD, and errors that resembled those produced by adults with AOS. In the first paper, they conclude that children with suspected CAS somehow differed from children with SD and took longer to normalize. The authors further noted that, without a diagnostic marker, there is the problem of circularity. How can one find diagnostic characteristics of CAS, if there are no diagnostic criteria available to convincingly identify the subjects of the study? The second and third papers tackled the problem of circularity in the diagnosis of children with suspected CAS as compared to SD. Shriberg *et al.* concluded that there was no simple pattern of segmental errors that formed a solid basis for diagnosing CAS. However, inappropriate stress, which was characteristic of 52 per cent of the children with suspected CAS as compared to 10 per cent of the children with SD of unknown origin, can be considered a diagnostic marker for CAS. Further elaborations of this characteristic resulted in the lexical stress ratio (Shriberg *et al.* 2003a) and the coefficient of variation ratio (Hosom *et al.*, 2004; Shriberg *et al.* 2003b).

Comprehensive studies analyzing segmental error patterns have been conducted by Maassen *et al.* (1997), Thoonen (1998), and Thoonen *et al.* (1994). A unique aspect of these studies was the inclusion of the phonotactic context in the analyses. Phonetic transcriptions of consonants produced in word and pseudo-word imitation tasks revealed overall increased substitution and omission rates for the children with CAS in comparison to children with normal speech. However, the overall profile of errors was not distinctive. Because errors were quantified, it was possible to correct for overall error rate, yielding profiles of relative error rates. While children with CAS produced a higher rate of phoneme anticipations, perseverations, and metatheses (i.e., syntagmatic errors) as compared to controls after correction for the overall higher error rate, the relative number of syntagmatic and paradigmatic errors1 appeared identical for both groups. In general, the error profiles showed very few differences between groups, suggesting that the speech of children with CAS can be characterized by a high rate of 'normal' slips of the

tongue. Even so, the children with CAS showed a particularly low percentage of retention of place of articulation in words, and inconsistency with respect to feature realization and feature preference. These two features, high rate of place-of-articulation errors and inconsistency, stood out as possibly diagnostic for CAS (see, however, Forrest and Morrisette, 1999, for counterarguments).

Clinical practice and future research

From the multi-level, multi-factorial model presented above, a series of conclusions can be drawn for clinical practice. One, CAS is not a categorical but a continuous diagnosis, varying from mild to severe. Second, different profiles of CAS should be found, in which the degree of involvement of each of the speech motor functions contributing to fluent speech can vary. Systematic inventories of such profiles could yield subtypes of CAS, for instance, the subtype a) with poor prosody (inappropriate stress), b) with poor co-articulation (syllabic structure errors), and c) with poor place of articulation (many phonemic substitution errors). Recognizing subtypes, as well as the motor aspects involved in each, yields important guidelines for therapy. Third, co-morbidity might be the rule rather than the exception, largely due to the multiple etiologies. For further discussion of this topic, see Nijland *et al.* (in press).

The developmental perspective leads us to approach CAS not as a dissociated, separate condition, but rather as a deficit with possibly far-reaching consequences for speech and language acquisition. As such, impairments of the developing speech motor control system may impact other, related domains, such as phonological and lexical development and auditory-perceptual functions. Thus, the underlying deficit in children with CAS may express itself in different signs and symptoms over the course of development. In the diagnosis of CAS, exclusion criteria should therefore be applied with great caution. Specifically, if speech motor planning difficulties can result in poor phonological development, then a phonological disorder at the behavioral level is not a valid exclusion criterion for CAS. Furthermore, a developmental perspective predicts changing symptomatology across ages.

A dynamic approach is advocated, both for diagnosis and for treatment. From motor theory we know that the acquisition of motor plans and programs requires much practice. The reason is that motor planning and programming are skill-specific: practicing the articulation of English, does not contribute to speaking Chinese, just as practicing playing the piano does not help much in playing the violin. [Note that different learning principles underlie the acquisition of the vocabulary and syntax of languages, as well as the fundamentals of music, as compared to motor learning.] Motor training therefore needs to be adapted to the developmental stage, as expressed in the motor patterns acquired thus far, to other cognitive functions, like memory and attention, and to higher-level language functions. An important aspect is that articulatory movement patterns are not acquired in a vacuum but can be related to meaningful lexical items, so that they can be anchored in lexical memory.

A prerequisite for dynamic diagnosis and treatment is a process-oriented approach. As was discussed above, the speech symptoms at the behavioral level are not transparent regarding the underlying processing deficit. The assumption is that treatment of processes is more effective and yields better generalization than treating symptoms. Recently, we started a process-oriented line of research that applies experimental techniques to directly manipulate speech production processes for dynamic assessment. For instance, the hypothesis that the speech production process of a child with CAS is characterized by overreliance on auditory feedback (Terband, 2011), could be tested in a speech task with auditory masking by presenting the child noise over headphones. If the speech becomes much poorer, then it can be concluded that, indeed,

auditory monitoring plays a role in normal speech production. If the speech barely deteriorates, apparently online feedback is not invoked. Dynamic treatment means that treatment results are continuously assessed and the treatment program is continuously adapted. This requires a dynamic integration of speech motor and phonological treatments.

Notes

1 As defined by Thoonen *et al.* (1994), syntagmatic errors are related to context, such as anticipations, perseverations, and transpositions; paradigmatic errors are speech sound errors of place, manner, or voice, not induced by the context.

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