Chapter 7

Comorbidity of Motor and Sensory Functions in Childhood Motor Speech Disorders

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Additional information is available at the end of the chapter

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Abstract

Subtypes of speech sound disorders (SSDs) with a sensorimotor origin are known as motor speech disorders (MSDs). The symptoms can be diverse, and the causes of the disorders in children are in many cases unknown. Examples of MSD are childhood apraxia of speech and dysarthria. MSD is often seen in neurodevelopmental disorders such as cerebral palsy, developmental coordination disorder (DCD) or autism spectrum disorders (ASD), or it is seen with no obvious diagnosis but usually with comorbid problems. Within all existing comorbidity dysfunctions, the motor and sensory systems are of interest for identifying possible underlying mechanisms of MSD. Namely, soft neurological signs such as hypotonia, decreased speed and low accuracy of motor skills and delayed motor development are given consideration by many researchers for better understanding of underlying motor mechanisms of MSD. Results from comorbidity studies highlight the relationship of MSD with complex sensorimotor tasks and sequential motor tasks. In this chapter, our aim is to frame findings from studies about comorbidity of sensory and motor dysfunctions in MSD in order to theorise affected mechanisms and propose an underlying global motor deficit. We will conclude with implications for therapy models.

Keywords: comorbidity, speech, manual, childhood apraxia of speech, dysarthria, motor function, sensory processing, sensorimotor, motor learning

1. Introduction

Childhood motor speech disorders (MSDs) are covered under speech sound disorders (SSDs) as described by Bowen in **Figure 1** [1] and are commonly identified by problems in speech sound production originated from dysfunctions in sensorimotor processing. The causes of the disorders are in many cases unknown, and the symptoms are diverse, though there is an ongoing search for diagnostic criteria that can help differentiate between different speech disorders in children addressed by several authors [2–9]. Based on genetic and environmental risk factors, Shriberg et al. [9] defined three types of MSDs in their classification system. These are (1) Motor Speech Disorder-Apraxia of Speech (MSD-AOS), (2) Motor Speech Disorder-Dysarthria (MSD-DYS) and (3) Motor Speech Disorder-Not Otherwise Specified (MSD-NOS). Dysarthria is defined as a motor speech disorder that results from a central or peripheral damage of the nervous system and is characterised with impaired execution and motor control of the muscles used for speech production, including the lips, tongue, vocal folds and/or diaphragm. There are several types of dysarthria depending on neurological localisation: *flaccid*, *spastic*, *ataxic*, *hypokinetic and hyperkinetic.* There can also be a mixture of types [10–12]. Childhood dysarthria is often defined as developmental and/or acquired; thus, dysarthria can also be 'idiopathic' without the influence of neurological damage [12, 13]. Childhood apraxia of speech (CAS) is defined as a motor speech disorder that is characterised by problems in planning of the movements (e.g. praxis of the lips, jaw, tongue and vocal folds) needed for speech [14]. The motor speech symptoms can intertwine and be mixed with perceptual and phonological dysfunctions, which makes it difficult to define exact diagnosis, and there are probably subgroups and definitions not yet found [1, 9].

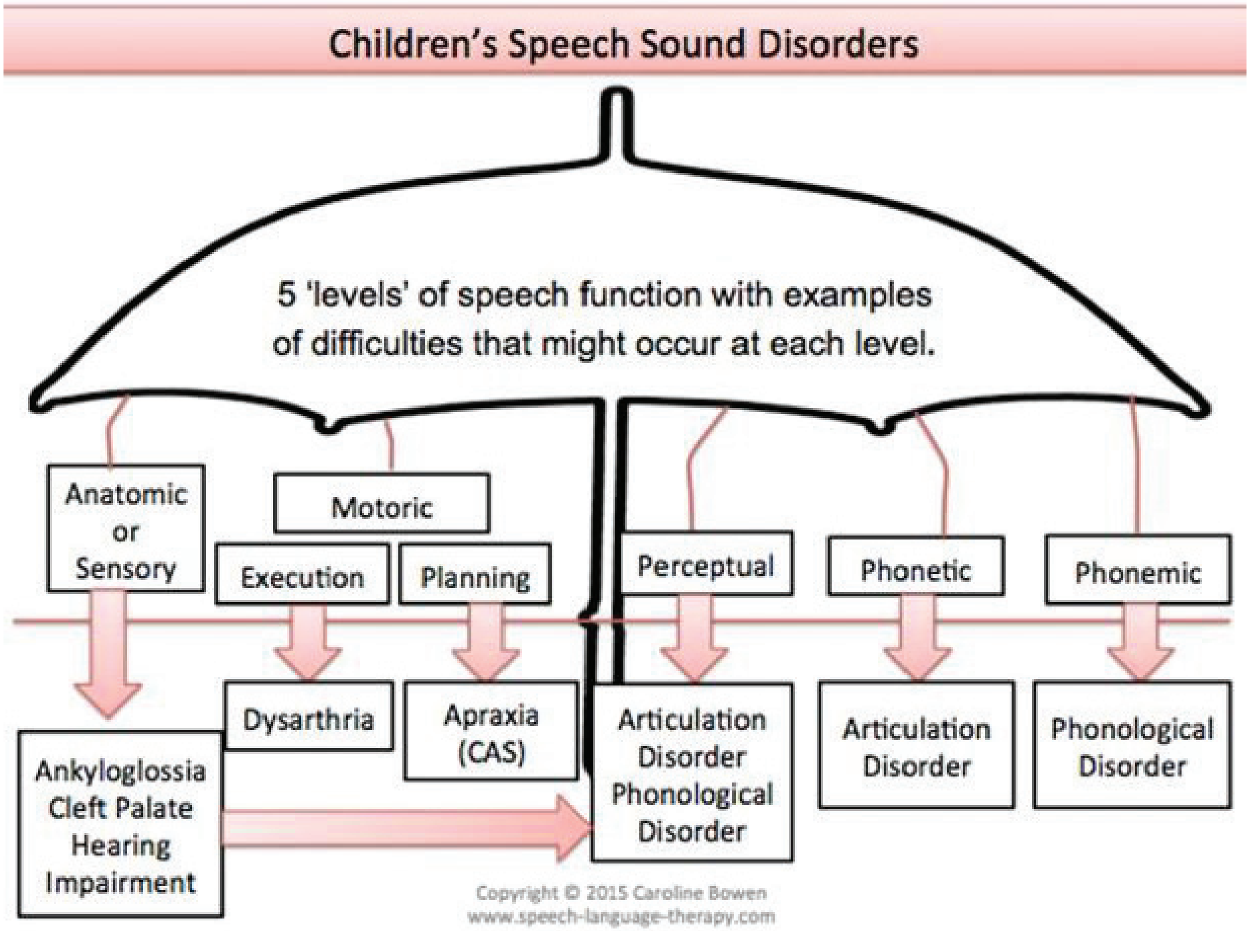

Figure 1. Five levels of speech function with examples of difficulties that might occur at each level [1] (Retrieved from www.speech-language-therapy.com/index.php?option=com_content&view=article&id=45 on 01.03.2017. Used by permission).

Comorbidity studies highlight the relationship of MSDs with dysfunctional sensory processing, complex sensorimotor tasks and sequential motor learning tasks as well as fine motor problems [15–17]. Findings also suggest some common neural correlates of speech and fine motor functions [18]. Soft neurological signs such as hypotonia, decreased speed, low accuracy of motor skills and delayed motor development have been considered by some researchers for better understanding of the motor origin of MSD [13, 17, 19]. MSD can also be part of symptomatic picture in specific neurodevelopmental disorders, genetic conditions or metabolic syndromes. Some but not all of these are cerebral palsy (CP), developmental coordination disorder (DCD), autism spectrum disorder (ASD), velocardiofacial syndrome and galactosemia. For example, dysarthria sometimes occurs in the context of CP. Isolated form of MSD seems to happen very rare; more often it occurs with subtle sensory, motor and cognitive problems. Hence, co-occurrence of subtle problems in MSD complicates the understanding of the disorder and planning of the intervention. In this chapter we will present our findings in MSD population with an integrative framework that draws on previous research including behavioural and neuroimaging studies as well as major motor control and learning theories. The main MSD diagnosis we present is CAS because more research findings exist and we have also conducted our previous study in this group [17]. It is important to mention that in clinical work we find that symptoms of 'idiopathic' dysarthria can be misinterpreted being symptoms of CAS or another developmental oral dysfunction due to similarities in speech deviances, for example, vowel distortions, nasality, imprecise consonants, rate control and abnormal prosody. In differential diagnosing of pure CAS from dysarthria, it is important to verify that the child has all three features that were presented and considered to have diagnostic validity in the technical report by ASHA [14]: (1) inconsistent error production on both consonants and vowels across repeated production of syllables and words, (2) lengthened and impaired co-articulatory transitions between sounds and syllables and (3) inappropriate prosody. The group of children with 'idiopathic' dysarthria has not yet been researched enough; therefore, we have chosen to broaden the view with inclusion of some examples from speech and language impairment (SLI) and neurodevelopmental disorders (NDDs). We have tried to dig in to co-occurrence of subtle or severe sensory and motor problems and shed light to this 'grey zone'. With support in our findings, we propose an underlying sensorimotor programming and motor learning vulnerability affecting more than one motor area and the presence of a global motor deficit.

2. General motor development and motor learning

Motor development is a biological process where genes and environmental factors interact. Children are born with a specialised repertoire of preprogrammed non-volitional movements, and during the first months of development, the newborns' interaction with the environment leads to integration of these motor patterns/schemas. The motor schemas become the foundation for development of volitional movements [20–22], and most of the skills that are unique to human beings such as speech or drawing can therefore be learned via practice implicitly throughout lifetime. Stages of motor learning, proposed by Fitts and Posner [23], are the *cognitive phase*, the *associative phase* and the *autonomous phase*. In *cognitive phase*, the child is new to the task and tries out different strategies to attain success. This period is characterised with trials and errors. While strategies leading to targeted action are retained, strategies leading to failure are discarded. This stage is very important for the development of sensory feedback and feedforward processes that eventually lead to the achievement of desired motor control in muscular organs. Here, muscle tone is an important aspect, and the contractibility thereby exertion of necessary force by muscles is essential for motor control. For example, hypotonia influences the ability of individual muscle groups to contract, and infants with low muscle tone put great effort into holding their heads up where oral-motor functions such as sucking can be affected as well as speech development [24]. The *associative phase* is characterised by refinement of the attained motor skill in terms of rate, consistency and energy expenditure. Further practice results in the *autonomous phase* where automatic motor response has small or no error and that defines the last phase of motor learning. Cognitive effort during motor learning decreases throughout the phases making *autonomous phase* the least cognitively demanding [25]. Typically developing children continue to learn motor skills almost in an effortless fashion despite the increasing complexity of the motor tasks as the child grows [26]. For children with sensorimotor dysfunctions as in MSDs, development of complex movements as in speech is somehow disrupted, and motor learning is highly affected by the nature and severity of the impairment [27]. More recent research has emphasised the importance of functioning cells in the 'mirror neuron system' being part of learning and execution of new sensorimotor skills as an interesting topic concerning MSDs [28].

3. Neural correlates of speech, fine and gross motor functions

During the last decade's computer models of brain function and technically advanced neuroimaging, studies have exploded as well as thorough literature on the topic [29]. Research has mainly drawn conclusions of which parts of the brain are involved during speech based on adults. Liegeois and Morgan [30] emphasised the lack of research computed on children and conducted a review searching for which kind of brain abnormalities children and adolescents with severe speech disorders had in developmental, progressive or childhood-acquired conditions. They found evidence that there is predominantly malfunction of neural networks bilaterally involved in severe childhood speech disorders not a leftward cortical dominance, which has often been reported in adults. The main brain regions affected in the children with apraxia of speech and dysarthria were the basal ganglia, thalamus, cerebellum as well as perisylvian and rolandic cortices. They do also suggest that their data show that neuroanatomical basis for speech production also to some extent overlaps in children and adults. One study using functional magnetic resonance imaging comparing children with DCD with typically developing peers demonstrated an under-activation in cerebellar-parietal and cerebellar-prefrontal networks and in brain regions associated with visual-spatial learning suggesting negative affects in learning motor skills [31]. Most of the studies we present below are made on an adult population, and for a child's brain, considerations on developmental and critical periods as well as plasticity should be taken. Before dominance between hemispheres is established, many sensorimotor tasks are processed bilaterally. For example, in childhood MSDs, the effect of bilateral structural anomalies results in functional reorganisation of cortical

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speech networks that is different from an adult population [32]. We have tried to highlight brain regions such as the cerebellum and cerebral cortex and functional connections in between that have been reported to relate with speech motor and visual-motor functions. We find cerebellum being one of the most interesting parts tickling our curiosity.

The cerebellum is most understood in terms of its contribution to motor control and learning, balance, posture and muscle tone; however, it is also involved in cognitive functions especially related to language. It consists of approximately 10% of the brain's volume; on the other hand, it contains over 50% of the total number of neurons in the brain showing the complexity of neural computations for its role in motor systems [33]. Motor commands are not initiated in the cerebellum; rather, the cerebellum modifies the motor commands of the descending pathways to make movements more adaptive and accurate via its functional subdivisions [34]. Neural activity within the cerebellum and also basal ganglia has found to be highly connected with motor areas for movements in the cerebral cortex and areas of prefrontal cortex related to cognitive functions [35]. Cerebellar subdivisions that are of interest for commonality of speech, fine and gross motor functions are the spinocerebellum and the cerebrocerebellum. While the spinocerebellum is involved in integration of sensory input with motor commands to produce adaptive motor coordination, the cerebrocerebellum, the largest functional subdivision with its extensive connections with cerebral cortex, is involved in the planning and timing of movements and the cognitive functions [33]. In 1998, Schmahmann and Sherman [36] defined the cerebellar cognitive affective syndrome (CCAS) in a group of 20 adult patients with disorders confined to the cerebellum. They found that not all deficits occurred in each patient, but decreased verbal fluency and visual-motor problems were particularly prominent, which was also replicated by others [37]. This is consistent with the outcome of our examination [17] of overall motor functions in 18 children with CAS where similar symptoms were found as in the group with CCAS though the children with CAS did not have any identified cerebellar dysfunction or lesion. Almost all the children had fine motor problems that required visual-motor integration, and an intriguing finding was correlation between visual-motor control, for example, drawing skill, and speech motor control which supports the hypothesis that cerebrocerebellar interconnections are involved in underlying speech motor control and complex eye-hand coordination tasks like drawing. The CCAS is described as a constellation of deficits affecting linguistic, visuospatial and executive functions. Similarly, in CAS, problems in executive functions are usually observed. However, not enough studies have explored the possible role of the cerebellum in apraxic disorders especially in CAS; the pathophysiological mechanisms remain to be elucidated [18].

The *cerebral cortex* has been mapped for functional regions in neuroimaging studies. For speech motor production, some models have been raised discussing functional networks instead of functional regions [38, 39] as it also was proposed for visual-motor functions [40, 41]. The neural correlates of speech motor control include the premotor and primary motor cortex, somatosensory cortex, auditory cortex, cerebellum and the basal ganglia as mentioned before [39]. Grossly similar regions were described as part of the so-called dorsal and ventral stream for visual-motor functions [40]. Brodmann area 6, which is located in premotor cortex, has been shown to be activated bilaterally during speech listening as well as speech production [38]. The same area was proposed to be involved in dorsal visual stream that is responsible for planning of visual-motor tasks [40]. Another cortical region that is of interest for communality between speech motor and hand motor functions is posterior parietal cortex (PPC). PPC receives cortical afferents from visual, auditory and somatosensory areas and projects mainly to frontal premotor areas [42]. Its role in planned movement execution has been mostly shown in visual-motor integration tasks [43–45]. However, it was also shown to be activated during auditory-motor integration tasks [46, 47]. Searching for the commonality of speech and hand motor learning, the supplementary motor area (SMA) and pre-supplementary motor area (pre-SMA), parts of medial premotor area, have been related to the learning of motor sequences. Both of them are shown to be sensitive to the order of movement elements within a sequence [48]. The pre-SMA is found to be important during the initial stage of motor learning [49] and when adapting to a new sequence [49, 50]. The SMA activation, on the other hand, increases with practice [51, 52]. It was proposed that pre-SMA is related to temporal control and the SMA integrates this temporal coding with the required motor output [53]. Recent studies report that, in addition to speech-related activation, Broca's area is also significantly involved during tasks devoid of verbal content. It has been shown that Broca's area is responsible for hand actions as well as integration of speech perception [54, 55].

3.1. Summary

Cortical and subcortical areas including the basal ganglia as well as the cerebellum are involved in complex sensorimotor functions like speech and fine motor tasks. Supporting studies in cognitive neuroscience propose that functional networks of speech motor and visual-motor functions do overlap especially in the parietal cortex, cerebellum and basal ganglia, where the cerebellum plays a central role in the emergence to be discovered. Highly skilled movements including speech and handwriting are unique for human beings, and there are still many question marks on how the brain and body interact concerning both typical development and when we meet dysfunctions. Here, we have tried to show that human motor functions are interacting with each other neurologically, and we hope for an interesting future research to come.

4. Comorbidities in motor functions within MSD and SLI

4.1. Background

Specific fine and gross motor milestones and locomotor skills such as head control, rolling, sitting, crawling, walking and pincer grip, should be reached by a certain age [22, 56–58]. The problem in different motor domains is commonly pronounced areas that accompany speech and language disorders, and the development of speech is associated with the ability to manoeuvre hand actions from the babbling stage and onwards [55]. Handling objects with vision, touch and speed of processing interact with the emergence of certain perceptive and cognitive abilities and also influence motor development [59, 60].

4.2. Motor functions in children with SSD and MSD

A few studies exist more or less recently exploring the possible correlation between oral motor, speech motor and fine and gross motor functions. Dewey et al. [61] found that children

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with CAS showed difficulties in sequencing both limb and orofacial gestures compared with children that had phonological disorders and discussed possible problems with general praxis. (Praxis involves motor programming and motor integration that is required to be able to execute complex learned and skilled movements.) General praxis dysfunctions were also revealed in a study by Poole et al. [62]. They compared children with developmental dyspraxia and adults having acquired apraxia, and both groups made similar movement error patterns when imitating oral and body gestures. Newmeyer et al. [15] evaluated fine motor skills, object manipulation, grasping and visual-motor integration in 32 preschool children between 2 and 5 years of age with severe speech sound disorders using Peabody Developmental Motor Scales. They also measured the ability to imitate oral-motor movements with Kaufman Speech Praxis Test. All children were lower than average in fine motor skills, and significant results was found between oral-motor imitation and visual-motor integration. (Visual-motor integration tasks can include measurements of precision in skills such as drawing, cutting or folding and drawings of geometric shapes.) The studies presented above show similarities in dysfunctional praxis of oral and limb functioning amongst children with CAS covering three decades. Even though there has been an evolution in assessments and diagnostic criteria for children with CAS and also NDD, an interesting concern is that united assessments and diagnostic markers covering both manual and oral/speech dyspraxia have not yet been invented as far as we know. An approach concerning these aspects could be beneficial in understanding the challenges children with CAS carry and in the development of therapies.

Dodd [19] looked for possible co-occurrence of deficiencies in oral-verbal and fine motor skills and compared speech-disordered children with a typically developed control group on tasks assessing volitional and non-volitional oral movements, fine motor skills and speech motor planning. The children (n = 51) were divided into five groups depending on which type of speech dysfunctions the child had. In comparison to other speech impairments and to the control group, the children with inconsistent speech errors but not diagnosed with CAS (n = 9) were poorer than controls in fine motor movements involving speed and dexterity. (Dexterity tasks can include specific activities such as reaching, grasping and bimanual coordination by using small objects, where accuracy in time is measured.) The children that were diagnosed with CAS (n = 5) had the same results as the previous group on speed and dexterity but also performed poorer on fine motor tasks involving deficiencies in integrating sensory information into a plan of actions such as visual-motor integration. Our own study [17] has similar findings as previous studies and shows a variety of motor problems that are moderately severe. The study involved 18 children with CAS between 4 years 5 months and 10 years 7 months (**Figure 2**). All the children suffered from a limited repertoire of speech sounds, inconsistent articulation of vowels and consonants and vowel distortions also affecting prosody. We measured correlations between three tests of the Verbal Motor Production Assessment for Children: *focal oromotor control*, *sequencing maintenance control* and *connected speech and language control* [63] and five tests from the Bruininks-Oseretsky Test of Motor Proficiency [64]: *overall body motor coordination*, u*pper limb coordination*, *manual dexterity*, *fine motor precision* and *fine motor integration.* The number of motor problems was investigated by using −1 SD as a cut-off point on the different subtests. Looking at **Figure 2**, a mosaic profile of motor problems is shown. Co-occurrence of non-speech, speech, manual and gross motor problems is seen in 15 of the 18 children (83%), and each child demonstrated problems in 2 of the 7different tests. Fourteen children had results lower than average in fine motor tasks including visual-motor integration. Correlations were found between *fine motor integration* and *focal oromotor control* which include non-speech and speech oral-motor movements (r = 0.54) and also with *connected speech and language control* (r = 0.51). The findings can be compared to a study by Rodger et al. [65] where the children with DCD performed lower than −1 SD on the *focal oromotor control* and *global motor control* (including oral reflexes) from the Verbal Motor Production Assessment for Children. Our study also gave interesting findings in the oral-motor area with significant results showing that the children had more deviant production of mandibular and lingual non-speech movements compared to non-speech labio-facial movements (p < 0.005) which is intriguing in the context of what Nijland et al. [66] found in their study with bite blocks. In that study, the children with CAS could not compensate their articulatory movements when the mandible was fixed in a bite block leading to more increased speech errors. No speech errors were made by typical developed children and adults performing the same exercise. The researcher established that they found evidence for an underlying motor programming deficits in children with CAS. Ho and Wilmut [67] also suggest a possible underlying motor deficit based on findings of atypical patterns of movement and inferior motor control of complex speech gestures in children with DCD.

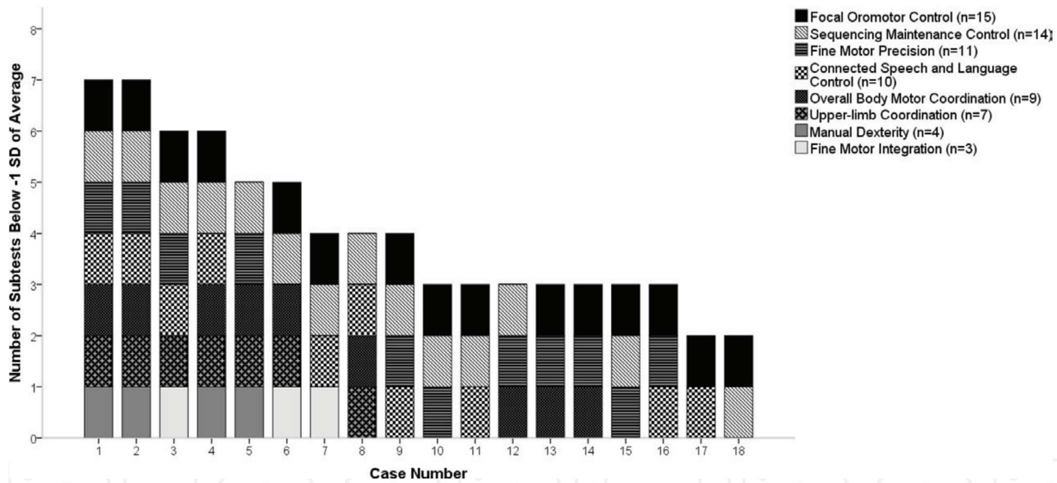

Figure 2. Co-occurrence of non-speech, speech, fine and gross motor problems (below −1 SD of average) in children with CAS. Each block presents one case in the group. Blue boxes are for problems in fine motor or overall body motor functions. Red boxes are for problems in speech motor functions. Only Cases 1 and 2 show pure speech motor problems, whereas other 16 cases show variety and different numbers of motor problems.

4.3. Motor functions in children with SLI

There is a rather large body of research on comorbidity of motor functions in children with SLI. The prevalence of motor disorder in children with SLI was found to be 18 out of 40 children where 12 out of the 18 had the diagnosis of DCD [68]. Similarly, Webster et al. [69] found that 52% of children with SLI had motor impairments. An exhaustive review of the literature on comorbidity between SLI and motor skill was conducted by Hill [70]. The review gives clear and substantial evidence that children with SLI had poorer fine and gross motor

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functions, poorer coordination as well as praxis functions than typical peers. Many of the children had similar symptoms as those seen in other neurodevelopmental disorders such as DCD. She suggests that SLI is part of a broader range of challenging symptoms where comorbidity of motor functions is one. More recent studies on this concept reveal similar outcomes as described above. Marton [71] found that children with SLI had poorer ability in imitating gross motor body postures, and in a study by Zelaznik and Goffman [72], the children had lower results in gross and fine motor skills both compared to age-matched peers. Very recent studies found that children with SLI have bimanual coordination dysfunctions and poor visual-motor sequencing learning correlated with lexical abilities [73, 74]. Also, children with mild expressive language disorder showed lower performance in manual dexterity, ball skills and static and dynamic balance [75]. This topic has been lifted in research for many decades, and in 1987 Bishop and Edmundson [76] described the poorer motor functions in children with SLI as a lag in maturation.

4.4. Comorbidity of speech and motor functions in children with NDD

NDDs such as CP, ASD, ADHD and DCD which are also to be elucidated with genetic causality usually affect more than one domain of development [77]. Even though the most prominent symptom is different in each, all of them present variety of symptoms in sensory, motor and cognitive domains of development. MSD can be one part of symptomatic picture in these specific neurodevelopmental disorders. Children with CP have delayed skills in communication related to severity in gross motor functions, and the severity of motor functions was also related with language dysfunctions and speech problems primarily in the motor speech domain [78, 79]. Children with ASD show disturbances in body movements and a non-natural shape of the mouth seen as early as at the age of 4–6 months [80], and it is common with a delay in motor development giving impact on never-reaching gross motor milestones [81]. Gernsbacher et al. [82] found a relationship between the development of oral and manual skills amongst toddlers and later dysfluency of speech in the children that was diagnosed with autism. The study was conducted in three steps: a caregiver landmark-based interview targeting motor skills, historical home videos confirming the information from step one and oral-motor assessment from the Kaufman Speech Praxis Test. The children were divided into two groups: one with minimal speech fluency and one with high speech fluency. The children with less fluent speech performed overall poorer in manual skills as well as in oral-motor items and had less capacity for communication with gestures which indicates that there is a deviant oral and manual development in children with autism giving impact on speech-language and communications skills. Authors suggest that the results are not a failure in understanding or lack of compliance based on the notion that the children succeeded performing some items. Diffuse symptoms including dysfunctional speech and motor skills have the last decade being discovered secondary to genetic disorders that often are very rare. One recent study found speech features of CAS amongst children with 16p11.2 deletions. Some of the children also performed low on fine and gross motor functions though there were large variations between the children in the motor function part [83]. We have presented a small range of articles in the NDD domain because the comorbid symptoms are well known amongst clinicians; nevertheless, it is important to highlight the challenges children with NDD can meet.

4.5. Summary

Comorbidity of general motor functions, speech motor and language skills is very common in children having interventions at SLP clinics. For some children the symptoms are severe and can be detected easily as often for children with NDD which can lead to therapy programmes from different professionals, physiotherapists, occupational therapists, psychologists, etc., sometimes working in teams. With children who have atypical patterns of development, the challenge for clinicians and educators is more camouflaged. Delayed or inappropriate speech production during preschool age is often the most visible problem but is frequently accompanied by unusual or clumsy fine and gross motor functions that can be considered less important to handle. However, when children reach school age, these subtle fine motor problems can lead to functional problems in academic skills. These children are somehow located in an undefinable 'grey zone' where not much of interventions for their entire dysfunctions exist. The question is what should be done for this type of children?

5. Comorbidities in sensory processing functions within MSD and SLI

5.1. Background

The sensory system is the foundation that makes it possible for our motor programmes to execute, coordinate and plan functional movements. The sensory system converts the environment into neural signals which the motor system through neural signals turns into muscle force [34]. Our abilities to extract an action relevant to the estimated goal rely on the ability of the nervous system to process the sensory information [84]. The brain decodes multisensory organs of our bodies, smell, taste, auditory, visual, touch, proprioceptive and vestibular, and serves us in every moment of life. If the ability to process sensory information is out of tune, it can turn into dysfunctional behavioural patterns [85, 86]. Children coming to SLPs often show different kinds of behaviours and have needs that have to be handled by the clinician. Some children have difficulties sitting on the chair due to restlessness or fatigue which could be caused by hyper- or hypotonicity of postural muscles; are easily disrupted by sudden sounds or visual objects in the room; are being sensitive to auditory and visual disturbance; are anxious and want to withdraw from the situation or have short intervals of concentration span. Dunn has proposed a conceptual model for how sensory processing functions affect daily life and the abilities to learn and develop new skills. The Sensory Profile Questionnaire [87] is a 125-item questionnaire for caregivers and school staff assessing a child's function in daily life. Different aspects of sensory functions are grouped into three main areas: sensory processing, sensory modulation and behavioural and emotional responses. Sensory processing measures the response and abilities in auditory, visual, vestibular, touch, multisensory and oral sensory systems. Sensory modulation refers to the ability to regulate sensory input in terms of facilitating or inhibiting responses and behavioural and emotional responses refer to the behaviours that the person shows as a result of abilities to process sensory information. Authors in the past have found relationship between sensory processing, speech production and language acquisition are Ayres [88], Ayres and Mailloux [89] and Fallon et al. [90] amongst others.

5.2. Sensory processing functions in children with MSD

This topic has not yet been vastly explored. The one and only existing article performed on children with CAS is by Newmeyer et al. [91] where the Sensory Profile Questionnaire was used for 38 children with severe CAS. The results reveal significant differences in several sensory processing areas compared to typical developed peers. The children with CAS had difficulties processing sensory information through their auditory, visual, tactile and multisensory systems. Differences were also seen in areas of modulation of movement affecting activity levels and sensory input. Factor clusters regarding oral sensitivity, fine motor/perceptual, emotional reactivity, sensory seeking and inattention/distractibility had strong significant differences compared to typical peers. The results from the Sensory Profile Questionnaire were also compared with Kaufman Speech Praxis Test which revealed significant positive correlations between oral-motor apraxia, abnormal imitations of oral movements and abnormal repetition of simple phonemes compared with sensitivity. Nijland et al. [16] found that complex sensorimotor and sequential tasks correlate significantly with the severity of the speech impairment in children with CAS indicating a comorbidity in non-verbal sequential functioning.

5.3. Sensory processing functions in children with SLI

In children with SLI, there has been some research connected to sensory processing. Taal et al. [92] found through the Sensory Profile Questionnaire that 116 children with SLI confirmed atypical behaviours in the sensory processing domains such as auditory processing, touch processing, vestibular processing, oral processing and visual processing compared to peers with neurotypical development. This interferes with normal development, and the effect generates multiple impairments in functions needed for speech and language learning. Sensory processing abnormalities also seem to negatively affect children with SSD not making the same progress in speech therapy compared to children with SSD without sensory processing disorders [93].

5.4. Sensory processing functions in children with NDDs and others affecting speech and language development

One recent article on sensory processing functions in children with CP found differences in 16 out of the 23 categories of sensory processing compared to typical developed peers evaluated by the Sensory Profile Questionnaire. The author accentuates the importance in sensory processing abilities for the development of motor and communication skills and functional behaviours that support in daily life activities [94]. Research on toddlers that suffered from non-organic failure-to-thrive and feeding problems found atypical performance in sensory processing areas of tactile, vestibular and oral compared with a control group. The children that had abnormal sensory processing in one or more area also showed delayed development in cognition, motor skills and language [95]. Atchison [96] has observed speech and language dysfunctions amongst children with sensory modulation disorder due to traumatic histories including adoption, foetal alcohol syndrome and abuse. He stresses that it is essential for SLPs to recognise the behaviours of children with a history of emotional and physical traumatic stress and how this affects their learning of speech and language.

5.5. Research on auditory and somatosensory processing

Through neurophysiological investigations, researchers have analysed sensorimotor commands behind speech and deviant speech. For example, Terband and Maassen ([97], p. 139) proposed that reduced oral sensitivity could be a core deficit in children with CAS. They discuss in a hypothesis that 'the lack of auditory feedback control is predicted to affect speech production more if the motor commands are inherently deviant/unstable (as is the case with degraded somatosensory information) since this can no longer be compensating by (over) reliance on auditory feedback'. Also in a later study computed with simulations in the DIVA model, Terband et al. [98] found that a combination of motor programming functions and the ability in auditory self-monitoring/auditory feedback affects to which extent phonemic representations develop on a phonological level. They hypothesise that deficiencies in these sensorimotor programmes can be part of the symptomatic picture in paediatric motor speech disorders. In another experimental study, the researchers claim evidence that somatosensory feedback which corporates proprioception has a central role in achieving the precision needed for acquisition of speech movements for both consonants and vowels independent of auditory feedback [99]. This gives strength to the previous study mentioned by Terband et al. [97] that oral proprioception, 'to read' somatosensory information, can be lacking in children with CAS. Other neurophysiological researchers have been interested in the relationship between sensory processing of auditory input and the ability to produce speech sounds in children with CAS. In a study by Groenen et al. [100], the children with CAS performed poorer than the control group in auditory discrimination of consonants. They also found a relationship between the frequency of substitutions in articulation and the degree of auditory processing deficit within some children. Studies have also showed that negatively affected perception of vowels in children with CAS indicated phonetic processing dysfunctions [101] and significant results were found between perception and performance [102].

5.6. Summary

Children with speech and language disorders process sensory information in a different way than typical peers which impact on their ability to learn and develop adequately and can also affect their behaviours in daily life. Oral-motor apraxia, abnormal imitation and oral movement correlated with increased sensory sensitivity in children with CAS, and one hypothesis is that deviant proprioception affecting somatosensory feedback could be a core deficit. It is of immense importance that SLPs working with children understand behaviours and demands that occur due to abnormal sensory processing and deeper knowledge in this area can support the choice of treatment. It seems that sensory processing dysfunctions amongst children affect their ability to develop speech, language and communication skills and also their quality of life [91–95].

6. Therapy models for children with MSD

6.1. Background

Children with MSDs often need to continue longer in therapy than for other types of articulatory speech dysfunctions. Patients usually have to exercise almost every day for several years in order to achieve results [103, 104]. This places high demands on both children and their caregivers. There are scarce with evidence that support treatments aimed for CAS and for dysarthria. There are some important reports available on intervention approaches that also discuss the outcomes and evidence of today's existing treatments [104–106].

6.2. Treatment with oral-motor exercises

Traditionally, treatment with oral-motor exercises has frequently been used as part of assessments and intervention programmes for children with MSD. The intention is to increase/decrease movement, relaxing/strengthening muscles, increase proprioception and increase/decrease dysfunctions in the sensory system of the lips, tongue and jaw to reinforce speech development. Oral-motor treatment is an umbrella term for different treatment models used for motor speech, feeding, orofacial myofunctional, oral awareness/discrimination and oral activities/exercises [107]. The application of non-speech oral movements (NSOMs) in treatment for speech has been questioned in several reports due to the absence of scientific research [108, 109]. Kent has in this debate contributed with a sober evaluation of the literature and with a very valuable summary of oral and speech muscle functioning [105]. And, Ruscello [110] gives credibility to clinicians to use their clinical judgement, proven experience and common sense together with empirical evidence [111]; indeed, more research is needed. As mentioned by other authors, not many studies exist where the effect on speech is measured after intervention with oral-motor exercises. The studies are mainly on children with dysarthria secondary to CP. Ray [112] examined speech intelligibility using orofacial myofunctional therapy (OMT) programme on 16 children with spastic CP and myofunctional disorder. OMT is a training programme for the tongue, lips and jaw muscles facilitating adequate posturing and function. After a 4-month therapy with OMT, speech intelligibility improved in words, and significant correlation was found between tongue functioning and speech intelligibility. Another study showed improvement of speech in some of the children with CP using orthodontic stimulatory plates together with oral and facial physiotherapy treatment according to Castillo-Morales orofacial regulation therapy. This therapy is based on the understanding that orofacial muscular functions are closely interlinked with postural control and breathing mechanisms [113]. Two studies on children with mild articulation disorder showed no significant improvements on speech output after the use of oral-motor exercises [114, 115]. No study on children with CAS has been found. Maybe the following proposition by Kent ([105], p. 777) can serve as a guideline in the search if oral-motor exercises ought 'to be or not to be'? 'To be useful, NSOMs may need only to resemble the target behaviours in some fundamental respect that can be used to modify a motor response, whether by strengthening or altering the basic motor pattern'.

6.3. Treatment with strategies based on principles of motor learning

Principles of motor learning (PML) strategies have gained ground in the last decade especially concerning treatment for CAS. PML is partly based on the target movement that is produced with high frequency (up to 100 times) in various conditions to support motor learning. When PML is incorporated in speech treatment, the target sounds are produced through modelling or being modelled by the facilitator (clinician/caregiver); successively, the facilitator reduces the support, all performed after a template. The goal is also to keep a standard of high motivation which can be supported by reinforcements [27, 116]. Murray and colleagues [104] found that two treatment programmes using PML aimed for CAS had suggestive or preponderant evidence, Integral Simulation/Dynamic Temporal and Tactile Cueing (DTTC) and Rapid Syllable Transition Treatment (ReST), with 14 children in each study. In a later study by Murray et al. [117] on a larger population of children with CAS comparing two treatments, effects were shown using ReST and the Nuffield Dyspraxia Programme-Third Edition (NDP3). CAS is a rare diagnosis, and at the clinic, the children often have co-occurring symptoms with other motor-based diagnoses including dysarthria features [13, 103], and a few single case studies on CAS inform that the children also were diagnosed with dysarthria [118–120]. In these studies, they performed an intervention with DTTC which showed less progress in some cases with mixed diagnoses though the studies did not problematise if the neuromuscular dysfunctions of dysarthria made an impact on the outcome. All the PML studies presented above had positive outcome on treated sounds and words which seems to be the biggest achievement this treatment strategy gives to children with CAS [121]. Less effect is noted on non-treated words, intelligibility and speech fluency though most studies did last for a fairly short period of time and acquisition of the new skills can prolong [105]. Pennington et al. [106] made a review on speech treatment for children with developmental dysarthria including the use of PML strategies. Pennington found that a few small observational studies had positive results on intelligibility and clarity of voice which gave impact on the children's participation in social and educational activities and quality of life. Also, Lee Silverman Voice Treatment (LSVT LOUD) works in the context of motor learning and activity-dependent neuroplasticity. Part of the goal is to strengthen muscles orally and for breath support to facilitate speech output. Research has shown positive outcomes indicating development of vocal loudness and intelligibility in children with dysarthria secondary to CP [122, 123].

6.4. Treatment strategies based on multisensory input

Motor learning principles are also involved in methods which try to incorporate input from several sensorimotor processing areas. Some are used for many decades, and others are emerging.

6.4.1. Multisensory cues for muscles used in speech

The Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) approach indicated that the use of multisensory input through auditory, visual and tactile-kinaesthetic-proprioceptive cues triggering specific facial muscles for each phoneme leads to a greater impact on speech development compared with children lacking sensorimotor input. In recent studies with children that had motor control issues as part of their speech disorder such as jaw sliding, jaw over extension and inadequate lip rounding/retraction, the PROMPT treatment showed to have effect on treated and untreated words, development of speech sounds and speech motor control [124–128]. PROMPT also seems to support positive development of motor speech movement patterns and intelligibility for children with CP [129, 130]. Another fairly new treatment method using biofeedback has been investigated in children with CAS. In biofeedback there is visual feedback with real-time ultrasound images of the tongue. Development of their ability to produce some of the practised target sounds was found, and a transfer into word production was also seen [131].

6.4.2. Sensorimotor integration approach

One model by Ayres [132] using exercises for development of sensory integration is based on the notion that the ability to process sensory information affects behaviour and learning. A few studies exist where speech development is measured. Ayres conducted a study with children suffering from apraxia and SLI, using sensory integration intervention, and found that the four children in the study increased their expressive language abilities [89]. And, another study involving children with ASD showed an increased complexity of utterance after sensory-based interventions [133]. One neurophysiological study on 17 children with CP using brainstem auditory evoked potential (BAEP) examination with significant improvement of transmissions in the auditory pathway section of the brainstem was measured after rehabilitation with Masgutova Neurosensorimotor Reflex Integration (MNRI) [134], a new form of a manual neuromodulation technique [135]. Hypothetically this improvement could lead to increased function in auditory processing. Several intervention studies with MNRI have recently been published mainly on children with NDD [136] however nonfocusing on speech outcome.

7. Conclusion and implications for therapy models used in MSD

There is still very little empirical evidence; however, a respectful volume of clinical experience for treatment addressed children with MSD. Recently, more studies have been published especially concerning treatments with motor learning strategies, and the future looks prosperous. As we have been discussing in this chapter, it is noted that children with MSD can suffer from comorbidity in several areas such as fine and gross motor including posture control dysfunctions and sensory processing disorders even when there is no NDD found. We have also presented neural correlates giving evidence for the co-occurrence and interaction of different motor skills in several brain regions which give strength to the hypothesis that motor, speech and language dysfunctions seldom come alone and can be symptoms of an underlying global deficit. This enhances the question if therapeutic models ought to work with a holistic perspective reinforcing sensorimotor processing functions (e.g. oral and manual proprioception, postural, visual, auditory and tactile) on several levels of the brain as part of a treatment approach premised on the notion that the functions of the extrapyramidal nervous system must be addressed in the case of abnormal sensorimotor integration [134]. Maybe if there is an underlying core deficit, it could be reached through corrections of the dysfunctional basic sensorimotor patterns enhancing development of adequate motor responses leading to functional behaviour [134] as also proposed by Kent [105] concerning oral-motor exercises as we earlier quoted under Section 5.2. Even though sensorimotor approaches like MNRI and others based on PML such as PROMPT, DTTC or ReST show some promising results in research and clinical experience, more studies are needed confirming evidence-based treatment models in MSD. Future case-control study designs and especially randomised-controlled trials would be very valuable. Our experience with MSD population is that it is a complex group with a variety type of subtle problems. Trying to achieve a study group as homogeneous as possible seems to be important for the statistical power of studies. However, such studies could affect the generalisation of the results. We suggest using clinical experience as an integral guide for inclusion criteria and the design of the studies. We welcome more research on existing as well as new treatment models increasing quality of life for children with MSD.

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