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# Apraxia of Speech and Grammatical Language Impairment in Children with Autism: Procedural Deficit Hypothesis

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## ABSTRACT

The presence and extent of motor speech and grammatical language impairments are very heterogeneous in children with autism spectrum disorders (ASD). Childhood apraxia of speech (CAS) is a prevalent concomitant motor speech disorder in children with autism affecting imitation skills and impairing speech acquisition due to motor sequencing deficits. Children with CAS also exhibit procedural learning impairments (Iuzzini-Seigel, 2021).

Ullman (2006) hypothesized in the ‘Procedural Deficit Hypothesis’ that procedural memory deficit underlies grammatical language impairments in ASD. This causes difficulty in formulating sentences on the basis of grammatical or word order sequencing rules. Therefore, children with ASD prefer to retrieve and use pre-learnt utterances stored as whole units in their semantic memory.

Literature review suggests disruption of a common FOXP2 (Forkhead Box P2) gene in both CAS and developmental language impairment (Morgan, Fisher, Scheffer & Hildebrand, 2016). Evidence also suggests that the same gene FOXP2 underlies functioning of procedural memory system (Ullman et. al., 1997).

From the review of literature it is hypothesized that procedural learning deficit is the plausible cause of grammatical language impairment and apraxia of speech in ASD. The potential implication of this review is that procedural memory training could enhance both speech motor planning/programming and grammar in children with ASD.

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## 1. INTRODUCTION

### 1.1. Communication, Language and Speech

Communication is crucial to strengthen human relationships and to realize the purpose of life. The process of communication involves sending and receiving both verbal and nonverbal messages. The input message is understood through a network of brain connections for receptive language. Subsequently, response or output message is formulated at expressive language areas in the frontal lobe based on linguistic rules. Motor planning and programming of the movements necessary for production of this planned utterance occurs. Then, command is given to the respiratory, phonatory, articulatory, and resonatory subsystems for speech production. Speech, the vocal expression of language, is finally executed by a controlled and precisely coordinated sequential movement of tongue, jaw, lips and soft palate.

### 1.2. Childhood Apraxia of Speech (CAS)

Sequencing is vital for both sentence formulation/ grammar and motor speech production. Childhood apraxia of speech (CAS) is a neurological speech sound disorder of motor sequencing and learning complex movements (Crary & Anderson, 1991) in the absence of neuromuscular deficits. The core impairment in planning or programming spatiotemporal parameters of movement results in errors in speech sound production. This affects child’s speech intelligibility, word formation, inflection or rhythm. Difficulty with intentional production of speech as in a speech imitation task, is commonly observed. Children with mild CAS may be able to repeat monosyllables like no/ baa/ moo, but may struggle repeating multisyllabic words like potato/ umbrella/ butterfly. This is because of the greater number of articulatory movements and coordination involved in production of multisyllabic utterances.

### 1.3. Autism Spectrum Disorders (ASD)

Autism (or autism spectrum disorders, ASD) is a complex neurodevelopmental condition characterized by persistent social communication deficits and restricted/repetitive behaviors or interests. Approximately 1/100 children are diagnosed with ASD around the world (Zeidan et al., 2022). Autism begins in early childhood and has a wide range of symptoms and severity. Many children show signs of autism in early infancy while others go through a period of regression between 18 and 24 months of age. They lose previously acquired language skills, eye contact or response to their name, and become socially withdrawn. On providing early intervention, they acquire new vocabulary but thereafter may exhibit grammar or word order sequencing deficits. Apraxia of speech is a prevalent concomitant disorder in many children with autism contributing to significant delay in emergence of speech. The presence and extent of motor speech and grammatical impairments are very heterogeneous in children with autism.

## 2. Literature findings

Although substantial research on the various types of speech and language impairments is evident, only few researchers have attempted to identify the link between memory, speech, language and its clinical implications. Review of literature reveals that both motor sequencing (speech) and grammar (language) are sustained by the procedural memory system in the long-term memory store.

### 2.1. Procedural and Semantic Memory

Procedural memory involves unconscious recollection of how to perform a task, and performing the task automatically, while it was acquired implicitly (without conscious recall) through repeated exposure to and practice of a task (Squire, 1986; Ewen & Mostofsky, 2013). Hence, it is called as implicit/non-declarative memory. Furthermore, Ullman (2001, 2004) explained that procedural memory sustains grammar/sentence formation because it’s a rule-governed sequential combination of units.

On the contrary, lexical knowledge about words, facts and events are rapidly and consciously acquired/used using the semantic/explicit/declarative memory, as distinguished by Paradis (2004) and Ullman (2004). Evidence suggests that semantic memory is rooted in temporal lobe structures while the procedural memory is rooted in frontal/basal-ganglia structures and they play critical roles in language production (Ullman et al., 1997). The discovery of the disassociation between declarative memory and procedural memory developed principally from the study of amnestic patients in the 1970s (Squire,1986).

### 2.2. Procedural vs Semantic Memory Impairments

Ullman (2013) distinguished procedural and semantic memory impairments in developmental and adult-onset disorders (Table 1). Developmental disorders like ASD and specific language impairment have impaired procedural memory and relatively intact semantic memory. Semantic memory may play compensatory roles in these disorders. On the contrary, procedural memory is intact and semantic memory is impaired in adult cognitive communication disorders like dementia and fluent aphasia.

**Table 1: Procedural vs semantic memory impairment across various disorders**

| Different Disorders | Procedural memory | Semantic memory |
|---|---|---|
| Specific language impairment, Autism spectrum disorder, Tourette syndrome, Parkinson’s disease, Huntington’s disease, and non-fluent aphasia | Impaired | Intact |
| Alzheimer’s disease, semantic | Intact | Impaired |
| dementia, fluent aphasia and | | |
| amnesia | | |

### 2.3. Procedural Memory in Neurodevelopmental Disorders

A systematic review of literature on procedural memory across various neurodevelopmental disorders was done and summarized in Table 2. Earlier research focused on exploring procedural memory impairments in autism spectrum disorder (ASD) and specific language impairment (SLI). More recently procedural memory has been studied in disorders like childhood apraxia of speech (CAS). Research also provides evidence for the compensatory roles of declarative memory across neurodevelopmental disorders (Ullman & Pullman, 2015) like SLI, dyslexia, ASD, Tourette syndrome, and obsessive-compulsive disorder.

**Table 2: Procedural memory in various neurodevelopmental disorders**

| No | Authors | Year | Disorder | Finding |
|---|---|---|---|---|
| 1 | Toichi & Kamio | 2002 | ASD | Superior "rote memory" |
| 2 | Ullman & Pierpont | 2005 | SLI | Procedural deficit causes grammar impairment; lexical or declarative memory is relatively spared. |
| 3 | Tomblin, Mainela-Arnold & Zhang | 2007 | SLI | Slower procedural learning when language impairment was defined in terms of grammar impairments. |
| 4 | Kemény & Lukács | 2010 | Language impairment | Impaired procedural learning |
| 5 | Romero-Munguía | 2013 | ASD | Faulty procedural memory causes deficits in several cognitive skills |
| 6 | Hedenius et. al | 2013 | Developmental Dyslexia | Impaired implicit sequence learning |
| 7 | Lum, Conti-Ramsden, Morgan & Ullman | 2014 | SLI | Impaired procedural learning in serial reaction time (SRT) task |
| 8 | Takács et. al | 2018 | Tourette syndrome | Enhanced grammar processing and procedural memory |
| 9 | Rochette, Soulières, Berthiaume, & Godbout | 2018 | ASD | Atypical relationship between frontal area and encoding of sensory-motor procedural memory |
| 10 | Bombonato et. al | 2022 | CAS | Implicit learning deficit (using SRT task). |

## 3. Discussions

### 3.1. Procedural Deficit Hypothesis and ASD

Walenski, Tager-Flusberg, and Ullman (2006) subsequently hypothesized in the 'Procedural Deficit Hypothesis' that procedural memory deficit underlies grammatical language impairments in children with autism. This causes difficulty in formulating sentences on the basis of grammatical or word order sequencing rules. However, lexical knowledge, which depends on the declarative memory system, remains relatively spared in children with ASD. Children with ASD compensate for procedural memory impairment by learning these words as whole lexical units using their intact semantic/declarative memory. Ullman and Pullman (2015) explained this as the 'See-Saw effect' in their 'dual-systems' model of language acquisition.

Procedural memory impairment and the 'See-Saw effect' in children with ASD was also explored by the present author (Treasa, 2021) in previous investigations. If a novel utterance has be learnt, repeated teaching trials (discrete trial training) with specific stimuli for each of the target utterance should be used for successful procedural learning. This was evident in a case study by Treasa and Chengappa (2014) who found that discrete trial training (which incorporates principles of procedural learning) and incidental teaching was useful in emergence of expressive grammatical morphology. In addition, comorbid impairments such as childhood apraxia of speech (CAS) is very common in ASD. Receptive language can be intact while expressive language can get confounded due to the presence of CAS.

### 3.2. Procedural Memory Impairment and CAS

Children with CAS exhibit struggle in smoothly and precisely sequencing speech sound into syllables, syllables into words, and words into sentences with appropriate prosody (Morgan, Fisher, Scheffer & Hildebrand, 2016). This is because of impaired production, sequencing, timing, and stress of sounds, syllables and words (American Speech-Language-Hearing Association, 2007). The core features of CAS include inconsistent speech errors, lengthened and disrupted co-articulatory transitions and inappropriate prosody. Children with CAS also exhibited procedural learning impairments (Iuzzini-Seigel, 2021) as supported by studies examining the number of practice trials. Maas et al (2019) reported that children with CAS showed greater benefit from the massed practice (many trials in a short period of time) conditions. Likewise, Edeal and Gildersleeve-Neumann (2011) found greater improvement for training in high (100-150 trials/session) practice as compared to low (30-40 trials/session) practice amount.

Iuzzini-Seigal (2021) used an adapted version of the classic serial reaction time task (Nissen & Bullemer, 1987) to test procedural learning of a five-step visuospatial sequence. They found that procedural learning impairment was more evident in CAS and those with poor grammar. Interestingly, their findings show that children with CAS ultimately demonstrated procedural learning although they had slower reaction times and required more trials to initially learn the sequence. Iuzzini-Seigal (2021) reviewed various studies and stated that "while typically developing children are able to acquire speech sounds, grammar, and motor patterns without being explicitly taught, children with CAS tend to require copious amounts of intensive treatment to make even minimal gains".

### 3.3. FOXP2: Linking CAS, language impairment and procedural memory

Review of the research literature suggests disruption of a common *FOXP2* (Forkhead Box P2) gene in both CAS and developmental language impairment. Morgan, Fisher, Scheffer and Hildebrand (2016) posited that CAS is the core phenotype of all *FOXP2*-related speech and language disorders, regardless of the underlying genetic alteration. This corroborates evidence from previous research by Laffin et al (2012) and Turner et al (2013). There can be co-morbid *FOXP2*-related language disorder. Watkins et al (2002) reported that these children have stronger nonverbal (performance) IQ compared to verbal IQ, and their fine and gross motor skills are intact. The phenotype of *FOXP2*-plus-related disorders tends to be more severe clinically and have features of oral motor deficits, global developmental delay or autism spectrum disorder (Morgan, Fisher, Scheffer & Hildebrand, 2016).

Review also indicates that the same gene *FOXP2* underlies functioning of procedural memory system which is affected in children with grammatical impairment and CAS. Takahashi, Liu, Hirokawa and Takahashi (2003) reported in their study that striatum is involved in procedural memory processing, and mutation of *FOXP2* results in neurological speech and language disorders. Their suggestion is consistent with the findings of Ullman et al. (1997) which indicated that procedural memory-dependent mental grammar is rooted in the basal ganglia and the frontal cortex.

## 4. Conclusion

To summarize, procedural memory deficit is causing sequencing difficulty in grammatical impairments (evident as word sequencing error) as well as childhood apraxia of speech (sound or syllable sequencing error). In addition, procedural deficit has been reported in children with autism spectrum disorders who commonly have co-morbid CAS and exhibit grammatical language impairment. Evidence also suggests that the same *FOXP2* gene which underlies functioning of procedural memory system is found to be disrupted in both ASD and CAS. From the review of literature, it is hypothesized that procedural learning deficit is the plausible cause of grammatical language impairment and childhood apraxia of speech in ASD. The potential implication for future studies is that procedural memory training can enhance both speech motor planning/programming and grammar in children with autism.

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