



RISK FACTORS OF STUTTERING

Thesis

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بِسْمِ اللَّهِ الرَّحْمَنِ الرَّحِيمِ

قَالَ

سُبْحَانَكَ لَا عِلْمَ لَنَا
إِلَّا مَا عَلَّمْتَنَا إِنَّكَ أَنْتَ
الْعَلِيمُ الْعَظِيمُ

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List of Abbreviations

Abb.	Full term
<i>ADHD</i>	<i>Attention deficit hyperactive disorder</i>
<i>APA</i>	<i>Auditory Perceptual Assessment</i>
<i>CWDNS</i>	<i>Children who did not stutter</i>
<i>CWS</i>	<i>Children who stutter</i>
<i>DAF</i>	<i>Delayed auditory feedback</i>
<i>DS</i>	<i>Developmental stuttering</i>
<i>DSM</i>	<i>Diagnostic and Statistical Manual of Mental Disorders</i>
<i>DZ</i>	<i>Dizygotic</i>
<i>EEA</i>	<i>Equal environment assumption</i>
<i>fMRI</i>	<i>Functional magnetic resonance imaging</i>
<i>GABAA</i>	<i>Gamma-aminobutyric acid A</i>
<i>GTS</i>	<i>Gilles de la Tourette syndrome</i>
<i>I.P.Ds</i>	<i>Intra-phonemic disruption</i>
<i>ICD</i>	<i>International Classification of Diseases</i>
<i>ICF</i>	<i>International Classification of Functioning</i>
<i>MEG</i>	<i>Magnetoencephalography</i>
<i>MZ</i>	<i>Monozygotic</i>
<i>NIH</i>	<i>National Institutes of Health</i>
<i>OCD</i>	<i>Obsessive-compulsive disorder</i>
<i>OD</i>	<i>Other Disfluencies</i>
<i>PANDAS</i>	<i>Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal infections</i>
<i>PDS</i>	<i>Persistent developmental stuttering</i>
<i>PET</i>	<i>Positron emission tomography</i>
<i>RFO</i>	<i>Right frontal operculum</i>
<i>SD</i>	<i>Standard deviation \</i>
<i>SLD</i>	<i>Stuttering-Like Disfluencies</i>
<i>SSI-3</i>	<i>Stuttering severity instrument third edition</i>
<i>VBM</i>	<i>Voxel-based morphometry</i>
<i>WHO</i>	<i>World Health Organization</i>

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INTRODUCTION

Speech is a natural means of self-expression and communication among humans. Fluency refers to the effortless production of long, continuous, and easy flow of speech. Dysfluency, in contrast, denotes involuntary interruptions that prevent the effortless and natural progression of speech. In speech language pathology, this condition is referred as stuttering speech disorder (*Weber-Fox et al., 2013*). The interruptions that break normal flow of speech includes dysfluency/dysfluent incidences such as repetition, prolongation, and interjection. The measure of these incidences in speech is essential for phoniatricians to clearly determine and establish the presence of disorder. In addition, it acts as a foundation for planning the treatment (*Guilar, 2013*).

Stuttering is a common neurodevelopmental disorder which in most cases starts before four years of age and has a lifetime prevalence of up to 8.5% (*Reilly et al., 2009*). Remission occurs before teenage in about 75% of cases. Studies designed to identify risk and associated factors for stuttering have examined child samples predominantly because this age-group has a higher chance of being affected than is the case with adult samples (*Kefalianos et al., 2014*).

Unfortunately, the conventional measuring techniques followed by phoniatricians are subjective, and different clinicians give variable estimates of dysfluency incidences on

the same speech sample. In addition, the techniques require a manual process, and hence it is costly and time consuming. Hence, dependable and efficient as well as objective approaches are necessary to support SLPs in the course of stuttering assessments (***Mahesha and Vinod, 2019***).

In stuttered speech, the disruptions that normally appear as different dysfluencies are considered as cessations of speech. It is perceived as an inability to initiate words, respiratory, and phonatory dysfunction, such as running out of air for speech, pitch raising, glottal fry, etc. It is significant to quantify the influence of respiratory and phonatory dysfunction to characterize different dysfluencies. The vocal source features have potential to estimate the respiratory and phonatory dysfunction (***Yairi and Seery, 2015***).

Research into stuttering epidemiology has two important drawbacks: 1) Comprehensive information about risk / associated factors and comorbid conditions obtained from adults and from population studies is sparse (***Heelan et al., 2016***). However, many potential vulnerabilities are only noticeable post childhood. 2) Epidemiological information has not been used in subtyping of stuttering, even though it has been used successfully to subtype other neurodevelopmental and early-onset neuropsychiatric disorders (***Rodgers et al., 2015***).

Clinical, linguistic and neurophysiological investigations are the predominant approaches in subtyping of stuttering (*Ambrose et al., 2015*). Information about the heterogeneity of stuttering subtypes, both in terms of different outcomes and in terms of etiopathogenetic pathways, is only become available recently (*Ajdacic-Gross et al., 2018*).

Gender (being male) is the dominant risk factor for stuttering, as also applies to other neurodevelopmental disorders. Examples include attention deficit hyperactive disorder (ADHD) (*Willcutt, 2012*), conduct disorder (*Zahn-Waxler et al., 2008*), tics and the Gilles de la Tourette syndrome (GTS) (*Rodgers et al., 2014*).

These neurodevelopmental disorders are the second-most prominent set of comorbidities with stuttering. The male to female ratio in adults who stutter is estimated at around 4:1 and this gender ratio is higher in those who persist, than in those who recover, from stuttering (*Ajdacic-Gross et al., 2018*).

Neuropsychiatric conditions such as parental anxiety and parental obsessive-compulsive disorder (OCD) are associated with stuttering (*Ajdacic-Gross et al., 2010*).

Stuttering is also associated with delays in speech development, other speech problems, and dyslexia. Additional risk / associated factors where the evidence is more limited include socioeconomic status, prenatal /perinatal / postnatal

brain damage, brain damage, low birth weight, head trauma and head injury, low intelligence, impaired child-parent interactions, conflicts with parents, negative parental reactions to normal childhood dysfluency, emotional reactivity and a sensitive temperament (*Yairi and Ambrose, 2013*). The most frequently investigated comorbid condition with stuttering is anxiety and, more particularly, social anxiety (*Smith et al., 2014*).

An alternative perspective about risk factors and comorbidities emerges from the PANDAS model (Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal infections) (*Murphy et al., 2012*). PANDAS focuses on infections caused by group A streptococci during the first few years of life. An autoimmune influence due to autoantibodies directed against basal ganglia tissue is postulated. PANDAS applies to neurodevelopmental disorders such as GTS and ADHD and a link with stuttering has been reported in a case study (*Toto et al., 2015*). In addition to streptococci, several other infectious agents have been investigated as causative factors in mental disorders, primarily in psychosis and anxiety disorders. These agents merit more consideration with respect to stuttering, as they may play a causative role in this condition too (*Torrey et al., 2012 and Witthauer et al., 2014*).

AIM OF THE WORK

The aim of this work is to study the different risk factors of stuttering in children in order to understand more about its nature, etiology and to help to decrease its incidence if possible.

REVIEW OF LITERATURE

Definition of stuttering

As defined by the Diagnostic and Statistical Manual of Mental Disorders (*American Psychiatric Association, 1994*), stuttering is “a disturbance in the normal fluency and time pattern of speech that is inappropriate for the individual’s age.” Stuttering manifests itself as repetitions of sounds, syllables, or words or as a speech block with prolonged pauses between sounds and words. Secondary behaviors such as eye blinking, jaw jerking, and head movements are learned approaches to minimize severity of stuttering and can lead to increased fear of speaking and embarrassment (*Costa and Kroll, 2000; Prasse and Kikano, 2008*).

Stuttering is a speech disorder in which sounds, syllables, or words are repeated or prolonged, disrupting the normal flow of speech. These speech disruptions may be accompanied by struggling behaviors, such as rapid eye blinks or tremors of the lips. Stuttering can make it difficult to communicate with other people, which often affect a person’s quality of life (*Cook et al., 2013*).

The impact of stuttering on a person's functioning and emotional state can be severe. This may include fears of having to enunciate specific vowels or consonants, fears of being caught stuttering in social situations, self-imposed isolation, anxiety,

stress, shame, being a possible target of bullying (especially in children), having to use word substitution and rearrange words in a sentence to hide stuttering, or a feeling of "loss of control" during speech and this may be leading to avoidance of speech. Stuttering is sometimes popularly associated with anxiety but there is actually no such correlation (though as mentioned social anxiety may actually develop in individuals as a result of their stuttering) (**Zerbeto and Chun, 2013**).

Stuttering is generally not a problem with the physical production of speech sounds or putting thoughts into words. Acute nervousness and stress do not cause stuttering but they can trigger stuttering in people who have the speech disorder, and living with a highly stigmatized disability can result in anxiety and high allostatic stress load (*i.e.*, chronic nervousness and stress) that reduce the amount of acute stress necessary to trigger stuttering in any given person who stutters, exacerbating the problem in the manner of a positive feedback system; the name 'stuttered speech syndrome' has been proposed for this condition. Neither acute nor chronic stress, however, itself creates any predisposition to stuttering (**Andrade et al., 2013**).

The disorder is also *variable* (not constant) which means that in certain situations, such as talking on the telephone or in a large group, the stuttering might be more severe or less, depending on whether or not the stutterer is self-conscious about their stuttering. Stutterers often find that their stuttering fluctuates and that they have "good" days, "bad" days and

"stutter-free" days. The times in which their stuttering fluctuates can be random (*Cook et al., 2013*).

Stuttering occurs in people of all ages, ethnicities, and cultures, but it is most commonly associated with young children as they develop and learn language and speech (*Büchel and Sommer, 2004*). Approximately 5% of all children will experience some form of speech dysfluency (*Guitar and Conture, 2007*). However, studies have shown that 50% to 80% of those who stutter will recover spontaneously by the age of 5 without the intervention of professional treatment (physiological dysfluency) (*Finn, 1996; Ashurst and Wasson, 2011*).

According to *Yairi and Ambrose (2005)*, 65% have the onset of stuttering before age 3 years and 85% before age 3.5 years. Onset of stuttering in hood is rare, but stuttering-like characteristics may develop late in life due to cerebral stroke, head injury, tumor or drug abuse (*Helm-Estabrooks, 1999*). Psychogenic stuttering may arise in hood after a traumatic deprivation (*Mahr and Leith, 1992*).

Self-reported population-based lifetime prevalence in Danish twins is about 8 % for males and 4 % for females (*Fagnani et al., 2011*). Twins might have a higher rate than 'single-born' children and *Yairi and Ambrose (2005)* mention that 5% of preschool-age children will have stuttering episodes. Most children recover from their stuttering and the prevalence