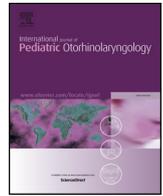




Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Auditory temporal processing assessment in children with developmental stuttering

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ARTICLE INFO

Keywords:

Auditory temporal processing
Auditory processing disorder
Developmental stuttering
Timing

ABSTRACT

Objective: Stuttering is a developmental disorder of speech production with a dynamic and multifactorial nature. Scientific theories mentioned the role of auditory processing disorder in stuttering. Investigating the auditory processing in stuttering would provide insights into the mechanisms of stuttering. The details of basic auditory processing in children with stuttering (CWS) continue to remain uncertain. This study aimed to investigate the auditory temporal processing (ATP) in CWS and also its relationship with the stuttering severity.

Methods: The participants of this comparative cross-sectional study were 54 CWS and 63 children without stuttering (CWOS). All children were between 7 and 12 years old. ATP ability of the participants was measured using the Backward Masking (BM), Duration Pattern (DP), and Gap in Noise (GIN) tests. Then, the groups were compared in terms of ATP results. The correlation between the scores of these tests and stuttering severity was assessed.

Results: According to the results, CWS showed poorer performance on DP, BM and GIN tests when compared with CWOS. Moreover, the stuttering severity had a significant negative correlation with the DP scores and percentage of correct identification scores in GIN; whereas it had a significant positive correlation with the BM thresholds.

Conclusions: The results showed that some CWS have ATP disorder, which could exacerbate their stuttering. These findings highlighted the role of ATP disorder in stuttering.

1. Introduction

Stuttering is a common childhood developmental disorder, which affects speech fluency [1]. It may recover spontaneously, otherwise requires long-term intensive therapeutic sessions, which typically have adverse impacts on the psychological and social health of children and their family [2,3]. Stuttering is characterized by repetition of a syllable or a part of a syllable, prolongation of sounds, and frequent interruptions in speech [4]. The prevalence of stuttering in childhood is between 4% and 5%; about 1% of this population will continue to have persistent stuttering until adulthood [5]. The underlying cause of stuttering continues to remain unknown. According to current theories, stuttering is defined as a dynamic and multifactorial disorder, which occurs due to the effects of complex interactions between atypical

speech motor control system, auditory processing, language and other cognitive processing, genetic, emotions, and social factors [6].

Auditory processing is a neural mechanism, which has been emphasized in numerous theories for its role in stuttering [7]. According to the Direction of Velocities into Articulators (DIVA) model, the auditory system has a role in activating and tuning speech sound maps and forming internal speech control models (feedforward and feedback models) through neural representation of sounds via processing others' speech, receiving self-produced speech feedback, and interacting with motor cortex [8]. Proposing the "internal model and feedback-biased motor control theory," Max et al. (2004) assumed that stuttering is due to a mismatch between predicted (feedforward) and actual (feedback) auditory consequences of speech. The researchers attributed this mismatch and speech error to the auditory feedback control pathway [9].

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<https://doi.org/10.1016/j.ijporl.2020.109935>

Received 25 November 2019; Received in revised form 3 February 2020; Accepted 4 February 2020

Available online 06 February 2020

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Civier et al. (2010) hypothesized that stuttering is due to the disruption in rapid and automatic feedforward control pathway which, in turn, increases dependence on the auditory feedback control system. This is intrinsically a slow pathway for speech control and causes errors and mismatches between the actual output and desired target speech. They reported that speech errors are mainly made in rapid speech components, such as syllables with rapid formant transition (F2 transition). As errors become larger, they are more likely to be corrected by the brain. To correct the errors, the brain restart the current syllable until the mismatch is resolved. Frequent articulation is associated with repetition of sound/syllable, which is seen in the majority of people with stuttering [10]. Similar to other relevant studies, Civier's study revealed the importance of ATP in the auditory feedback control system [11,12].

ATP is crucial for the perception of prosody components, including rhythm, and rapid speech elements, and is a prerequisite for superior skills in speech perception and spoken language processing. These temporal components are essential in the auditory feedback pathway to produce fluent speech [12]. Through the feedback of speech temporal information, the auditory system allows the brain to predict the temporal properties of the next speech. There is evidence showing that speech is actively monitored and adjusted through manipulating the timing of auditory feedback [13]. Since speech is a rapid dynamic motor process, all of its fundamental neural components should work together in precise timing [14]. It is thus expected that ATP disorder can affect fluent speech production and cause or exacerbate stuttering [9]. There are some evidences that people with stuttering have ATP disorder. Beal et al. (2010) investigated auditory evoked magnetic fields during application of different passive listening and active speaking tasks for patients with stuttering and found significant prolongation of M50 and M100 peak latencies in response to passive listening and suppression latencies of these waves during speech production. They argued that difference in auditory cortex timing in different stimulating conditions in this population may cause a difference in the timing of feedforward or feedback speech control elements [14]. Other studies of magnetoencephalography (MEG) also reported timing deficit of the left auditory cortex and that signaling from the brainstem to the left primary auditory cortex was slower in patient with stuttering than fluent speakers [15]. Ismail et al. (2017) showed delayed absolute latencies of the late auditory evoked potentials in CWS, specifically in those with more severe stuttering [16].

Howell et al. (2002) performed a behavioral study, in which one aspects of ATP, including temporal masking, was investigated in 8-12-year-old children with stuttering using the BM method. They found that the BM thresholds were higher in CWS than CWOS [17]. Howell et al. (2006) reported that "backward masking performance at teenage is one factor that distinguishes speakers who persist in their stutter from those who recover" [18].

ATP findings in CWS provide a clear understanding of the mechanism of this disorder. Regarding that there are some ATP rehabilitation techniques, these findings can be helpful in effective stuttering rehabilitation. There are insufficient ATP studies on children with stutter and contradictory results are observed regarding the relationship between ATP and stuttering severity. One reason for these contradictory results may be due to the heterogeneity of case groups in different studies. This is because stuttering is intrinsically a complex disorder, which is affected by environmental, biological, and psychological factors [11]. Moreover, the majority of studies are based on limited data. Therefore, more studies are needed for a precise investigation into the relationship between different ATP components and stuttering. This study aimed to investigate ATP in a large group of CWS and a homogeneous control group using the GIN, DP, and BM tests.

2. Methods

2.1. Subjects

This study was conducted on 54 children with stuttering (CWS group) and 63 children without stuttering (control group). Moreover, 90.7% and 93.7% of the CWS and control groups were boys, respectively. CWS were selected from the children's Medical Center hospital and private speech and language therapy clinics in Tehran, using convenience sampling. The controls were selected from different schools in Tehran among eligible students. Both groups were age- and sex-matched and it was tried to match them also in terms of socioeconomic status [13,19].

The inclusion criteria for CWS group were: 1) diagnosis of stuttering by an experienced speech-language therapist in aforementioned centers and SSI-3 (very mild); 2) age range between 7 and 12 years old; 3) pure-tone audiometry thresholds within normal limits (> 20 dBHL) for air- and bone-conduction pathways at 250–8000 Hz; 4) normal otoscopy; 5) normal IQs (≥ 85 on Wechsler's Revised Intelligence Scale for children [20] administered by a psychologist; 6) being right-handed (using the Edinburgh handedness questionnaire); 7) native speakers of Persian; 8) no history of taking drugs affecting the central nervous system, neurological problems, psychological problems, head trauma, ear surgery, and ear diseases according to self-reports, learning and language impairment according to the speech-language pathologist, and ADHD according to the psychiatrist diagnosis; 9) no cognitive activities like music-related activities; and 10) mother's educational attainment between 12 and 16 years and Hollingshead score between 4 and 6 [47].

The inclusion criteria for the control group (CWOS) were: 1) fluent speech without any physical signs mentioned in SSI-3 based on the reports by parents and teachers; 2) items 2–10 above; 3) not susceptible to ADP based on Auditory Processing Domain Questionnaire (APDQ) [21].

2.2. Procedures

First, the research methodology and stages were completely explained to the children and their parents. The eligible children were enrolled after completion of the consent forms. Participants could leave the study at any time. All children were screened for peripheral hearing ability and all tests were conducted in a soundproof room with a noise level of lower than 30 dBA. The stimuli were given using a laptop and a headphone (Sennheiser HD202). The output of the headphone was calibrated using a sound level meter (Bruel & Kjaer, 2250 L) and artificial ear type 4153 (Denmark) connected to weight or adapter of the circumaural headphone.

2.3. Stuttering assessment

Participants were assessed using the Riley's (1994) Stuttering Severity Instrument, version three (SSI-3). During the assessment, interviews of about 20-min duration were recorded. The recording included a reading of a text and spontaneous speech (narration) containing a minimum of 200 syllables for each child. Any associated physical concomitant was also recorded separately by two evaluators. These interviews were used to assess the frequency and duration of stuttering and any associated secondary and physical concomitants, and scoring was according to the guidelines specified in Riley (1994) [22]. Assessment and scoring were done by an experienced and certified speech-language pathologist and a trained assistant, who was a master's student. This study used the SSI-3 score and percent syllables stuttered (SS %) to investigate the correlation of ATP tests with stuttering severity.

Table 1

Means and SDs of auditory temporal processing tasks for CWS and CWOS. The results show significant differences between the two groups for all tasks.

Variables	CWS		CWOS		P
	M(SD)		M(SD)		
	R	L	R	L	
DPT	62.17(15.36)	62.34(14.75)	79.99(11.51)	79.73(11.37)	< 0.001
BM	62.59(10.43)		46.7(6.59)		< 0.001
GINth	6.37(1.37)	6.24(1.31)	4.88(1.01)	4.96(0.89)	< 0.001
GINp	62.98(9.66)	63.5(9.88)	70.68(7.81)	70.19(7.23)	< 0.001

2.4. ATP tests

2.4.1. Backward masking

In this test, the listener should detect the tonal stimulus, which comes right before the masking noise at different intensity levels from suprathreshold to threshold levels. Noise reduces the sensitivity to preceding tone even in normal individuals; however, people with APD perform weaker in this regard, that their threshold of tone exceeds the normal range. This test was developed using MATLAB, the sampling rate of 44100 Hz and 16 bit. The test comprised of a 1 kHz probe tone with 20-ms duration, initial intensity of 94-dB SPL, noise frequency in the range of 600–1400 Hz and bandwidth of 1.16-octave band, the central frequency of 916 Hz, 300-ms duration, spectral level of 40-dB, and all stimuli were gated with 10-ms. cosine-squared envelopes [17,18].

2.4.2. BM procedure

Stimuli were presented monaurally in the right ear. The test was done using a three alternative forced choice paradigm [18,23]. Each item had three stimuli with 800-ms interstimulus interval, two 300-ms noise bursts and one random noise-tone stimulus (target signal) [18]. The examinee should select the target stimulus from three different choices correspond to cards marked with numbers of 1, 2, and 3. Prior to the test, 10 items with intensities easily applicable to the child (94-dB SPL) were generated to make him/her familiar with the stimulus. Thresholds were derived with the 2-down 1-up tracking procedure using a 2-dB step size to find the psychometric functions for a target level of 71% correct. Each threshold was estimated based on the mean of four last reversals out of ten reversals. In total, the threshold was evaluated twice and repeated if the difference between the thresholds was more than 2 dB. Finally, the mean of three assessments was regarded as the final threshold [18].

2.4.3. Duration pattern

This test is composed of three tone patterns presented with frequency of 1000 Hz and two different durations (short: 250 ms and long: 500 ms). In each pattern, there are 300-msec intertone interval and 6-sec interpattern interval. In general, there are six patterns (LLS, LSL, LSS, SLS, SLL, and SSL), which are generated randomly. In this pattern, S and L are used to denote short and long durations, respectively. It is a monaural test with the presentation level of 50-dB HL or 70-dB SPL. The test includes 60 items (30 to each ear) [24,25].

2.4.4. Gap in Noise

This test comprises of a series of 6-sec segments of broadband noise. Each segment contains zero to three silent intervals and there is a 5-sec interval between every two segments. The segment's intervals had different durations of 2, 3, 4, 5, 6, 8, 10, 12, 15, and 20 msec. The location and duration of the intervals in the noise segments were distributed in a quasi-random sequence. Prior to the main test stage, ten pilot tests were provided to make participants familiar with the test. GIN test has two measurement criteria: approximate gap threshold defined as the shortest length of the gap identified correctly on four out of six

attempts; the percentage of correct gap identification defined as a proportion of correct responses to the sum of the gap intervals. GIN included four test lists, each list was randomly presented in one ear [25,26].

2.5. Data analysis

Data were analyzed in SPSS 23. The mean, standard deviation, and percentage were used to describe data. The multivariate analysis of variance (MANOVA) was used to compare the between-group data. In addition, Pearson correlation was used to determine the correlation of the SSI-3 score and SS% with ATP tests. The significance level was considered to be $p = 0.05$ throughout the study [27].

3. Results

All CWS had been diagnosed before school age. There was no significant difference in age between the CWS group (mean 9.913 ± 1.67 years; range 7 years and 5 months–12 years and 8 months; 1, 15, 9, 11, 7 and 11 children at the age of 7, 8, 9, 10, 11 and 12 years respectively) and the CWOS group (mean 9.98 ± 1.48 years; range 8–12 years; 16, 11, 10, 12 and 14 children at the age of 8, 9, 10, 11 and 12 years respectively) ($p = 0.748$).

3.1. Stuttering severity scores

The mean SSI-3 and SS% was 16.5185 ± 6.5638 and 3.0179 ± 1.8585 for the CWS group, respectively. In total, 13, 22, and 19 participants in the CWS group had very mild (24.1%), mild (40.7%), and moderate (35.2%) scores on the SSI-3, respectively.

3.2. Auditory temporal processing tests

Table 1 presents the means and standard deviations of the ATP tests for both CWS and CWOS groups. These results show that the performance of CWS group was significantly poorer than CWOS in these tests ($p < 0.001$). As compared to the control group, the BM thresholds increased, DP scores reduced, GIN thresholds increased, and the percentage of correct identification scores on the GIN test reduced in the CWS group.

3.3. Correlation between ATP tests and stuttering severity scores

The correlation coefficient between the stuttering severity scores and ATP results is presented in Table 2. According to these results, there was a significant negative correlation between DP scores of the right ($r = -0.269$, $p = 0.049$) and left ($r = -0.329$, $p = 0.015$) ears and percentage of correct identification scores of the right ($r = -0.391$, $p = 0.003$) and left ($r = -0.385$, $p = 0.005$) ears with SSI-3 scores. There was also a positive significant correlation between the BM thresholds ($r = 0.394$, $p = 0.003$) and SSI-3 scores. In other words, with increasing the stuttering severity, the DP score and the percent of correct GIN responses of both ears reduced, and the BM threshold

Table 2
Correlation coefficients for the three main auditory temporal processing assessments, and the SSI-3 and SS% in CWS.

Variables	DPT		BM	GINth		GINp	
	R	L	R	R	L	R	L
SSI-3	-0.269*	-0.329*	0.394**	0.218	0.225	-0.391**	-0.245**
SS%	-0.125	-0.065	0.336*	0.138	0.207	-0.375	-0.263

**Correlation is significant at the 0.01 level (two-tailed).

* Correlation is significant at the 0.05 level (two-tailed).

increased in the right ear. Moreover, there was a significant positive correlation between the BM threshold and %SS. In other words, the BM threshold in the right ear increased with increasing the percent syllables stuttered. There was no significant correlation between GIN thresholds (GINth) of the right and left ears with SSI-3 and %SS scores ($p > 0.05$). There was no significant correlation between the DPT scores of the right and left ears, GIN correct responses of right and left ears, and SS% ($p > 0.05$).

4. Discussion

The findings of the present study showed that the CWS group had poorer scores in ATP tests, specifically BM and DP, and higher distribution of responses than the CWOS group. BM deficit in CWS was also reported by Howell et al. (2000). They showed that CWS had higher BM thresholds than the controls. They argued that probably the neural processes involved in speech fluency, which are related to the detection of synchrony between planning and execution of speech, are common with those in BM-related temporal processes. Since the performance of both BM and speech depends on the synchrony detector, it can be concluded that any disruption in BM tests is related to fluency disorder [17]. Howell and Williams (2004) studied a wider age range and did not observe any difference between CWS and CWOS in BM. However, there was some evidence showing between-group differences in this age range in the rates of threshold alterations. They reported that development of BM in participants with stuttering differs from fluent participants [28]. Their study differed with the present study in the sample size and age range, which can be the cause of contradictory results. Moreover, they did not provide any information about the inclusion criteria and stuttering severity. Differences in the inclusion criteria, the participant's homogeneity, and the disorder severity can affect the results. Howell, Davis, and Williams (2006) investigated a group of people with recovered and persistent stuttering and concluded that the BM thresholds were higher in a group with persistent stuttering than the group who recovered. They regarded the BM deficit as an effective factor and sufficient condition for persistent stuttering [18]. Howell, Davis, and Williams (2006) attributed these contradictory results in their previous studies (2000 and 2004) to the number of participants with persistent stuttering. Probably, Howell (2000) investigated more participants with persistent stuttering.

The findings of the present study were consistent with other studies on multiple aspects of auditory processing, including ATP, in people who stutter. Andrade et al. (2008) investigated a broad spectrum of verbal and nonverbal ATP tests, including DP and Pitch Pattern Sequence Test (PPST), in people with stuttering. Results showed that 92.85% of these people, the majority of whom were in the age group of 8–11 years, had a disorder in these tests, specifically in nonverbal tests [29]. Valame and Kekade (2014) showed that CWS had a deficit in the DP test and the Random Gap Detection Test (RGDT) [12].

Some other studies, which used non-behavioral methods, such as MEG, to assess the performance of the auditory system in people with stuttering have reported auditory cortex timing disturbance. The researchers attributed these timing differences to a deficiency in accessing the neural representations of speech sounds in the feedforward and feedback control pathways. They argued that the inability to produce

stable and correct neural representations of the sounds in childhood or maintain it persistently has a role in stuttering [14].

In the present study investigations into the relationship of stuttering severity and ATP tests showed a correlation only between BM thresholds, DP scores, and percent of correct GIN responses with the stuttering severity (SSI-3). Howell also reported a correlation between BM test results and stuttering severity [17].

In addition to the total stuttering severity score (SSI-3), we used SS% to investigate data correlation. Because in speech production and control modeling studies, a syllable unit is used to explain these processes and speech errors in stuttering [8,10]. Moreover, the transition of syllable-related information in central nervous system requires accurate and rapid temporal processing [11]. As a result, it was argued that there is a relationship between temporal processing results and SS%, and more accurate data on the relationship of ATP and stuttering can be obtained. In the present study, only the BM threshold was significantly correlated with SS%. It is probable that the neural mechanisms of BM is directly correlated with stuttering or its severity. Civier et al. (2010) hypothesized that people with stuttering are more dependent on the auditory feedback pathway for speech control. Due to the intrinsic delay of the auditory feedback pathway, the mismatch and error occur between the actual speech feedback and desired target speech. They reported that speech errors are mainly made in rapid speech components, such as syllables with rapid formant transition (F2 transition) [10]. Wright et al. (1997) assumed that the backward masking deficit interferes with the processing of rapid speech components and can cause phonological problems in children with dyslexia and language disorders (30). In the present study, the authors assumed that in the BM deficit cases, the vowels in syllables such as consonant-vowel with formant transition, have a great masking effect on the formant transition [30,31]. As a result, the syllable transition does not occur properly in the auditory feedback pathway, and thus the mismatch between actual auditory outcome and desired auditory targets increases. More error rate is associated with a higher chance of correction by the brain, thereby increasing stuttering severity [10]. This argument can justify the relationship between BM deficit and stuttering severity (SS%) in this study.

Other studies on the DP test in CWS have not reported any relationship between DP scores and SSI-3 scores [12,29,32]. The present study showed a significant relationship between DP test and SSI-3 scores. According to similar studies, abnormal processing of temporal patterns, such as duration pattern, interfere with the perception of suprasegmental aspects of speech (prosody), such as rhythm, and can affect speech fluency [11,12]. Wieland et al. (2015) reported that CWS had auditory rhythm discrimination disorder. They attributed the auditory rhythm discrimination disorder to a deficit in internal timing neural network, called basal ganglia-thalamocortical (BGTC), which includes the auditory cortex. Any disturbance in this network interferes with the internal timing patterns of speech perception and fluent speech production. In fact, normal balance between rhythmic components of feedforward and feedback mechanisms is disturbed during speech production and perception [13]. Chang et al. (2016) also argued that the major deficit in CWS is related to the perception of temporal patterns of sound sequence. This skill is the base of rhythm processing and speech perception and production [33]. Therefore, it is expected that

any deficit in auditory temporal pattern perception affects the stuttering severity.

An interesting finding of the present study is a significant correlation of GIN correct scores with stuttering severity (SSI-3); whereas no significant relationship was observed between the GIN approximate thresholds and stuttering severity. Other studies did not report similar results because they used a different tests for the assessment of temporal resolution ability [11,12,32]. The GIN test shows relatively good sensitivity to central auditory nervous system lesions and can accurately assess temporal resolution [26]. Musiek et al. (2005) stated that other tests, such as Auditory Fusion Test-Revised (AFTR) and RGDT run for the assessment of temporal resolution, are actually aiming to assess auditory fusion and have a neural mechanism different from the GIN. According to them, two GIN components (threshold and percent of correct responses) can produce different information in different populations [26]. It is then recommended to conduct further studies into auditory temporal resolution ability using the GIN test in CWS, so as to determine the relationship of this disorder with different components of the test.

To summarize, results showed that the auditory temporal processing disorder is correlated with the incidence and/or exacerbation of stuttering. It is recommended to consider this important processing component in routine assessment of children with stuttering. Regarding that there are some rehabilitation methods for temporal processing disorders, it is hoped that future studies investigate the effectiveness of these methods in improving stuttering in these children. Future studies are recommended to investigate other processing components of the auditory system in this group of children to achieve a better understanding of the relationship between the auditory system and stuttering.

Declaration of competing interestCOI

We have no conflict(s) of interest to declare. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Acknowledgment

All participants, their parents and Ms. Alikarami, speech-language pathologist and personnel of the.

Speech language pathology department of Children's Medical Center hospital are thanked for their contribution. This study was supported by the University of Social Welfare and Rehabilitation Sciences of Tehran and approved by the Ethics Committee of this university under the code IR.USWR.REC.1397.1.

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