Impaired neural entrainment to low frequency amplitude modulations in English-speaking children with dyslexia or dyslexia and DLD

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Abstract

Neural synchronization to amplitude-modulated noise at three frequencies (2Hz, 5Hz, 8Hz) thought to be important for syllable perception was investigated in English-speaking school-aged children.The theoretically-important delta-band (~2Hz, stressed syllable level) was included along with two syllable-level rates. The auditory steady state response (ASSR) was recorded using EEG in 36 7-to-12-years-old children. Half of the sample had either dyslexia or dyslexia and DLD (developmental language disorder). In comparison to typically-developing children, children with dyslexia or with dyslexia and DLD showed reduced ASSRs for 2Hz stimulation but similar ASSRs at 5Hz and 8Hz. These novel data for English ASSRs converge with prior data suggesting that children with dyslexia have atypical synchrony between brain oscillations and incoming auditory stimulation at ~2Hz, the rate of stressed syllable production across languages. This atypical synchronization likely impairs speech processing, phonological processing, and possibly syntactic processing, as predicted by Temporal Sampling theory.

Key words

Dyslexia, DLD, auditory steady state response, speech envelope, neural oscillations

1. Introduction

Developmental dyslexia is a pervasive neurodevelopmental disorder in which children fail to learn age-appropriate reading skills despite otherwise normal intellectual functioning, normal hearing and an adequate learning environment (Snowling, 2000; Vellutino et al., 2004). Dyslexia affects approximately 5-10% of school aged children (Vellutino et al., 2004). The majority of children with dyslexia have difficulties with phonological processing (the ability to identify and mentally manipulate speech sounds). Developmental language disorder (DLD) affects around 7% of school aged children (Tomblin et al., 1997). Children with DLD have persistent difficulties in learning oral language, which are not associated with a known condition such as sensori-neural hearing loss or Autism Spectrum Disorder (Bishop et al., 2017). Children with DLD typically have difficulty with the accurate processing and production of syntactic structures in speech (Marchman et al., 1999). However, the overlap between dyslexia and DLD can be as high as 50% (McArthur et al., 2000), suggestive of some shared underlying sensory factors. In the current study, we assess the potential importance of neural synchronization to low-frequency amplitude modulations (AMs) in these developmental disorders.

Both DLD and dyslexia have been associated with atypical discrimination of amplitude envelope rise times, which could be expected to relate to atypical neural sampling of speech signals (Temporal Sampling [TS] theory, Goswami, 2011; 2015; 2019). Atypical neural sampling of speech input could be expected to hinder the perceptual representation of speech units, as the perceptual organization of speech information by an affected child (assigning acoustic elements to the groupings comprising words or prosodic phrases in a particular language) would differ from other children, possibly from infancy. In turn, this could lead to the development of poor phonological processing skills in dyslexia (Goswami, 2018), and poor processing of syntax in DLD (Cumming et al., 2015; Richards & Goswami, 2019). Regarding dyslexia, these differences in perceptual organization would be expected to impede the efficient development of grapheme-to-phoneme conversion skills, thereby negatively affecting reading acquisition (Giraud & Poeppel, 2012a, 2012b; Goswami, 2015, 2018; Goswami et al., 2014). Regarding DLD, differences in the perceptual organization of speech-based information would also affect the extraction of prosodic phrasing, the tightly integrated hierarchies of meter and syntax, which highlight the grammatical structure of a language, thereby potentially leading to deficits in the development of syntactic knowledge and grammatical competence (Cumming et al., 2015; Richards & Goswami, 2015, 2019).

The Temporal Sampling framework integrates data on (i) the temporal envelope of speech, i.e., amplitude fluctuations over time (Shannon et al., 1995), and (ii) brain oscillations, i.e., rhythmic shifting of neuronal ensembles between high and low excitability states. Regarding the temporal envelope, the amplitude modulations (AM) nested in the amplitude envelope of speech between 20 and 50 Hz are thought to carry linguistic attributes at the time scale of individual phonemes (e.g., indexing formant transitions that distinguish place of articulation, /b/ vs. /d/, and voice onset time differences that distinguish voicing /p/ vs. /b/, etc.); AMs between 4 and 8 Hz are thought to be relevant to identifying syllables; and AMs from 1 to 3 Hz are thought to reflect lexical and phrasal or prosodic units (for review, Giraud & Poeppel, 2012a, 2012b). During speech perception, the phase of ongoing brain oscillations is known to re-set to reflect the phase of the amplitude envelope of the speech stimulus (Gross et al., 2013). This speech-brain synchrony facilitates perception by synchronizing periods of maximal neural excitability to the time windows in the speech input that contain the most useful information (Peelle & Davis, 2012). Temporal sampling of speech signals by brain oscillations at delta (<4 Hz), theta (4-8 Hz) and gamma (>25 Hz) rates thus helps to package the incoming information into the linguistic phrasal, syllabic, and phonemic scales respectively (Giraud & Poeppel, 2012a). Further, according to the "asymmetric sampling in time" theory (Poeppel, 2003), the left and right auditory cortices show oscillations at different preferred rates: gamma (25-45 Hz) in the left hemisphere and delta-theta (1-8 Hz) in the right. This asymmetry has been observed in adult participants regardless of the nature of the input (e.g., speech vs. non-speech), and independently of the involvement of higher-level speech processing (Giraud & Poeppel, 2012a; Morillon et al., 2010).

There are differing views on the level at which amplitude envelope processing is impaired in dyslexia. Giraud and Poeppel (2012a) hypothesized that children with dyslexia might not show typical left hemisphere specialization for gamma oscillations, which would lead to difficulties in processing phonemic units, in turn leading to phonological deficits. On the other hand, Goswami (2011, 2015, 2018) hypothesized that children with dyslexia and DLD have atypical oscillatory entrainment in the delta and theta ranges, primarily in the right hemisphere, leading to processing deficits in the prosodic (<4 Hz) and syllabic (~4-8 Hz) ranges, which may be causally linked to language and reading difficulties. There is evidence supporting both theories in individuals with dyslexia based on both behavioural and neurophysiological methods, although most of these data come from adults (e.g., Menell et al., 1999; Lehongre et al., 2011; Hämäläinen et al., 2012; Marchesotti et al., 2020; see Lizarazu et al., 2021, for review). In neurophysiological studies with *children* that have used speech inputs, neural synchronization to delta-band information appears most affected (Power et al., 2013, 2016; Molinaro et al., 2016; Destoky et al, 2020; Keshavarzi et al., 2022, Mandke et al., 2022). There are no prior studies of neural synchronization for children with DLD, for either speech or non-speech inputs. Studies of children with dyslexia are reviewed briefly below.

Many psychophysical studies use the temporal modulation transfer function (TMTF) as a measure of sensitivity to amplitude modulations. TMTF determines the minimum depth of AM required to discriminate amplitude-modulated white noise from unmodulated white noise (i.e., AM detection threshold) as a function of the modulation frequency. Lorenzi, Dumont and Füllgrabe (2000) found that French-speaking 10-year-old children with dyslexia showed higher AM detection thresholds (reduced sensitivity) at a range of AM rates compared to age-matched control children and adults, with the largest effect seen at 4 Hz. Deficits in the processing of 4 Hz AMs have also been reported in 12-year-old French-speaking children with dyslexia (Rocheron et al., 2002). Goswami and colleagues (Goswami et al., 2016; Leong & Goswami, 2014) have investigated envelope processing using speech rather than white noise AM stimuli. Goswami et al. (2016) presented nursery rhymes to 10-year-old English-speaking children with dyslexia. The rhymes were band-pass filtered so that they were missing either low frequency (<4 Hz) or high frequency (22-40 Hz) AMs. Children with dyslexia showed similar recognition performance as control children; however, they also showed poorer acoustic learning during the experiment compared to the controls in the low frequency condition only. These child data converge in suggesting perceptual differences in dyslexia in processing low-frequency AM information.

An electrophysiological measure that has the potential to study AM sensitivity is the auditory steady state response (ASSR). ASSRs are neural responses to amplitude modulations in the auditory stimulus, and they reflect how well the auditory system phase locks to the amplitude envelope of the stimulus (Picton et al., 2003). Unlike the TMTF method, which focuses on the AM detection threshold, ASSR provides additional information regarding the degree to which brain oscillations match the amplitude modulations of the incoming stimulus. It is believed that the ASSR generated by amplitude modulations below 40 Hz have their origin in the auditory cortex whereas the ASSR generated by modulations above 40 Hz have subcortical and brainstem origins (Picton et al., 2003). Traditionally, ASSR was used to assess the integrity of the auditory pathway in hearing assessments (for a detailed review see Korczak et al., 2012). More recently ASSRs have been widely used to study the neural bases of speech perception. Because the amplitude envelope of the stimuli evoking ASSRs can be adjusted to match the temporal rates at which meaningful phonological components occur in speech, ASSRs potentially provide an objective measure of the encoding of different time scales in the speech envelope (Miyazaki et al., 2013; Tang et al., 2016). In support of this notion, a number of studies have shown significant correlations between ASSR and speech perception abilities in adults (typically using speech-in-noise measures; Alaerts et al., 2009; Dimitrijevic et al., 2004; Poelmans, Luts, Vandermosten, Boets, et al., 2012; Poelmans, Luts, Vandermosten, Ghesquière, & Wouters, 2012) as well as impaired ASSRs to 2 Hz AM-noise but not 20 Hz AM-noise in adults with dyslexia compared to controls (Hämäläinen et al., 2012). There are only a few ASSR studies to date with children.

Several of these developmental studies have demonstrated atypical ASSRs in school-aged children with dyslexia, but findings have been inconsistent across languages with regards to temporal rate. Furthermore, this body of research has not yet included English-speaking children. Working in Spanish, Lizarazu et al. (2015) presented children (8- to 14-year-olds) and adults with dyslexia with amplitude-modulated noise at 2 Hz, 4 Hz, 7 Hz, 30 Hz and 60 Hz using MEG. The dyslexia group had enhanced (hence atypical) ASSR at 4 Hz compared to controls, irrespective of age. Moreover, the right hemisphere dominance of 4 Hz ASSR observed in controls was absent in the dyslexia group. Greater right-lateralized responding to 4 Hz AMs was significantly related to reading rate, but only for the control participants (reading accuracy was already at ceiling in the transparent Spanish orthography, even for the dyslexic children). Meanwhile the dyslexic group showed right hemisphere dominance for the 30 Hz ASSR, whereas the controls showed no hemispheric bias. No group differences were found at the delta rate of stimulation (2 Hz). A longitudinal study of Dutch-speaking children with and without family risk of dyslexia (who were tested at 5, 7, and 9 years of age), found that neither family risk of dyslexia nor age had any effect on the ASSRs recorded for syllable rate (4 Hz) modulations (De Vos et al., 2017a). However, the onset of reading development was found to be associated with increased responses to phoneme rate (20 Hz) modulations. Additionally, those children (13 from the family risk group plus one control child) who later developed dyslexia had atypically larger ASSRs for phoneme rate modulations (20 Hz) after one year of reading instruction (at age 7). These data suggest that it is learning to read itself that affects the ASSR to faster AMs in dyslexia. In a second study with Dutch teenagers (15-year-olds), the ASSR at 4 Hz, 10 Hz, 20 Hz and 40 Hz was compared in groups with and without dyslexia (De Vos et al., 2017b). A significant group difference in ASSR was found for 10 Hz stimulation only, with dyslexic participants showing a reduced response. This contrasts with the English language study of ASSRs in university students using MEG (Hämäläinen et al., 2012), in which there was a trend for enhanced responding at 10 Hz for dyslexic participants. For 20 Hz stimulation, the Dutch participants with dyslexia showed an increased ASSR compared to controls for both left ear and right ear stimulation, but not when both ears were stimulated, contradicting earlier findings with Dutch dyslexic adults reported by Poelmans, Luts, Vandermosten, Ghesquière, et al. (2012). Accordingly, the ASSR data seem to vary by language and potentially by method

(MEG, EEG). The former may suggest that affected frequencies vary by the nature of the language (e.g., stress-timed vs. syllable-timed languages) and/or after learning a particular orthographic code for language. Despite atypical delta band responding being a key feature of TS theory (Goswami, 2011, 2015, 2018), it is also notable that only one ASSR study with children (Lizarazu et al., 2015) has included 2 Hz stimulation.

However, as the ASSR is a non-speech measure, it is also possible that the brain may respond differently to AMs that reflect real syllables or real stressed syllable patterns when speech is the input. Seven studies of neural entrainment to speech by children with dyslexia are now available in the literature, which concur on suggesting low frequency AM processing deficits, converging on ~2 Hz. Power, Mead, Barnes, and Goswami (2013) and Keshavarzi, Mandke, Macfarlane, Parvez, Gabrielczyk, Wilson, and Goswami (2022) used an auditory-visual rhythmic speech paradigm and EEG with English-speaking children with dyslexia based on the rhythmic repetition of the syllable "ba" at a 2 Hz rate. Power et al. (2013) reported that, compared to age-matched controls, children with dyslexia showed a *different preferred phase* of entrainment in the delta band. A different preferred phase of entrainment implies enhanced neuronal excitability in dyslexia at *less informative* temporal points in the speech signal, which could be expected to affect phonological representation. Keshavarzi et al. (2022) replicated the different preferred phase effect with younger children with dyslexia, again for the delta band only, and also reported that pre-stimulus angular velocity was significantly different in the dyslexic group in the delta band, suggestive of atypically fast neural responding to rhythmic 2 Hz speech input in this band only. In Spanish, Molinaro, Lizarazu, Lallier, Bourguignon, and Carreiras (2016) reported an MEG study of sentence processing by adults and children with dyslexia. Many of the participants were the same individuals studied by Lizarazu et al. (2015)

who had not shown a 2 Hz ASSR difference compared to controls. Molinaro et al. found that both the adults and children with dyslexia showed impaired oscillatory entrainment to speech in the delta band, with reduced delta synchronization originating in the right primary auditory cortex (Molinaro et al., 2016). Similar findings were reported in an MEG study by Mandke, Flanagan, Macfarlane, Gabrielczyk, Wilson, Gross, and Goswami (2022) using a story listening task with English-speaking children. Using lagged speech-brain coherence measures, Mandke et al. reported group differences (dyslexic vs. CA controls) for AM information <5 Hz, which in their speech materials corresponded to both prosodic and syllable-level information. An MEG study with French-speaking dyslexic children using a speech-in-noise task reported atypical cortical tracking at the phrasal rate for their speech materials (0.2 – 1.5 Hz, Destoky et al., 2020). Power, Colling, Mead, Barnes, and Goswami (2016) tested the same sample of English dyslexic children as Power et al. (2013), using a reverse engineering approach. The speech envelopes of noise-vocoded sentences were estimated from children's neural (EEG) response (via envelope reconstruction) in a sentence recognition paradigm. This enabled a direct measure of the quality of children's speech envelope representations. Power et al. reported that the children with dyslexia showed significantly poorer speech encoding for the band < 2 Hz, compared to both age-matched control participants, and also in comparison to *younger reading-level matched* (RL) control children. Inclusion of an RL-matched control group helps to determine whether observed differences in neural activity are a cause of dyslexia or instead a consequence of the atypical (severely reduced) reading experience that accompanies having dyslexia (Goswami, 2015). Despite being matched for reading experience and being able to report correctly the same number of words in the sentences as the RL controls, the children with dyslexia showed significantly poorer encoding of speech envelope information in the delta band than the younger

RL children, suggestive of a fundamental representational deficit. Finally, Di Liberto et al. (2018) used a similar reverse engineering approach with English-speaking children with dyslexia but presented natural speech in a passive listening paradigm. They also included an RL control group, as well as an age-matched control group. Di Liberto et al. reported that the two control groups showed very similar scalp patterns regarding cortical tracking of speech, whereas the dyslexic group did not. Group differences were most marked when both delta-band and theta-band phase locking responses were combined (1 – 8 Hz). The largest group differences were found for a region of interest in the right hemisphere, replicating the findings by Molinaro et al. in Spanish. Individual differences in cortical tracking also showed significant correlations with phonological awareness and phonological memory.

Accordingly, when speech stimuli are used as input, there is greater consensus regarding the AM rates predicted by TS theory concerning which temporal rates may exhibit atypical neural entrainment in dyslexia. The seven studies using real speech all found atypical neural responses in the delta band, for both stress-timed (English) and syllable-timed (Spanish, French) languages. In the non-speech literature, there is a notable absence of any ASSR studies using 2 Hz stimulation with children in stress-timed languages like English and Dutch. In the present study, we include 2 Hz stimulation and study English-speaking children, thus enabling a more comprehensive evaluation of TS Theory. We investigate auditory neural synchronization using the ASSR with 7- to 12-year-old English-speaking children with and without dyslexia or dyslexia and DLD. Three AM oscillatory frequencies were employed, 2 Hz, 5 Hz and 8 Hz, selected to correspond to the rate of prosodic and syllabic cues in the speech envelope (Varnet et al., 2017). The syllable envelope range was divided into two frequencies because some previous acoustic studies using the TMTF have shown an impairment only at the lower end of syllable envelope frequency (Lorenzi et al., 2000).

2. Methods

2.1. Participants

Eighteen 7- to 12-year-old children with dyslexia or with dyslexia and DLD (M=8.67, SD=1.78, 4 girls) and 18 age-matched control children (M=8.48, SD=1.63, 4 girls) volunteered for the experiment. This study was part of a larger study of 70 children investigating auditory processing in typically developing children and children with dyslexia and/or DLD, and the current volunteers either met the criteria for dyslexia (N=11) or for dyslexia and DLD (N=7, details in Table A1 in the Appendix, see also Di Liberto et al., 2018). Parental questionnaires were used to determine that children were not at-risk for other developmental disorders such as autism. Hearing and handedness were not assessed formally at the time of testing. However, participants who reported having hearing difficulties or recurrent hearing infections were excluded at the time of recruitment. The ethics committee for human research at [blinded for review] approved all the experimental methods in the study (approval number: H9660). Informed consent was obtained from parents for all participants, and children also gave verbal assent. The participants were given $30 compensation for their participation. Participants completed the behavioral and EEG tasks either in a single testing session lasting approximately 2 hours (n=30) or in separate behavioral and EEG sessions less than 7 days apart (n=6). All participants completed the behavioral tasks first followed by the EEG task. Five children in the dyslexia group had a formal diagnosis of dyslexia. Fewer children than anticipated volunteered for this study; hence, we did not divide the atypical group into dyslexia versus dyslexia and DLD as this would have reduced statistical power.

2.2. Behavioral measures

Group assignment (dyslexia or dyslexia and DLD versus control) was determined based on children's performance on tests from the screening battery set out below. Children were assigned to the dyslexia group if a) they obtained a score of at least 1*SD* below the age-appropriate mean in at least one reading task, *and* at least one phonological awareness task or memory task, *and* b) had a non-verbal IQ score within the normal range and no indications of Autism Spectrum Disorder (ASD) or attention deficit hyperactivity disorder (ADHD). Individual scores obtained by the children are shown in Table A1 in the Appendix. Two standardized tests typically used to diagnose DLD were also administered, the Test of Reception of Grammar and the Clinical Evaluation of Language Fundamentals Repeating Sentences subscale. Children who had scores below 1 *SD* on one or both of these measures were assigned to the dyslexia with DLD group (see Table A1). We did not collect any information about potential remediation being experienced by individual participants. Children were assigned to the control group if they obtained average scores (±1*SD*) on all the tasks of the screening battery and had no indications of ASD or ADHD.

Word and non-word reading: The sight word efficiency and the phonemic encoding efficiency sub-tests of the Test of Word Reading Efficiency (TOWRE; Torgesen et al., 2012) were administered. The TOWRE consists of two lists, 66 words and 66 non-words. In separate tests for each, children are required to read as many items as possible from each list in 45 seconds. A standardized score (M=100, SD=10) is computed based on how many words are read accurately in this time for each test.

Phonological awareness: Four sub-tests of the phonological awareness battery of the Comprehensive Test of Phonological Processing (CTOPP; Wagner et al., 2013) were administered as follows. *Elision* –children are required to pronounce a word while omitting one of its component sounds, e.g., “say cup without /k/”. *Blending words* – children hear two parts of a word and are asked to combine them and produce the resulting word, e.g., “/pen/ and /səl/ make pencil”. *Sound matching* – children are shown two images of objects and are required to point to the object whose label contains a target sound, e.g., when shown the objects ‘sun’ and ‘ball’, the child is asked to point to the one that starts with /s/. *Phoneme isolation* – children are required to listen to a word and identify one of its component sounds, e.g., “what is the second sound of the word train”. A composite standardized score for phonological awareness is then computed (M=100, SD=10).

Phonological memory: All children completed the digit and non-word repetition subtests of the CTOPP (Wagner et al., 2013). Children were presented with sequences of digits or non-words that increased in complexity on each trial and were required to repeat them in the same order as they were presented. This yields a composite standardized score for phonological memory (M=100, SD=10).

Rapid Symbolic Naming: The rapid digit naming and rapid letter naming subtests of the CTOPP (Wagner et al., 2013) were administered. In these, children are presented with a list of 36 items (digits or letters) on a card and are required to name as many as possible in a 2-minute period. The number of accurately named items in that time is used to calculate a standardized composite rapid symbolic naming score (M=100, SD=10).

Working memory: Children completed the forward and backward number repetition subtests of the Clinical Evaluation of Language Fundamentals test (CELF; Semel et al., 2006). This measures the ability to repeat random number sequences of graduated length. A composite standardized working memory score is obtained based on the number of items that the child could successfully recall in each subtest (M=10, SD=3).

Grammatical competence: The Test of Reception of Grammar (TROG: Bishop, 2003a) and the Recalling Sentences subtest of the CELF (Semel et al., 2006) were administered. In the TROG, children are shown a card with four images and hear a sentence. They are required to point to the image on the card that is described by the sentence. The total number of correct responses is used to calculate the standardized reception of grammar score (M=100, SD=10). In the Recalling Sentences subtest, children hear a sentence and are required to repeat it *verbatim*. Responses are scored according to the number of errors made in each repetition and used to compute a standardized score for this subtest (M=10, SD=3).

Non-Verbal Intelligence: Children completed the matrices subtest of the Kaufman Brief Intelligence Test (KBIT: Kaufman & Kaufman, 2004). The number of matrices completed correctly out of a maximum of 46 is used to compute a standardized non-verbal intelligence score (M=100, SD=10).

Parental questionnaires: In addition to the screening battery, children's parents completed the Children's Communication Checklist (CCC-2, Bishop, 2003b) and the Swanson, Nolan, and Pelham rating scale (SNAP-IV; Swanson, 1992). The CCC-2 is used to assess children's general communicative abilities and identify communicative deficits characteristic of SLI or Autism Spectrum Disorder (ASD). The SNAP-IV is used to identify behavioral patterns characteristic of ADHD or other behavioral disorders. No children in the final sample showed any indications of ASD or ADHD.

2.3. Auditory steady state response (ASSR) recording

2.3.1. Stimuli

The stimuli consisted of 30 seconds duration white noise amplitude-modulated at three frequencies 2 Hz, 5 Hz and 8 Hz with a modulation depth of 100%. These were presented in three separate blocks, one for each of the AMs, and within each block the 30-second stimulus was repeated eight times. The inter-stimulus interval within blocks was 5 seconds. All stimuli were presented at an intensity of 75 dB SPL through 2 speakers placed directly in front of the participant (RMS amplitude measured using Brüel & Kjær sound level meter type 2250 and a microphone placed at the point of space representing the middle of the participant's head).

2.3.2. EEG recording

Participants sat on a comfortable chair 1 m away from an LCD screen and watched a silent video of their choice. They were instructed to ignore the sounds they heard and concentrate on the video. While they watched the video and ignored the sounds, their continuous EEG was recorded using 129 channel (128 channels plus a reference electrode) Hydrocel Geodesic Sensor Net (HCGSN), NetAmps 300 amplifier and NetStation 4.5.7 software (EGI Inc.) at a sampling rate of 1000 Hz with the reference electrode placed at Cz. No online filtering was performed during the acquisition. The electrode impedances were kept below 50 kΩ. The continuous EEG was saved for offline analysis.

2.3.3. EEG analysis

The EEG data were analysed offline using EEGLAB (Delorme & Makeig, 2004) and letswave 6 (http://nocions.github.io/letswave6/) toolboxes in MATLAB 2014a (Mathworks, Natick, USA). The EEG was first band pass filtered between 0.3 and 30 Hz using Hamming windowed sinc FIR filter ('pop_eegfiltnew' function in EEGLAB). It was then divided into epochs between 0 and 30 seconds relative to stimulus onset. The entire stimulus duration was selected as the epoch because ASSRs require several cycles of stimulation to become entrained (Nozaradan et al., 2016). After epoching, noisy electrodes (typically electrodes in the outer ring of the net) were identified visually and removed from the data (average: 4 electrodes; range 1 to 6). Ocular artifact correction was performed using independent component analysis (the 'run_ica' function in EEGLAB). Independent components with known properties of eyeblinks and horizontal eye movements were identified and removed. Noisy EEG channels were then interpolated. The EEG data were then re-referenced to the average of the mastoid electrodes (Alaerts et al., 2009; Dimitrijevic et al., 2004). Trials exceeding ± 100 μV were removed from the dataset. All participants had at least 6 accepted trials per block (average number of accepted trials: Control – 2 Hz M = 6.83, SD = 0.98; 5 Hz M = 6.94, SD = 0.93; 8 Hz M = 6.83, SD = 0.98; Dyslexia or dyslexia with DLD – 2 Hz M = 6.83, SD = 0.78; 5 Hz M = 6.77, SD = 0.80; 8 Hz M = 6.55, SD = 0.85). A 2 x 3 mixed ANOVA on the number of accepted trials with the between-subject factor group (control, dyslexia or dyslexia with DLD) and within-subject factor condition (2 Hz, 5 Hz, 8Hz) showed no significant main effects or interactions (all $F<1$) suggesting there was no systematic signal to noise ratio differences across conditions or groups. The epochs were averaged across trials for each participant and condition. This time domain averaging improves the signal to noise ratio of the EEG activity time locked to the amplitude modulation cycles (Nozaradan et al., 2011; Nozaradan et al., 2016).

The averaged waveforms were then transformed to the frequency domain using a discrete Fourier transformation resulting in a frequency spectrum of amplitudes between 0 and 30 Hz with a frequency resolution of 0.033 Hz. The obtained sprectum contained both the EEG activity elicited by the stimuli as well as the residual background noise unrelated to the stimuli. The background noise was removed by subtracting, at each time point in the frequency spectra, the average amplitude measured in the neighboring frequency bins (second to fifth bin on both sides, see Mouraux et al., 2011; Nozaradan, Peretz, et al., 2016; Nozaradan, Peretz, & Mouraux, 2012). To ensure that the background noise that was subtracted did not vary between control children and children with dyslexia or dyslexia with DLD, thereby potentially affecting group-level ASSR results, we calculated the average amplitude in the neighboring frequency bins (second to fifth bins around the frequencies of interest, 2 Hz, 5 Hz and 8 Hz). These noise amplitudes were then subjected to a 3 x 2 mixed analysis of variance (ANOVA) with the factors stimulation frequency (2 Hz, 5 Hz, 8Hz) and group (control, dyslexia or dyslexia with DLD). The ANOVA did not reveal any significant effects (all Fs < 2), showing that there is no systematic difference in the background EEG activity around the stimulation frequencies. EEG spectra of the individual participants were averaged to produce the grand averaged spectrum per condition and group.

The ASSR amplitude was calculated from the electrodes in the fronto-central scalp region as this location generates ASSRs with high amplitude for auditory stimuli (Nozaradan, Peretz, et al., 2016). We averaged the response from 18 fronto-central electrodes and the amplitude of the response at the frequencies of interest (2 Hz, 5 Hz, and 8 Hz) was computed. Figure 1 shows the electrodes used for the analysis.

[Insert Figure 1 about here]

2.3.4. Statistical analysis

The first analysis sought evidence for selective neural entrainment in the EEG response. This was done by computing z scores at the frequency of interest (2Hz, 5 Hz, 8 Hz) as the difference in amplitude between that frequency and the mean on 20 neighboring frequency bins (second to 11th frequency bin on either side), divided by the standard deviation of the 20 neighboring bins. In line with previous studies, we considered z scores greater than 3.1 ($p < .001$, one-tailed, i.e., signal > noise) to be significant (Peter et al., 2022; Quek et al., 2018). Levene's tests of homogeneity of variance showed that the variance in ASSR amplitude was equivalent for all the frequencies. Differences between dyslexic and control groups were thus evaluated using a mixed ANOVA with the within-subject factor of condition (2 Hz, 5 Hz, 8 Hz) and the between-subject factor of group (control, dyslexia or dyslexia with DLD). Note that a previous exploratory ANOVA had included the additional factor of hemisphere and did not show any significant hemispheric effects. Partial η^2 was computed as a measure of effect size. In case of more than one degree of freedom in the numerator, the Greenhouse-Geisser correction was applied to account for the potential violation of sphericity. Pearson correlations between ASSR amplitude and behavioral measures were computed.

3. Results

3.1. Behavioral tasks

Table 1 presents the means, standard deviations, and results of independent-sample t-tests for each task from the screening battery. Children in the control group outperformed the children with dyslexia or dyslexia with DLD on all tasks. Although the control group also had higher non-verbal IQ scores, all the children in the dyslexia or dyslexia with DLD group had non-verbal IQ scores within the normal range (i.e., not more than 1SD below the standardized mean) and sat close to the normalized mean of 100.

Table 1. *Mean scores for the tasks of the screening battery and independent-samples t-test values for comparison between dyslexia or dyslexia with DLD and control group performance. Standard deviations are in parentheses.*

	Dyslexia/Dyslexia with DLD	Control	*t* (*df* = 34)	*p*
Age (Months)	104.13(21.46)	101.80(19.61)	0.34	.736
Non-Verbal Intelligence	100.50(10.65)	112.94(9.55)	3.69	.001
Word reading	80.50(11.61)	103.67(15.13)	5.15	.001
Non-Word Reading	79.61(8.50)	100.39(13.41)	5.55	.001
Phonological Awareness (Words)	85.00(11.21)	102.50(10.34)	4.87	.001
Phonological Awareness (Non words)	75.11(18.83)	97.50(10.72)	4.38	.001
Phonological Memory	83.11(12.94)	102.44(13.78)	4.33	.001
Rapid Symbolic Naming	85.88(12.77)	104.44(12.57)	4.39	.001

Working Memory	7.05(2.89)	9.78(2.53)	2.99	.005
Test of Reception of Grammar	95.44(11.32)	106.67(8.58)	3.35	.002
Recalling Sentences	8.27(2.76)	11.05(2.21)	3.33	.002

3.2. ASSR

The spectra of responses to amplitude-modulated noise at different modulation frequencies for each group are shown in Figure 2. As can be seen, all conditions generated a peak in the spectrum at the frequency of the amplitude envelope of the stimulus, 2 Hz, 5 Hz, and 8 Hz. The z scores comparing ASSR amplitude at the stimulation frequency and the nearby frequencies showed a significant response at the stimulation frequencies for both groups (Table 2). This shows that a significant ASSR was generated in both groups and for all frequencies.

[Insert Figure 2 about here]

Table 2. Results (z scores) of the significant testing of the ASSR.

	Control	Dyslexia/ Dyslexia with DLD
2 Hz	37.63*	19.53*
5 Hz	11.96*	12.16*

8 Hz	19.17*	20.96*

* p < .001

ASSR amplitudes across frequencies and groups are shown in Figure 3. The two-way ANOVA on ASSR amplitude with the factors condition (2 Hz, 5 Hz, 8 Hz) and group (control, dyslexia or dyslexia with DLD) showed a significant main effect of condition F(1.31, 44.64)=63.05, p=.001, partial ŋ^{2}=.65. Bonferroni corrected pairwise comparisons revealed that the ASSR for 2 Hz (M=0.553, SE=0.055) was significantly higher than the ASSR for 5 Hz (M=0.117, SE=0.022) and for 8 Hz (M=0.119, SE=0.019). While the main effect of group was not significant F(1, 34)=1.26 p=.268, partial ŋ^{2}=.036, there was a significant interaction between group and condition, F(1.31, 44.64)=5.91, p=.013, partial ŋ^{2}=.15.

To interpret the condition x group interaction, separate one-way ANOVAs were computed for each frequency with the between-subject factor group (control, dyslexia or dyslexia with DLD). For 2 Hz ASSR, there was a significant effect of group, F(1,34)=4.53, p=.040, partial ŋ^{2}=.12, with a significantly larger ASSR for the control group than in the dyslexia or dyslexia with DLD group (M=0.669, SE=.077 vs. M=0.437, SE=0.077). There was no effect of group for ASSR amplitude at 5 Hz, F(1,34)=1.07, p=.307, partial ŋ^{2}=.03, nor at 8 Hz, F(1,34)=0.28, p=.596, partial ŋ^{2}=.008.

[Insert Figure 3 about here]

Accordingly, the children with dyslexia or with dyslexia and DLD showed a reduced ASSR amplitude compared to control children in the 2 Hz condition only. ASSRs at 5 Hz and 8 Hz were equivalent for both groups.

3.3. Correlation between ASSR and behavioral measures

Table 3 shows the Pearson correlation coefficients (and, in parentheses, partial correlation coefficients controlling for children's non-verbal IQ scores) between the ASSR and behavioral measures of phonological processing, reading, and language pooled across groups. Children's non-word reading ability and rapid symbolic naming scores were significantly correlated with higher 2 Hz ASSR amplitudes. Therefore, children with higher 2 Hz ASSR amplitudes read non-words more accurately and named more items correctly in the allotted time on the rapid symbolic naming task. However, only the correlation with rapid symbolic naming remained significant when the false discovery rate correction for multiple comparisons was applied (Benjamini & Hochberg, 1995). Regarding the partial correlations controlling for non-verbal IQ scores (shown in parentheses in the table), the correlation for rapid symbolic naming did not survive correction for multiple comparisons. It can further be noted that neither non-verbal IQ nor age showed significant correlations with ASSR amplitudes at any frequencies.

Table 3. *Correlations and (partial correlations in parentheses) of ASSR amplitudes with behavioral measures.*

	2 Hz	5 Hz	8 Hz

Word reading	0.29 (.22)	-0.14 (-.07)	-0.16 (-.23)
Non-Word Reading	0.34* (.25)	-0.23 (-.15)	-0.15 (-.24)
Phonological Awareness (Words)	0.02 (-.11)	-0.14 (-.06)	0.09 (.03)
Phonological Awareness (Non words)	0.20 (.04)	-0.13 (-.01)	-0.08 (-.03)
Phonological Memory	0.19 (.04)	-0.08 (.05)	-0.13 (-.26)
Rapid Symbolic Naming	0.45**† (.37*)	-0.20 (-.12)	-0.19 (-.29)

*p<.05, **p<.01, †p<.05 after correcting for multiple comparisons

4. Discussion

The objective of this study was to use ASSRs, neural responses to amplitude modulation measured via electrophysiology, to assess how well the child's auditory system phase locks to AM-noise stimuli when the child has a developmental language disorder. Prior ASSR studies with children in a range of languages have provided an inconsistent picture regarding which modulation frequencies are affected in dyslexia, and only one prior ASSR study has included the theoretically important rate of 2 Hz (Lizarazu et al., 2015). Some developmental ASSR studies reported atypical phase locking to low frequency stimulation (e.g., 4 Hz in Lizarazu et al., 2015) and others to high frequency stimulation (e.g., 20 Hz in De Vos et al., 2017b). There are no prior ASSR nor cortical speech tracking studies to our knowledge regarding children with DLD. We presented English-speaking children with dyslexia or with dyslexia and DLD and age-matched control children with amplitude-modulated noise at three stimulation frequencies related to extracting linguistic information from the speech envelope: 2 Hz (prosody), 5 Hz (lower range of syllable rate), 8 Hz (upper range of syllable rate). The results revealed that English-speaking children with dyslexia or with dyslexia and DLD have a specific deficit in synchronizing brain oscillations with amplitude-modulated noise presented at a 2 Hz modulation rate, but show intact synchronization for 5 Hz and 8 Hz AM-noise stimuli. The finding that the ASSR to syllable rate modulations (5 Hz and 8 Hz) did not differ between groups is consistent with some prior ASSR studies conducted with amplitude-modulated noise in another stress-timed language, Dutch (De Vos et al., 2017a, b).

The finding of a selective deficit for 2 Hz stimulation in the English-speaking children with dyslexia or with dyslexia and DLD tested here provides support for the TS framework, a sensory-neural theory of the basis of language disorders in children (Goswami, 2011, 2015, 2018, 2019). TS theory was originally confined to dyslexia, and proposed impairments in cortical oscillatory phase locking mechanisms for AM information in speech at frequencies below 10 Hz, that is delta band (0.5 – 4 Hz) and theta band (4 – 8 Hz) synchronization. Following speech modelling work showing the importance of both delta- and theta-rate bands of AMs for speech rhythm perception (Leong et al., 2014; Leong & Goswami, 2015), TS theory was extended to DLD via the prosodic phrasing hypothesis (Cumming et al., 2015a). As children with DLD exhibit the same rhythmic synchronization (tapping) and linguistic stress perception difficulties found in dyslexia, it was proposed that difficulties in processing prosodic phrasing and prosodic hierarchies dependent on perceiving sensory cues to speech rhythm and stress patterning may underlie the syntactic difficulties that characterize children with DLD (Cumming et al., 2015a, b; Richards & Goswami, 2015, 2019).

The current findings indicate that the deficit in cortical oscillatory entrainment for the children with dyslexia or with dyslexia and DLD was specific to 2 Hz (prosody level). We did not find a deficit at 5 Hz nor 8 Hz (syllable level). This pattern supports behavioral studies showing impaired use of 2 Hz modulations in *speech perception* by English-speaking children and adults with dyslexia (Goswami et al., 2016; Leong & Goswami, 2014), as well as DLD studies showing rhythmic synchronization and rhythmic perception impairments at 2 Hz (Corriveau & Goswami, 2009; Cumming et al., 2015b). It is also consistent with the prior studies of oscillatory entrainment to speech inputs by children with dyslexia reviewed earlier, which all showed atypical delta-band synchronization and encoding, using either EEG or MEG (Destoky et al., 2020; Di Liberto et al., 2018; Keshavarzi et al., 2022; Mandke et al., 2022; Molinaro et al., 2016; Power et al., 2016, 2013). These data complement an fNIRS study with dyslexic children using 2 Hz AM noise (Cutini et al., 2016), which showed that neural synchronization and amplitude rise time perception were significantly related. It is notable that atypical delta-band entrainment in dyslexia has now been found for *both* stress-timed (English) and syllable-timed (Spanish, French) languages. It is also notable that while Molinaro et al. (2016) reported atypical synchronization in Spanish in the delta band in dyslexia when natural speech was the input, some of the same participants took part in the ASSR study conducted by Lizarazu et al. (2015) where no deficit in the ASSR to 2 Hz stimulation was reported. Accordingly, for Spanish, speech and non-speech inputs produced different results in the theoretically-important delta band. By contrast, this was not the case for English, as some of the participants in the current study also participated in the study by Di Liberto et al. (2018), which used natural speech. However, Di Liberto et al. found the strongest group differences in neural entrainment to speech when *both* delta-band and theta-band synchronization were considered together. Accordingly, at least for Spanish and English, atypical neural synchronization in children with language impairments (oral and written language) is best characterized by impairments in cortical oscillatory phase locking mechanisms for AM information in speech at frequencies below 10 Hz. This is in line with the original proposals made by the TS framework (Goswami, 2011).

One limitation of the current study is that children with dyslexia and children with dyslexia and DLD were considered together when assessing the ASSR, despite these two developmental disorders of language presenting with different cognitive profiles. This decision was taken to maximise statistical power, and because delta band differences were expected for both disorders. The importance of delta-band neural synchronization for efficient speech processing by children with language disorders may be understood in light of advances in computational modelling of the AM structure of the amplitude envelope of child-directed (English nursery rhymes) and infant-directed speech (IDS). Recent modelling studies have revealed that AMs at both ~2 Hz and ~5 Hz nested in the speech envelope of these genres play a key role in the perceptual experience of speech rhythm (Leong & Goswami, 2014, 2015; Leong et al., 2017). The speech modelling, which used both Bayesian probability and PCA approaches, showed important *phase relationships* between slower AMs (centred on ~2 Hz and ~5 Hz) in the amplitude envelope that determined whether participants were hearing a trochaic or iambic rhythm pattern (Leong & Goswami, 2014). Modelling of the AM-structure of English nursery rhymes showed that when these two rates of AM are in phase and are both peaking, then a strong syllable is heard (Leong & Goswami, 2015). When the theta band AM is peaking but the delta band AM is in a trough, then a weak syllable is heard. Accordingly, accurate neural synchronization to either the 2 Hz modulation rate and/or the 5 Hz modulation rate would seem to be important for the experience of speech rhythm. Rhythmic sensitivity is widely considered a precursor of language acquisition, with the earliest representations of the speech signal encoding its rhythmic structure (Mehler et al., 1988). Subsequent aspects of language, such as phonology and syntax, may thus be scaffolded onto these rhythmic representations. A further modelling study compared the AM structure of IDS to adult-directed speech (ADS, Leong et al., 2017). Here it was found that IDS had significantly more modulation energy in the delta band (~2 Hz) than ADS, while ADS had significantly more modulation energy in the theta band (~5 Hz) than IDS. Further, the phase synchronization between these two slower-rate bands was significantly greater in IDS than in ADS. This suggests that the AM structure of IDS facilitates 2 Hz entrainment to the AM patterns in speech, as indeed has been demonstrated for infants aged 4, 7 and 11 months (Attaheri et al., 2022). IDS thus emphasizes the acoustic information related to accurate neural delta-theta phase alignment.

These modifications in IDS would support the extraction of speech rhythm patterns by infants which, in turn, would support the development of both phonological and syntactic representations in multiple ways. In addition, infants are sensitive to low-frequency speech information before birth (Spence & DeCasper, 1987) and have been found to use this information to discriminate between rhythm classes of languages at birth (Mehler et al., 1988; Nazzi et al., 1998). Accordingly, rhythmic sensitivity has direct influences on infants’ processing of their native language, which could be expected to affect both the quality of phonological representation and the extraction of the prosodic hierarches which help to specify syntax.

Atypical neural synchronization during infancy for 2 Hz AM speech information could thus lead to deficits in the development of early phonological and syntactic competence and the impaired formation of phonologically-well-specified representations in infants' early lexicons. No study of the ASSR in infants at family risk for dyslexia or DLD is currently available. If atypical neural synchronization is present from before birth, then over developmental time, affected children may develop phonological and/or syntactic deficits long before they enter school and begin learning to read. There is already behavioral infant and toddler data consistent with such a developmental trajectory from studies of participants at family risk for dyslexia (Kalashnikova et al., 2018, 2019b, 2019a).

Regarding brain-behavior correlations between behavioral tests administered to the current sample and ASSR amplitude at 2 Hz, only the significant relationship for rapid automatized naming (RAN) survived correction for multiple comparisons, and only when the analyses did not control for children's non-verbal IQ scores. RAN is one of the three areas of phonological processing typically impaired in individuals with developmental dyslexia (the others are phonological awareness and phonological memory, see Ziegler & Goswami, 2005). Although RAN is thought by some to be purely an index of the speed of access to familiar lexical items (Denckla & Rudel, 1976; Wolf & Bowers, 1999), it is also known to be affected by the quality of children's phonological representations. For example, RAN speed differs for highly familiar items drawn from dense versus sparse phonological neighborhoods (Guardia, 2010). Neighborhood density effects are typically attributed to the quality of the phonological representations of words held in long-term memory (see Clarkson et al., 2017), suggesting that RAN tasks are also an index of the quality of the child's lexical phonological representations. While it is surprising that the 2 Hz ASSR did not correlate with any other of the measures administered to our sample, Table 3 shows that the correlations between these measures and the ASSR for 2 Hz stimulation was always systematically positive and typically larger than the correlations for the other stimulation rates. As noted earlier, when some of these same children participated in the EEG entrainment study using natural speech, then significant relations between atypical neural synchronization and both phonological awareness and phonological memory were found (Di Liberto et al., 2018).

In summary, our data indicate that children with dyslexia or dyslexia and DLD have a specific deficit in synchronizing brain oscillations with incoming non-speech auditory rhythmic stimulation when that stimulation is in the delta band. Moreover, the degree of brain-stimulus synchronization at 2 Hz was correlated with a behavioral measure of phonological processing that is impaired in developmental dyslexia across languages, RAN. There is a clear confluence of findings here linking brain responses and linguistic processing. Children with dyslexia or dyslexia and DLD in this study were shown to have reduced brain-stimulus synchronization at 2 Hz. Previous research shows that children with atypical brain-stimulus synchronization at 2 Hz have poorer phonological skills; and children with atypical brain-stimulus synchronization at 2 Hz have poorer linguistic skills compared to those without language difficulties (Di Liberto et al., 2018; Keshavarzi et al., 2022; Mandke et al., 2022; Molinaro et al., 2016; Power et al., 2013, 2016). These results support the hypothesis that atypical neural entrainment to speech envelope information at low frequencies is central to the linguistic difficulties that characterize children with developmental dyslexia and DLD across languages.

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Figure captions

Figure 1. Scalp location of the electrodes used for the analysis.

Figure 2. FFT spectra and the topography of ASSR for different modulation frequencies for control (A) and dyslexia/dyslexia with DLD (B) groups. Asterisks show significant response at the frequency of amplitude modulation.

Figure 3. Rain cloud plots (Allen et al., 2019) depicting boxplots, individual data points and probability density of the ASSR amplitudes across stimuli and groups. The rhombus represents the mean.

Appendix

Table A 1. *Individual scores for each behavioural measure completed by the 18 children assigned to the Dyslexia (D) or Dyslexia with DLD (DDLD) group. Refer to the main text for a full description of the administered sub-tests, procedures, and scoring criteria.*

ID	Age	Sex	Group	Non-Verbal IQ[1]	Word reading[2]	Non-Word Reading[2]	Phon. Awareness (Words)[3]	Phon. Awareness (Non words)[3]	Phon. Memory[3]	Rapid Symbolic Naming[3]	Working Memory[3]	Grammar[4]	Recalling Sentences[4]
1	8,6	M	D	100	77	64	73	61	113	95	14	95	10
2	8,1	M	D	100	93	84	75	70	98	92	12	104	12
3	10,4	M	D	124	80	83	98	88	88	92	5	111	9
4	6,5	M	DDLD	85	94	80	92	75	73	61	6	72	6
5	10,4	F	D	103	65	81	96	61	92	75	7	116	12
6	9,0	F	D	112	63	78	114	128	98	67	9	106	13
7	7,3	F	DDLD	87	79	74	75	58	64	82	7	83	5
8	6,1	M	D	87	70	75	96	49	79	76	3	99	7
9	10,8	M	D	96	100	95	71	73	88	82	5	88	10
10	8,2	M	DDLD	103	65	73	75	73	64	88	8	85	4
11	7,2	M	D	107	76	74	86	73	85	70	6	97	8
12	9,1	F	D	107	90	82	88	82	92	79	3	104	12
13	11,6	F	DDLD	115	80	73	90	82	88	92	9	102	6
14	7,5	M	DDLD	85	70	69	84	67	79	101	5	88	5
15	8,5	M	DDLD	103	95	85	80	83	70	88	5	79	7
16	9,0	M	D	103	73	80	75	58	79	98	6	95	8
17	11,9	M	D	99	92	99	80	64	70	107	7	97	9
18	6,0	M	DDLD	93	87	84	82	107	76	101	10	97	6

1. Kaufman Brief Intelligence Test (KBIT: Kaufman & Kaufman, 2004); 2. Test of Word Reading Efficiency (TOWRE; Torgesen, Wagner, & Rashotte, 2012); 3. Comprehensive Test of Phonological Processing (CTOPP; Wagner, Torgesen, Rashotte, & Pearson,

2013); 4. Clinical Evaluation of Language Fundamentals test (CELF; Semel, Wiig, & Secord, 2006); 5. The Test of Reception of Grammar (TROG: Bishop, 2003a).