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REVIEW ARTICLE

Music Therapy in Childhood Neurodevelopmental Disorders

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Abstract

The global prevalence of neurodevelopmental disorders (NDDs) has been on a rising trend in the past decade. Currently, no definitive treatment exists and the prime focus stays on rehabilitation strategies when it comes to managing children with developmental disabilities. Alternative interventions, therefore, have gained considerable interest among researchers. One such form of intervention utilizes music therapeutically to target diverse neural networks pertaining to cognitive, sensorimotor and language functions. Although various studies have reported conflicting results regarding the therapeutic value of use of music therapy in children with NDDs, the existing body of evidence is encouraging. Largely, children with autism spectrum disorders, cerebral palsy and specific learning disability (dyslexia) have demonstrated a favorable response to some extent. However, no uniform consensus exists among the published studies with respect to acoustic features, type of musical instrument employed, and duration of intervention and outcome measures. Neurophysiological correlates of music training explored by the limited available imaging studies have provided further insight into this complementary treatment approach. In this article, we aimed to provide a comprehensive review of the effect of music in the modulation of various developmental processes and evidence from literature regarding the role of music therapy in the rehabilitation of common neurodevelopmental disorders.

Background

Neurodevelopmental disorders (NDDs) are a group of heterogeneous conditions characterized by delay or disturbance in the acquisition of skills in a variety of developmental domains, including motor, social, language and cognition [1]. Although etiology remains unidentified in many cases, various factors such as perinatal complications (asphyxia, infections, hypoglycemic injury), genetic causes, metabolic errors, exposure to environmental toxins and socio-economic factors can lead to an aberrant development trajectory. Very often, the chronic disabling course of NDDs are complicated by multiple behavioral and medical comorbidities. Rehabilitation in the form of sensorimotor training and tailored behavioral interventions form the standard of care for the majority of the developmental disabilities. None of the approachesoffer a definitive cure and hence complementaryalternative therapies and novel therapeutic approaches are worth exploring.

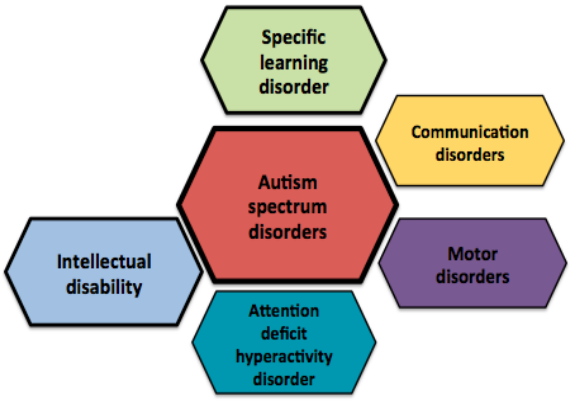


Figure 1: Classification of NDDs as per DSM-5, 2013[1]

Increased survival and thus an increased prevalence of NDD's has been observed in the recent years. A multicentric study by Arora et al. (2018) assessed the prevalence of NDDs in children aged 2-9 years. [2] These included neuromotor impairments including cerebral palsy, ASD, ID, hearing impairment, language disorders, vision impairment and epilepsy. All-site-pooled estimates for NDDs were 9.2% and 13.6% in children of 2-5 and 6-9 year age categories, respectively. Nearly one-fifth of these children had more than one NDD.

According to a systematic review by Zeidan J et al. (2022), one in 100 children worldwide has autism. [3] One in 44 children aged 8 years in the United States was estimated to have ASD as per CDC's Autism and Developmental Disabilities Monitoring (ADDM) network, 2018.[4] The global prevalence rate of SLD ranges from 17.5 percent to 21 percent. [5] Of children with specific learning disorders, 80% present with dyslexia. INCLEN data in 2011 revealed 1.8 million children aged 2-9 years old suffer from learning disorders. [6] Cerebral palsy is the leading cause of childhood disabilities with an estimated pooled prevalence of 2.95 cases per 1000 children in India. [7]

The upsurge in the number of children with developmental disabilities is confounded by multiple factors; such as increased public awareness, standardized universal screening for developmental delay at well-child visits and increased availability of early intervention programs.

The diversified needs of people living with NDDs are reflected in the fact that some of the affected children manifest specific deficits, such as ADHD and specific learning difficulties, which enables them for a self-sustaining life. Others such as children with ASD, ID however may need lifelong assistance even with basic living skills depending on the degree of disability.

Can music benefit children with NDDs?

NDDs have multiple associated co-morbidities apart from the core features, which prohibit optimal performance by these children. Traditional therapy in these children is generally not curative and the focus is on improving quality of life via training and limiting associated co-morbidities. The chronic disabling nature of NDDs poses a significant health and economic burden on families and society, raising the demand for the need of more effective and efficient rehabilitation strategies. Evidence from the literature suggests that music therapy could offer a platform for neuro-rehabilitation.

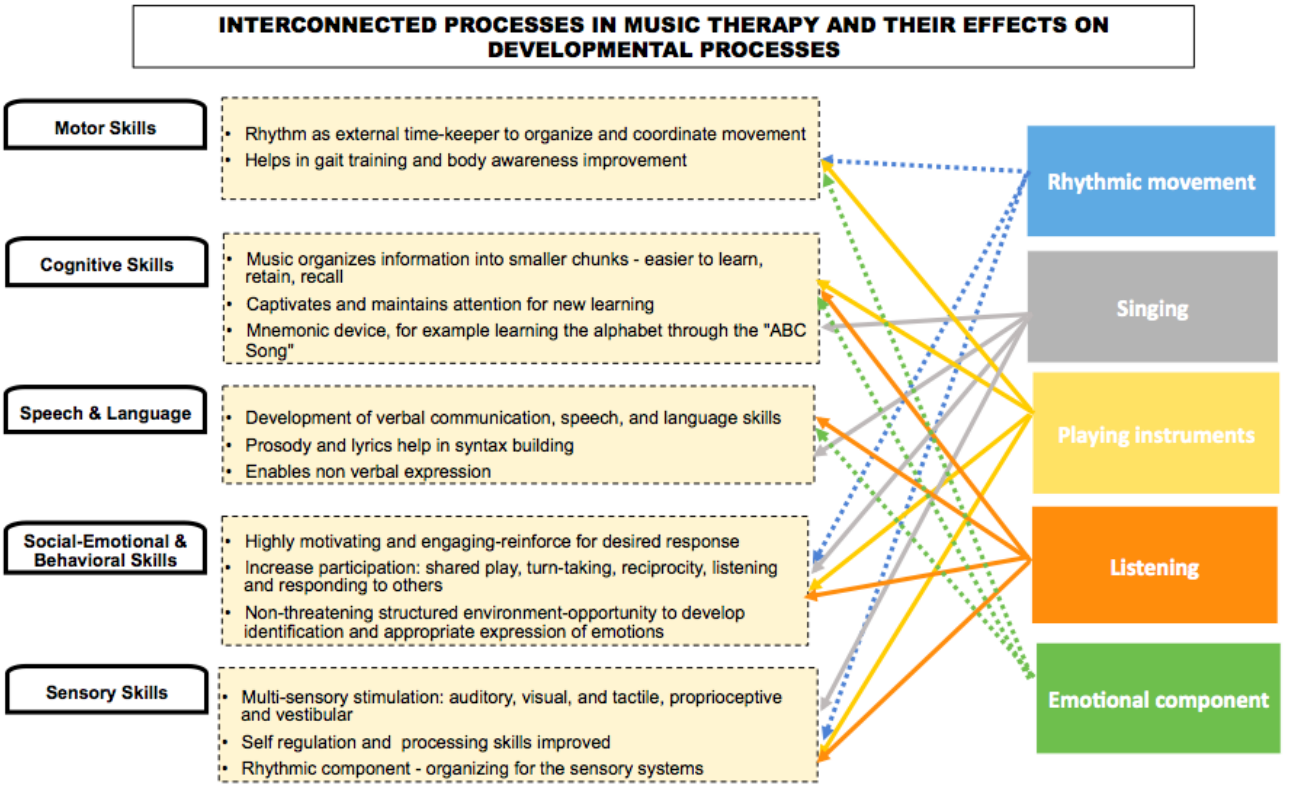
The networks and processes involved in understanding, producing and appreciating music[8], can be used therapeutically to engage and shape non-musical perceptual, cognitive, language, social[9] and sensorimotor functions in these children. (Refer to Figure 2 for a detailed illustration of the effects of music on developmental [10] processes).[11] Animal studies have shown that a music-rich environment induces long-term changes in neurotransmitters and neuroplasticity. [12] Considerable research in musicians and instrumentalists has demonstrated the long-term effect of music therapy in enhancing sensory integration with increased volume and density in the cortex especially

fronto-temporal region and cerebellum as well as improved connectivity [13] of white-matter tracts. [14] Even short-term practice in musical modalities like piano helped improve verbal intelligence and executive function in preschool children. [15] A parallel relationship is also theorized to exist between syntax processing in language[16]and usage of harmonics in music and hence training in music improves speech perception and even pronunciation of non-native languages. [17]

How is music processed in human brain?

Music is an art or rather a scientific process in which sounds and tones are arranged in combinations or sequenced temporally such that they create unity and continuity. These could be vocal, instrumental, mechanical sounds and tones and this arrangement has a rhythm, melody and harmony (Merriam-Webster et al, 2021).

The processing of these harmonious sounds involves extensive complex brain networks. Figure 3 shows a pictorial representation of the activation of cognitive networks during the processing of music. It starts with sound perception by inner ears carried via auditory nerves largely to inferior colliculi of midbrain where basic acoustic features like intensity and periodicity of music are picked up. There onwards there is a further refinement of this information into sound pitch, temporal and spatial variation of notes, via the thalamus to auditory cortex majorly. Keeping continuous track of music requires attention and working memory. Familiar notes involve episodic memory and recall via the hippocampus and parietal regions. Perception of rhythm and active involvement via movement, singing or playing an instrument also involves the somatosensory cortex, basal ganglia and cerebellar networks.



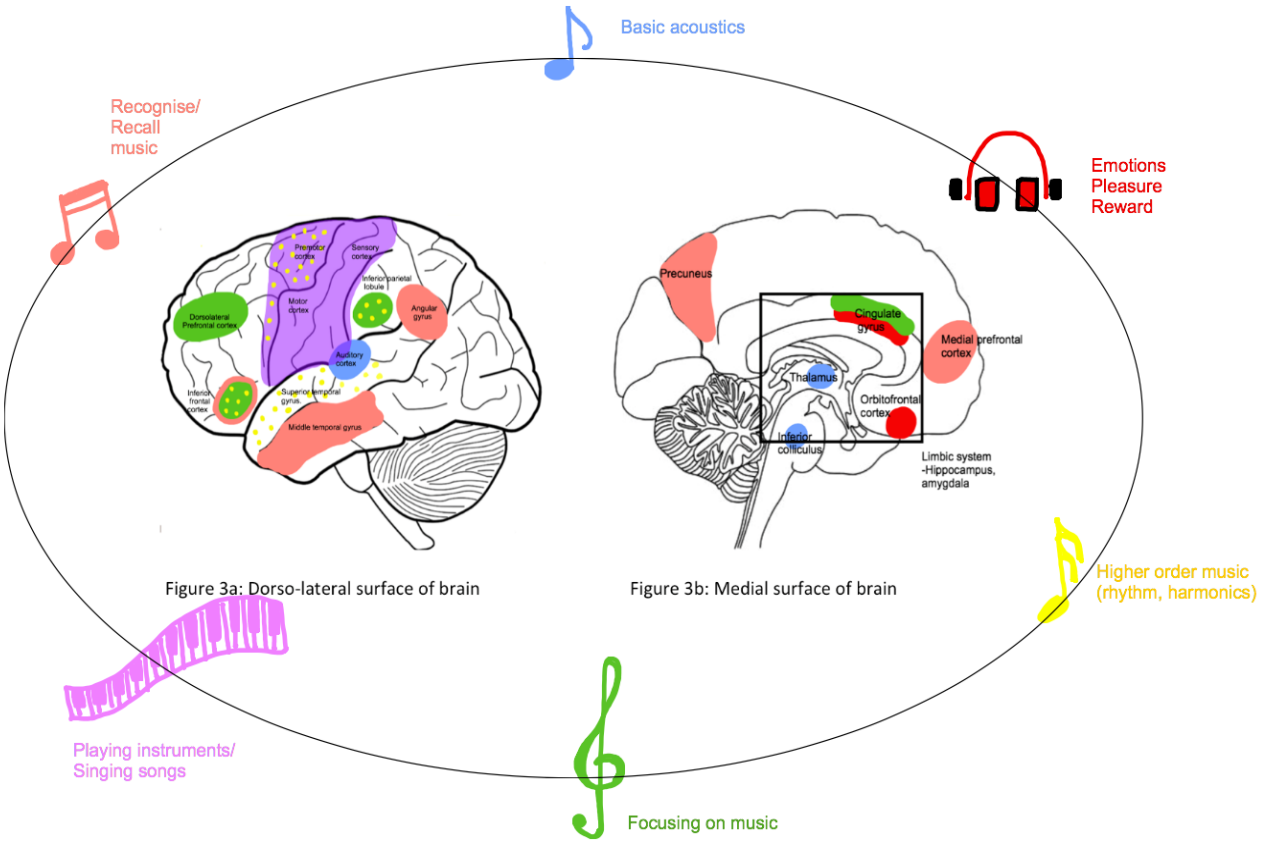


Figure 3a: Dorso-lateral surface of brain

Figure 3b: Medial surface of brain

Basic acoustics: frequency, loudness	Auditory cortex, thalamus, inferior colliculi
Higher order music: rhythm, harmonics	Inferior frontal gyrus, inferior parietal lobule, superior temporal gyrus, premotor cortex
Focusing music	Dorsolateral prefrontal cortex, cingulate gyrus, inferior frontal gyrus, inferior parietal lobule
Recognising and recalling music related memories	Angular gyrus, middle temporal gyrus, hippocampus, precuneus, medial prefrontal cortex
Playing, singing and dancing to music beats	Motor and somatosensory cortices
Music induced emotions and pleasure	Limbic system (hippocampus, amygdala), cingulate gyrus, orbitofrontal cortex

Figure 3: Pictorial illustration of key cognitive and sensorimotor areas activated during music processing

[Adapted from Särkämö T et al. Music perception and cognition: development, neural basis, and rehabilitative use of music. Wiley Interdiscip Rev Cogn Sci. 2013 Jul;4 (4):441-451][10]

Music entails more than just a summation of these acoustic features and other connections to limbic areas (amygdala, orbitofrontal cortex) trigger various emotional processes, which make the experience holistic. [10]

Evidence from literature about music therapy in the rehabilitation of children with neurodevelopmental disorders

Autism spectrum disorder

Autism spectrum disorders are characterized

by core deficits in social communication and interaction associated with repetitive behaviors with restricted interests and activities.[1] Children with ASD often can have other debilitating behaviors (ADHD, challenging behaviors, anxiety disorder, etc.) and medical co-morbidities (epilepsy, sleep issues and gastrointestinal problems). The core features as well as the comorbidities have been traditionally targeted using applied behavior analysis (ABA) therapy primarily with pharmacotherapy which may help alleviate associated symptoms. These traditional approaches are tedious and time consuming and are not curative. This nudges caregivers and adults living with ASD towards complementary alternative therapies. [18] Among these, music therapy has been shown to be an emerging alternative with a growing body of evidence in its favor.[19]

Individuals with ASD have different processing abilities for music as compared to neurotypicals. They show specific interests and talents pertaining to musical modalities with reduced habituation to musical stimuli. They exhibit a strong preference for musical stimuli as compared to verbal stimuli and have superior recognition of pitch. [20] There are alternate mechanisms of processing music as evident from imaging studies (MRI), where people with ASD activated temporal networks during perceptionof sung word similar to controls; but have reduced integrity in language processing networks (fronto-temporal tracts, inferior frontal gyrus, and supramarginal gyrus). In addition to providing an enjoyable, safe outlet for expression it will help other non-musical skills too. Group [21] activities and activities with parents [25] also help in bonding [22] and improve social[21] expression in these children.

Many studies have been published recently on the role of musical therapy and musical training in people with autism. Variable improvements in social aspects, parent-child interactions and bonding, attention span, and improved repetitive behaviors have been described, although large randomized controlled trials fail to replicate the same.[23]Studies have also shown that there are changes in neural circuitry and improved overall core symptoms of ASD. Even population surveys demonstrate the role of antenatal music in preventing ASD like symptoms, although robust data to back these are not yet available[24] A recently published systematic review by Applewhite et al. (2022) has also shown significant heterogeneity in methodologies and outcome variables used in published studies and conclusive evidence for supporting music therapy still is lacking although trend towards beneficial role does exist. The major studies illustrating the effect of music therapy in children with ASD have been tabulated below (Table 1).

Although there have not been any head-to-head trials or designs comparing music therapy with a combination of music and dance therapy, older studies in 2013 by Mateos et al.[25] did not demonstrate any significant benefit. The recent noteworthy study by Bergmann et al. (2021) [26] subjected ASD children to music and combined music-dance therapies, following which significant improvement was noted in social and emotional behavior scales. Another study by Lakes et al. (2019) [27] also reported improved obsessive, compulsive and stereotyped behaviors with the combination of therapies. (Table 2)

Table 1: Brief overview of major studies on music therapy in children with ASD

Author	Methodology (Type of study, sample size, diagnosis)	Intervention	Outcome measures	Results
Ghasemtabar et al. (2015) [28]	Longitudinal study design Children with ASD (n = 27)	MT programs for 45 days in 12 sessions (two sessions of (1-hour/week), whereas the control group received no intervention	SSRS-P: Social Skills Ration System Scale for elementary period	Significant increase in social skills scores post-test ($p < 0.001$)
Srinivasan et al. (2015) [29]	Randomised controlled trial Children with ASD (n = 36)	Two novel interventions - rhythm and robotic therapies, vs standard-of-care	SCQ: Social Communication Questionnaire ADOS: Autism Diagnostic Observation Scale-2 VABS: Vineland Adaptive Behaviour Scale RBS-R: Repetitive Behaviour Scale-Revised	With training, the rhythm group showed a reduction in negative affect and an increase in interest affect and positive affect.
Schwartzberg & Silverman (2016) [30]	Randomized control trial Children with ASD (n = 29)	Cluster randomized to experimental (sung short story) or active control (read aloud short story) groups Daily scheduled music therapy	CC scores: Comprehension check (CC) question scores	Mean CC scores increased from day one to day 3 for both the control and experimental groups. Mean change in CC scores post-intervention: $p < 0.05$

Bieleninik et al. (2017)[23] .	Randomised controlled trial Children with ASD (n = 364)	Enhanced standard care vs Enhanced standard care plus improvisational music therapy in a 1:1 ratio	ADOS- social affect domain Parent-rated social responsiveness.	Children with ASD in MT did not result in significant improvement in mean symptom scores compared to enhanced standard care
Crawford et al. (2017)[31]	Randomised controlled trial Children with ASD, 4-7 years age (n = 364)	Ratio 1 : 1 : 2 Standard care+ One (low-frequency) session of IMT per week Or Standard care+ Three (high-frequency) sessions of IMT per week, Or Standard alone	Social affect score derived from the Autism Diagnostic Observation Schedule (ADOS) at 5 months.	No significant difference in mean scores of ADOS social effect and parent-rated social responsiveness score.
Lense et al. (2020)[32]	Mixed design with survey and experimental design Children with ASD (n = 14) Neurotypical children (n = 14)	Parent-child integrated music class program Serenade Program : 10-week program	ADOS-2, 14-item program evaluation survey, semi-structured interview, video recordings	Improved family well-being with enhancing the value of integrated community participation experiences at the level of the family structure.

Cibrian et al. (2020)[33]	Pilot randomised controlled trial Children with ASD (n = 22)	NMT sessions for 2-month Random allocation to either use an elastic touch-display (experimental group) or tambourines (control group).	DCDQ: Developmental Coordination Disorder Questionnaire PiT: Personality Item Test, timing, synchronisation assessment, the strength control assessment	Significant improvement in coordination with greater control of their movements, p = 0.003
Rabeyron et al. (2020)[34]	Randomised controlled trial Children with ASD (n = 36)	Comparing music therapy (MT) to music listening (ML) ASD aged 4 to 7 years	CGI: Clinical Global Impression CARS: Childhood Autism Rating Scale (CARS) ABC: Aberrant Behavior Checklist (ABC)	Change in CGI scores: p < 0.001; Change in CARS and ABC scores for both groups: p = 0.001
Pedregal& Heaton (2021)[35]	Pilot randomised controlled trial Children with ASD (n = 11)	Adolescents with ASD completed 5 music sessions and pre and post-tests of Alexithymia, emotion recognition and language	BPVS-III: British Picture Vocabulary Scale: Third Edition EAQ: Emotion awareness questionnaire ER: Emotion recognition test	Change in vocal and facial emotional recognition scores, p < 0.01 Change in not hiding emotions score, p < 0.01

Table 2: Brief overview of major studies on music and dance therapy in children with ASD

Author	Methodology (Type of study, sample size, diagnosis)	Intervention	Outcome measures	Results
Mateos-Moreno &Atencia-Dona (2013)[25]	Experimental design Children with ASD (n = 16) Neurotypical children (n = 8)	36 sessions of combined MT and DMT	ECA-R: Evaluation of Autistic Behaviur	Positive trend towards a reduction in scores in both control and experimental groups
Lakes et al. (2019)[27]	Longitudinal study Design Children with ASD (n = 12)	4 week intervention of Creatively Able, a music and movement intervention for children with ASD Given two times per week	RCS: Response to challenge Scale RBS-R: Repetitive Behaviour Scale-Revised; PACES: Physical Activity Enjoyment Scale	Group-level and individual level reduction in stereotypies and compulsive behaviours
Bergmann et al.(2021)[26]	Experimental design Adults with ASD (n = 12)	Autism-Competence-Group (AutCom)-psycho-educative approach with music and dance/ movement. 16structured 90-minute sessions	AutCom: Autism-Competence-Group intervention and Questionnaire Primary outcome variables - social and emotional competence, Secondary outcome-challenging behavior and quality of life	Significant group differences in AutCom questionnaire, p = 0.024

Cerebral palsy

Cerebral palsy (CP) is a group of permanent disorders affecting the development of movement and posture, resulting in activity limitation, which is attributed to the non-progressive disturbances that occurred in the developing fetal or infant brain. A child with cerebral palsy faces tremendous lifetime challenges such as physical disabilities, neurobehavioral concerns, sensory impairment, epilepsy and secondary musculoskeletal problems. The management goals in CP are primarily directed toward a multimodality strategy that focuses on improving functionality and capacities in order to achieve independence.

Sensorimotor training forms an essential building block in the rehabilitation of cerebral palsy. Growing evidence indicates that music-based interventions can accomplish the same by recruiting multiple brain areas that are involved in coordination of extremity function with audio-visual-tactile feedback mechanism during active music playing. Imaging studies have delineated various anatomical, functional, and neuroplastic correlates of aberrant developmental trajectories after brain injury in children with cerebral palsy. These changes affect corticospinal tracts and sensorimotor networks as well. Long-term music practice has been demonstrated to cause structural differences (increased thickness/volume in motor cortex, auditory tracts, corpus callosum, cerebellum), increased white matter connectivity and enhanced functional networking among various sensory modalities, particularly between auditory and somatosensory systems. [36]

Music-supported motor training can be used to achieve a more functional and balanced gait pattern while simultaneously motivating children for active participation in physical therapy sessions. There has been a surge of interest in the functional restoration of the upper extremity in individuals with stroke, cerebral palsy, and Parkinson's disease through musical instrument training. [37] Acoustic musical instruments such as piano, guitar, and percussive instruments have been most commonly used for such rehabilitation purposes. [37] With a focus on hand function, Alves Pinto et al. were the first to employ task-based functional MRI to study the neural correlates of rehabilitation following musical training in CP children (n=10). Increased functional connectivity between the left primary motor cortex and right cerebellum was observed during a finger-tapping test after 18 months of supervised piano training, signifying the neuroplastic effect of music on the neural networks. [38] Recently digital musical devices, such as electronic keyboards, drum pads, and tablets/iPads with commercial music software, paved the way for the development of innovative rehabilitation techniques.[39] Schaffert and colleagues developed and evaluated a novel music-based therapeutic device (SONATA) for upper limb movement training in 21 healthy subjects. [37] This device proved to be feasible and hence opened up novel opportunities for use in restoring motor function.

Table 3: Brief overview of major studies on music therapy in children with cerebral palsy

Author	Methodology (Type of study, sample size, diagnosis)	Intervention	Outcome measures	Results
Alves Pinto et al.(2016) [38]	RCT N=16 (6-16 years) Neuromotor impairments affecting hand function	Individualized and supervised piano lessons twice weekly for 18 months (cases = 10)	Task based functional MRI (finger tapping task) Variability in keystroke of piano	Increased functional connectivity between the left primary motor strip and right cerebellum in patients who received piano training
Caballero E et al. (2018)[40]	RCT N=27 (4-16 years) Severe bilateral cerebral palsy	Neurologic music program of therapeutic instrumental music (TMT) for 16 weeks in addition to its regular physiotherapy (cases = 18)	Overall and specific "Chailey levels of Ability" Locomotor stages	Significant improvement in functionality: overall and specific "arm and hand position", activities from the "Chailey Levels of Ability" and the locomotor stages were observed in the intervention group, persisting for more than 4 months.
Santonja-Medina CS et al. (2022)[41]	Analytic quasi-experimental study N=17 Severe cerebral palsy	Motor learning through therapeutic instrumental music performance (TIMP), using percussion instruments Once weekly for 4 months(total 13 sessions)	Hoisan software video recording for quantifying participation in creating music	Significant improvements: "visual contact", "motor participation", "motor participation repetition". Significant differences were also observed in the subcategories: "reaching and stroke," "hitting with the hand" and "grasping and hitting."

Specific learning disability

Dyslexia is defined as an unexpected difficulty in reading in an individual who has the intelligence to be a much better reader; dyslexia reflects a difficulty in getting to the individual sounds of spoken language which typically impacts speaking (word retrieval), reading (accuracy and fluency), spelling, and often, learning a second language.[42]Dyslexia is, at its core a problem with phonological processing: that is getting to the elemental sounds of spoken language (phonemes), affecting both spoken and written language. Interestingly this phonological deficit has been attributed to faulty temporal processing skills by many researchers. Children with developmental dyslexia may benefit from music based on the hypothesis of a positive correlation between musical perceptual abilities and phonological awareness and reading skills.

The majority of the evidence regarding the role of educational music therapy as a remedial tool for children with learning difficulties came from two randomized controlled trials that evaluated. Register and colleagues showed that short-term instructional program incorporated with music for 4 weeks improved reading abilities, particularly reading comprehension, word vocabulary and word knowledge in 8 students with dyslexia.[43]Children with dyslexia showed enhanced phonological awareness and reading abilities (pseudoword accuracy, working memory, auditory attention) after participating in school-based music therapy group sessions for 30 weeks (Flaugnacco et al. 2016).[44]A favorable response in the blending subset of phonological awareness showed a positive correlation with rhythmic skills.

Table 4: Brief overview of major studies on music therapy in children with specific learning disability (dyslexia)

Author	Methodology (Type of study, sample size, diagnosis)	Intervention	Outcome measures	Results
Register et al.(2007)[43]	RCT N=33 (8-dyslexia, rest 2nd grade students) SLD	Musical program thrice weekly for 4 weeks (cases) Traditional program (controls)	Vocabulary and reading comprehension tests	Significant improvement in reading comprehension in SLD group

Flaugnacco et al.(2016) [44]	RCT N=48 (8-11 years) Dyslexia	Music (percussive instruments) &sensorimotor training (cases): twice weekly for 30 weeks Painting (controls)	Phonological awareness (phonemic blending/ segmentation) Reading abilities (reading loud: text/single words/ pseudowords)	Better improvement in text reading, pseudowords accuracy, working memory, auditory attention and phonological abilities in cases as compared to controls.

Attention deficit and hyperactivity disorder

The core deficit in ADHD is related to a primary deficit in behavioral inhibition and executive control. Children with ADHD who struggle with impulsivity and inattentiveness can benefit from the rhythmic, emotional, and motivational elements of music therapy. The results of a survey of 268 music therapists regarding the role of the use of music in the treatment of ADHD in preschool and school-aged children demonstrated a favorable response. [45]In a study by Montello and Coons (1998), educational rhythm-based music interventions reduced hostility and inattention in school children with problem behavior as per teacher reports.[46] Rothman et al. (2014) showed similar results of increased attention span and better quality of life in ADHD children who received music sessions with percussion instruments and musical games for 18 weeks.[47]

Table 5: Brief overview of major studies on music therapy in children with ADHD

Author	**Methodology (Type of study, sample size, diagnosis)**	**Intervention**	**Outcome measures**	**Results**
Montello and Coons (1998)[46]	Pre-post evaluation N=16 (11-14 boys) Attention deficit disorder, learning disability, emotional issues	Rhythm based music intervention once weekly for 12 weeks (cases)	Teachers report: inattention, hostility and motivation	Significant improvement in rhythm based intervention group
Jackson et al. (2003)[45]	Survey (questionnaire based) among 268 music therapists	Several types of music interventions	Perceived effectiveness of music in ADHD	Music therapy was effective in ADHD in the majority of cases

Rothman et al. (2014) [47]	Case control study N=101 (5-10 years) ADHD	Music exercises using percussion instruments and musical games once a week for 18 weeks (cases)	Attention Quality of life ratings ADHD/conduct disorder symptom checklist	Significant improvement in attention and quality of life

Intellectual Disability

Intellectually disabled children have significant impairment both in intellectual functioning and in adaptive behavior domains as reflected in personal, social-adaptive and academic/occupational skills. Similar to other NDDs, no curative treatment exists.

A handful of studies, mainly pre-post and observational studies are available in the literature, which assessed the effect of music therapy in children with developmental delay or intellectual disability. Williams et al. (2012) reported an improvement in social skills and parent-child interactions but with persistent child problem behavior following a 10-week music therapy session.[48] Educational music therapy improved social communication skills when applied in a classroom setting for 15 weeks in a study by Mendelson et al. (2016).[49] On the contrary, observational studies (Aldridge et al. 1995, Duffy and Fuller et al.2001) failed to show any positive effect of music in children with developmental disabilities.

Table 6: Brief overview of major studies on music therapy in children with intellectual disability

Author	**Methodology (Type of study, sample size, diagnosis)**	**Intervention**	**Outcome measures**	**Results**
Williams et al. (2012) [48]	Pre-post evaluation, N=201 (3-60 months) Global delay/ASD/ Specific language impairment	Singing songs, playing instruments: weekly for 10 weeks	Parent reported assessments: child parent interactions, social play skills, receptive communication skills, parenting	Improvement in child social engagement and child-parent bonding

Mendelson et al.(2016) [49]	Pre-post evaluation N=33 ID	Interactive music therapy (songs), weekly for 7 weeks or 15 weeks	Teachers' ratings: Social Skills Improvement System-Rating Scale (SSIS-RS) Behavioral observations	SSIS-RS: no significant results. Social/ communicative responses improved during long-term therapy
Duffy and Fuller et al. (2001) [50]	Case control study, N=32, ID	Group music therapy twice weekly for 8 weeks (cases) Non-music therapy (controls)	Social skills evaluation by analysis of videotaped sessions	No significant difference between cases and controls

Scope for future

There has been a surge of interest in this field of alternative therapy and exploratory evidence does suggest some improvement. However, there is a major lacuna of well-powered studies with meticulous design to conclusively recommend or refute the use of music in NDDs. Largely no major harms have been reported in any of the studies.

Music therapy to a large extent is individualized but good practice guidelines for planning of therapy, structured recommendations for the type of therapy to be used, type of music, music combined with movement therapy, use of artificial intelligence vs. therapists role in music therapy is still unclear at this point of time. These are a few of the many variables, which require further exploration.

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