

Review

Therapeutic Use of Music, Dance, and Rhythmic Auditory Cueing for Patients with Huntington's Disease: A Systematic Review

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Abstract.

Background: Studies have assessed the therapeutic effect of music, dance, and rhythmic auditory cueing for patients with Huntington's disease (HD). However, the synthesis of evidence in support of their positive impact on symptoms is lacking.

Objective: We conducted a systematic literature review to evaluate the potential benefits of music, dance, and rhythm on the cognitive, psychiatric and motor function in patients with HD.

Methods: Two- and three-keyword searches and a manual search identified medical literature published from 1999 through 2019. We considered literature that assessed outcomes of art-based rehabilitation programs or individual modalities for persons with early, middle, or advanced HD. Structured analysis was conducted using data entry tables with categories for patient health status, art methods, and outcomes.

Results: Seven articles and six abstracts met eligibility criteria, of which nine evaluated art-based rehabilitation programs. Studies mainly assessed cognitive, psychiatric, and motor functions through music, dance, or rhythm modalities. Although results were conflicting, in summary improvements to motor function were dependent on disease severity and more responsive to art therapy programs than rhythm-motor synchronization. Benefits to global cognition that resulted from rhythmic training correlated with microstructural changes. Qualitative data verified a positive impact on language production, chorea, behavior, and quality of life.

Conclusion: Our review has shown a potential benefit of music, dance, and rhythm for patients with HD, which is particularly important for a disease that has no cure. Art forms seemed to affect cognitive, psychiatric, motor, psychosocial, and neuroanatomical domains. However, evidence is preliminary, warranting further investigation to establish the foundation for this field.

Keyword: Music therapy, dance therapy, art therapy, Huntington's disease

INTRODUCTION

Huntington's disease (HD) is an autosomal dominant neurodegenerative disorder characterized by striatal and basal ganglion atrophy [1–4]. Cogni-

tive, psychiatric, and motor dysfunctions progress through early, middle, and advanced phases that correspond to five HD stages on the Shoulson and Fahn Total Functional Capacity (TFC) scale [5–7]. Striatal neuropathology is associated with cognitive and psychiatric functions [8–10] that often precede motor deficits [8–12]. Cognitive impairments are observed in executive function, memory, affective processing, and psychomotor speed [7, 13–15]. Psychiatric

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symptoms, including irritability, depression, anxiety, apathy, disinhibition, and emotion dysregulation, are common in HD [16–19]. As the clinical hallmark of HD [20], motor impairments initially impact voluntary motor functions such as gait and balance [21] and evolve to involuntary choreiform movements [20]. The triad of HD symptoms are interconnected as a result of cortical loops that coordinate cognitive, emotional, and motor functions [22].

Factors of disease presentation, such as age at onset, rate of decline, and severity of symptoms vary among the HD cohort [7]. However, since there is currently no cure or disease-modifying therapy [23], there is a consistent need for long-term, multidisciplinary care through pharmacological [24, 25] and non-pharmacological [26, 27] interventions. These methods may reduce or stabilize disease progression in other progressive neurodegenerative disorders, such as Parkinson's disease (PD) [7]. Recent studies have investigated the use of physical therapy for HD patients [28–32]. Multidisciplinary rehabilitation programs are safe and feasible interventions that positively affect verbal learning and memory, anxiety and depression, quality of life, balance, spatial-temporal gait parameters, and gray matter changes [33–36].

In addition to physical exercise, there may be beneficial health effects of using art such as music and dance in patients with neurological diseases [37–39]. Some of these effects could potentially arise from enhanced neuroplasticity; however, empirical data is lacking. Specifically, pathways of emotion are activated in localized brain areas such as the amygdala, cingulate cortex, and hypothalamus [38]. In addition, the use of sensorimotor processing in art interventions may strengthen synaptic connections between auditory and motor cortices and increase localized areas such as the corpus callosum and basal ganglia [37]. Neuroplastic features are specific for the therapeutic modality. For example, listening to music improves global neuroconnectivity and atrophic recovery, while playing a musical instrument, singing, or dancing increases gray and white matter in frontotemporal areas and alters neurochemical levels including endorphins and dopamine [37, 38, 40]. As a result, music (MT) and dance movement therapies (DMT) in conjunction with conventional clinical interventions have a potential to positively affect cognitive, psychiatric, and motor functions for persons with progressive neurodegenerative disease. Studies give a particular focus on PD patients and associated benefits from dance exercise programs that use MT and rhythmic auditory cueing [39, 40].

Findings show improved motor control, gait, spatial memory, functional well-being, and quality of life [41–44]. The rationale for using MT and DMT in rehabilitation programs is the idea that music and dance are specialized auditory and motor forms that induce specific neuroplastic changes [37]. Therefore, the therapeutic outcomes associated with art-based rehabilitation are potentially distinct to that of traditional physiotherapy [37, 45–48].

Given the positive effects of art-based rehabilitation programs for persons with other neurodegenerative diseases [49], there may be similar benefits for patients with HD. However, few studies have examined MT, DMT, or rhythmic auditory cueing in clinical practices specific to HD, rendering us unable to define potential therapeutic outcomes associated with art-based rehabilitation in patients with HD. In the past decade, there has been a slight increase in the number of HD studies investigating MT protocol proposals [50], MT assessment tools [51], clinicians' experiences of MT [52–54], and music perception abilities [55]. But yet, there lacks a greater focus on MT/DMT outcomes for persons with HD in comparison to other neurodegenerative diseases.

In 2015, a systematic review on the use of MT for patients with HD was published [56]. However, it reviews studies published from 1976 to 2001, including one study published in 2007, and focuses primarily on the history of MT and MT methods. As such, there is a need for an up-to-date systematic review of the literature in the past two decades that includes the cognitive, emotional, and motor outcomes of MT, DMT and rhythmic auditory cueing for HD. We did a systematic review to study the formats and effects reported in the literature on the therapeutic use of music, dance, and rhythmic auditory cueing for patients with HD.

MATERIALS AND METHODS

The systematic review adhered to the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) statement guidelines [57].

Search strategy for study selection

A literature search limited to English-language studies published from 1999 through October 2018 was first performed in November 2018 using PubMed, Scopus, and Web of Science electronic medical databases. Additionally, we conducted a manual search of included studies and systematic

reviews in EMBASE. We repeated the literature search in April 2019, which was limited to English-language studies published from 1999 through 2019. This yielded one additional study for inclusion. The search strategy utilized a two- and three-keyword search. The two-keyword search used the following terms: (“Huntington’s [disease]”) AND (“music” OR “music therapy” OR “dance” OR “dance exercise” OR “dance rehabilitation” OR “[rhythm] [auditory] cueing”). The three-keyword search used the following terms: (“Huntington’s [disease]”) AND (“motor [function]” OR “cognitive [function]” OR “psychiatric [function]” OR “quality of life”) AND (“music [therapy]” OR “dance [therapy]” OR “[rhythm] [auditory] cueing”). A total of 278 searches were gathered, of which 184 were duplicates, yielding 94 unique search results.

Eligibility criteria

Included studies met the following criteria: 1) human participants received a genetically or clinically confirmed HD diagnosis 2) the therapeutic effect of art-based rehabilitation was evaluated quantitatively or qualitatively and 3) published research was an article, book chapter, or conference abstract.

This review considered protocols that incorporated MT, DMT, and rhythmic auditory cueing modalities for patients with prodromal, early, middle, or advanced HD and their caregivers. The rationale for including MT, DMT, and rhythmic auditory cueing is that each art intervention incorporates different methodologies. For example, MT and DMT utilize music and dance, respectively, to target domains of function. Conversely, rhythmic auditory cueing is a form of rhythmic training that synchronizes specific functional domains, such as speech and motor movements, with rhythmic patterns.

Studies that were carried out in the community (i.e., public dance studios), patients’ homes, rehabilitation centers, or hospitals were also considered. Quantitative syntheses included cognitive, psychiatric, and motor outcomes. Outcome measures of cognitive function included verbal communication, psychomotor speed, and executive functions. Outcome measures of psychiatric function included emotional regulation and levels of depression, anxiety, apathy, and aggression. Outcome measures of motor function included balance, spatial-temporal gait parameters, and fine motor skills. Studies that reported microstructural out-

comes and psychosocial outcomes (i.e., quality of life) were also considered. Qualitative syntheses included patient and/or caregiver feedback regarding the efficacy, safety, and feasibility of the program.

Exclusion criteria were proposals of study designs, assessments of clinical protocols, evaluations of novel measurement tools, and systematic reviews or meta-analyses. Of the 94 unique literature searches, 81 met at least one exclusion criterion, yielding 13 total studies for inclusion in the systematic review (Table 1). The flow chart of the search process and outcome is shown in Fig. 1.

Data extraction

After reviewing 36 abstracts and 58 full texts, 30 and 51 studies were excluded, respectively. 56 excluded studies neglected to examine therapeutic effects of art. Participants of 49 excluded studies were either clinicians, persons with other neurodegenerative diseases, or HD animal models. 9 studies were excluded due to literature format (i.e., study protocol proposal, systematic review). Data was extracted from published conference abstracts as a result of the lack of literature.

Outputs for keyword combinations in each database were recorded in structured tables during the search process. Data entry forms had the following categories: research setting, aims, study design, procedure, participant sample (sample size), participant age and gender (mean \pm SD), art form, measurement tools, outcome measures, and results.

Data synthesis and analysis

Due to the limited number of studies that assessed the cognitive, psychiatric, and motor outcomes of MT, DMT, or rhythmic auditory cueing for HD patients, findings from included studies were synthesized and summarized as descriptive results.

RESULTS

This systematic review included seven research articles published between 1999 and 2019 in English-language journals (Table 1). Data was collected from six conference abstracts (see Tables 1–3) but missing information led to its exclusion from the primary synthesis.

Table 1
Design and study population of seven articles and six abstracts on the therapeutic use of art

Source (first author, year publication reference number)	Descriptive characteristics	Literature format	Country	Setting	Study design	Outcome measures and assessment method	Participant information	Sample (sample size); age and gender	HD stage
Brandt M., 2016 [58]		Conference abstract	Netherlands		Case report	<p>Outcomes:</p> <p>1) Cognitive: language processing</p> <p>2) Psychiatric: levels of anxiety or fear pertaining to the daily use of speech</p> <p>3) Motor: components of language production (i.e., articulation, fluency, use of voice, speaking volume, breathing)</p> <p>4) Psychosocial: quality of life as a result of daily use of speech</p> <p>Tools: Subjective assessment of program efficacy and perceived benefits through patient feedback</p>	HD patients	Patients were able to speak but had verbal communication deficits (speech unintelligible); had intact language comprehension; were motivated and not easily distracted; had affinity for music	
Delval A., 2008 [59]		Article	France		Cross-sectional study	<p>Outcomes:</p> <p>1) Motor: spatial kinematic gait parameters (i.e., stride length, velocity); temporal kinematic gait parameters (i.e., cadence, ratio of single-limb support time to double-limb support time); kinematic angle parameters (i.e., sagittal plane excursion angles for hip, knee, and ankle joints); frequency of the center of mass of knee and ankle joint trajectories; time between heel strikes and metronomic cue</p> <p>Tools: Video motion system: (VICON video system, Oxford Metrics, Oxford UK)</p>	<p>1) HD patients ($n = 15$): mean age = 43.9 ± 9.8 years; sex ratio = 0.52</p> <p>2) Healthy controls matched by age and gender ($n = 15$): mean age = 40.5 ± 10.5 years; sex ratio = 0.52</p>	Patients could perform dual-motor and motor-cognitive tasks	

Hyson C., 2005 [60]	Conference abstract	United States of America (USA)	Non-randomized 6-week pilot clinical trial	<p>Outcomes:</p> <p>1) Cognitive: memory and attention</p> <p>2) Psychiatric: mood</p> <p>3) Motor: balance and posture, fine motor skills, vocalization</p> <p>Tools: Subjective assessment of MT and program tolerability through survey</p> <p>Cognitive, Psychiatric, Motor: UHDRS</p>	HD patients ($n = 5$), 4 male	Patients could adhere to and participate in program
Johnson K.A., 2000 [61]	Article	Australia	Cross-sectional study	<p>Outcomes:</p> <p>1) Motor: bimanual in-phase and anti-phase hand movement parameters (i.e., variation in and accuracy of co-ordination pattern and velocity)</p> <p>Tools: Bimanual cranks were used to perform rotary hand movements; automatic algorithms used displacement data gathered by laptop computer to determine hand movement parameters</p>	<p>1) HD patients ($n = 12$): mean age = 53.7 ± 8.23 years; 9 male</p> <p>2) Healthy controls ($n = 12$): mean age = 54.2 ± 7.82 years; 9 male</p>	Patients were in stages I, II, or III (TFC scale)
Kloos A.D., 2013 [62]	Article	USA	Cross-over, controlled, single-blinded, six-week trial	<p>Outcomes:</p> <p>1) Psychiatric: balance confidence</p> <p>2) Motor: spatiotemporal gait parameters (i.e., speed, maneuverability, forward and backward variability); balance and mobility</p> <p>3) Psychosocial: quality of life</p> <p>4) Other: program feasibility, adherence, acceptability, safety</p>	HD patients ($n = 18$); mean age = 50.7 ± 14.7 years; 7 male	Ambulatory patients represented "a wide range of disease severity"; patients could walk 10 m without assistance

(Continued)

Table 1
(Continued)

Source (first author, year publication reference number)	Descriptive characteristics	Literature format	Country	Setting	Study design	Outcome measures and assessment method	Participant information	Sample (sample size); age and gender	HD stage
Lagessen S., 2011 [63]		Conference Abstract	Norway	NKS Olaviken Hospital (specialized nursing home for persons with advanced HD)	Case report	<p>Tools:</p> <p>Psychiatric: Activities-Specific Balance Confidence Scale</p> <p>Motor: GAITRite walkway system (CIR Systems, Inc, Haverton, PA), FSST and TT</p> <p>Psychosocial: WHO-QoL</p> <p>Other: subjective assessment of program acceptability through patient feedback; physiological parameters assessed program safety</p> <p>Outcomes:</p> <p>Case 1:</p> <p>1) Psychiatric: behavioral control</p> <p>Case 2:</p> <p>2) Motor: Speech and articulation, choreiform movements</p> <p>Tools: Subjective assessment of program efficacy and perceived benefits through clinician feedback</p>	HD patients (n=2)	Patients were in advanced phase	
Metzler-Baddeley C., 2014 [64]		Article	United Kingdom	Patients' homes and the Cardiff University Brain Research Imaging Centre	Non-randomized 8-week pilot clinical trial	<p>Outcomes:</p> <p>1) Cognitive: Executive functions (i.e., multi-tasking, attention switching and inhibition, verbal working memory, verbal and category fluency, processing speed)</p> <p>2) Neuroanatomical: Subcortical basal ganglia volume</p> <p>Tools:</p> <p>Cognitive: standard dual task, TMT, Stroop task, DSST, Delis and Kaplan executive function battery</p>	HD patients (n = 10); age range = 24–64 years; 5 male	Patients had prodromal HD (n = 1) or early to advanced HD (n = 9)	

Muto K., 2005 [65]	Conference Abstract	Japan	Pilot observation study	<p>Neuroanatomy: Signa HDx3.0T/M MRI system (GE Medical Systems, Milwaukee)</p> <p>Outcomes: 1) Psychiatric: levels of tension and fear related to developing or having HD motor symptoms; caregivers' awareness (empathy) of patients' emotional wellbeing 2) Psychosocial: participant satisfaction</p> <p>Tools: Subjective assessment of participant satisfaction through survey and perceived benefits through structured discussions and clinicians' observations</p>	<p>Workshop groups included patients with HD, persons at risk for HD, and their family members ($n = 13-15$ per group); age range = 3-70 years</p> <p>Patients were able to adhere to workshop protocol and provide verbal feedback</p>
Orejas M.E., 2008 [66]	Conference Abstract	Spain	6-month RCT	<p>Outcomes: 1) Psychiatric: UHDRS-behavioral scale functions (<i>outcomes unspecified</i>) 2) Motor: UHDRS-motor scale functions (<i>outcomes unspecified</i>) 3) Psychosocial: life quality satisfaction, functional ability</p> <p>Tools: subjective assessment of satisfaction through questionnaire Psychosocial: PDQ-39, NHQ, FIM and Barthel scales</p>	<p>1) HD patients in treatment group ($n = 7$) 2) HD patients in placebo group</p> <p>Patients were able to adhere to exercise programs and provide feedback</p>
Salgues J., 2016 [67]	Conference Abstract	France	Pilot observation study	<p>Outcomes: 1) Cognitive: development of consciousness 2) Psychiatric: level of relaxation</p>	<p>Workshops included HD patients and their relatives or caregivers</p>

(Continued)

Table 1
(Continued)

Source (first author, year publication reference number)	Descriptive characteristics	Literature format	Country	Setting	Study design	Outcome measures and assessment method	Participant information	Sample (sample size); age and gender	HD stage
Thaut M.H., 1999 [68]		Article	USA	Gait training area of Neurologic Rehabilitation Centre	Cross-sectional study	<p>Tools: Subjective assessment of program efficacy and perceived benefits through instructor's observations</p> <p>Outcomes:</p> <p>1) Motor: uncted and cued gait patterns (i.e., modulation of gait velocity as a function of step rates and stride length, synchronization between step cadence and rhythmic cue)</p> <p>Tools: Stride analysis system (Infotronic CDG, Eindhoven, The Netherlands) assessed stride parameters (i.e., gait velocity, cadence, stride length swing symmetry)</p>	HD patients ($n = 27$); mean age = 47 ± 10.7 years; 13 male	Patients had prodromal HD ($n = 4$) or HD of various phases ($n = 23$) (Shoulson-Fahn scores of degree of chorea and functional disability ranged from 0–2.5 on a 0–3 scale)	
Trinkler L., 2019 [69]		Article	France	Contemporary Dance Studio	Randomized Controlled pilot study	<p>Outcomes:</p> <p>1) Cognitive: UHDRS cognitive score (i.e., literal fluency)</p> <p>2) Psychiatric: Apathy and Problem behavior</p> <p>3) Motor: UHDRS motor score</p> <p>4) Neuroanatomical: brain volume and structural changes</p> <p>5) Psychosocial: quality of life, patient experiences of DMT</p>	<p>1) HD patients ($n = 19$); age range 43–78 years; eight male</p> <p>2) Healthy age- and education matched controls ($n = 12$); age range 44–72 years; four male</p>	Patients had mild to moderate HD (TFC range 7–13, UHDRS motor score range 3–58)	

<p>Van Bruggen-Ruff M.C., 2017 [70]</p>	<p>Article</p>	<p>Netherlands</p>	<p>Long-term care facilities</p>	<p>Multi-center RCT</p>	<p>Tools: Cognitive: Stroop task, Symbol Digit Code, MDRS, TMT Psychiatric: LARS, PBA-s Neuroanatomy: sMRI and VBM Psychosocial: QLI, semi-structured interview Outcomes: 1) Cognitive: social-cognitive functioning (i.e., communicative and expressive skills) 2) Psychiatric: behavioral problems (i.e., mental rigidity, aggression) Tools: Cognitive: BOSH Psychiatric: PBA-s and BOSH</p>	<p>1) HD patients in intervention group (<i>n</i> = 32); mean age = 54.5 years; 10 male</p>	<p>Patients had advanced HD (<i>n</i> = 63)</p>
<p>2) HD patients in control group (<i>n</i> = 31); mean age = 54.3 years; 10 male</p>							

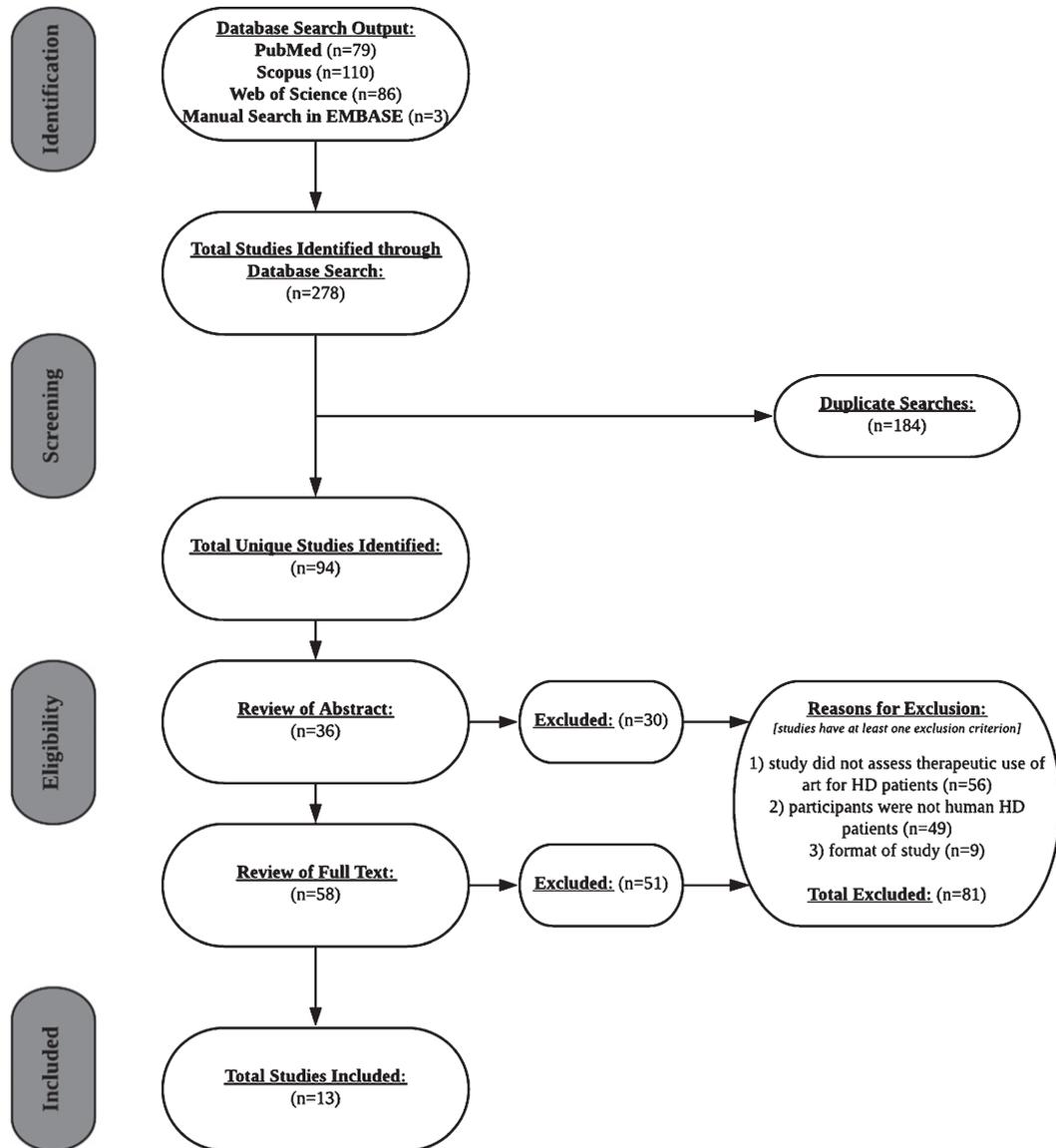


Fig. 1. PRISMA flow chart of the selection process of included studies.

Descriptive characteristics of included studies

Studies originated from North American, European, and Australian countries. Research settings included participants' homes ($n=2$), a dance studio ($n=1$), long-term care facilities ($n=1$), and rehabilitation centers ($n=1$). Two studies did not specify research setting. Cross-sectional studies [59, 61, 68], randomized controlled trials (RCT) [69, 70], cross-over [62], and non-randomized [64] pilot clinical trials were included. Studies assessed a combination of cognitive ($n=3$), psychiatric ($n=3$), motor ($n=5$), psychosocial ($n=2$), and microstructural

($n=2$) outcomes. One study evaluated program feasibility and safety [62]. Outcomes were quantitatively [59, 61, 62, 64, 68–70] and/or qualitatively [62, 69] obtained.

Participants

Studies included patients with HD [62, 64, 68] or compared patients to age-matched healthy control subjects [59, 61, 69, 70]. Early, middle, and advanced phases of HD were represented [61, 64, 68–70]. Two studies did not specify disease phase but provided information on patient functionality.

Participants included both males and females and were in early-to-middle adulthood (Table 1).

Research aims and procedures

The effect of MT, DMT, and rhythmic auditory cueing for patients with HD was examined by evaluating cognitive [64, 69, 70], psychiatric [62, 69, 70], motor [59, 61, 62, 68, 69], psychosocial [62, 69], and/or neuroanatomical [64, 69] changes in response to art-based clinical programs [62, 64, 69, 70] or external rhythmic cueing [59, 61, 68] (Tables 2 and 3).

Functional outcomes of two programs were compared to control treatments without art [62, 69, 70]. All studies that independently assessed rhythm compared gait parameters [59, 68] or bimanual hand coordination [61] across cueing conditions and health status.

Features of MT, DMT, and rhythmic auditory cueing

Art modalities, genres, and instruments

Studies evaluated a combination of music ($n = 4$), dance ($n = 1$), and rhythm ($n = 5$) modalities (Table 2). Rhythmic patterns included Brazilian samba, Spanish rumba, West-African kuku, and Cuban son [64] as well as folk music [68] and contemporary dance [69]. All rhythmic patterns were external auditory cues since they required the use of an external source, such as an instrument or metronome, as opposed to an internal source, such as singing. One study allowed participants to select their preferred music genres [62]. Studies used instruments such as bongo drums [64], metronomes [59, 61, 68], or computerized melodies [62]. These instrumental features distinguished the use of musical from non-musical stimuli, which are considered to be independent forms of auditory information [37]. In addition, the use of instruments to create live music or computerized music may influence the emotional salience of auditory input [37]. One study lacked this information [70].

Neurologic art therapy techniques

Included studies lacked information on the use of art therapy techniques. However, specific MT protocols were identified in abstracts [58, 60, 63]. Of these studies, two used MT for speech and language rehabilitation via melodic intonation therapy (MIT) [63, 71] and modified-MIT in the form of the Speech Music Therapy for Aphasia (SMTA) program

[58, 72]. MT was additionally used for sensorimotor rehabilitation by combining rhythmic auditory stimulation (RAS), patterned sensory enhancement (PSE), and therapeutic instrumental music performance (TIMP) techniques into one clinical program [60, 73, 74].

Methods (structured direction or improvisation)

All studies required participants to follow structured directions administered by an in-person instructor [69, 70], a pre-recorded tape [62, 64], or set rhythmic patterns for motor synchronization [59, 61, 68]. Two clinical programs combined structured direction and improvisation techniques by using a structured protocol that allowed participants to engage in artistic creativity through dance [69] and music [70].

Methods (individual or group sessions)

Studies that evaluated rhythmic auditory cueing independently from a clinical program administered protocol individually to participants [59, 61, 68]. In studies that assessed the efficacy of a DMT program, individual [62] and group sessions [69] were utilized. Two of the included studies chose individual or group sessions based on patients' needs [64, 70].

Methods (instructor and participant roles)

Instructor roles ranged from dictating rhythmic patterns for participants to follow through movement [59, 61, 68] and MT [64] to guiding program safety [62] and structure [69, 70] as participants took the lead. Participants coordinated movements to external cues [59, 61, 68], danced [62, 69], listened to music [70], or played instruments [64].

Cognitive function outcomes

One non-randomized pilot clinical trial evaluated changes to verbal working memory and attention, multi-tasking, verbal/category fluency, and processing speed that resulted from participation in a rhythmic drumming exercise program [64] (Table 1). Measurement tools included the standard dual task [75], Trails Making Test (TMT) [75], Stroop task [76], Digit Symbol Substitution Test (DSST) [77], and Delis and Kaplan executive function battery [78]. Findings showed significant improvements to memory and attention, verbal/category fluency, and DSST post-baseline accuracy scores as well as TMT and Stroop task response times [64] (Table 3). Another multi-center RCT compared MT to recreational

Table 2
 Characteristics of art forms in seven articles and six abstracts

Source (first author, year of publication reference number)	Art modality	Art genres	Instruments	Methods	Individual OR group	Instructor and participant roles
	Music	Dance	Rhythm auditory cueing	Structured direction OR improvisation		
Brandt M., 2016 [58]	X		Piano	Structured Direction: participants dictated phrases in coordination with repetitive rhythmic melodies	Individual	Instructor: music therapist performed piano melodies while speech therapist dictated phrases for patient to verbalize Participant: patient produced speech by following music therapist and speech therapist
Delval A., 2008 [59]			Metronome	Structured Direction: participants synchronized gait to set rhythmic tempos	Individual	Instructor: investigators provided rhythmic cues to participants Participant: patients synchronized gait to cues while performing motor and cognitive tasks; selected comfortable tempo for cues through their baseline pace
Hyson C., 2005 [60]	X	X	Musical instruments (details unspecified)	Structured Direction: RAS used to synchronize gait to external auditory cues Improvisation: PSE technique applied acoustical properties of music to non-rhythmic body movements; TAMP allowed participants to engage in the free-form playing of musical instruments	Individual and Group	Instructor: led rhythmic auditory cueing exercises and supervised PSE and TAMP Participant: synchronized gait to rhythmic cues and played music instruments
Johnson K.A., 2000 [61]			Metronome	Structured Direction: participants synchronized hand movements to set rhythmic tempos	Individual	Instructor: investigators provided rhythmic cues to participants Participant: synchronized hand movements to cues

Kloos A.D., 2013 [62]	X	X	X	Computerized melodies on video game software; footpad with arrows were used to coordinate dance movements with arrow visual cues on screen	Structured Direction: dance movements were directed by arrow visual cues and auditory rhythmic cues	Individual	Instructor: investigators assisted in selecting level of difficulty by helping participants choose song speeds Participant: chose preferred songs and song speeds, followed visual and auditory cues while dancing Instructor: led clinical MT according to patients' specific needs
Lagesen S., 2011 [63]	X	X	Voice (<i>details unspecified</i>)	Structured Direction: patients listened to familiar songs and completed MIT, which connected their speech to melodic features Improvisation: program included clinical improvisation and song writing	Individual	Participant: patients listen to, composed, and performed music; patients had agency to select songs and use artistic creativity Instructor: led rhythmic exercises through pre-recorded CD	
Metzler-Baddeley C., 2014 [64]	X	X	Bongo drums	Structured Direction: through a pre-recorded training program, participants followed specific rhythmic exercises	Individual (patients' caregivers or relatives were involved when it was necessary)	Participant: played musical instrument as they followed instructions on CD Instructor: led workshop by providing Buto dance imagery for participants Participant: recreated imagery through dance	
Muto K., 2005 [65]	X	X	Brazilian samba, Spanish rumba, West-African kuku, Cuban son	Structured Direction: participants followed specific dance movements	Group	Participant: played musical instrument as they followed instructions on CD Instructor: led workshop by providing Buto dance imagery for participants Participant: recreated imagery through dance	

(Continued)

Table 2
(Continued)

Source (first author, year of publication reference number)	Art modality	Art genres	Instruments	Methods	Individual and Group	(details unspecified)
Orejas M.E., 2008 [66]	X	Ballroom dance		(details unspecified)	Group	Instructor: manipulated participant's body posture
Salgues J., 2016 [67]	X	Contemporary dance		(details unspecified)	Individual	Participant: followed instructors' dance movements Instructor: administered auditory cues to participants
Thaut M.H., 1999 [68]	X	Folk music	Computerized music; metronome	Structured Direction: participants synchronized gait to set rhythmic tempos	Individual	Participant: synchronized gait to cues; selected comfortable tempo of cues Instructor: led and supervised structured dance classes
Trinkler L., 2019 [69]	X	Contemporary dance		Structured Direction: there were four features of workshops that were consistent throughout participation Improvisation: participants improvised dance movements for individual body parts and in collaboration with other participants	Group	Participant: used artistic creativity through improvisation of dance movements; had the choice to participate in at least one of three dance class options per week Instructor: had the agency to dictate content of each MT session
Van Bruggen-Ruff M.C., 2017 [70]	X		Structure Direction: instructors used an established MT protocol	Individual and Group Improvisation: content of music therapy program was based on instructors' and patients' current goals; patients engaged in "expressive musical interaction"	Instructor: led program by "loosely" adhering to MT protocol	

Table 3
Aims, interventions, and outcomes in seven studies and six abstracts on the therapeutic use of art

Source (first author, year publication reference number)	Research aim	Procedure	Art form methods	Data analyses and outcomes
Brandt M., 2016 [58]	To evaluate the efficacy of combining MT and speech therapy methods as Huntington Speech Music Therapy (HSMT) for HD patients based on speech production and perception outcomes	Speech therapist assisted patients with verbal production of automatic sequences (i.e., greetings, alphabet etc.), patient-related words and sentences (i.e., names of family and friends, personal hobbies etc.) and conversations while music therapist accompanied patient with repetitive melodic sequences on the piano	HSMT used repetitive, short piano melodies to accompany patients as they produced words and sentences	HSMT effectively structured incoming and outgoing verbal information, and therefore, facilitated ease for patients when engaging in daily complex conversations; HSMT stimulated patients' use of speech Conclusion: HSMT improved speech production of automatic sequences, patient-related words and sentences, and of general conversation.
Delval A., 2008 [59]	To assess the impact of rhythmic cues on gait parameters during a walking task (free gait) and dual task paradigms involving motor (gait+motor task) and cognitive (gait+cognitive task) domains	Task 1: Participants walked at their baseline cadence (free gait) without rhythmic cues and then walked with cues set at 120% of their baseline cadence. Three trials were completed Task 2: Participants walked at their baseline cadence while carrying a tray of filled glasses (gait+motor task) and then walked with cues set at 100% and 120% of their baseline cadence. Three trials were completed Task 3: Participants walked at their baseline cadence while counting backwards (gait+cognitive task) and then walked with cues set at 100% and 120% of their baseline cadence. Three trials were completed	During gait tasks, rhythmic cues from a metronome were set at three frequencies: subjects' baseline cadence, 100% of subjects' baseline cadence, 120% of subjects' baseline cadence	Task 1: Healthy controls increased their gait speed and cadence but not stride length when cues were set at 120% of their baseline cadence ($p < 0.05$). No observed differences in gait parameters with added cues for HD patients. Step synchronization was more impaired for HD patients Tasks 2 and 3: Healthy controls increased their gait speed and cadence but not stride length when cues were set at 120% of their baseline cadence ($p < 0.05$) compared to when cues were set at 100% of their cadence. No observed differences in gait parameters with added cues of various frequencies for HD patients. Step synchronizations was more impaired for HD patients Conclusion: Rhythmic cues were ineffective for improving kinematic gait parameters of HD patients. Although there was an observed "trend towards improvement" in gait parameters, significance was not achieved. Attentional deficits associated with HD may explain synchronization deficits.

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Table 3
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Source (first author, year publication reference number)	Research aim	Procedure	Art form methods	Data analyses and outcomes
Hyson C., 2005 [60]	To evaluate whether a novel MT program is accepted by HD patients and effectively impacts mood and motor symptoms	Patients participated in a six-week MT program that involved one individual session and one group session each week. Patients were evaluated with the UHDRS at baseline and post-MT and completed a post-MT feedback questionnaire	MT program combined methods of RAS, PSE, and TIMP	Tolerability of MT program was achieved (100% adherence, 98% attendance). The majority of HD patients perceived a positive benefit of the use of MT. Improvements in UHDRS scores were observed (i.e., finger tapping, pronation/supination, Luria), though not significant Conclusion: MT is a tolerated clinical method with positive perceived benefits for HD patients. Preliminary evidence suggested a positive impact of MT on UHDRS motor score.
Johnson K.A., 2000 [61]	To determine whether rhythmic cues facilitate improved coordination of bimanual in-phase and anti-phase hand movements	Participants performed bimanual in-phase and anti-phase hand movements at fast and slow speeds. A bimanual crank was used to perform circular hand motions. In the "cue on" trials, participants tried to synchronize in-phase and anti-phase movements with rhythmic cues (i.e., one hand rotation per beat). In the "cue off" trials, participants tried to remember the beat frequencies after exposure as they completed hand movements	During in-phase and anti-phase hand movement tasks, rhythmic cues from a metronome were set at two frequencies: fast (1.5 Hz) and slow (0.5 Hz)	Fast and slow in-phase hand movements were unimpaired for controls and HD patients. HD patients' bimanual coordination was less accurate [F(1,22) = 6.58, $P < 0.018$] and more variable [F(1,11) = 14.044, $P < 0.003$] at fast speeds. Performance was not influenced by the use of rhythmic cues Fast and slow anti-phase movements were unimpaired for controls but impaired for HD patients [F(1,22) = 12.39, $P < 0.002$]. Both controls and HD patients had high coordination variability [F(1,22) = 7.72, $P < 0.011$] at fast speeds. Performance was not influenced by the use of rhythmic cues Conclusion: While in-phase hand movements are less impaired than anti-phase hand movements, rhythmic cueing is ineffective for improving both forms of coordination patterns for HD patients. This finding is inconsistent with that of other patient populations such as PD, which may be due to HD ganglionic atrophy.

<p>Kloos A.D., 2013 [62]</p>	<p>To evaluate the feasibility, acceptability, and safety of a DDR video game exercise program and to determine whether it affects gait, psychiatric, and psychosocial outcomes</p>	<p>Patients completed either six weeks of DDR or a handheld video game (i.e., Bingo, Blackjack, Solitaire) (45 minute sessions, two days a week). DDR exercise sessions increased in difficulty as patients improved synchronization accuracy. Handheld video games worked to stimulate cognitive networks. After six weeks, patients completed the opposing exercise intervention for another six weeks. Outcome measures were assessed at baseline and post-treatment for each intervention</p>	<p>DDR required patients to synchronize their dance movements, guided by visual cues (arrows) and auditory cues (melodic beats). Patients and instructors controlled song selection and tempo</p>	<p>DDR demonstrated:</p> <ol style="list-style-type: none"> 1) Feasibility: patients could increase level of difficulty of video game while maintaining accuracy scores 2) Adherence: 100% participation for all participants 3) Acceptance: patient feedback showed high motivation to participate in program 4) Safety: normal physiological parameters, no adverse events or injuries reported <p>For all patients, certain gait parameters significantly improved after completing DDR: double support percentage (2.54% reduction, $p = 0.03$), backward walking (4.18% reduction, $p = 0.01$). For patients with minor motor impairments, significant improvements in forward heel-to-heel base of support</p> <p>No significant difference in Activities-Specific Balance Confidence Scale, TT, or WHO-QoL scores between treatment groups</p> <p>Conclusion: DDR, which combined music and dance into one cohesive exercise program, benefited gait and was a feasible and safe method of physical exercise.</p> <p>Case 1: clinicians' observations showed that MT reduced patient's challenging behaviors during morning-care procedures</p> <p>Case 2: clinicians' observations showed that MT improved speech and articulation and reduced choreiform movements. MT additionally benefited patient's relationship with clinical staff</p> <p>Conclusion: The clinical use of MT may individually benefit patients with advanced HD.</p>
<p>Lagesen S., 2011 [63]</p>	<p>To determine the clinical benefits of MT for two patients with advanced HD</p>	<p>Case 1: a patient who presented with difficult behaviors during morning-care procedures was provided four weeks of MT</p> <p>Case 2: a patient who presented with speech and articulation deficits and chorea was provided six weeks of MT</p>	<p>MT included improvisation, song writing, listening to familiar songs, and modified MIT. MT features were altered for each patient, based on symptoms and specific goals</p>	<p>Conclusion: DDR, which combined music and dance into one cohesive exercise program, benefited gait and was a feasible and safe method of physical exercise.</p> <p>Case 1: clinicians' observations showed that MT reduced patient's challenging behaviors during morning-care procedures</p> <p>Case 2: clinicians' observations showed that MT improved speech and articulation and reduced choreiform movements. MT additionally benefited patient's relationship with clinical staff</p> <p>Conclusion: The clinical use of MT may individually benefit patients with advanced HD.</p>

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Source (first author, year publication reference number)	Research aim	Procedure	Art form methods	Data analyses and outcomes
Metzler-Baddeley C., 2014 [64]	To assess the efficacy of a rhythmic drumming exercise program according to changes in executive functioning and microstructure of cortical areas	Patients were provided with rhythmic drumming exercises on CD (22 different exercises, 15 minutes each), bongo drums, and a training diary. Patients exercised 15 minutes/day five days/week for eight weeks by following drumming exercises on CD and recording each session in diary	Rhythm exercise patterns varied in genre (i.e., Brazilian samba, Spanish rumba, West-African kuku, Cuban son). Although drumming exercises progressed in speed and rhythmic complexity, patients were able to progress through levels of difficulty at their own pace	<p>Significant improvements were observed in cognition ($t(4) = 3.30, p \leq 0.03$):</p> <ol style="list-style-type: none"> 1) Dual task mean(SD) accuracy scores (baseline: 9.4(3.1), post-baseline: 14.0(3.7)) 2) DSSST mean(SD) scores (baseline: 42.4(15.4), post-baseline: 43.6(13.6)) 3) TMT mean(SD) response times in seconds (baseline: 39.0(18.9), post-baseline: 31.4(9.4)) 4) Stroop task mean(SD) response times in seconds (baseline: 111.0(20.1), post-baseline: 93.8(24.1)) 5) Verbal fluency mean(SD) accuracy scores (baseline: 28.0(10.4), post-baseline: 39.8(8.8)) 6) Category fluency mean(SD) accuracy scores (baseline: 30.4(11.3), post-baseline: 39.2(15.4)) <p>Significant microstructural changes in fractional anisotropy (FA), axial diffusivity (AD) and radial diffusivity (RD) indices were observed in white matter:</p> <ol style="list-style-type: none"> 1) segment 1 of corpus callosum: significant increase [FA $t(4) = 5.2, p = 0.006$]; AD $t(4) = 5.1, p = 0.007$]; significant decrease [RD $t(4) = -4.6, p = 0.011$] 2) segment 2 of corpus callosum: significant increase [AD $t(4) = 3.03, p = 0.04$] 3) cortico-spinal tract: significant increase [FA $t(4) = 3.3, p = 0.03$]; AD $t(4) = 2.9, p = 0.04$]; significant decrease [RD $t(4) = -4.2, p = 0.014$] 4) anterior thalamic radiation: significant increase [FA $t(4) = 3.7, p = 0.02$]; AD $t(4) = 4.04, p = 0.016$] <p>Significant positive correlations were obtained for changes in cognition and microstructure of corpus callosal segments.</p> <p>Conclusion: A two-month drumming training program improved executive function and changed white matter microstructure in HD patients. Improvements in executive function resulted from microstructural changes in the corpus callosum, suggesting that long-term training induced neuroplasticity.</p>

Muto K., 2005 [65]	To assess the satisfaction level of a dance workshop for patients, persons at risk for HD, and their caregivers. To determine whether dance effectively targets psychiatric issues related to the HD diagnosis and creates mutual understanding amongst patients and caregivers on the illness experience	Participants and instructors engaged in an initial friendly discussion before dance workshop. Participants subsequently danced for two hours while following instructors' Buto dance movements. After the workshop, participants completed a satisfaction survey	Buto dance incorporated "dark, slow, and contorted" imagery as an artistic representation of one's "inner unconscious space."	Patients' fears and tensions related to hiding motor impairments were diminished while dancing. Dancing allowed persons at risk for HD to let go of their fears about developing motor impairments. For caregivers, dance allowed them to better relate to the HD illness experience Conclusion: HD patients, individuals at risk for HD, and their caregivers were satisfied with a Buto dance workshop as a way to improve mood and emotional issues. Participation of caregivers or individuals without HD may additionally result in an empathic understanding of the HD experience.
Orejas M.E., 2008 [66]	To evaluate the therapeutic effect of ballroom dancing for HD patients according to motor, functional capacity, behavioral, and quality of life outcomes	Treatment group: HD patients completed weekly one hour sessions of DMT for six months (<i>details unspecified</i>) Control group: HD patients completed a home-based exercise program without music or dance (<i>details unspecified</i>) All patients were assessed at baseline and post-treatment with motor, functional, behavioral, and quality of life scales. All patients completed a post-treatment satisfaction questionnaire	DMT used classical ballroom dancing (<i>details unspecified</i>)	<i>Results of quantitative measurements not available.</i> Patients in treatment group showed high levels of motivation to participate and compliance compared to patients in control group. Patients in treatment group reported feeling better overall. Conclusion: DMT effectively motivated patients to engage in exercise and while complying to protocol. Dance is a feasible intervention in HD rehabilitation programs and may target feelings of apathy that make it difficult for patients to achieve long-term and consistent participation in exercise.
Salgues J., 2016 [67]	To examine the therapeutic effect of a contemporary dance workshop for patients and persons indirectly affected by HD	Dance instructors led creative dance exercises that required participants with and without HD to experiment with different body movements. Participants learned how body posture, regardless of chorea, directly reflects their consciousness	Contemporary dance workshops combined artistic methods (i.e., imagination, poeticism, invention, multiple sensory modalities) with therapeutic approaches that target the development of consciousness and empathy through manipulation of body posture	Observations showed that dance workshops helped HD patients feel less isolated as a result of their diagnosis (<i>details unspecified</i>) Conclusion: There is a positive impact of DMT on patients, their relatives, and their caregivers because dance served as a medium for understanding the illness experience (<i>details unspecified</i>).

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Source (first author, year publication reference number)	Research aim	Procedure	Art form methods	Data analyses and outcomes
Thaut M.H., 1999 [68]	To evaluate the impact of external rhythmic cues on HD patients' ability to modulate gait velocity at faster and slower speeds	<p>Patients walked on a 26-meter walkway without rhythmic cues for four trials with the following pacing conditions:</p> <p>(1) normal speed (pre-test baseline), (2) slower than baseline, (3) faster than baseline, (4) normal speed (post-test baseline).</p> <p>In the next four trials, patients walked with rhythmic cues set at the following pacing conditions:</p> <p>(1) metronome 10% slower than baseline, (2) metronome 20% faster than baseline, (3) music tempo 20% faster than baseline (4) normal speed (post-test baseline)</p> <p>Gait parameters were collected for each trial</p>	Rhythmic auditory cueing utilized metronomic beats and musical beat patterns	<p>Modulation of gait velocities was achieved with and without rhythmic cueing ($p < 0.05$) but not with music. A carry-over effect was observed after cued trials, as unpaced gait velocity maintained a significantly greater value than baseline velocity ($p < 0.05$). A carry-over effect was not observed for trials without rhythmic cues</p> <p>While severity of HD did not impact modulation, greater disability and chorea scores significantly lowered gait velocity and impaired synchronization of movements to cues</p> <p>Conclusion: HD patients can modulate gait velocity with and without external rhythmic cues, though not with music. However, external rhythmic cues were ineffective for achieving gait synchronization, regardless of disease severity.</p>
Trinkler L., 2019 [69]	To assess the impact of contemporary dance on motor, neuropsychiatric and cognitive functions, including apathy, quality of life, and brain structure for patients with HD	<p>Group 1: five months of contemporary dance, participated in at least one two-hour class per week. Evaluation of outcomes occurred after five months of treatment</p> <p>Group 2: five months of usual care. Evaluation of outcomes occurred after five months of treatment. Participants continued with contemporary dance treatment for another five months (at least one two-hour session per week).</p> <p>Evaluation of outcomes occurred after DMT</p>	<p>Four features of contemporary dance workshops:</p> <p>1.) warm-up session incorporated relaxation and body consciousness exercises</p>	<p>Patients' UHDRS motor, cognitive, (i.e., MDRS, Stroop, TMTA), and neuropsychiatric scores (i.e., "irritability" and "lack of initiative" subscales on PBA) were significantly worse than healthy controls. Apathy and quality of life scores were not significantly different amongst patient and control groups</p> <p>Motor impairments (median[IQR]) decreased in contemporary dance group from 28 [6–51] to 27 [7–33]. Motor impairments increased in those who received usual care from 19 [13–35] to 25 [14–42] ($Z = -2.44, p = 0.015$). Significant differences in cognitive and behavioral scores were not observed across treatment groups</p>

<p>Van Bruggen-Ruffi M.C., 2017 [70]</p> <p>To compare the therapeutic effect of MT and recreational therapy on quality of life based on communication and behavior outcomes</p>	<p>Treatment Group (MT): Group sessions followed MT protocol with the aim of "improving and stimulating communication and self-expression" (one-hr/week, 16 weeks)</p> <p>Control Group (Recreational therapy): Group activities involved communication between patients (i.e., arts-and-crafts, puzzles, reading, cooking). Musical activities were excluded</p> <p>Communication and behavior outcomes were gathered at baseline and after 8, 16 and 28 weeks</p>	<p>2.) participants improvised movements with and without music based on themes (i.e., "machines," "ocean," "neck," etc.)</p> <p>3.) group improvisation of dance movements based on same themes</p> <p>4.) massage exercises on the floor</p> <p>MT was partially structured, as each session started and ended with the same songs. However, overall content of each session depended on the goals of patients. Patients' musical experiences varied (level of experience did not impact ability to participate in MT)</p> <p>Group activities involved communication between patients (i.e., arts-and-crafts, puzzles, reading, cooking). Musical activities were excluded</p> <p>Communication and behavior outcomes were gathered at baseline and after 8, 16 and 28 weeks</p>	<p>Increase in medial superior parietal and paracentral lobule volume post-DMT</p> <p>Conclusion: Contemporary dance workshops that incorporate "spatial and bodily representations" were accepted and adhered to by patients. They significantly improved motor functions and induced neuroplastic effects that reflected intact compensatory mechanisms and aligned with positive patient feedback.</p> <p>Significant difference in social-cognitive subscale scores between treatment and control groups ($p=0.042$), with a positive effect observed for control group</p> <p>An additional benefit of MT group sessions on communication and behavior compared to recreational therapy was not observed. No significant difference in mental rigidity/aggression mean scores on BOSH ($p=0.125$) was observed</p> <p>Conclusion: RCT research design and communication outcomes were not optimal features of clinical trials that aimed to evaluate the impact of art-based interventions on quality of life for HD patients, especially for studies that included patients with limited communication at the advanced phase of disease.</p>
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non-MT therapy by assessing changes to social-cognitive functioning [70] on the Behavior Observation Scale Huntington (BOSH) [79] (Table 1). Improvements to social-cognitive subscale scores achieved significance for participants of recreational therapy but not MT (Table 3). In another RCT, a contemporary dance workshop was evaluated by observing changes to UHDRS cognitive scores including Stroop task [76], Symbol Digit Code [6], Mattis Dementia Rating Scale (MDRS), a measure for global cognitive functioning [80], and TMT [75]. Changes to cognitive scores were insignificant.

Cognitive outcomes were additionally examined in conference abstracts, which included language processing [58], memory and attention [60], and development of consciousness [67]. Positive patient feedback was collected on a novel MT program for speech rehabilitation [58].

Psychiatric function outcomes

A cross-over pilot clinical trial [62] evaluated the efficacy of a novel video-game DMT program by assessing balance confidence with the Activities-Specific Balance Confidence Scale [81] (Table 1). Changes to baseline scores did not reach significance. Another multi-center RCT used the Problem Behavior Assessment (PBA-s) [82] and BOSH [79] to compare improvements to mental rigidity, aggression, and behavior severity across MT and recreational therapy programs [70]. Improvements to individual estimated mean scores were not significantly different across treatment groups (Table 3). Another RCT that used the PBA-s [82] and Lille Apathy Rating Scale (LARS) [83] to evaluate the efficacy of contemporary dance workshops did not find significant improvements to behavior.

Other psychiatric outcomes were represented in conference abstracts, which included overall mood [60, 67], fear and anxiety [58, 65] and behavioral control [63, 65]. Preliminary qualitative findings showed that MT facilitated ease of speech production [58], increased patients' motivation [60], and reduced challenging behaviors of an in-patient during morning-care procedures [63]. Additionally, DMT reduced fears and tensions associated with HD progression and increased caregivers' empathic understanding of the illness experience [65].

Motor function outcomes

Two cross-sectional studies compared gait parameters [59] and modulation of gait velocity [68] across

external rhythmic cueing and no cueing conditions (Table 1). A significant benefit of auditory cues for kinematic gait parameters and velocity modulation was not observed [59, 68] (Table 3). Although one study found a trend towards improvement in gait, significance was not achieved [59]. In fact, in one study that included musical cues, music, but not rhythmic patterns, impeded gait velocity modulation [68]. In another study that evaluated the effect of contemporary dance workshops on motor function using the UHDRS [5], findings showed a decrease in motor impairments post-DMT and an increase in motor impairments after usual care that did not involve art forms [69]. Another clinical trial [62] evaluated the efficacy of a novel video-game DMT program by assessing changes to spatiotemporal gait parameters through the GAITRite walkway system [84]. Significant improvements to certain gait parameters were dependent on the severity of HD, such as forward heel-to-heel base of support [62].

The clinical trial [62] also compared balance outcomes between DMT and non-DMT treatments through the Four Step Square Test (FSST) [85] and Tinetti Mobility Test (TT) [86] (Table 1). Significant differences in balance scores were not observed across treatment groups. Additionally, a cross-sectional study used bimanual cranks [87] to evaluate fine motor skills by comparing bimanual in-phase and anti-phase hand movements across external rhythmic cueing and no cueing conditions [61]. Findings showed that coordination accuracy and velocity were not influenced by the use of rhythmic cues [61] (Table 3).

Motor outcomes such as language production [58, 60, 63], fine motor skills [60], posture [60], and choreiform movements [63] were also examined in conference abstracts (Table 1). Clinical observations collected in one case report showed improvements to motor control and speech production [63]. Patient feedback from another study was positive for the use of music in speech therapy [58]. Additionally, an MT clinical trial showed an improvement trend in fine motor sub-scores on the Unified Huntington's Disease Rating Scale (UHDRS) [5]; however, significance was not achieved [60] (Table 3).

Psychosocial outcomes

Quality of life

One clinical trial examined the impact of DMT compared to non-DMT programs on patients' quality of life by utilizing the World Health Organization

Quality of Life-Bref scale (WHO-QoL) [88, 89]. Significant differences in WHO-QoL post-baseline scores across treatment groups were not observed. Another RCT did not find a significant impact of contemporary dance workshops on quality of life based on the Quality of Life Index (QLI) [90].

Two conference abstracts also evaluated quality of life connected to language production [58] and overall functionality [66]. Assessment methods included the Parkinson's disease Questionnaire (PDQ-39) [91], the Nottingham Health Questionnaire (NHQ) [92], the Functional Independence Measure (FIM) [93], and Barthel Scales [94]. Preliminary patient feedback confirmed the positive impact music-based speech therapy had in the daily navigation through complex conversations [58].

Participant satisfaction

One RCT evaluated patients' experiences of DMT through qualitative interviews [69]. Patient feedback suggested that "contemporary dance altered the way [patients] 'felt and lived in their bodies.'" Two conference abstracts assessed participant satisfaction of DMT programs through surveys [65, 66]. Preliminary findings suggested high satisfaction levels from patients [65, 66] and their caregivers [65], though specific outcomes were inconclusive.

Neuroanatomy

One non-randomized pilot clinical trial evaluated the effect of a rhythmic drumming exercise program on subcortical basal ganglia volume [64]. Magnetic resonance imaging (MRI) showed significant changes to corpus callosal white matter microstructure, or myelinated nerve fiber bundles that positively correlated with improvements in global cognition. Another RCT evaluated the effect of contemporary dance workshops on brain volume [69]. Structured MRI (sMRI) and voxel-based morphometry (VBM) showed a volumetric increase in medial superior parietal and paracentral lobule structures post-DMT that is associated with compensatory mechanisms for spatial and somatosensory processing.

Program feasibility, adherence, acceptability, and safety

One cross-over clinical trial made comparisons between a video-game DMT program and a non-DMT program based on feasibility, adherence, acceptability, and safety [62]. Feasibility of DMT was

confirmed through significant improvements to accuracy scores. High attendance and completion of both DMT and non-DMT indicated the adherence of programs. Additionally, positive feedback from patients suggested high acceptability of DMT and no adverse health events or injuries confirmed its safety. Another RCT found that contemporary dance workshops were adhered to through high attendance [69].

DISCUSSION

To our knowledge, this systematic review is the first to evaluate outcomes of MT, DMT, and rhythmic auditory cueing for HD patients. We synthesized quantitative and qualitative analyses from 7 prospective articles and 6 conference abstracts that assessed the impact of established art-based clinical programs or individual art forms on functioning. There was heterogeneity across all studies in the use of art modalities, methodologies, and therapeutic outcomes. The current review confirmed a potential positive effect of using art forms in conventional HD clinical practices. Half of the included articles found benefits of music, dance, and rhythm to cognitive, motor, and microstructural outcomes. Abstracts confirmed additional benefits to psychiatric symptoms and quality of life. Art forms were determined to be feasible and safe methods for HD patients. However, it is worth mentioning that limited studies led to the extraction of data from abstracts. Therefore, the field is currently underdeveloped, making it difficult to conclusively define the collective and distinctive benefits that result from MT, DMT, and rhythmic auditory cueing.

We found that improvements to cognitive function scores [64, 69] such as memory and attention, verbal and category fluency, and processing speed were significant. These changes were contradicted by insignificant changes to executive function subscales on the UHDRS [60]. Additionally, positive patient feedback of benefits to speech processing [58] conflicted with insignificant changes to social-cognitive baseline scores [70]. Inconsistencies in findings regarding cognitive outcomes may be explained by the comparisons between dissimilar art programs—rhythmic training [64] versus MT [60] and HSMT [58] versus general MT [70]. Additionally, outcome comparisons between dissimilar research designs—case report [58] versus RCT [70]—could inhibit consistencies in the synthesis. Our review may therefore suggest that the

therapeutic effects of music, dance, and rhythmic training are unique to the art form, which potentially results from underlying differences in the neuroplastic effects of varying art interventions [37, 38, 40]. For example, HSMT required persons to listen to music, which potentially stimulated global neuroconnectivity and atrophic recovery of speech production [58]. Conversely, social-cognitive scores improved upon persons improvising dance movements [70]. These findings may result from changes to dopamine levels that facilitated dopaminergic signaling in the amygdala, which is essential for social-cognitive functioning [95]. Findings additionally confirmed that a rhythmic drumming exercise program, which required sensorimotor processing, led to an overall volumetric increase in the corpus callosum [64]. These neuroplastic changes positively correlated with significant improvements in cognitive functions [64]. The results concur with findings from previous studies [37, 38], which suggest that there are potential neuroplastic effects of art interventions, specifically in subcortical basal ganglion areas, that positively affect functioning in cognitive domains. Rhythmic drumming may have additionally induced stronger synaptic connections between auditory and motor cortices that facilitated an intact exercise performance. It is worth mentioning that this was a within-study comparison that was not examined in other studies in the review.

We also found consistent patient feedback on perceived benefits for psychiatric functions. Studies demonstrated that both MT and DMT reduced fears and tensions associated with speech and motor dysfunctions, respectively [58, 65]. MT and DMT also motivated patients to continue a consistent exercise regime [60, 62]. These findings may result from the ability of MT and DMT to activate emotional pathways in the amygdala, cingulate cortex, and hypothalamus that improve mood and reduce anxieties related to HD progression [38]. However, qualitative syntheses were inconsistent with changes to balance confidence [62], depression, and apathy scores [69]. We predict that inconsistencies in findings manifest from our synthesis of one patient's feedback in a case report [58] or that of a very small sample size [60] with outcomes that represented larger samples [62, 69]. Additionally, studies' lack of information on HD phase represented in participant samples potentially led to outcome cross-comparisons amongst patients with varying degrees of disease progression. As functionality of patients changes with severity of HD, so should the goals of art therapy. Therapeu-

tic effects of art for HD patients should therefore be uniquely interpreted for each disease phase. While certain studies in the review indicated a relationship between feelings of fear and anxiety and motor dysfunctions [58, 65], other studies reported mood symptoms of depression and apathy without reference to an underlying cause [69]. There may be value in defining the causal relationship between features of HD pathology and psychiatric symptoms so that informative between-study comparisons can come to fruition.

In addition, the review has shown consistent benefits of art forms for motor function, which was found through positive clinician feedback and significant improvements in UHDRS scores [63, 69]. These findings complimented the consistent positive feedback from clinicians and patients on the use of MT for speech production [58, 63]. We expected to see a positive effect of art on motor control, considering that published studies have validated the evidence-based clinical use of dance to improve motor impairments of PD patients [41–44]. Additionally, we expected to find that improvements in UHDRS motor scores were accompanied by an increase in medial superior parietal and paracentral lobule volume as a result of participating in contemporary DMT [69]. These neuroplastic effects may result from sensorimotor processing, which is integral to dance [45]. Although HD and PD pathologies differ, various art forms provide similar benefits to these patient cohorts. MT, DMT, and rhythmic auditory cueing may therefore be pertinent for persons with other forms of neuromuscular disease. Our review also found positive effects of DMT on gait [62], which was not observed in studies that evaluated rhythmic auditory cueing [59, 68]. Generally, studies in the review found that synchronization of gait and hand movements to external auditory cues was impaired for all patients, yielding comparable outcomes to that of free movement [59, 61, 68]. Therefore, our review has demonstrated a distinctive purpose of DMT for gait function that may be more adaptable to the HD phase.

We did not expect to synthesize psychosocial outcomes from less than a third of the included studies in the review. Considering that the concurrence of conventional and alternative treatment methods aims to promote quality of life and patient satisfaction, we expected to synthesize psychosocial functions across the majority of the included studies. Furthermore, to our surprise, findings showed inconsistencies in the comparison of positive patient feedback [58] to insignificant changes to life quality

scores [62, 69]. It is worth mentioning that items on the WHO-QoL and QLI are not specific to the HD illness experience, which may account for observed inconsistencies. However, levels of satisfaction were comparable between patients and their caregivers [65, 66], suggesting the potential for art forms to transcend healthcare roles.

In the synthesis of positive outcomes represented in the review, we present probable explanations that are supported by the included studies. First, the therapeutic use of art for persons with HD promotes neuroplastic effects to the brain that may reduce cognitive, psychiatric, and motor impairments or perhaps restore function. Because artistic activities such as music performance and dance require specialized cognitive and motor abilities to that of non-artistic activities, these changes may be distinct from that of conventional HD rehabilitation. Only two studies investigated neuroplastic effects as a result of art-based rehabilitation, so our understanding of this comparison is currently inconclusive. Second, music, dance, and rhythmic auditory cueing are conduits for motivating HD patients to engage in exercise. For a disease in which apathy is a clinical hallmark, it is essential to develop strategies to promote patients' long-term and consistent engagement in exercise regimes. Our review has shown that art forms can maintain excellent attendance and achieve high satisfaction. Last, we have synthesized studies that represented patients across the entire spectrum of HD severity. As such, art methods work regardless of patients' functionality, suggesting that it can be a consistent method of treatment that is flexible for change.

It is worth addressing limitations of this review. First, approximately half of the included studies were conference abstracts, limiting our access to vital details on art methodologies, participants, and general outcomes. For example, some conference abstracts provided minimal information on participants' HD phase and the art therapy designs involved. We included conference abstracts because of the small number of relevant studies currently available. As a result, we had to synthesize studies with potentially few characteristic consistencies, making it difficult to gather informative comparisons. Without informational access, outcome comparisons between different art forms and patients at different HD stages was inevitable. Second, many of the studies we reviewed included small sample sizes, weak protocols, and a general span of investigated outcomes. This also contributed to between-study

comparisons that involved unaccounted variability, hindering a collective synthesis. Although these limitations made it difficult to synthesize the therapeutic effects of art for patients with HD, recognizing these confounders allows us to understand the factors that may influence outcomes and therefore, avenues for improvement in future studies. Third, we limited our search to medical databases, excluding sources from artistic databases. Although this may have led to an unawareness of other relevant art-based programs and methodologies, we aimed to focus on the integrity of neurological information and scientific research designs, which is optimally supported by medical resources.

We present considerations for future research. First, art-based exercise programs should be tailored to individual phases of HD so that music, dance, and rhythm benefit rather than impede functioning. The development of phase-specific protocols may give patients with HD consistent access to clinical art forms that produce meaningful outcomes. Second, studies should determine the ways in which to effectively incorporate variety within one art method. Utilizing different artistic genres, instruments, and activities, for example, may increase patients' affinity for art-based programs by catering to their preferences. As a result, programs can maximize benefits by directly targeting apathy. Third, the investigation of the therapeutic use of art should take the form of different study designs. As such, we can establish ideal methodological features to implement for the HD cohort. For example, one study concluded that an RCT was undesirable for evaluating cognitive functioning of patients with advanced HD [70]. Therefore, research designs must be altered to accommodate different disease presentations. Additionally, the investigation of differential outcomes of music, dance, and rhythm through neuroimaging techniques may better inform the future development of art-based rehabilitation programs. Lastly, few studies in the review assessed the impact of art therapy on the quality of life of family members or utilized caregiver feedback to determine the impact of music, dance, and rhythm on patients. Understanding caregiver roles may be particularly useful for characterizing outcomes for patients at middle-to-advanced disease phases as well as the ways in which these outcomes transcend patients to indirectly influence caregiver burden.

In conclusion, the review has provided preliminary support for the clinical use of music, dance, and rhythm for patients with HD, with a particular

benefit to gait, global cognitive function, and subcortical plasticity. While it is evident that art forms are feasible and safe methods that garner caregiver and patient satisfaction, limited research has rendered us unable to conclusively determine the manner in which various neurologic art therapy techniques produce therapeutic outcomes. We recommend that future research compares responses of functional domains to art forms by evaluating the mechanistic differences of music, dance, and rhythm that may account for therapeutic outcomes.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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