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Case Report

The use of rhythmic auditory stimulation for functional gait disorder: A case report

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Abstract.
BACKGROUND: Functional gait disorders (FGD) are a common and disabling condition. Consensus-based rehabilitation techniques for treating FGD and other functional neurological disorder presentations at large utilize a variety of therapeutic strategies, including distraction, novel approaches to movement, entrainment, stress/hypervigilance modulation, and psychotherapy.
CASE REPORT: Here we present a case of a 24-year-old woman with a complex history of anxiety, depression, left frontal astrocytoma, postural orthostatic tachycardia syndrome (POTS) and FGD. During a multidisciplinary inpatient rehabilitation stay for FGD, the patient underwent rhythmic auditory stimulation (RAS) delivered by a neurologic music therapist in conjunction with physical therapy, occupational therapy, and psychotherapy.
RESULTS: The RAS intervention appeared to play a significant role in symptom resolution for this patient. Improvement in the patient's truncal displacement, foot dragging, and well as overall gait speed occurred following serial RAS trials performed over a single treatment session. Benefits persisted immediately following the intervention and upon subsequent reassessment. Although at four-year follow-up the patient's FGD symptoms remained resolved, fatigue continued to limit her ambulatory capacity and overall endurance.
CONCLUSION: RAS represents a unique therapeutic approach for treating FGD, complementary to existing consensus-based rehabilitation recommendations, and may warrant further consideration by the field.

Keywords: Functional neurological disorder (FND), functional gait disorder, rhythmic auditory stimulation (RAS), neurologic music therapy (NMT), gait training

1. Introduction

Functional gait disorders (FGD) are a common and disabling condition (Nonnekes et al. 2020). As is the case for functional neurological disorder (FND) at large, symptom inconsistency and incongruity with other neurological conditions remain key diagnostic features of FGD (Perez et al. 2021; Stone and Carson 2015). Additional positive clinical signs of FGD may include unergonomic posture, exaggerated truncal sway, sudden knee buckling, monoplegic limb dragging, and/or excessive caution or slowness (Baik

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and Lang 2007; Nonnekes et al. 2018; Sokol and Espay 2016; Sudarsky 2006). Kinematically, FGDs have been associated with alterations in base of support, center of gravity, stride length, symmetry, velocity, and variability (Breitkopf et al. 2018; Lin et al. 2020). Functional mobility is frequently limited by FGDs, with some individuals becoming dependent upon walkers, forearm crutches, or even wheelchairs in severe cases (Baizabal-Carvallo, Hallett, and Jankovic 2019; Jordbru et al. 2014).

Multidisciplinary rehabilitation appears to be effective for treating FGDs (Czarnecki et al. 2012; Jordbru et al. 2014; Matthews, Brown, and Stone 2016; Petrochilos et al. 2020; Speed 1996). Recent physical therapy (PT) and occupational therapy (OT) consensus recommendations offer technical advice for treating therapists (C. Nicholson et al. 2020; Nielsen et al. 2015). Recommended strategies include modulation of excessive self-directed attention via distraction, revision of abnormal movement patterns through novel approaches to motor tasks, entrainment to suppress aberrant movements, as well as relaxation and sensory grounding skills (C. Nicholson et al. 2020; Nielsen et al. 2015). As described in the PT and OT consensus recommendations (T. R. Nicholson et al. 2020; Nielsen et al. 2015), a patient with FGD may be invited to speed up or slow down, exaggerate movements, walk backwards, or move to a set rhythm among other potential therapeutic strategies.

The present case study describes the application of rhythmic auditory stimulation (RAS), a form of neurologic music therapy (NMT), to treat FGD (Thaut and Abiru 2010). In RAS, an external rhythmic stimulus—e.g., the beat of a metronome or metronome-enhanced music—is used to facilitate intrinsically rhythmic movements such as walking (Hayden et al. 2009). Underlying RAS theory is a complex intertwining of auditory and motor systems across cortical, subcortical, and spinal levels (Thaut 2015; Thaut and Abiru 2010). RAS in particular capitalizes on the phenomenon of entrainment, using rhythm and music to facilitate motor initiation and motor control (Rossignol and Jones 1976; Thaut 2015).

The standardized RAS protocol begins with assessing the patient's baseline gait parameters and kinematics, and ability to entrain to music or rhythm. Following entrainment, exercises of progressive difficultly are undertaken and frequency modulation is initiated, in which the tempo of the rhythm or music is increased by 5–10% until the patient achieves a more normal cadence. Lastly, carryover effects in the absence of music are evaluated (Thaut and Rice 2014). While each step in the protocol is important to determine the appropriate direction of treatment, the time spent on each step is not strictly delineated. Decisions to progress or regress through the protocol depend upon the patient's responsiveness to the intervention and ability to maintain changes in gait. A summary description of RAS is provided in Table 1.

Table 1
Standardized RAS protocol (Thaut and Rice 2014)

Step 1: Assess baseline gait
Step 2: Match baseline cadence with rhythmic stimulus to achieve resonant frequency entrainment
Step 3: Modulate rhythmic frequency by 5–10% increments and monitor for normalization of gait parameters
Step 4: Simulate real-world ambulation conditions (starting, stopping, weaving around objects) for advanced gait exercises
Step 5: Gauge carryover of intervention by fading rhythmic stimuli

Abbreviation: RAS, rhythmic auditory stimulation.

When delivered in rehabilitation settings, RAS has been shown to improve gait parameters of velocity, cadence, stride length, and symmetry across a range of neurologic conditions, including Parkinson disease, stroke, and traumatic brain injury (Hausdorff et al., 2007; Hayden et al., 2009; Hurt et al., 1998). Anecdotally, at Spaulding Rehabilitation Hospital, neurologic music therapists (NMTs) working in conjunction with PTs have observed short-term immediate improvements in gait parameters using RAS to treat FGD as well. Below we describe one patient's positive response to the intervention and offer a supplementary video for further visual characterization. We speculate on possible reasons underlying the patient's positive response and consider ways in which RAS may represent a novel treatment approach for FND that is complementary to existing consensus-based rehabilitation strategies.

2. Case presentation

Nancy is a 24-year-old woman with a musical background of singing and playing the piano. She has a complex history including adverse life experiences at a young age with early memories of panic attacks, subsequent emergence of post-traumatic stress symptoms, and self-injurious behaviors leading to a hospitalization for suicidal ideation at age 17. In 2016, Nancy entered her first semester of college and began experiencing difficulties with fatigue, light-headedness, episodic tachycardia, balance, and falls.

She was shortly thereafter admitted to a community hospital for medical evaluation. Records of a neurological exam were not available for review. However, an MRI brain was performed, showing an undefined left frontal white matter abnormality. MRI of the cervical cord showed a possible small syrinx at C5–C7. Lumbar puncture results were normal. The overall assessment was non-diagnostic, and Nancy was discharged to inpatient rehabilitation for two weeks, resulting in modest improvements in stamina and strength. She thereafter continued with outpatient PT as well as psychotherapy, but her symptoms and gait difficulties continued to worsen. Within several weeks she was relying upon a wheelchair for mobility. At an outpatient neurology follow-up visit several months later, Nancy was directed to the Emergency Room at a tertiary care hospital for urgent re-evaluation. She was then admitted to the neurology service.

On neurologic examination, mental status and cognitive testing were within normal limits. Cranial nerves were intact. A coarse, arrhythmic tremor observed when arms were outstretched, resolved when the patient was cognitively distracted by performing the alphabet backwards. Strength was full throughout. Sensation was diffusely intact to light touch, temperature, pinprick, vibration, and position senses. Reflexes were graded as 3 + throughout, with a slightly more brisk response on the right (reportedly lifelong per the patient's mother). Nancy's bilateral lower extremities became rigid when passively flexed or extended during bedside testing, while at other times her tone was noted to be completely normal. During stance and gait testing, she exhibited a high degree of incoordination and truncal sway without falling. Use of distraction resulted in greater fluidity of ambulation.

Hemodynamic monitoring during hospitalization showed a maximum heart rate of 118 beats per minute. Vital signs did not change with posture. Thyroid, vitamin, and autoimmune laboratory studies were unremarkable. Imaging was again notable for a left orbitofrontal and insular non-enhancing lesion (Fig. 1) Cervical spine MRI confirmed a stable, benign cervical syrinx at C5–C7. The thoracic MRI was normal.

The neurology team diagnosed Nancy with "high suspicion" for a left orbitofrontal and insular low-grade glioma, possible postural orthostatic tachycardia syndrome (POTS), and a concurrent FGD. Though it is impossible to say whether the low-grade glioma had caused the patient's gait symptoms,

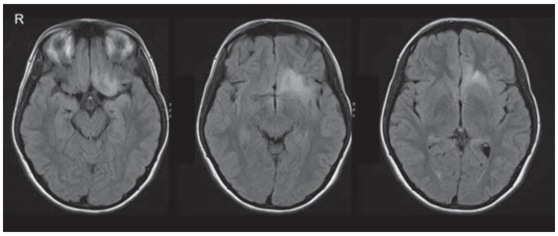

Fig. 1. Non-enhancing T2/FLAIR (fluid-attenuated inversion recovery) hyperintensity on MRI approximately $3.9 \times 3.5 \times 2.0$ cm, centered in the left inferior frontal lobe. The lateral border of the lesion extends into the left insula. R, right.

the neurology team did not believe that this was the case. Of note, FND is commonly comorbid with other neurological conditions, e.g., electrographic seizures, multiple sclerosis, Parkinson's disease (Walzl, Carson, and Stone 2019; Walzl, Solomon, and Stone in press). Ultimately, the presence of functional signs, including symptom inconsistency and variability, phenotypic functional gait characteristics (exaggerated truncal sway), and improvement of gait with distraction led to the FGD diagnosis (Finkelstein et al. 2021).

In further support of this diagnosis, Nancy's glioma was not located in a region associated with ataxic gait disorders—the orbitofrontal cortex is better known for its role in sensory integration, behavior and reward (Rolls 2004) —and the insula for its role in sensory-motor, affect-processing, and cognition (Chang et al. 2013). Additionally, if a structural brain lesion such as a glioma were to cause a gait disorder, it would be unlikely that distraction techniques would so strikingly and immediately improve gait, as occurred repeatedly on neurologic examination. As such, the patient's gait pattern appeared both *incongruent* with what would be expected from a lesion affecting the orbitofrontal cortex and insula and *inconsistent* with how structural lesions typically affect the brain given its waxing and waning quality, two classic hallmarks of FGD (Nonnekes et al. 2020).

A plan was made for Nancy to eventually follow up with outpatient neurosurgery and neuro-oncology for further work-up of her glioma, and cardiology for further evaluation of POTS. In the meantime, Nancy attended inpatient rehabilitation for her FGD, which we detail below. Of note, after her inpatient rehabilitation stay, Nancy was diagnosed with POTS by cardiology and underwent brain biopsy, revealing a WHO II isocitrate dehydrogenase-mutant astrocytoma. As part of Nancy's glioma diagnosis and treatment planning, a multidisciplinary tumor board subsequently convened. Members of this team further believed that Nancy's gait disorder was unlikely to be explained by her glioma given its anatomic location.

Upon admission to the inpatient rehabilitation facility, Nancy agreed with the diagnosis of FGD. Her stated rehabilitation goals included a desire to return to college and ambulate with "as little assistance as possible." Her rehabilitation team set goals for independence with activities of daily living and supervision level for ambulation by the time of discharge. Although her insurance program initially approved a one-week rehabilitation stay, she ultimately stayed at the inpatient rehabilitation facility for eight days. During this time Nancy worked with PT, OT, speech and language pathology, psychology, and NMT.

On initial psychology consultation, Nancy was noted to have a dysphoric, tearful affect and expressed the greatest distress regarding her gait and health-insurance difficulties related to her rehabilitation stay. She also reported that she had recently begun acknowledging her prior traumas and abuses and that this work seemed to correspond with the emergence of her physical symptoms.

Admission PT evaluation confirmed deficits in strength, motor control, balance, and endurance. Most notably, when ambulating short distances (∼50 feet, or 15 meters), Nancy exhibited excessive movements in her trunk and foot dragging, which appeared to improve with cognitive dual tasking distractions (times tables and serial subtractions). Scores on functional outcome measures, including the Berg Balance Scale, Dynamic Gait Index, and 10-Meter Walk Test, were all well below age-matched normative values. While symptoms improved during dual tasks, they did not completely resolve, and would return following cessation of the distraction technique. Minimal carryover was noted within individual PT sessions and across PT sessions. Overall, Nancy did not improve in dynamic balance, gait biomechanics, gait speed nor endurance while working for several consecutive days with PT.

Nancy's speech therapy evaluation demonstrated average to above average cognitive communication skills in domains of attention, memory, executive function, and problem-solving/reasoning. With the Speech-Language Pathologist she worked on compensatory strategies (e.g., schedules, notes, etc.) to aid in instances of fatigue.

On the sixth day of the patient's inpatient rehabilitation stay, a NMT, who commonly co-treats a variety of neurologic conditions with PT, began a RAS intervention to assist with gait retraining. At this point, functional gait features were still present and Nancy's cadence was below age-matched norms (Tudor-Locke et al. in press) at approximately 55 steps per minute (Video).

Nancy underwent four RAS trials over a single 30-minute treatment session. Within each trial the NMT and PT assessed the quality of Nancy's gait pattern. The first RAS trial utilized a metronome played out loud and Nancy was instructed to "step on each beat." Nancy quickly entrained to the metronome during the first trial and rapidly improved in coordination, balance, and trunk stability, walking 250 feet with a cadence of 80 beats per minute (bpm). Trial 2 utilized patient-preferred music, with the NMT playing a live version of one of the patient's favorite songs. Again here, the patient entrained well and walked to a cadence of 90–95 bpm sustained for 250 feet (Video). In this RAS trial, the patient's affect brightened, and she began to smile.

Subsequent RAS trials included intermittent fading of music to assess for immediate carryover effect without the rhythmic stimulus. Immediate carryover was achieved with cadence sustained at 90–95 steps per minute. The standardized protocol for RAS typically recommends tempo increases of 5–10% per trial (Thaut, McIntosh, and Rice 1997; Thaut and Rice 2014). However, due to Nancy's initial positive response, the tempo of the rhythm advanced more rapidly to a normative cadence on the latter trials. Regarding level of supervision for each of these trials, the PT started with hand-held assist x 1 and progressed to close guard supervision.

Approximately 10 minutes after these RAS trials, Nancy's gait was reevaluated without any rhythmic stimulus and she continued to demonstrate delayed carryover, with generally sustained improvements in gait parameters, albeit to a slightly less robust degree (Video). During the delayed carryover assessment, Nancy's right hand and index finger moved in time to a rhythm. At the end of the video, Nancy tells her PT, "I just needed to have a metronome in my head and then it works." Table 2 compares Nancy's gait features before, during, and after RAS.

Nancy was discharged home two days after the RAS trial. On discharge assessment, her PT noted overall improvements in gait with resolution of excessive truncal and limb dragging following "cues to internalize walking to the beat." Nancy was however still noted to fatigue quickly following ambulation of longer distances. Subsequent outpatient follow-up with a neurologist/neuropsychiatrist specializing in

Table 2
Comparison of gait features before, during, and after RAS

	Baseline	RAS trial	Carryover
Music/rhythm	Absent	Live music	No external rhythm/music is playing; however, the patient moves her hand in time to an inaudible internal rhythm
Phenotypic gait features	Narrow base of support, excessive truncal sway, foot dragging, rapidly changing gaze directionality	Gait appears normalized	Appearance of a mild sideways tilt and slightly greater variability in base of support
Cadence	55 bpm	90–95 bpm	90–95 bpm
Level of supervision	Hand-held support	Hand-held support progressing to contact guard	Contact guard
Affective display	Euthymic, neutral expression	Bright, smiling	Bright, smiling

Abbreviations: bpm, beats per minute; RAS, rhythmic auditory stimulation.

FND redocumented ongoing resolution of her FGD with particular benefit from the music-based intervention.

The rapid resolution of Nancy's symptoms during the last two days of her inpatient rehabilitation stay speaks further to the likelihood that her gait symptoms were functional, as in the absence of glioma-specific treatment, tumor burden would not be expected to improve in that short duration of time. While it was not ultimately believed that Nancy's gait disorder was directly caused by her glioma, it remains possible that her lesion represented an underlying structural vulnerability for the development of a functional neurological symptom (Perez et al. 2017). Orbitofrontal and insular lesions can alter sensory integration affect processing (Chang et al. 2013; Rolls 2004), which are known predisposing factors for FND (T. R. Nicholson et al. 2020).

Three months after discharge from inpatient rehabilitation, Nancy's follow-up neuroimaging confirmed stabilization of her astrocytoma. However, over subsequent years Nancy's tumor progressed, and she has since undergone proton beam radiation therapy. Since her RAS intervention until the present—four years later—Nancy's FGD remains resolved. She continues to ambulate with a normal gait pattern, though fatigue and pain limit her endurance and she still requires a wheelchair for long-distance mobility. Nancy has since been graduated from college.

3. Discussion

A 24-year-old woman with a FGD showed resolution of truncal displacement, foot dragging, and normalization of gait speed during and following a RAS intervention delivered during an inpatient rehabilitation stay. Her FGD symptoms remain remitted four years later. There are many perspectives through which we can interpret this patient's symptom improvement.

3.1. Mechanisms unrelated to RAS

First, it is possible that RAS training may have been irrelevant to Nancy's symptom resolution. During RAS training, Nancy may have coincidentally experienced a rapid, spontaneous remission of her FGD. FND onset (Cock and Edwards 2018; Lidstone et al. 2020), resolution (Nielsen et al. 2015; Stephen et al. 2021), and symptom relapse following a period of recovery (Fobian and Elliott 2019) can all occur abruptly. Sudden recovery can follow resolution of a perpetuating factor such as a psychological stressor (Baker et al. 2021). On a population level, the literature most often associates rapid remissions with shorter duration of functional symptoms on the order of days to weeks (Gargalas et al. 2017; Gilmour and Jenkins 2021), though rapid remissions of longer-lasting symptoms are also possible. It could have been mere coincidence that Nancy's symptom resolution occurred at the same moment she was undergoing RAS treatment.

Second, it is possible that resolution of Nancy's FGD reflected the several hours of rehabilitation therapy she received in the few days prior to the RAS intervention. Relatively abrupt and lasting remissions of chronic FND symptoms have also been documented following a brief, intensive rehabilitation treatment lasting on the order of days to weeks (Czarnecki et al. 2012; Jacob et al. 2018; Nielsen et al. 2017). While this is certainly possible in Nancy's case, it may be slightly less probable given that her treating PT and OT had documented minimal carryover gains during their preceding therapy hours.

3.2. RAS-specific mechanisms

It is also possible that RAS aided Nancy's rehabilitation by acting upon specific neurobiological networks. RAS has been shown to be effective for gait retraining across a range of neurological disorders including Parkinson disease (PD), stroke, and traumatic brain injury (Hausdorff et al. 2007; Hayden et al. 2009; Hurt et al. 1998). With these populations, RAS has been shown to improve coordination, gait symmetry, stride length and cadence (Schaffert et al. 2019; Thaut and Abiru 2010). In PD, RAS may enhance motor timing and thus counter basal ganglia dysfunction (Schaffert et al. 2019). Following a paretic stroke, RAS may facilitate voluntary movement due to improved coordination of muscle activation (Richards, Malouin, and Dean 1999; Yoo and Kim 2016) or greater synchronization of motor planning and execution (Cha et al. 2014; Yoo and Kim 2016).

The pathophysiology of gait dysfunction in a FGD differs greatly from the pathophysiology of gait dysfunction due to PD or stroke. Most notably, gait changes due to FGD broadly reflect dysfunctional connectivity rather than neurodegeneration or structural injury to the brain. Functional magnetic resonance imaging studies (fMRI) on FND have implicated networks underlying attention, agency, motor control, and emotion regulation (Edwards et al. 2011; Espay et al. 2018; Lin et al. 2020; Nahab et al. 2017). Given that functional rather than structural changes predominantly underlie FGD, this condition may also possess a greater capacity for reversibility with rehabilitation efforts compared with other neurologic disorders.

3.3. Modulation of attention

Multiple elements of RAS could benefit individuals with FGD. At the very beginning of the RAS session, Nancy was invited to ambulate while watching a NMT playing live music. The dynamic auditory-visual stimulus of the NMT—a live interacting person—may have functioned as an elaborated distraction mechanism. Neurobiological models of FND describe excessive self-directed attention as a core pathophysiologic mechanism (Edwards et al. 2012; Edwards and Rothwell 2011). Correspondingly, distraction techniques, which temporarily reduce this excessive self-focus, are a well described therapeutic tool for FND rehabilitation (Breitkopf et al. 2018; Nielsen et al. 2015; Scott and Stone 2014).

We can hypothesize that excessive self-directed attention was a feature of Nancy's FGD given her repeated positive response to distraction techniques of cognitive dual tasking (alphabet backwards, serial subtraction, times tables) during physical examination and rehabilitation sessions preceding the RAS intervention. The human, multimodal stimulus of RAS may have been sufficiently compelling to momentarily help to shift Nancy's attention from an internal to an external focus, simultaneously facilitating improvement in gait parameters. With RAS, the cognitive load of ambulation may have thus been reduced.

3.4. Acoustic biofeedback

One of the next early steps of RAS is entrainment. To achieve entrainment an NMT matched a rhythmic stimulus to Nancy's natural cadence. The "tap function" on a smart-phone metronome is typically utilized, with the NMT tapping the metronome each time a patient steps. The metronome thus records and calculates the patient's own cadence and thereafter can be used to produce an external sound at the same rate.

Through this process, a few new tools for gait retraining may have been available to Nancy. For one, Nancy was now essentially receiving acoustic biofeedback—the sound of Nancy's feet contacting the ground during ambulation were amplified by the sound of the metronome, or later by musical tones. Sound is well-known to enhance motor learning during motor tasks. Natural movement sounds are believed to provide one with information about agency and facilitate discrimination between one's own movements and that of others (Schaffert et al. 2019).

Accordingly in sport, depriving athletes of auditory feedback during performance and training (the sound of hitting a tennis ball with a tennis racket; the sound of hurdling, etc.) decreases athletic precision, whereas providing auditory feedback enhances performance (Schaffert et al. 2019). A reduced sense of agency over motor control is also a common feature of FGD (Espay et al. 2018). It is possible that hearing an augmented auditory signal of one's own movement during RAS entrainment, enhanced Nancy's sense of motor control.

Table 3
Entrainment and rhythmic frequency modulation for functional tremor versus functional gait disorder

	Functional tremor	Functional gait disorder
Functional movement	Limb shaking	Abnormal gait
Target of entrainment	Unaffected limb	Gait
Alteration of external rhythm	Slowing down rhythm	Speeding up rhythm
Desired outcome	Movement suppression	Gait symmetry, coordination, speed

3.5. Entrainment and frequency modulation

Beyond providing multimodal feedback, RAS also fundamentally utilizes rhythmic entrainment. Neurobiologically, entrainment involves priming the auditory-motor pathways, in which rhythmic patterns generate expectations for when auditory events will next occur. Through auditory-motor coupling, a primed motor system anticipates an upcoming movement, preparing the body for motor execution (Schaffert et al. 2019; Thaut 2015). RAS works particularly well for movements that are oscillatory and intrinsically rhythmic, such as walking, helping to enhance the quality, precision, and consistency of movements (Thaut and Abiru 2010). Entrainment was achieved in Nancy's case (Video), because close matching between Nancy's cadence and the musical rhythm played by the NMT was visually apparent.

After entrainment is established, the next step of RAS training involves modulating the rhythmic frequency. In fact, frequency modulation of an entrained rhythm has already been diagnostically and therapeutically used for other subtypes of FND, namely functional tremors (Bartl et al. 2020). Entrainment is diagnostically considered a clinical "rule-in" sign for functional tremor. In this case, a patient is asked to rhythmically move an unaffected body part while the examiner monitors for entrainment or suppression of movement in the affected body part (Kim, Pakiam, and Lang 1999; Roper et al. 2013; Stone 2009). Therapeutically, OT consensus recommendations (C. Nicholson et al. 2020) describe a strategy in which rehabilitation therapists invite a patient to superimpose a new voluntary rhythm onto the functional tremor, then gradually slow down the rhythm until the functional tremor is suppressed. Correspondingly, an experimental study that evaluated "retraining" functional tremors with external rhythmic cueing showed benefits to a proportion of participants (Espay et al. 2014).

RAS and functional tremor rehabilitation share many similar elements, namely entrainment of a bodily movement, followed by manipulation of an external rhythm for the purpose of altering a bodily movement. In Nancy's case, these efforts delivered more formally through RAS appeared to positively benefit a FGD. Table 3 identifies key similarities and differences between entrainment and frequency modulation that are applied to functional tremors and FGD.

Formal standards for delivering RAS were largely maintained during Nancy's treatment with one notable exception. Nancy's NMT advanced the rhythmic stimuli more quickly than the rate of 5–10% recommended in standard RAS protocols designed for treating individuals with structural neurological conditions (Thaut and Rice 2014). Across diagnoses, as an individual approaches a more normative cadence during RAS (speeding up in the present case), improvements in balance, symmetry, and coordination are often seen. During her single RAS session, Nancy progressed rapidly from a cadence of 55 bpm to 90–95 bpm. Such a rapid progression likely had a higher probability of occurring with Nancy's FGD compared with a structural gait disorder because no accompanying abnormalities in strength or tone were involved in Nancy's case.

Given that ambulation involves greater complexity of motion and coordination than a functional limb tremor, one may also conjecture that a more standardized treatment approach, using RAS formally rather than informally applying its principles as is done at the bedside for functional tremors, may be deserving of further scientific inquiry.

3.6. Carryover

A final stage of RAS involves assessment for carryover—the extent to which gait improvements persist once the rhythmic stimulus ends (Thaut and Rice 2014). Across studies, immediate, delayed (minutes later) and longer-term carryover effects are often measured. In stroke, studies have shown benefits of a three month course of rhythm and music therapy, including improvements in balance and gait, persisting through to follow-up six months later (Bunketorp-Käll et al. 2017). In PD, training has resulted in carryover lasting minutes to weeks (Hausdorff et al. 2007; McIntosh et al. 1997; Nieuwboer et al. 2009), though these effects may later deteriorate with disease progression, as can be expected for a neurodegenerative disorder (Bella et al. 2018).

It has been hypothesized that carryover may reflect some degree of motor function restoration, involving neuroplasticity in networks underlying motor control of gait (Grau-Sánchez et al. 2013; Hausdorff et al. 2007). In Nancy's case, carryover was observed immediately, after a 10-minute delay, and then continued to persist. The carryover section of the video demonstrates Nancy's application a rhythmic compensatory strategy moving her hand in time to an inaudible internal rhythm, after a 10-minute post-RAS delay. Ultimately, Nancy's carryover occurred more quickly, robustly, and permanently than is typical for other neurologic conditions such as stroke or PD. Again this likely represents the predominance of a functional rather than a structural pathophysiology of FGD and its potential for rapid reversibility (Gargalas et al. 2017; Gilmour and Jenkins 2021; Nielsen et al. 2015; Stephen et al. 2021). Following the RAS intervention, an internalized rhythm subsequently became an effective self-management tool, during which Nancy would imagine a beat playing inside her head to assist in her walking.

3.7. The significance of music

Given that the RAS intervention involved music, rather than just rhythm, a few final considerations are worth stating here. Music processing is implicated to involve many brain regions, ranging from phylogenetically older areas to the neocortex (Koelsch 2018). These systems in turn overlap with circuits involved with FGD. As the effector systems compete for behavioral expression, sufficiently powerful contributions from music lead to associated action tendencies (dancing, tapping, singing, etc.), which may ultimately override mechanisms of FGD.

Music historically has also served for social bonding. Playing or engaging in music together encourages self-other merging by interpersonal synchrony and strengthening relationships (Tarr, Launay, and Dunbar 2014). Patients with FND often have insecure attachment styles (Luyten and Fonagy 2020). From an interpersonal perspective, the therapeutic benefit of socioemotional bonding with the NMT may be particularly pronounced within the FGD population, with subsequent reduction in symptoms. This would explain why the interaction with the NMT may have greater therapeutic yield than the patient simply listening to recorded music or a metronome. The interaction and formed relationship can then be internalized for later recall during the carryover phase.

In addition to social bonding, affective responses to music may also contribute to FGD symptom alleviation in this case (Pick et al. 2019). The music's effect on the Nancy's affective state (bright, smiling) suggests a change in mood and motivation compared to her previously recorded neutral expression at baseline. Reduced perceived threat and associated anxiety, whether on a conscious or unconscious level, could also facilitate improvements in Nancy's gait.

4. Conclusion

In the present case, a patient exhibiting a classic FGD pattern benefited from a RAS intervention. When ambulating in the presence of an NMT playing patient-preferred music, the patient entrained to a rhythm with normalization of multiple gait parameters that persisted into a carryover period. Use of internalized rhythmic cues subsequently remained a strategy for helping the patient sustain treatment effects. Four years following her initial presentation, the patient's FGD remained improved, though fatigue and pain continued to limit her endurance. RAS may hold potential as a tool for treating FGD given its inherent ability to reinforce a "normal" or more automatic movement pattern for walking with a rhythmic stimulus, while simultaneously creating a distracting, or even psychologically pleasant environment. The theories and practice of RAS are complementary to existing consensus based FND rehabilitation recommendations. More research is needed to determine the possible benefit of RAS in the treatment of FGD.

Conflict of interest

C.H. is a salaried employee of MedRhythms, Inc., which contracts neurologic music therapy services for Spaulding Rehabilitation Network. D.L.P. has received honoraria for continuing medical education lectures in functional neurological disorder and is on the editorial board of Epilepsy & Behavior. G.P. has received funding from the Foundation for Physical Medicine and Rehabilitation to study the role of Rhythmic Auditory Stimulation for Functional

Neurologic Disorder. There are no other financial or non-financial conflicts to disclose.

Supplementary material

Video: https://youtu.be/eZUyChzrI7s

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