The effect of internal cueing strategies on gait in Parkinson's disease and underlying mechanisms: A structured review

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A thesis submitted to Auckland University of Technology in partial fulfilment of the requirements for the degree of Master of Health Practice (MHPrac)

2021

Faculty of Health and Environmental Sciences

Abstract

People with Parkinson's (PWP) typically present with postural instability and gait impairment leading to a high number of falls. Gait rehabilitation that incorporates movement guidance training with sensory cueing and augmented feedback has been successfully investigated in the past to facilitate gait improvement in PWP. Despite showing clinical efficacy, there are limitations to the use of external cues such as device cost and the implementation of cue strategies in daily functional activities. An alternative approach is to use internal cueing which, although less commonly used, may yield similar benefits. This structured review examines the underlying mechanisms and efficacy for use of internal cues to reduce gait impairment among people living with Parkinson's disease. Based on 14 studies, findings suggest that internal cueing improves gait speed and step length. Findings also suggest a selective response of cueing strategies on gait outcomes. While further research is required, internal cueing may offer a low-cost self-management tool to improve gait outcomes.

Keywords

Parkinson's disease; gait performance; internal cueing

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Attestation of Authorship

"I hereby declare that this submission is my own work and that, to the best of my knowledge and belief, it contains no material previously published or written by another person (except where explicitly defined in the acknowledgements), nor material which to a substantial extent has been submitted for the award of any other degree or diploma of a university or other institution of higher learning."

Co-Authored Works

This dissertation is the primary work of Meetu Wadhera. The supervisory team of Professor Denise Taylor and Dr Sue Lord made valuable contributions to stream line the concept of the research question and methodology adopted in the dissertation. They also offered valuable improvements to this process through the process of iterative editing.

Acknowledgements

I would like to thank my supervisors Dr Sue Lord and Professor Denise Taylor for providing guidance and feedback throughout this project. Thanks to Dr Sue Lord for your comments and feedback throughout this project which inspired me to think from multiple perspectives and made this project both enjoyable and enriching. I would also like to thank Auckland District Health Board for supporting this project. Thank you to my loving family, my husband and children (especially my daughter) for their unconditional support.

Introduction

Over the last decade, the global burden of Parkinson's disease (PD) has more than doubled from 2.5 million in 1990 to 6.1 million in 2016 (Rocca., 2018). In New Zealand alone the number of People with Parkinson (PWP) will double over the next 25 years (Myall et al., 2017). PD is associated with loss of dopaminergic neurons in the basal ganglia which play an important role in the production and control of automatic movements by facilitating the initiation of movement sequences (Muthukrishnan et al., 2019). Disruption in the basal ganglia lead to movement disorders manifesting as tremor, rigidity, bradykinesia or hypokinesia (slowness of movement), freezing of gait (FOG; feeling of feet being glued to the ground) and abnormality in posture and gait (Muthukrishnan et al., 2019; Olson et al., 2019). These symptoms predispose individuals to an increased number of falls in PD (Muthukrishnan et al., 2019; Olson et al., 2019). Pharmacological treatment via dopaminergic replacement therapy is effective in managing certain motor and non-motor symptoms associated with PD, although the effect of medication on postural instability and gait asymmetry is either inconsistent or minimal (Muthukrishnan et al., 2019). The short-stepped, shuffling, forward stooped gait with asymmetrical arm swing is a hallmark feature in PD affecting an individual's functional tasks and participation in social and community activities (Morris et al., 2010).

Physical therapy involving gait training is used extensively in the management of PD to improve walking (Morris et al., 1996; Harrison et al., 2018). One of the rehabilitation approaches developed over the years is movement guidance training (Morris et al., 2010; Keus et al., 2004; Samyra et al., 2007; Nieuwboer., 2015). This training incorporates provision of sensory cues (external signal that can be extracted by sensory input such as visual, auditory or tactile cues) and augmented feedback (like verbal prompts or visual feedback to guide/reinforce movement) to promote goal oriented mode of motor control, posture and gait parameters (Morris et al., 2010; Keus et al., 2004; Samyra et al., 2007; Nieuwboer., 2015). Cueing is defined as "using external temporal or spatial stimuli to facilitate movement initiation and continuation" (Nieuwboer., 2007) (p 134).

The series of experimental studies conducted by Morris and colleagues in the 1990's demonstrated that movement guidance gait training with the use of external cueing enabled PWP to improve their gait quality by walking with longer steps at near normal cadence (number of steps per minute) (Morris et al., 1994 a,b; 1996). Application of movement guidance strategies through cueing is also supported by the best practice physical therapy guidelines in Parkinson's disease (Keus et al., 2007, 2014; Grimes et al., 2019). Various types of cueing strategies are used and can broadly include either external or internal cueing strategies. The following section will outline and review the literature concerning external cues to understand the potential use of internal cues as an alternative.

External cueing strategies

External cueing modalities include auditory cueing (Nieuwboer et al., 2007); visual cueing (Shen & Mak., 2012; Donovan et al., 2011), somatosensory or tactile cueing (Wegen et al., 2006; Stuart & Mancini., 2020), and 'on demand' cueing through external devices such as laser shoes and smartphone (Velik et al., 2012; Ginis et al., 2016). Visual and auditory cueing are popular cueing therapies (Spaulding et al., 2013). Several proposed mechanisms relate to improved cue based gait performance. Cue modalities act selectively on features of gait but broadly act similarly to suppress pathological basal ganglia activity through activation of unaffected visuomotor and corticostriatal pathways (Azulay et al., 1999; Sarma et al., 2012). Despite their reported beneficial effects, external cues are not universally applicable in all stages of PD and not all PWP respond well to external cueing. Primary barriers for reduced responsiveness to external cues include cognitive overload and lack of perceptual rhythmicity (Nieuwboer., 2015).

Visual cues require attentional or cognitive resources and optic flow to increase stride length (Morris et al., 1996; Azulay et al., 1999) which is not always present, especially in more advanced stages of PD due to cognitive impairment. Auditory cues such as rhythmic auditory stimulation (RAS) activate corticostriatal pathways to compensate for the impaired internal rhythm of the basal ganglia and positively influence cadence, velocity and stride length (Chen et al., 2006, 2008; Grahn & Rowe., 2009; Spaulding et al., 2013). Introducing these cues in early PD can interfere with the intact internal basal ganglia rhythm. This increases gait variability resulting in a less consistent gait pattern. However, auditory cues are beneficial as the disease progresses by acting as an external pacemaker for the impaired basal ganglia rhythm (Lirani-Silva et al., 2019). The lack of portability of external cues limits their applicability in functional tasks. Cost is also a potential barrier. Most of the traditional external cues only provide continuous cueing (Nieuwboer et al., 2007; Donovan et al., 2011). This results in cue dependency and habituation as reported by Spildooren et al. (2012). They found that the positive effect of cues leading to gait improvement reduced dramatically following the removal of cues.

‘On demand’ cues

‘On demand’ cueing is an alternative approach where the cue is presented either externally through a device, or internally through self-generation when gait deviates from normal (Velik et al., 2012). This helps to redirect attention back to the walking task which prolongs cue efficacy (Velik et al., 2012). Ginis et al. (2017) measured efficacy of four different input cueing modalities including ‘on demand’ cues in PWP with respect to their cognitive status, subjective preference and FOG. Participants with and without FOG received gait training with *continuous cueing* (continuous metronome beats matched to comfortable cadence), *intelligent cueing* (bouts of eight beats indicating comfortable cadence), *intelligent feedback* (instruction to adapt gait speed when cadence deviated from the comfortable target) or *no input*. Participants 'with FOG' produced their most stable gait during continuous cueing, whereas the gait of participants 'without FOG' improved with intelligent cueing. PWP with lower cognitive scores and FOG were associated with worse gait with intelligent input. Results from the study suggest that 'on-demand' cues enable individuals to modify the gait through self-adaptation via attention. This is effective for PWP in the early stages of the disease, when individuals present with less cognitive and gait deficit. These cues potentially help to counteract the monotony of an external cue, avoid habituation and free attentional resources for more efficient use (Nieuwboer., 2015; Velik et al., 2015; Ginis et al., 2017).

'On demand' internal cues

The substitute of 'on demand' cueing without an external device is through self-generation of cues internally. Examples include, internal attentional cues by focussing on step length (walking while saying 'big step, big step'), or internal musical cues by singing aloud or mental singing while matching footsteps. The evidence of this approach dates back to mid-1990 when Morris et al. (1996) reported similar improvement in gait velocity and double limb support duration (DLS) in the PD group with both attentional and visual cues. The more recent study by Harrison et al. (2018), examined the effects of internal musical cues (singing) and externally generated musical cues (music) on forward and backward walking in three groups of people, including healthy young, healthy old and PWP. PWP showed greater improvement in gait characteristics compared to their healthy counterparts improving in both forward and more challenging backward walking direction with internal musical cues.

One of the proposed advantages of internally generated self-cues is the elimination of the need for adjustment of every foot step to synchronise to external cues, which helps reduce cognitive load and attentional resources (Harrison et al., 2018; Peterson &

Smulders., 2015). Internal cues offer a more cost effective and practical cueing replacement in daily functional tasks such as turning around in the corridor or backward stepping to sit in the chair due to their availability through ‘oneself’. Despite the potential advantages of on-demand internal cueing strategies, they are not used to the same extent clinically as external cues. The barriers may include reduced awareness amongst clinicians regarding the potential use of internal cues, and a limited number of studies reviewing the clinical efficacy of internal cues for improving gait performance in PD. Additionally huge demand on the public health system may lead to delayed referrals for physiotherapy assessment and rehab missing the therapeutic window to incorporate ‘internal cueing strategy’ due to more pronounced cognitive and gait deficit as PD progresses.

This structured review seeks to answer ‘is internal cueing effective in improving gait in people with PD?’ The review has two aims. The first aim is to explore current literature concerning the underlying mechanisms of internal cues as a strategy for gait performance in PD. The second aim is to examine efficacy of internal cues. If internal cueing is found to be effective, it has the potential to be used as a low cost, patient-controlled strategy in PWP to improve mobility. Before discussing the mechanisms of how cueing might help improve gait dysfunction in PD it is important to understand the role and functions of the basal ganglia in relation to gait and cognition, in particular attention.

Basal ganglia

Neuro physiology and circuits

The term ‘basal ganglia’ refers to a group of sub cortical nuclei in the brain, primarily composed of four main components (striatum, globus pallidus, subthalamic nuclei and the substantia nigra) and is mainly responsible for execution of automatic movements and timed responses without conscious effort (Lanciego et al., 2012). The striatum includes the caudate nucleus and putamen, and the globus pallidus consists of internal and external globus pallidus; GPi and GPe respectively (Lanciego et al., 2012). The most widely accepted classic basal ganglia model proposes that the circulation of signals between the basal ganglia and the cortex runs through two neural pathways with opposing effects on each other for effective execution of movement (see Figure 1) (Lanciego et al., 2012). Depending on the information flow, the nuclei in the basal ganglia are broadly categorised into input, output and intrinsic nuclei (Lanciego et al., 2012). The input nuclei receive incoming information; the input unit of the basal ganglia is the striatum consisting of inhibitory neurons (caudate nucleus and putamen) which receive innervation from the entire cerebral cortex (Lanciego et al., 2012). The output nuclei send information to the thalamus and consist of the GPi and the substantia nigra pars reticularis and also consist of inhibitory neurons (Lanciego et al., 2012). The intrinsic nuclei such as the GPe, the subthalamic nuclei and the substantia nigra pars compacta are located between the input and output nuclei and support the relay of information (Lanciego et al., 2012).

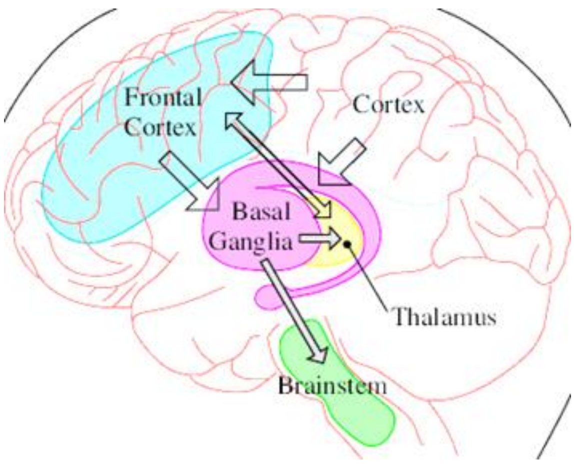


Figure.1. Cortico-basal ganglia-thalamic loop

Girard, B. et al., (2006)

Function

Contribution towards a motor set

Basal ganglia help the preparation and maintenance of motor plans and enable the motor functions to be carried out functionally and appropriately (Lewis et al., 2000; Morris et al., 1996). This is done through the cortico-basal ganglia-thalamic loop. In the direct cortico-basal ganglia-thalamic loop (see Figures 1 and 2), the cortex provides excitatory input to the striatum inhibiting the GPi and substantia nigra influence on thalamus. This results in excitation of thalamic activity which in turn excites the cortex leading to cortical activation and execution of voluntary movement (Lanciego et al., 2012). On the other hand, the indirect cortico-basal ganglia- thalamic pathway takes a longer and more indirect route. It excites inhibitory neurons of the GPi and substantia nigra which subsequently leads to suppression of the thalamic and cortical activity and movement (Lanciego et al., 2012).

The balance of activity between the direct and indirect pathway is critical for motor control and execution of functional voluntary movement and is modulated by dopaminergic neurons present in the substantia nigra (Lanciego et al., 2012). Through this loop, the basal ganglia contribute to the actions of the cortical motor set (Lewis et al., 2000; Morris et al., 1996). This specific role of the basal ganglia also helps to consolidate proprioceptive feedback of body position to guide an on-going movement towards a target (Almeida et al., 2005).

Impairment of basal ganglia in PD disrupts the cortico-basal ganglia-thalamic circuitry loop. This leads to an inability of the motor set to generate sufficient force and movement to initiate a normal stepping gait causing gait hypokinesia (Morris et al., 1996).

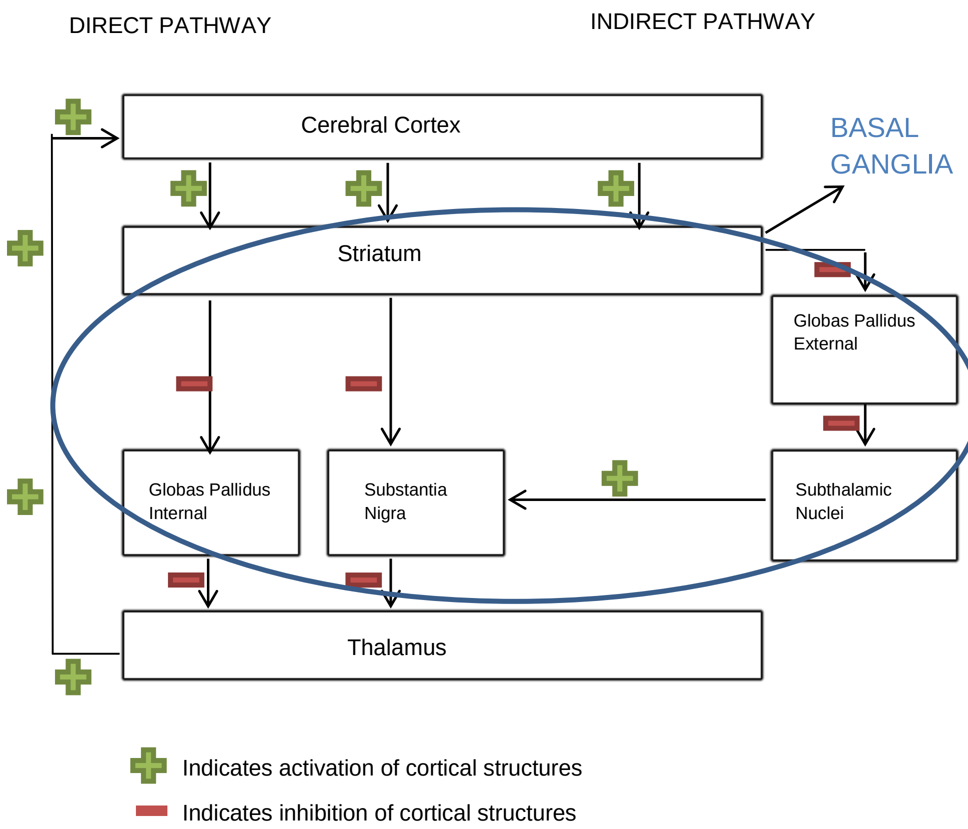


Figure.2. Diagrammatic illustration of Cortico-basal ganglia- thalamic loop

Provision of internal cue

The second significant role of the basal ganglia is to provide an internal cue or trigger for well learned sequential movements such as eating, writing, talking and walking to be internally regulated and carried out automatically (Lewis et al., 2000). This was evident in sequential movement research studies conducted in both primates and humans (Brotchie et al., 1991 a,b; Seitz & Roland., 1992). Seitz and Roland (1992) conducted upper limb positron emission topography (PET) in order to observe areas of activity in the brain while participants were learning a new task. During the early stages of learning the task, high levels of metabolic activity in the cortex, pre motor and supplementary motor areas (SMA) were observed. This was associated with minimal activity in the basal ganglia. However, repeated practice of learning a novel task coincided with a marked increase in basal ganglia activity, and a decline in activation of the cortical regions. This evidences the significant role the basal ganglia played in allowing a shift of movement performance from conscious control to automaticity.

The above finding also matches with the Fitts and Posner proposed model of skill acquisition. This model states that people pass through three stages of learning when acquiring a new skill, namely cognitive, associative and autonomous stages (Taylor & Ivry., 2012). The cognitive phase consists of employing conscious attentional strategies to gain an overall understanding of the task. This implies an increased cognitive load resulting in enhanced cortical activation (Taylor & Ivry., 2012; Morris et al., 1996). This is followed by an associative phase where there is a generalised understanding of the task, implying a slight reduction in the cognitive load (Taylor & Ivry., 2012; Morris et al., 1996). In the last autonomous phase, motor skill is well established; movements are accurate, consistent and efficient with limited demand on attentional resources (Taylor & Ivry., 2012; Morris et al., 1996). This implies less cognitive load and activation of cortical regions.

Experimental studies including primates by Brotchie et al., (1991 a,b) looked at basal ganglia's role in triggering automatic movements. They detected a gradual increase in neural activity in the SMA during the pre-movement period, but once the external signal to movement took place, the neural activity in SMA suddenly stopped. The sudden drop in SMA neural activity was the result of interaction between the basal ganglia and SMA for a well learned sequential movement. During a movement sequence, neurons in the globus pallidus of the basal ganglia are discharged phasically at the end of each movement in the sequence (Morris et al., 1994 a). This phasic output from the basal ganglia inhibits the thalamus which has projections to the SMA (Lewis et al., 2000). Through this indirect thalamic connection, the basal ganglia is able to turn off preparatory activity in the SMA. Instead, it acts to trigger each sub movement in the sequence enabling well learned sequential movements to run automatically (Morris et al., 1994 a; Lewis et al., 2000). Morris et al. (1996) also noted that the internal or phasic cue from the basal ganglia is only present for the well learned sequential movements and absent for new or complex tasks. This suggests that the interaction between the basal ganglia and SMA only occurs in well learned automatic movements such as writing, walking and speaking where motor skills are acquired from a young age. Basal ganglia impairment in PD causes loss of internal cues for automatic movements affecting reciprocal gait pattern and arm swing while walking.

Internal rhythm

Basal ganglia also help generate subconscious timed responses for daily functional tasks through the internal clock (Ferrandez et al., 2003). In order to understand this, we need to review ‘temporal’ and ‘spatial’ processing; a brief explanation is provided of what each entail. ‘Temporal processing’ was first introduced by neurophysiologist Karl Lashley in 1951 in one of the chapters of his book ‘The problem of serial order in behaviour’ (Rosenbaum et al., 2007). It was further elaborated on by Mauk and Buonomano. (2004). In simple terms, in order to process sensory and motor information, our nervous system processes time in the range of tens to hundreds of milliseconds (ms) and in order to do so is reliant upon the spatial and temporal patterns of the action potentials. Incoming spatial stimuli are processed by sensory neurons; whilst temporal patterns such as interval, duration and motion discrimination are processed through a complex neural mechanism (Mauk & Buonomano., 2004). For example, the flash of a light or the pitch discrimination between two high frequency tones can be processed in a snapshot by the activation of sensory neurons, retinal ganglia neurons in the case of a light flash, and different hair cells in the cochlea in the case of two different frequency tones (Mauk & Buonomano., 2004). By contrast, patterns such as the duration of a flashed bar of light or the interval between two high frequency tones are unable to be processed by a snapshot of neural activity, and instead require the nervous system to process the underlying temporal information through a complex neural pathway (Mauk & Buonomano., 2004).

‘Temporal processing’, ‘temporal integration’ and ‘timing’ are used interchangeably and describe a wide range of timescales over which humans process time or generate timed responses (Mauk & Buonomano., 2004). Most of the timed responses generated in our daily life activities occur at a subconscious level (Ferrandez et al., 2003). For example, whilst waiting at traffic lights when driving in a known area we can anticipate the duration of light changes to start moving again, or when making a cup of tea we can anticipate the duration for boiling water in the kettle.

There are three stages of temporal information processing based on animal studies. (Church & Broadbent., 1990; Grondin., 2010; Gibbon et al., 1984). These are the internal clock mechanism that measures subconscious duration; memory mechanism compares the current duration with the duration stored in reference memory and a decision mechanism selects the appropriate response (Church & Broadbent., 1990; Grondin., 2010; Gibbon et al., 1984). These different stages involve different regions of the brain and are modulated by different neurotransmitters (Meck., 1996). The internal clock mechanism is associated with dopamine function in the basal ganglia, while the temporal memory and attentional mechanisms appear to be linked to acetylcholine function in the frontal cortex (Ferrandez et al., 2003). Basal ganglia provide internal cues for the execution of well learned automatic movement sequences and generate subconscious timed responses through the internal clock. Basal ganglia dysfunction disrupts the internal clock or internal rhythm, which affects the fluency and smoothness of the voluntary movement.

In summary, the basal ganglia play a pivotal role by contributing towards a motor set for whole movement sequences, and providing internal cues and rhythm for the execution of well learned, sequential and automatic movements such as gait. Disruption of the basal ganglia causes impairment of well learned movement sequences, resulting in movement disorders and gait dysfunction, including gait symmetry and stability. External and internal cueing strategies increase sensory and attentional drive through by passing the defective basal ganglia. This helps to generate adequate force and amplitude resulting in increased step length, stride and walking speed in PWP (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007; Werner et al., 2010). The next section explores the underlying pathophysiology of gait dysfunction in PD.

Pathogenesis of gait impairment in Parkinson's

Gait hypokinesia resulting in a short- stepped shuffling, gait is a hallmark feature of PD (Morris et al., 1996). Past studies investigating gait parameters in PWP have linked gait hypokinesia to reduced stride length, decreased cadence and an increase in DLS (Bowes et al., 1990). Although the locomotor deficits were described early in PD research literature, the underlying mechanism of gait impairment was poorly understood and documented. To determine the underlying motor control mechanisms responsible for gait hypokinesia in PD, Morris and colleagues conducted a series of laboratory experiments in 1994 and 1996 (Morris et al., 1994 a,b; Morris et al., 1996). These studies now form the basis of our understanding and provide an insight into the pathogenesis of hypokinesia in PD.

In the first study spatial and temporal gait parameters (velocity, cadence, stride length and DLS) were compared in PD subjects and age/height matched control subjects in three walking conditions; slow, normal and fast walking. PD subjects had a significantly slower walking velocity compared to their age matched counterparts in all three walking conditions. Further, PD subjects had less than normal stride length for each condition, and lower mean cadence than control subjects for normal and fast walking. There were no significant differences between the PD and control group for DLS duration in all three walking conditions. Linear regression analysis indicated that for any given velocity, the stride length was shorter and cadence higher in PD subjects. The second study investigated the differences in the three main gait parameters; stride length, cadence and DLS between the PD subjects and the age matched control group. Gait velocity was not changed, the control group was asked to walk at the same speed as their PD counterparts. The results demonstrated that with velocity controlled, the mean stride length in the PD group was significantly smaller (PD=0.924 m, control=1.10 m) and mean cadence was significantly greater (PD=102.05 steps/min, control=88.3 steps/min) than the control group. This implies that PWP walk with smaller steps and at a faster pace than those without PD, which is primarily used as a compensatory strategy to maintain gait speed. There was no significant difference observed in DLS between groups at the controlled velocity.

The above studies showed that gait hypokinesia results in a shorter stride and higher cadence in PWP compared to their healthy counterparts for any given velocity, but the authors were unable to pinpoint the underlying cause of this pathology. For this reason, the third study consisted of a series of experimental conditions with external cueing (auditory cueing through metronome and visual cueing through floor markers) to compare spatial and temporal gait parameters, primarily focusing on stride length and cadence between PD subjects and healthy controls. PD subjects had difficulty adjusting their stride length when reliant on the internal motor mechanisms. They achieved an improvised stride length with external cues, although the range of increase was still considerably lower than the healthy counterparts. On the other hand, PD subjects encountered little difficulty in controlling their cadence either through internal control or with the assistance of external cueing. Additionally, they were able to match the timing of their footsteps to the cues to achieve as near normal cadence as their healthy counterparts.

Taken together, this series of studies established the underlying cause of gait hypokinesia in PD is linked to the internal inability to generate steps of adequate amplitude or stride length and outlined two possible causes for this impairment. As explained earlier, one of the functions of the basal ganglia is to provide an internal cue for sequential, well learned movements such as walking. Disturbance in the basal ganglia would mean disruption of the SMA preparatory activity and absence of the phasic cue, leading to impaired motor preparation and difficulty in stringing each sub movement together, causing shorter steps (Morris et al., 1996). Gait hypokinesia may also be due to impairment of the cortical motor set activity which is influenced by the direct and indirect cortico-basal ganglia- thalamic pathway. Dysfunction of basal ganglia disrupts the circuitry loop and inability of the motor set to generate sufficient force and movement to initiate a normal stepping gait (Morris et al., 1996).

In order to determine the underlying mechanisms of gait hypokinesia and reduced stride length, Morris and colleagues conducted another series of experiments in 1996. The primary aim this time was to explore motor control mechanisms that contribute to the normalisation of stride length using two different cueing strategies. The first study examined whether external cues (visual cues) used similar or different motor control mechanisms to training with an attentional strategy. Primary outcome measures were gait velocity (meters/minute), walking cadence (steps/minute), mean stride length (meters) and DLS (% gait cycle). The visual cues were laminated white floor markers, and the attentional cue was delivered through cognitive training until the subjects developed a mental picture of the ‘correct step size’. The same PWP participated in both visual and attentional cueing strategy conditions with experiments conducted on separate days. Outcome measures were completed prior to the intervention (baseline phase), immediately post intervention (intervention phase) and two hours after the training stopped (retention phase). At baseline PWP in visual and attentional cueing conditions mobilised with a shorter stride, reduced velocity and increased DLS compared with the control group. A statistically significant difference was found in the velocity and DLS in the visual cue group from baseline 49.7 m/min and 35.9 % gait cycle to the intervention phase 72.2 m/min, p=0.001 and 30.2 % gait cycle, p=0.001 respectively. Gait training using attentional cueing strategy produced similar beneficial effects on gait parameters with a statistically significant increase in the velocity and DLS from baseline 52.3 m/min and 35.9 % gait cycle to the intervention phase 71.4 m/min, p=0.001 and 29.9 % gait cycle, p=0.001.

Gait parameters post visual and attentional cue training were similar to gait parameters of the control groups in both experimental studies. PWP were able to maintain the improvised gait parameters for a short period of time even when the cues were removed. The second study measured the same gait parameters with the addition of a secondary task, resulting in poor gait quality due to increased cognitive load. These results indicate that both visual and attentional cueing strategies enhance stride length. This leads to increased gait velocity during single task conditions by directing attention, supporting the view that impairment in stride length is linked to the cortical motor set and cortico-basal ganglia- thalamic pathway, rather than automatic stringing of sub-movements through internal cueing.

To summarise the results of this seminal and influential body of work, gait hypokinesia in PD is caused by an inability to generate an adequate stride length which is linked to the inadequate contribution by the basal ganglia to the cortical motor set. PD is also associated with defective internal rhythm resulting in festination and FOG (Satoh et al., 2008). FOG as explained in the earlier section, is an inability to initiate or continue intended locomotion resulting in the feeling of ‘feet being glued to the ground’, affecting almost a third of PWP (Horin et al., 2020 a; Moore et al., 2007). PWP with FOG have higher gait variability than their counterparts without FOG (Horin et al., 2020 a). In recent times, gait hypokinesia and increase gait variability is broadly included in the umbrella term of postural instability and gait disturbance (PIGD). These combined symptoms of gait dysfunction associated with slower gait speed predispose PWP to a higher risk of falls, loss of independence and increased mortality (Hausdorff., 2009; Gurevich & Hausdorff., 2003; Yogev et al., 2007; Mirelman et al., 2019).

PWP retain the ability to normalise their gait pattern through bypassing the defective basal ganglia. They can generate adequate stride length and normal stepping patterns by using cueing and attentional strategies. Incorporation of goal based cognitive and motor approaches in the early to moderate stages of PD offers the most effective rehabilitation by inducing neuroplastic changes (Ferrazzoli, Ortelli, Madeo, Giladi, Petzinger & Frazzitta., 2018). The following section elaborates on the underlying mechanisms of commonly used external cues to better understand the rationalisation for an alternative internal cue approach.

Proposed cue mechanisms

Mechanism of action for visual cueing

As reported earlier, the beneficial effect of visual cueing on gait parameters in PWP has been established (Morris et al., 1996; Azualy et al., 1999; Lewis et al., 2000). Two possible explanations for this effect relate to an increase in attentional drive to enhance motor control, and increased optic flow during gait (Morris et al., 1996; Azualy et al., 1999).

Increase attentional drive

The first hypothesis suggests the use of external visual cues (such as stripes placed on the walking surface) act to focus attention on the stepping process which enhances the conscious cortical preparatory activity (Morris et al., 1996; Azulay et al., 2006). Walking is a rhythmic and automatic process, which in a predictable environment does not require significant involvement of cortical structures. This leads the basal ganglia take over and provide internal cueing for smooth automatic movement to take place (Lewis et al., 2000). The administration of external cues and employment of conscious attentional strategies to the stepping process helps to by-pass defective basal ganglia in PD to improve stride length (Morris et al. 1996; Azulay et al., 2006).

Enhancement of optic flow

PWP rely heavily on visual information for gait control to compensate for the reduced kinaesthetic feedback from the lower limbs (Azulay et al., 1999). Sensory input within cortical networks play an important role to guide locomotion and maintain correct posture (Raffi & Piras., 2019). This is done with the integration of three different sensory systems; somatosensory input originates from proprioceptive signals from muscles and joints, vestibular input arises from linear and rotational acceleration of the head relative to the body, and visual input or optic flow emerges from relative motion between an individual and their environment (Raffi & Piras., 2019).

Optic flow influences the sense of self motion, postural orientation, veering of gait and visuospatial cognition (Bardy et al., 1999; Putcha et al., 2014). Azulay et al. (1999) suggested that the positive effect of visual cues on gait in PWP is a combination of attentional strategy and enhancement of optic flow activating the visual-motor pathway. In order to understand the influence of optic flow, the authors conducted an experiment on PD gait with and without visual cues (stripes on the floor) under two visual conditions; normal lighting and stroboscopic/flashing light to supress the optic flow completely. Thirty-two participants were included in the study with an equal number of PWP and healthy age matched controls. The baseline data showed that PWP walked with reduced step length and slow velocity gait, with similar findings as that earlier reported by Morris et al. (1994 a,b) in their experimental series. Under normal lighting PWP walked significantly faster with visual cues (0.82 ± 0.19 m/sec) than without (0.76 ± 0.2 m/sec), with an average of 9.4% m/sec improvement in gait speed primarily due to increased stride length. The effect of stroboscopic lighting was significantly different in PWP but not in controls, with a marked reduction in velocity and stride length both without visual cues (velocity from 0.76 ± 0.2 m/sec to 0.70 ± 0.2 m/sec, stride length from 925 ± 175 mm to 857 ± 166 mm) and with cues (velocity from 0.82 ± 0.2 m/s to 0.74 ± 0.2 m/s, stride length from 968 ± 176 mm to 893 ± 193 mm) in PWP. The fact that PWP reverted back to their baseline measurements even with visual cues in the presence of stroboscopic lighting suggests that PWP, and not healthy controls, are heavily reliant on optic flow for locomotion.

Both mechanisms have limitations and can minimise the efficacy of visual cues. Dual tasking may lead to dividing attention from maintaining gait secondary to cognitive over load. Disruption in optic flow may occur if vision is impaired or when walking through a narrow or confined space such as a doorway or threshold steps, where there is insufficient light reflection.

Mechanism of action for auditory cueing

Timed responses in daily activities are generated at the subconscious level (Gibbon et al., 1984; Ferrandez et al., 2003). This is done either through the internal clock or memory mechanism involving the basal ganglia and complex neural activity as explained in the earlier section of this review. This phenomenon also highlights one of the unique capabilities of humans to appreciate musical rhythms (Cannon & Patel., 2020). Hearing music often results in spontaneous rhythmic movements such as foot tapping or nodding along that feels automatic and is evidenced in young children and adults of all ages without any musical training (Grahn., 2009). Rhythmic perception is a complex neural mechanism that involves perceiving one of the primary characteristic components of the musical rhythm, called 'beat' which is broadly classified as a perceived pulse that marks equally spaced points in time (Grahn., 2009). A musical piece in theory is only composed of sounds so listening to music should only activate the auditory system, but this is not always the case. Listening to musical rhythms often results in spontaneous rhythmic movements, suggesting a strong correlation between motor area recruitment with beat perception (Cannon & Patel., 2020).

Neuroscientific studies have established the engagement of cortical and subcortical motor regions of the brain during movement synchronisation with auditory rhythms, primarily including the premotor cortex (PMC), SMA, basal ganglia and cerebellum (Rao et al., 1997; Jancke et al., 2000; Lewis et al., 2004; Chen et al., 2006, 2008; Grahn & Rowe., 2009; Kung et al., 2013). The functional magnetic resonance (fMRI) study conducted by Chen et al. (2008) on the effects of beat perception in human subjects provided more insight into motor area recruitment and revealed the engagement of premotor regions in auditory-motor rhythm processing. The study also affirmed that motor regions such as the PMC, SMA and cerebellum continue to be activated when one only listens to musical rhythms, concluding that beat perception engages the motor system even in the absence of movement.

The regeneration of beat is hypothesised to use the same direct pathway that generates voluntary movement in human body namely the cortico-basal ganglia-thalamic loop (Calabresi et al., 2014). Listening to a beat-based rhythm leads to searching for temporal regularity or beat finding in the cortex (Grahn & Rowe., 2013). Once a beat is found this temporal pattern of neural activity gets stored in the cortex for regeneration (Cannon & Patel., 2020). When people listen to music the cortico-basal ganglia- thalamic loop is activated leading to regeneration of the beat via stored memory (Cannon & Patel., 2020). Grahn. (2009) established a direct link between basal ganglia activity and beat processing by conducting tests on PWP. Participants were asked to determine whether two auditory rhythmic stimuli were the same or different in the discrimination task to establish ability to discriminate changes in rhythm between healthy controls and PWP. Results showed PWP have reduced awareness to accurately engage with beat timing. Although the capacity for beat processing in PWP is not completely lost and when a beat is perceived with more rhythmic characteristics, such as using a metronome or through music (volume, pitch), this impairment is reduced.

Auditory cues increase gait variability in healthy controls by interfering with the normal gait rhythm generated by the basal ganglia (Baker et al., 2008). Rhythmic auditory cues are more effective during later stages of PD by acting as an external pacemaker. They help to reduce cognitive load when walking to match footsteps to an external metronome beat, improving stride length and walking speed in PD (Rochester et al., 2011; Lirani-Silva et al., 2019). Cue frequency is also a significant determinant of auditory cue efficacy. Studies report increased gait variability in PD with an auditory cue delivered at 20% below preferred cadence (Ebersbach et al., 1999). The optimal benefit of auditory cue is noted at 10% below and above normal cadence depending on PD stage and cognition (Lohnes & Earhart., 2011)

Efficacy of visual and auditory cues

Visual and auditory cueing remain widely implemented cueing therapies in the community. Nieuwboer et al. (2007) investigated the effectiveness of multi-modality cue training in the community. Although old, this remains highest level of evidence for the efficacy of external cues. In a single blinded randomised crossover trial 153 PWP with Hoehn and Yahr stage two to four (scale used to quantify stages of PD, refer to Appendix C) received a three week home cueing programme using three cueing modalities. The participants received auditory cue (a beep delivered through an earpiece); visual cue (light flashes delivered through a light-emitting diode attached to a pair of glasses); and somatosensory cue (pulsed vibrations delivered by a miniature cylinder worn under a wristband). The participants trialled all cueing modalities in the first week, but trained with their preferred modality. Majority of the participants preferred auditory cues (67%) followed by somatosensory cues (33%) as a well-tolerated and discrete alternative. Participants underwent cued gait training at home during gait initiation and termination, heel strike and push-off, and sideways and backwards stepping. They also received cued practice in familiar environmental situations such as walking while dual tasking and walking over various surfaces and long distances. The study found short lasting (up to 12 weeks) statistically significant improvements in posture and gait scores, improved by 4.2% after intervention ($p = 0.005$). Secondary outcome measures such as gait speed and step length improved by 5 cm/sec ($p = 0.005$) and 4 cm ($p < 0.001$), respectively in all the participants. Although these beneficial effects reduced considerably following cue removal over six to twelve months.

Auditory cueing in particular has emerged as more efficient and cost effective cue modality. A meta-analysis of 28 studies conducted by Spaulding et al. (2013) compared the effect of visual and auditory cueing on gait in PWP concluded an increased efficacy of auditory cue in treating temporal parameters of gait disorder in PD. The meta-analysis included studies until September 2011 that evaluated the effect of auditory and/or visual cueing of gait in people with PD; objectively reported and evaluated the impact of cue practice on gait parameters and were published in English. Unpublished papers alongside studies evaluating cognitive cueing or effect of mobility aids were excluded from the meta-analysis. The data was reported as Hedge g and confidence interval to improve the validity of the study results. Auditory cueing demonstrated significant improvement of cadence (Hedge g=.556; 95% confidence interval [CI], .291–.893), stride length (Hedge g=.497; 95% CI, .289–.696), and velocity (Hedge g=.544; 95% CI, .294–.795). In contrast, visual cueing significantly improved stride length only (Hedge g=.554; 95% CI, .072–1.036).

A more recent meta-analysis by Ghai et al., 2018 analysing the effects of auditory cues on young healthy controls and elderly participants provided more insight into the mechanism of auditory cues. They suggested the beneficial effects of auditory cues are linked to perception of stimuli with shorter reaction times (20–50 ms) as compared to its visual or tactile counterparts (Ghai et al., 2018). Additionally these cues simultaneously activate supplementary motor areas and sub cortical structures due to rich connectivity within the cortical neural network (Ghai et al., 2018). The meta-analysis included 34 studies until May 2017, involving 854 (499 young/ 355 elderly) participants. The study reported significant positive small to large standardised effects on spatiotemporal gait parameters with rhythmic auditory cues in 88% of included studies, especially stride length (Hedge g= 0.61) and gait velocity (Hedge g= 0.85).

Limitations of visual and auditory cues

Gait rehabilitation in PWP involving visual and auditory cueing strategies in a clinical setting involves delivery of these cues as a set of lines or markers on the floor or the use of metronome beats respectively. PWP alter their gait parameters by either stepping on to visual markers or walking to a predetermined beat in order to achieve improved gait quality. Visual cues may also be provided by projection lights embedded on a walking stick or the walking frame, although these devices are expensive (device cost listed in Appendix A). There is an upcoming market for laser guided shoes where the device can be installed in standard footwear for high functioning PWP who are not reliant on mobility aids. This product is not available in New Zealand and in UK retails at a high cost (Appendix A). Visual cues require PWP to physically look at the cues which might affect their postural stability and balance while completing certain functional activities (for instance backward stepping to sit in the chair) increasing the risk of falls. Auditory cues, although considerably cheaper to purchase or downloaded free of cost on the mobile app (Appendix A), can increase cognitive load when trying to synchronise stride length to the metronome beat. Step synchronisation to auditory cues is not feasible in many every day activities; one such example is attending to personal care needs in compact bathroom space. Auditory cues also have the potential to get masked in loud environments such as in busy supermarket or if provided through ear phones reduce the awareness of surrounding conditions for example while walking outdoors or crossing the road. These interventions cannot be implemented in PWP with visual and hearing impairment. While external cues are beneficial in improving gait quality in PD they also pose logistical challenge for ambulation in a real life setting as outlined above.

Internal cues - an alternative approach

Continuous external cues use conscious attentional strategies which are effective for gait improvement due to increased attention and focus on stride length but immediately lose their efficacy due to cue dependency and habituation (Nieuwboer et al., 2007; Spildooren et al., 2012). The decline in beneficial effects of cueing intervention following its removal critically underscores the need for permanent cueing devices. ‘On demand’ external cues improve cue efficacy (Nieuwboer., 2015; Velik et al., 2015; Ginis et al., 2017) although comes with an additional device cost, and lack of device portability in functional activities as explained in the earlier section. ‘On demand’ internal cueing is an alternative cost effective patient controlled cueing approach due to the ability to internally generate and control the application of the cues. This strategy enables incorporation of cues only when deviation from the normal gait occurs via attentional resources. Internal attentional cues to focus on improving step length (such as walking while saying repeatedly big step, big step) are found to have similar beneficial effects on gait parameters as visual cues (Morris et al., 1996).

Another promising approach is internal-generation of musical cues in the form of singing aloud or mental singing in one's own head to improve gait stability in PD. As already explained earlier, one of the functions of the basal ganglia is to maintain 'internal rhythm' or a steady gait in healthy individuals by controlling internal regulation of movement amplitude and timing (Nombela et al., 2013). This internal rhythm however, is affected in PWP leading to a more variable and less efficient gait predisposing the individuals to a high risk of falls (Harrison et al., 2017). Musical cueing works in similar manner to auditory cues through bypassing the defective internal rhythm of basal ganglia and activate multiple cortical structures (Harrison et al., 2017, 2018). The beneficial effects of musical cues reach far beyond gait improvement as they are also identified as mood elevators by increasing endorphin release in the normal healthy population (Dunbar et al., 2012). The efficacy of musical cues on motor and emotional response was tested in PD by Pacchetti et al. (2000). The randomised controlled trial included thirty two PWP who received weekly sessions of music training and physiotherapy. The study findings reported an improvement in motor performance bradykinesia items ($p < .0001$), improved happiness measure ($p < .0001$), improved quality of life ($p < .0001$) and improved rigidity ($p < .0001$). The above evidence suggests internal musical cueing can help elevate mood through the endorphine system and likely has a positive effect on motivation, thereby enhancing quality of life.

The exact mechanism is still unclear though singing internally is believed to work in a similar manner as musical or auditory cueing as explained above. The above internal cueing strategies show encouraging results in the limited number of studies conducted so far. Improving gait performance via cueing strategies mandates a change or modification in behaviour which require new learning strategies. Literature supports that motor learning is challenging for PWP, especially learning automatic responses, and PWP need practise to learn new strategies and how to adapt to new and changing environments (Nieuwboer et al., 2009). Neurophysiological studies demonstrate increased motor activity in the premotor cortex and improved movement performance under different cued conditions (Sarma et al., 2012) suggesting PWP may retain cognitive capacity to learn new strategies, although at a slower rate and with more practise.

There is limited clinical evidence to date investigating the effectiveness of internal cues on gait parameters in PWP. In light of this, the aims of this structured review are:

1) Review current literature and outline the underlying mechanisms of internal cue strategy to improve gait performance in PD

2) Examine efficacy of internal cues on gait features

Method

Methodology

A structured review has been selected to answer the proposed research question because it allows a structured approach to appraising the literature but is not confined to selecting only very limited research designs such as randomised controlled trials (RCTs). There are two aspects to this review which overlap and emerge from the same body of literature.

The first aspect focuses on the question ‘what are the mechanisms underlying internal cueing in people with PD’. This aspect of the review takes a broader approach and is based on qualitative and narrative components of the included studies and opinions expressed in the publications, and as such do not require a quality check. The above synthesis analyses the current literature to gain an insight into the proposed underlying mechanisms of internal cueing strategy and addresses the first aim of this structured review. The underlying mechanisms of internal cues are further elaborated in the discussion section of the review.

The second aspect focuses on the question ‘is internal cueing effective in improving gait in people with PD’. This aspect required quantitative analysis of experimental and quasi experimental research designs. Differences in outcomes are reported as well as associations between gait and features that may impact on outcome are analysed. The quality of each study is considered with respect to the McMaster Critical Tool for quantitative studies which helped guide the synthesis. Because this is a structured rather than a systematic review, a score is not given to each study. Studies were included if they used experimental designs (for example RCTs), quasi experimental research designs (such as pre-post, correlational studies) or cohort studies.

Search strategy

Five databases were used for the literature search: Medline via EBSCO, Ovid and Web of Science, Scopus and CINAHL. The chosen databases provided published peer reviewed academic journal articles in the scope of physiotherapy practise across the world. Each search used the key terms “Parkinson’s disease”, “Cues” and “Gait”. For each key term, a list of synonyms was entered into the search as described below (Table 1). The use of broader search terms ensured relevant article retrieval from the databases, for example, MESH heading “Gait” automatically included studies with gait parameters such as posture and balance and “Cues” included rehabilitation and physical therapy. The search was limited to published papers on humans until the present time (November 2020), written in the English language and restricted to full journal articles. References of the articles were also reviewed for further potential reading.

Table 1

Key terms search table used for structured search

	Medline via EBSCO	Medline via Ovid (Mutilfield search)	Scopus (Advanced search)	CINAHL	Medline via Web of science
Parkinson disease	MESH headings: Parkinson’s disease or Parkinsons or parkinson's	Keyword search: Parkinson$	ALL“Parkinson disease”	MESH headings: Parkinson's disease*	TS (Parkinson*)
Gait	MESH headings: AND gait or ambulation or mobility or locomotion	Keyword search: gait or ambulation	Keyword search: AND gait OR mobility OR ambulation	MESH headings: AND gait or locomotion or walk or ambulation	Keyword search: AND Gait
Cues	MESH headings: AND cues or cue or cueing	Keyword search: cue or cueing	Keyword search: AND internal cues OR intrinsic cueing	MESH headings: AND cueing or cues or cue	Keyword search: AND Cues

Table includes five databases used, individual terms for each cohort and MESH headings where applicable. The term “internal cueing” is interchangeably used with “cognitive cueing” or “self-generated cueing”.

Inclusion and exclusion criteria

The inclusion and exclusion criteria for article selection are listed below. Article selection was an extensive process, more detail is provided in the search yield section. Articles were included upon initial screening of the title and abstract of the published paper. When in doubt, the full text was read to determine whether the cueing strategy implemented inferred external cueing through a device or internal cueing through a self-generated technique. More information for title screening and exclusion reasons post full text screening are listed in the results section and presented in the PRISMA flow chart in Figure 3.

Inclusion criteria

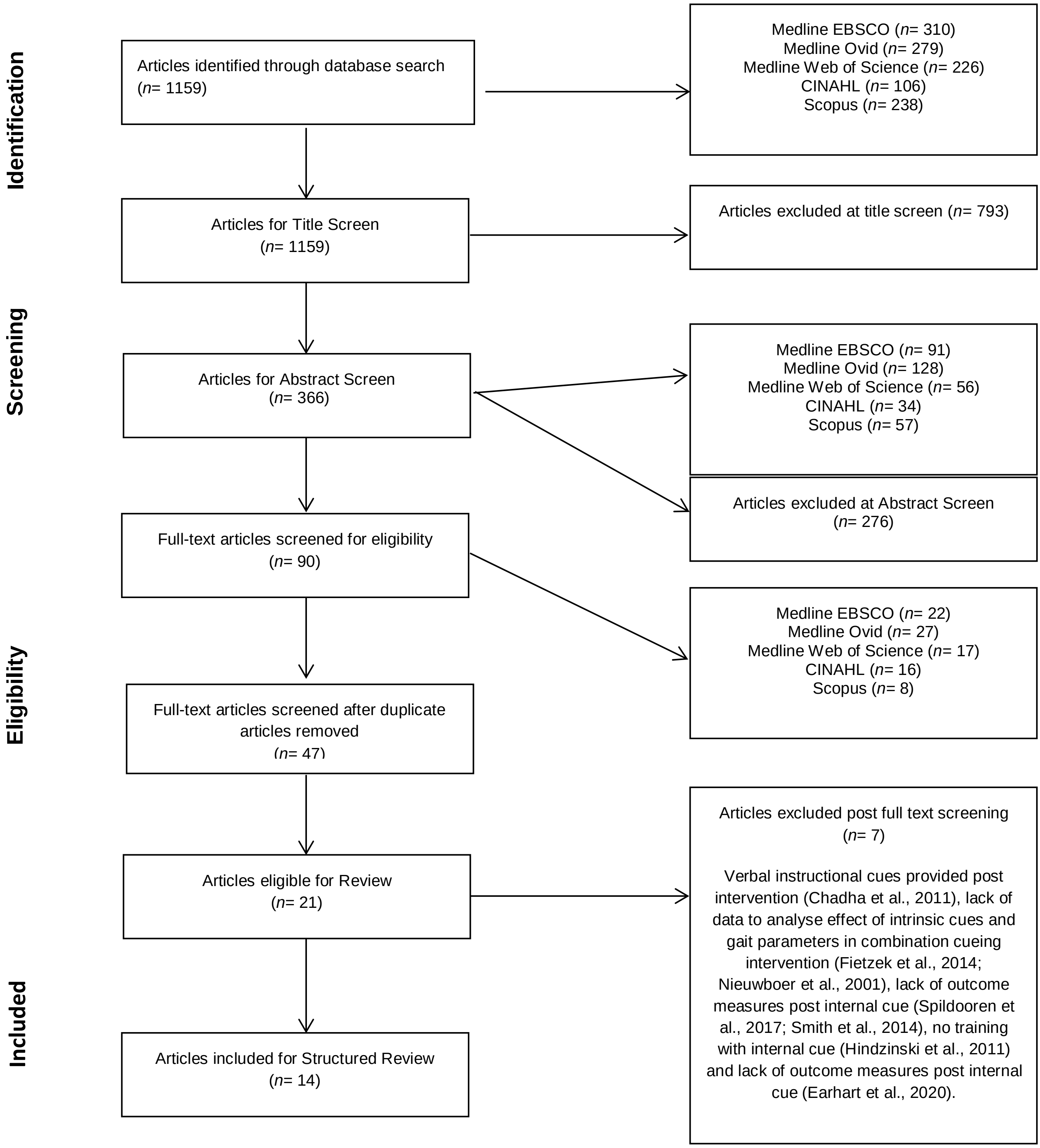
The studies included in this review had to meet the following inclusion criteria:

1. Population: Study participants with diagnosis of PD
2. Study design: Experimental, quasi-experimental or cohort studies

3. Intervention: Application of gait training with internal cues as a single intervention modality. Studies should have completed gait assessment pre and post internal cueing interventions under single task conditions on stable medication regime in the *on* phase. For the purpose of this structured review, internal cueing is defined as *any self-employed technique that does not require the use of any external device or instructions provided by an external person (caregiver or examiner) post training session.*
4. Comparison: Studies were also included if internal cueing strategies were given in conjunction with other rehabilitation interventions.
5. Outcomes: Any validated measures of gait parameters or aspects associated to gait such as spatiotemporal parameters (speed, cadence, step length) or gait variability measures.

Exclusion criteria

1. Studies recruited PD participants with a diagnosis of dementia or marked cognitive impairment. Due to the nature of the intervention, PD participants were required to have adequate attentional and cognitive reserve to follow instructions and participate in 'internal cueing' gait practise.
2. Studies recruited PD participants with psychiatric disorder/depression due to possibility of reduced compliance, attendance and engagement in the cued gait training programme.
3. Studies implemented self-supervised home exercise programme without providing any gait training with internal cueing strategies.



Information for title screening and exclusion reasons post full text screening are listed in the Results Section.

Figure.3. PRISMA diagram presenting search yield for structured review

Data extraction

Full text of included papers was screened for data extraction. At this point the papers not meeting the complete inclusion criteria were excluded from data extraction. Extracted data from the included studies was transferred into a table. Table 2 describes the key variables including study design, participant characteristics, intervention protocol, gait parameters, main study findings and results.

Data analysis/ synthesis

Another table was created for data synthesis. Table 3 is colour coded and summarises the effect of internal cues on different gait domains in PD. Where applicable, the studies that outlined their individualised domains were matched to the most relevant domain adopted in this review. Through this table, analysis of key variables was conducted that allowed to explore the data within gait domains and between different interventions. This structured synthesis highlighted the robustness of internal cueing interventions and suggested a selective response of cueing strategies on gait outcomes. The diagrammatic representation of internal cueing interventions and their selective response on gait domains in PD is illustrated in Figure 4.

Table 2

Main characteristics of the studies assessing effect of internal cueing on gait domains in Parkinson's disease.

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
1. Morris et al. (1996)	HC: (n=16); 9M & 7F, Age: 72.7 (63-82), STMS>21 PD: (n=16); 9M & 7F, Age: 72.7 (64-81), Webster rating scale: 14.8 (9-24), STMS>21 *Med: On*	Stride length *(m),* Gait velocity *(m/min),* Cadence *(steps/min),* DS *(% gait cycle)*	*Tool:* Stride analyzer (12mts)	Repeated measures cross sectional study (Study 1)	20 min uncued baseline trials in HC and PD followed by 20 min repeated 10 mt cued trials in intervention phase followed by repeated 10 mt trials in the retention phase in PD for 2 cueing conditions: *Visual cue (Ext cue)*: step over the floor markers *Attentional strategy (Int cue)*: focus attention on walking with target stride length in mind	*Study 1 findings* *Pre to post test:* ↑ gait velocity and ↓ DLS for both visual (Ext cue) and attentional cueing strategy (Int cue). *Delayed retention test (motor learning):* Improved gait velocity correlated with both cueing conditions after 2 hours. No significant within group variability found in PD for visual and attention cueing	Velocity (*p*= 0.001), DLS (*p*= 0.01)
2. Lehman et al. (2005)	PD: (n=11); 8M & 3F, Age: 75.81 (67-83), Disease duration: 6.51 (1-13), MMSE: 28 (27-30), H&Y scale 2, 2.5 *Med: On*	Gait velocity *(cm/sec),* Step length *(cm),* Cadence *(steps/min),*	*Tool:* GaitRite systems walkway (30 foot)	Randomised controlled trial (Part 2)	Uncued baseline trial followed by 10 day x 3 sessions x 1800 feet per day gait training programme for 2 groups: *Treatment group using verbal instruction cue training:* take long steps (self generated post training) *Control group:* similar intervention without cue, walk at normal, comfortable, everday pace	*Part 2 findings:* ↑gait velocity and step length and ↓ cadence associated with treatment group (Int cue) *Delayed retention test:* Improved gait parameters correlated with treatment group after 4 weeks. No significant improvement in control group	Velocity (*p*< 0.05), Stride length (*p*< 0.05)

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
3. Baker et al. (2007)	HC: (n=12); 5M & 7F, Age: 71.50± 2.58, MMSE: 28.6± 1.8 PD: (n=15); 6M & 9F, Age: 68.83± 3.30, UPDRS motor subscale: 23.4± 9.2, Disease duration: 6.5± 3.2, MMSE: 27.9±2.17, H&Y scale 2-3 *Med: On*	Walking speed *(cm/sec),* Step amplitude *(cms),* Step frequency *(steps/min),*	*Tool:* GaitRite systems walkway (8 mts)	Repeated measures experimental study	3 uncued baseline trials for ST and DT conditions followed by cued training trials for 3 different cue types followed by final uncued trial for both ST/DT conditions: *Auditory cue (Ext cue)*: walk to metronome beat *Attentional strategy (Int cue)*: focus attention on taking big steps *Auditory cue+Attentional cue (combination):* Walk to beat while taking big steps	*ST findings:* ↑walking speed and step amplitude associated with both attentional cues (Int cue) and combination of auditory/attentional cues (Int/Ext cue). Improved gait parameters not correlated to auditory cue alone	Velocity (*p*< 0.001), Stride length (*p*< 0.001)
4. Baker et al. (2008)	HC: (n=12); 5M & 7F, Age: 71.50± 2.58, MMSE: 28.6± 1.8 PD: (n=14); 5M & 9F, Age: 69.3± 3.36, UPDRS motor subscale: 22.86± 9.2, Disease duration: 6.64± 3.2, MMSE: 27.71±2.16, H&Y scale 2-3 *Med: On*	Walking speed *(cm/sec),* Step time *(sec),* DS *(sec),*	*Tool:* GaitRite systems walkway (8 mts)	Repeated measures experimental study	3 uncued baseline trials for ST and DT conditions followed by cued training trials for 3 different cue types followed by final uncued trial for both ST/DT conditions: *Auditory cue at 90% walking cadence (Ext cue)*: walk to metronome beat *Attentional strategy (Int cue)*: focus attention on taking big steps *Auditory cue+Attentional cue (combination):* Walk to beat while taking big steps	*"ST" findings:* ↓gait variability (step time CV and DLS CV) was associated with both attentional cues (Int cue) and combination of auditory/attentional cues (Int/Ext cue). ↑gait stability with cueing only correlated in PD. No improvement with auditory cue alone	DLS CV (*p*= 0.001)

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
5. Satoh et al. (2008)	PD: (n=8); 5M & 3F, Age: 64± 9, Disease duration: 4.25, Nil to Mild CI, H&Y scale 2-3 *Med: On*	Walk time *(sec),* steps *(number)*	*Tool:* 5 mt walkway	Pre-test post-test	Single session gait training few times with 7 progressive tasks as described below with a final goal of walking on a straight path and turning while mentally singing *Progressive tasks:* Listen to song, clap hands while listening to the song, sing song, clap hands while singing song, sing song while walking and sitting in chair, walk with singing song, *Mental singing (Int cue)*: walk while singing song in mind	*Pre to post test:* ↓ walk time and number of steps walking on straight path and during turns associated with mental singing (Int cue)	Walk time (*p*= 0.008), Number of steps (*p*= 0.008)
6. Lowry et al. (2010)	PD: (n=7); 6M & 1F, Age: 70.28, Disease duration: 6.18, MMSE ≥ 24, H&Y scale 2-3 *Med: On*	Walking speed *(mts/sec),* Stride length *(mts),* Cadence *(steps/min),* Stride time CV, AP HR, vertical HR, ML HR	*Tool:* Gait walkway with triaxial accelero meter (18 mts)	Pre-test post-test	Uncued baseline trials for preferred and fast pace walking followed by cued training trials followed by final uncued trial for 3 different cueing conditions: *Visual cue at 20% longer step length (Ext cue)*: walk on the marked strips *Cognitive cueing (Int cue)*: think big steps and focus on taking longer steps *Verbal cues (Ext cue):* Experimenter prompting big step during swing phase of every 3rd step	*Pre to post test:* ↑walking speed and stride length associated with verbal (Ext cue) and cognitive cue (Int cue), ↑gait stability in AP motion also associated with verbal and cognitive cueing. No improvement in gait stability or spatial/ temporal parameters with visual cue	Velocity (*p*= 0.043), Stride length (*p*= 0.018), AP HR (*p*= 0.018)

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
7. Werner et al. (2010)	PD: (n=12); 10M & 2F, Age: 60-75, Disease duration: 2 or 3 H&Y scale, Nil/Mild CI *Med: On*	Stride length *(m),* Gait velocity *(m/sec),* Cadence *(steps/min),* Stride length CV, Gait velocity CV	*Tool:* Motus 2000 Real time system (7.5mts)	Pre-test post-test	Uncued baseline trial followed by 4 training sessions each lasting 90 minutes, twice/week, 15 trials/session for 2 cueing conditions: *VI (Int cue):* take a big step, self generated post training *VI + Video Feedback (VI+FB):* same protocol as above with additional video feedback	*Pre to post test:* ↑ Stride length and gait velocity asscoiated with both VI (Int cue) and VI+FB (combination cue). No change in cadence. *Delayed retention test (motor learning):* Improvement in stride length and gait velocity maintained 3, 6 or 12 months post training in both groups. No significance within subject variability found	Velocity (*p*< 0.01), Stride length (*p*< 0.01)
8. Rochester et al. (2011)	PD: (n=50); 31M & 19F, Age: 69.22± 6.6, Disease duration: 8.69± 5.19, MMSE: 28.22± 1.57, H&Y scale 2, 3 or 4 *Med: On*	Gait velocity *(m/min),* Stride length *(m),* Step frequency *(steps/min),* Stride time CV, DS CV	*Tool:* Stride analyzer (6mts)	Repeated measures experimental study	3 uncued baseline trials followed by 2 trials at home in "on" and "off" phase in PD for 2 cueing conditions: *Auditory cue (Ext cue)*: take large step to the beat *Attentional strategy (Int cue)*: focus attention to increase step length	*On phase findings:* ↑ gait velocity and stride length corelated with both auditory (Ext cue) and attentional cues (Int cue), ↑ gait stability only associated with auditory cue (Ext cue)	Velocity (*p*< 0.001), Stride length (*p*< 0.001), Step frequency(*p*< 0.001)
9. Lohnes & Earhart .(2011)	Young HC: (n=11); 4M & 7F, Age: 24.09± 0.83 Age matched HC: (n=11); 4M & 7F, Age: 70.82± 10.44 PD: (n=11); 4M & 7F, Age: 70.27± 6.80,	Stride length *(cm),* Gait velocity *(cm/sec),* Cadence *(steps/sec)*	*Tool:* GaitRite CIR systems walkway (5mts)	Pre-test post-test	3 uncued baseline trials for ST and DT conditions followed by cued training trials for 3 different cue types followed by final uncued trial for both ST/DT conditions: *Auditory cue (Ext cue)*: walk to metronome beat at 90% and 110% of walking cadence	*ST findings:* ↑gait velocity and stride length associated with both attentional cues (Int cue) and combination of auditory/attentional cues (Int/Ext cue). No additional benefit of combination cue observed. Improved gait parameters not correlated to an auditory cue alone	Did not achieve statistical significance

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
	UPDRS motor subscale: 21.55± 6.71, Disease duration: 9.09± 5.39, Nil CI *Med: On*				*Attentional strategy (Int cue)*: think about taking large strides *Auditory cue+Attentional cue*		
10. Harrison et al. (2017)	PD: (n=23); 13M & 10F, Age: 69.5± 7.6, UPDRS motor subscale: 30.5± 11.8, Disease duration: 3.8± 4.2, MMSE≥ 24, H&Y scale 2-3 *Med: On*	Stride length *(cm),* Gait velocity *(cm/sec),* Cadence *(steps/min),* Step time CV, SST CV, Step length CV	*Tool:* GaitRite CIR systems walkway (5mts)	Pre-test post-test	Single session initial baseline trial followed by 3 x training trials for 5 different cue conditions: *Uncued:* walk at preferred cadence *Music only (Ext cue)*: walk to beat of song *Sing only (Int cue)*: sing aloud while walking *Music+Sing (combination)*: walk to beat of song while singing aloud *Verbal dual task:* Excluded	*Pre to post test:* ↑gait stability with improved step time and single support time associated with singing (Int cue). ↓ gait stability correlated with combination of music and sing (Ext cue).	Step time CV (*p*= 0.008), SST CV(*p*= 0.031)
11. Harrison et al. (2018)	Young HC: (n=30); 15M & 15F, Age: 25.8± 2.8, MMSE: 30 Old HC: (n=30); 15M & 15F, Age: 64.9± 7.2, MMSE: 30 PD: (n=30); 15M & 15F, Age: 65.8± 6.5, UPDRS	Stride length *(m),* Gait velocity *(m/sec),* Cadence *(steps/min),* Stride length CV, Stride time CV, SST CV	*Tool:* GaitRite CIR systems walkway (5mts)	Repeated measures cross sectional study	3 uncued baseline trials followed by 3 cued trials in forward and back ward walk in Young HC, Old HC and PD for 2 cueing conditions: *Music (Ext cue)*: walk to beat of song *Sing (Int cue)*: sing aloud while walking (self generated vocal cues)	Singing (Int cue) associated with ↑gait velocity, cadence and stride length in back ward walk and ↑gait stability in both forward and back ward walk. ↓ gait stability correlated with music (Ext cue). % gait improvement in PD>HC	Cadence (*p*< 0.001), Forward walk; Stride length CV (*p*< 0.001), Stride time CV (*p*< 0.001), SST CV (*p*< 0.002), Backward walk; Stride length CV (*p*< 0.021), Stride time CV

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
	motor subscale: 24.9, Disease duration: 5.77± 3.79, MMSE: 29 *Med: On*						(*p*< 0.018), SST CV (*p*< 0.011)
12. Harrison et al. (2019)	HC: (n=30); 15M & 15F, Age: 64.9± 7.2, MMSE: 30 PD: (n=30); 15M & 15F, Age: 65.8± 6.5, UPDRS motor subscale: 24.9, Disease duration: 5.77± 3.79, MMSE: 29 *Med: On*	Stride length *(m),* Gait velocity *(m/sec),* Cadence *(steps/min),* Stride length CV, Stride time CV, SST CV	*Tool:* GaitRite CIR systems walkway (5mts)	Repeated measures cross sectional study	3 uncued baseline trials followed by 3 blocks of cued trials at 90%, 100%, 110% of walking cadence in HC and PD for 3 cueing conditions: *Music (Ext cue)*: walk to beat of song *Sing (Int cue)*: sing aloud while walking *Mental (Int cue)*: sing in own head while stepping	↑ gait stability at 100% and 110% cue rate was correlated with mental singing (Int cue). ↓ gait stability was associated with music (Ext cue) at slower and faster cue rate in both HC and PD.	Stride time CV at 100% cue rate (*p*< 0.001)
13. Horin et al. (2020 a)	HC: (n=24); 5M & 19F, Age: 66.0± 7.3, MMSE> 24 (25-30) PD: (n=33); 18M & 15F, Age: 67.7± 8.4, UPDRS motor subscale:	Stride length *(cm),* Gait velocity *(m/sec),* Stride length CV, SST CV	*Tool:* GaitRite CIR systems walkway (5mts)	Repeated measures experimental study (only gait peformance measures included)	3 baseline trials followed by gait trials in HC and PD for 3 conditions: *Uncued:* walk at preferred cadence *Music (Ext cue)*: walk to beat of song at 100% of walking cadence *Mental Sing (Int cue)*: sing in own head while stepping	*Gait performance findings:* ↑ stride length gait variability correlated to music (Ext cue) in both HC and PD. ↓ gait variability (SST) associated with self generated mental singing (Int cue)	Did not reach statistical significance

Study	Participant characteristics	Gait variables measures	Gait analysis tool	Study design	Intervention	Main study findings	Statistical significance for Internal cueing (*p* value)
	24.6, Disease duration: 8.2± 5, MMSE> 24 (26-30) *Med: On*						
14. Horin et al. (2020 b)	HC: (n=24); 5M & 19F, Age: 66.0± 7.3, MMSE> 24 (25-30) PD: (n= 44); 23M & 21F, Age: 67.94, UPDRS motor subscale: 25.79, Disease duration: 7.34, H&Y scale 2, 3, MMSE> 24 *Med: On*	Stride length *(cm),* Gait velocity *(m/sec),* Cadence *(steps/min),* Step time CV, Stride length CV, SST CV	*Tool:* GaitRite CIR systems walkway (5mts)	Repeated measures cross sectional study	3 baseline uncued trials followed by gait trials in HC and PD for 2 cueing conditions: *Music (Ext cue)*: match footfalls to beat of song at 100% of walking cadence *Mental Sing (Int cue)*: match footfalls to beat of metal singing	↑ gait variability (step time CV) correlated to music, PD-FOG> PD+FOG & HC. Both music and mental cueing (Ext & Int cue) correlated with ↑gait stability in more variable gait. Self generated/mental cue not associated with ↑gait variability as compared to uncued gait	Did not reach statistical significance

Abbreviations as follows; PD, Parkinson's disease; H & Y scale, Hoehn & Yahr disease severity classification; CI, cognitive impairement; Med, medication; HC, healthy controls; UPDRS, unified Parkinson's disease rating scale; CV, coefficient of variation; VI, verbal instruction; Ext cue, external cue; Int cue, internal cue; STMS, short test of mental status dementia; DLS, double limb support duration; ST, single task; DT, dual task; AP, anterior-posterior; ML, mediolateral; HR, harmonic ratio; SST, single support time; PD-FOG, PD without freezing of gait; PD+FOG, PD with freezing of gait

Table 3

Colour correlation table to display effect of internal cueing on gait domains in Parkinson's disease.

Gait Domains	Internal cueing	Retention (Memory/Motor learning)
Pace		
Step velocity (m/sec)	●1 ●2 ●3 ●6 ●7 ●8 ●9 ●11 ●12 ●14 ●10 ●13	●1 ●2 ●7
Step length (m)	●1 ●2 ●3 ●6 ●7 ●8 ●9 ●11 ●12 ●14 ●10 ●13	●2 ●7
Rhythm		
Double limb support time (sec)	●1 ●8	
Step time (sec)	●5 ●8 ●14 ●6	
Cadence (steps/min)	●2 ●4 ●6 ●11 ●12 ●1 ●7 ●9 ●10 ●14	●2
Gait variability		
Step time variability (sec)	●8 ●4 ●6 ●10 ●11 ●12 ●14	
Single support time variability (sec)	●10 ●11 ●12 ●13 ●14	
Step length variability (m)	●10 ●11 ●12 ●13 ●14	
Double limb support time variability (sec)	●4 ●8	

Green indicates postive effect was found, red indicates no effect was found. (1) Morris et al. (1996); (2) Lehman et al. (2005); (3) Baker et al. (2007); (4) Baker et al. (2008); (5) Satoh et al. (2008); (6) Lowry et al. (2010); (7) Werner et al. (2010); (8) Rochester et al. (2011); (9) Lohnes & Earhart. (2011); (10) Harrison et al. (2017); (11) Harrison et al. (2018); (12) Harrison et al. (2019); (13) Horin et al. (2020 a); (14) Horin et al. (2020 b).

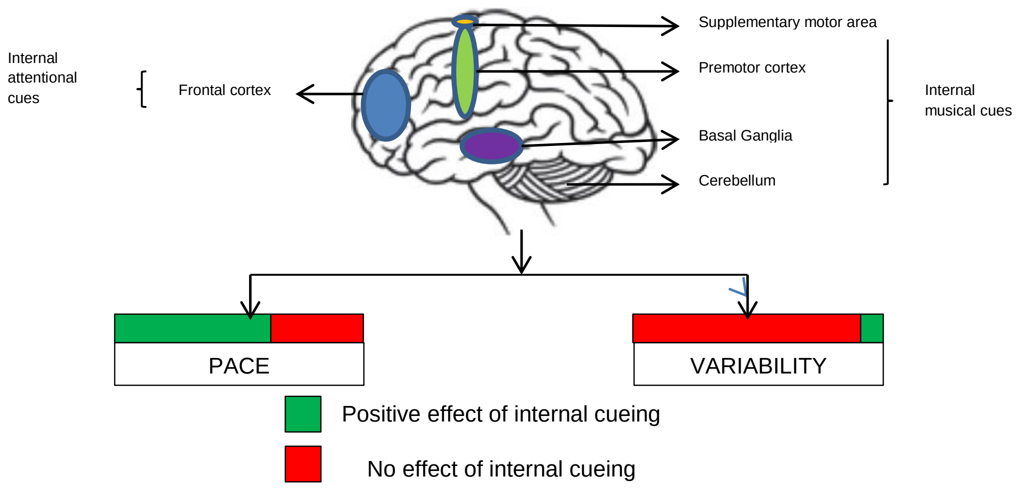


Figure.4. Diagrammatic representation of effect of internal cues on gait domains in PD

Results

Search yield

The search strategy generated a total of 1159 articles with limiters (‘full text’, ‘English’ and ‘humans’); modified searches were applied for two electronic databases, Medline Ovid and Scopus. Medline Ovid: the key words search for “Parkinson$” and “gait” and “cues” with the above limiters resulted in retrieval of only 35 articles, therefore the search was modified with less limiters (only English language) resulting in 279 papers; Scopus: the key words search for “Parkinson’s disease” AND “gait” AND “cues” resulted in a huge volume of articles (4886), therefore the search was modified to “Parkinson’s disease” AND “gait” AND “internal cues” resulting in the generation of 238 papers. The search yield is demonstrated in the PRISMA flow chart in Figure 3.

As mentioned in the previous section, the challenges encountered while searching and selecting articles included a limited number of studies on internal cueing, lack of uniformity defining external and internal cues and an interchangeable use of terminology for internal cueing (also known as cognitive, intrinsic, internally generated or self-generated cues). Verbal instructional strategies were included if the examiner did not provide verbal prompts post intervention. While the intent was to keep the search specific to internal cueing there was also potential risk of insufficient capture of evidence with such limited search. Overall, it was concluded that it was relevant to review an extensive list of articles for appropriate article selection, therefore broader search terms were used.

All articles were title screened to retrieve relevant articles resulting in 366 papers of interest. After an abstract screening, 90 papers were eligible for review, and then duplicates were removed. The remaining 47 articles were screened resulting in the final tally of 21 articles to be included for data extraction. Full text articles were reviewed during abstract screening when there was doubt whether the cueing strategy that was implemented inferred external cueing through a device or prompts provided by an external person, or whether internal cueing through self-administration strategies was used.

After data extraction, 7 papers were excluded from the review. The reasons for excluded studies are elaborated on the PRISMA flow chart in Figure 3.

Participant characteristics

A total of 504 subjects participated in the 14 studies included in this review (Table 2). This includes 304 PD participants, 159 age matched healthy controls in eight studies (Morris et al., 1996; Baker et al., 2007, 2008; Lohnes & Earhart., 2011; Harrison et al., 2018, 2019; Horin et al., 2020 a,b) and 41 young healthy controls in two studies (Lohnes & Earhart., 2011; Harrison et al., 2018). The average age ranged between 64-72 years for PD participants, 64-72 years for age matched healthy controls and 24-26 years for young healthy controls. All studies included PD participants with independent mobility, minimal cognitive impairment and mild to moderate functional disability, as indicated by the Hoehn and Yahr scale of two to three (Clarke et al., 2016), except

Rochester et al. (2011) where the PD sample group had more pronounced physical dysfunction with a Hoehn and Yahr scoring of two to four inclusive.

Study design

All studies used quasi experimental research design (such as pre-post-test, repeated measures correlational study, repeated measures experimental study) other than one which was a randomised controlled trial (Lehman et al., 2005) (Table 2). PD participants were recruited using convenience sampling from local movement centres or outpatient's neurology clinics, age matched healthy controls were recruited via local advertisement, emails or through social media. Young healthy controls in two studies (Lohnes & Earhart., 2011; Harrison et al., 2018) were recruited through a volunteer health database by Washington University school of Medicine. Half of the studies had a small number of PD participants < 15 (Lehman et al., 2005; Baker et al., 2007, 2008; Satoh et al., 2008; Lowry et al., 2010; Werner et al., 2010; Lohnes & Earhart., 2011) while the other half had more than 25 PD participants (Morris et al., 1996; Rochester et al., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b).

Intervention protocol

Internal cue gait training interventions comprised an attentional strategy in the form of *think about taking long steps or big steps* or a internal musical cue strategy by singing aloud or mental singing with simultaneously matching footsteps (Table 2). Eight studies employed an attentional strategy (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007, 2008; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Lohnes & Earhart., 2011) and six studies used a internal musical cue strategy (Satoh et al., 2008; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b).

Measurement of gait - methodological comparisons

Gait measurement tools in most studies used gait walkway systems (pressure sensitive walkway machine that provides information on spatial and temporal gait analysis)

(Lehman et al., 2005; Baker et al., 2007, 2008; Satoh et al., 2008; Lohnes & Earhart., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b). One study used a combination of a walkway system and trunk accelerometer (wearable sensors that provide information on mechanical motion through electrical signals) (Lowry et al., 2010); two studies used a stride analyser (machine that provides information on gait pattern) (Morris et al., 1996; Rochester et al., 2011); and one study used a real time optokinetic system (visual motion tracking system that determines location in real time) (Werner et al., 2010). All studies assessed spatial and temporal characteristics of gait and eight studies reviewed additional gait variability measures to report their findings (Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b).

Study findings - Effect of internal cueing on gait parameters

The effect of internal cueing was explored with respect to independent gait domains and statistical significance in PD (illustrated in Table 2). Individual gait characteristics such as spatial and temporal parameters were mapped onto their respective broader gait domains to explore the relationship (presented in Table 3). Spatial gait characteristics, such as step velocity and step length were loaded onto pace domain whereas temporal characteristics for example DLS, step time and cadence were reviewed under the rhythm domain (Morris et al., 2017). Variability measures, including step time variability, single support time (SST) variability, step length variability and DLS variability were reviewed under gait variability domain. Studies outlining their individualised domains were matched to the most relevant domain adopted in this review. The purpose of the review is to examine efficacy of internal cues on gait domains in PD therefore the results are only reported for PD participants. The diagrammatic representation highlighting the selective response of internal cues on gait domains in PD is illustrated in Figure 4.

Pace

Step velocity and step length were the most commonly assessed pace characteristics in 13 of 14 studies (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007, 2008; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Lohnes & Earhart., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b). One study did not outline pace domains, instead it reported rhythm characteristics such as step time in outcome measures (Satoh et al., 2008). The positive effect of internal cueing on step length and step velocity was evident in the majority of studies (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Lohnes & Earhart., 2011; Harrison et al., 2018, 2019; Horin et al., 2020 b).

Statistical significance was found in five studies for both step velocity and step length (Lehman et al., 2005; Baker et al., 2007; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011). The *p* value for these studies is reported in Table 2. An additional study reached statistical significance for the step velocity (Morris et al., 1996). Two studies did not report any change in step length and velocity post internal cueing (Harrison et al., 2017; Horin et al., 2020 a) and one did not include step length and step velocity as an outcome measure (Baker et al., 2008).

Rhythm

DLS and step time were assessed in five of 14 studies (Morris et al., 1996; Satoh et al., 2008; Lowry et al., 2010; Rochester et al., 2011; Horin et al., 2020 b). Out of two studies testing DLS, one found a beneficial effect of internal cueing reaching statistical significance (Morris et al., 1996) while the second reported no change post internal cueing (Rochester et al., 2011). The positive effect of internal cueing on step time was found in two studies; both reached statistical significance (Satoh et al., 2008; Harrison et al., 2017). One reported no change in step time with internal cueing (Lowry et al., 2010).

Five studies found a beneficial effect of internal cueing on cadence (Lehman et al., 2005; Baker et al., 2008; Lowry et al., 2010; Harrison et al., 2018, 2019) however only one reached statistical significance (Harrison et al., 2018). Five reported no change in cadence with internal cueing (Morris et al., 1996; Werner et al., 2010; Lohnes & Earhart., 2011; Harrison et al., 2017; Horin et al., 2020 b) and three did not outline cadence in outcome measures (Baker et al., 2007; Rochester et al., 2011; Horin et al., 2020 a).

Gait variability

Characteristics of gait variability included step time variability, SST variability, step length variability and DLS variability which were assessed in eight of 14 studies (Baker et al., 2008; Lowry et al., 2010; Rochester et al., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b). Seven studies tested step time variability (Baker et al., 2008; Lowry et al., 2010; Rochester et al., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 b); three reached statistical significance (Harrison et al., 2017; 2018; 2019). One study reported increased step time variability with internal cueing (Rochester et al., 2011).

Five studies assessed SST variability and step length variability (Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b); two reported statistical significance (Harrison et al., 2017; 2018). Only one study reached statistical significance for step length variability with internal cueing (Harrison et al., 2018). Two studies tested DLS variability (Baker et al., 2008; Rochester et al., 2011) and only one reported statistical significance post internal cueing (Baker et al., 2008).

Motor learning post-internal cueing

Three of 14 studies measured both the short and long term effect of internal cueing on motor learning in a delayed retention test (Morris et al., 1996; Lehman et al., 2005; Werner et al., 2010). Lehman et al. (2005) reported a maintained improvement in step length reaching statistical significance four weeks post internal cueing. Werner et al.

(2010) reported maintenance of step velocity and step length of statistical significance for a few months up to a year following internal cue training. Morris et al. (1996) reported improved step velocity but it did not reach statistical significance.

Discussion

To the author's knowledge this is the first structured review to summarise the efficacy of internal cues on gait performance in PD. The aims of this review were to firstly explore current literature concerning the underlying mechanisms of internal cues as a strategy to improve gait performance in PD and secondly to examine efficacy of internal cues. Overall, current evidence from this review suggests the use of internal cues increases conscious cortical activity through by passing the basal ganglia and activates supplementary cortical areas which help to substitute for the impaired internal rhythm of the basal ganglia. Results from this review indicate a positive effect for internal cueing on the pace domain of gait (primarily gait speed and step length).

The following section elaborates and summarises the key findings from this review.

Internal cueing interventions

The two main internal cueing interventions included internal attentional strategy in the form of *think about taking big steps* or internal musical cues and matching footsteps to the cues. Internal musical cues were delivered through mental singing or singing aloud and used a musical version of *Row row row your boat* due to its salient beat and participants' familiarity with the song (Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b). Satoh et al. (2008) used the Japanese nursery song *A rabbit and a turtle* due to its familiarity with the population group. Both approaches used a self-generated technique without the use of an external device post training session.

Eight of 14 studies in this review employed an attentional strategy (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007, 2008; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Lohnes & Earhart., 2011). Six studies used internal musical cues delivered through mental singing or singing aloud (Satoh et al., 2008; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b). Both internal cueing approaches have their pros and cons and it is likely one approach is better suited than another depending on an individual's personal preference and stage of PD, surrounding environment and provision of cues. The suitability of these two approaches is elaborated in the summary of key findings in the latter part of the discussion.

Internal attentional cues

Out of eight studies that employed attentional cueing, four (Baker et al., 2007, 2008; Rochester et al., 2011; Lohnes & Earhart., 2011) compared attentional cueing to rhythmic auditory cueing via a metronome or combination strategy of attentional and auditory cueing. Two (Baker et al., 2007; Lohnes & Earhart., 2011) tested pace domains and reported improved walking speed and stride length with attentional and combination strategies. Both interventions were equally effective in improving pace domains in PD while auditory cueing alone was found to be ineffective. One study (Baker et al., 2008) tested the effectiveness of attentional, auditory and a combination of attentional and auditory cueing on gait variability measures. It was found that attentional cueing alone was ineffective in improving gait fluctuations and variability in PD, and that maximum benefit is obtained by combining the two strategies (internal and external cueing) together. Attentional cueing is effective in selectively modifying pace domains (step length and step velocity) in PD but on its own is ineffective in improving gait stability (Baker et al., 2008). This is especially true in complex situations where attention is divided, for example when multi-tasking or walking in a busy shopping mall. An external auditory cue in this situation may act to produce a more consistent gait and improve gait stability, whilst reducing attentional load. Baker et al. (2007) and Lohnes & Earhart. (2011) indicated external auditory cues increase gait variability in healthy adults due to interference with the intact basal ganglia and internal rhythm as a result of increased attentional demand. Taken together, this body of research suggests that external auditory cues are beneficial in advanced stages of PD to compensate for the impaired internal beat timing of the basal ganglia.

The fourth study by Rochester et al. (2011) examined the effect of medication on gait and compared attentional cueing to external auditory cueing in PD participants in the *on* and *off* medication. Stride time gait variability increased significantly with internal cues in the *on* phase, whereas external cues reduced stride time variability and were more effective than medication. The authors proposed the selective effect of medication on gait is unable to modify gait variability fluctuations and attributed the effectiveness of external cueing in improving gait variability to the activation of additional neural circuits involving the cerebellum and brain stem (Olmo et al., 2006). These additional cortical structures would normally not get activated with internal generation of cues or conscious cognitive control. The finding of this study contradicts earlier research carried out in this field (Baker et al., 2008; Lohnes & Earhart., 2011) which does not report a negative impact of internal cueing on gait variability in PD in the *on* phase. This contrast finding (from the same research group) may be attributed to the patient sample recruited in this study which had a high percentage of PD participants with freezing and falling. Both these characteristics are related to worse internal beat timing and cognitive impairment in PWP (Rochester et al., 2011; Horin et al., 2020 a), and therefore a person’s inability to generate internal cues. The three year longitudinal study conducted by Lirani-Silva et al. (2019) also confirmed greater responsiveness to external auditory cueing in PWP as the disease progresses to compensate for the impaired attentional role.

Two studies (Morris et al., 1996; Lowry et al., 2010) compared attentional cueing to visual cues and found attentional cueing alone is as equally effective as visual cues in improving stride length and walking speed in PD. This finding is significant in PWP with visual impairment that may benefit from employing internal cues to improve gait performance. Can verbal instructions transition to self-prompted cues? This question was considered in two studies employing the verbal instruction *focus on big steps* (Lehman et al., 2005; Werner et al., 2010). Results showed improved stride and walking speed in post training sessions when no verbal cues were provided, suggesting the practise and instructions provided during the training session led to the translation of verbal cues into self-prompted attentional strategies to maintain step length. In both these studies the improved gait parameters were maintained for short (four weeks) and long term post training for up to a year. The outcome of Werner et al. (2010) study also found no additional benefit with the provision of videotape performance feedback, indicating that verbal instructional cueing alone was sufficient.

Internal musical cues

As noted in the earlier section, internal musical cues are the second main approach to delivery of internal cueing, with six studies examining their efficacy. Four of these (Satoh et al., 2008; Harrison et al., 2019; Horin et al., 2020 a,b) compared internal mental singing to external auditory musical cues and showed reduced gait variability with mental singing with minimal improvement in spatial and temporal parameters of gait. Satoh et al. (2008) found improved gait rhythm in the form of smoothness of movement and reduced number of steps while walking and turning. Harrison et al. (2019) compared internal ‘overt’ singing to mental singing and found improved gait variability with mental singing. Two further studies (Harrison et al., 2017, 2018) supported this finding by comparing *internal singing aloud* with external musical cues. Results showed statistical significance in gait variability measures with *internal singing aloud* in both these studies. No improvement in pace domains of gait was found in Harrison et al. (2017), in contrast to results of Harrison et al. (2018) which found positive effects on step length and step velocity, although it did not reach statistical significance. Taken together, these studies suggest the application of internal musical cues are beneficial in reducing gait variability in PD with no additional beneficial effect on the pace domain.

Effect of internal cues on gait performance

Gait speed and step length

A key finding of this research is that internal cue gait training has a positive effect on gait speed and step length in PD compared with non-cued gait. This is clinically important given that a slower gait with short steps is linked to an increased risk of falls and poor balance (Espy et al., 2010; Cromwell et al., 2004). An improvement of 1.03 cms/sec in walking speed and 2 cms in step amplitude constitutes clinically meaningful differences in these parameters, beyond the normal variance in PD gait (Baker et al., 2007).

The strategy used by most of the studies that reported an improvement in pace domain (via increases in step or stride length) employed internal attentional cues by focusing attention on *big steps* (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Lohnes & Earhart., 2011). The easily accessible intervention can be used by PWP, and although this strategy alone does not improve gait stability (Baker et al., 2008; Rochester et al., 2011), it has an immediate effect on the key component of gait (stride length).

Gait variability

PWP with freezing walk with increased gait variability, reflecting reduced automaticity of gait (Harrison et al., 2017). Strategies to reduce gait variability are important to improve gait performance and reduce the incidence of falls in PD; eight of 14 studies in this review assessed gait variability characteristics (Baker et al., 2008; Lowry et al., 2010; Rochester et al., 2011; Harrison et al., 2017, 2018, 2019; Horin et al., 2020 a,b). The majority of the studies tested step time variability which is a strong predictor of falls in PD (Schaafsma et al., 2003). Other variability measures tested included SST variability, step length variability and DLS variability. Results were contradictory, with authors either reporting no improvement (Rochester et al., 2008; Lowry et al., 2010; Horin et al., 2020 b) or improvement which was statistically significant (Harrison et al., 2017, 2018, 2019). However Rochester et al. (2011) showed increased step time variability which requires further consideration.

External cues selectively activate independent systems such as dorsolateral cerebellar circuits and the brain stem, which are unrelated to cognitive control and modify gait variability measures to reduce gait instability and falls in PD (Rochester et al., 2011). Increased gait variability to internal cues was only noted in Rochester et al., (2011). As discussed in the previous section, the PD sample group recruited in this study with more advanced disease severity may have contributed to this finding. Two studies reported improvement in step length and gait speed without any negative impact on gait variability measures (Baker et al., 2008; Lowry et al., 2010). This implies that PWP are capable of using internal cues to improve their gait quality without increasing attentional demand and affecting gait stability.

Effect of internal cues on function

The effect of internal cues in functional activities of daily life was tested in three studies (Satoh et al., 2008; Baker et al., 2007, 2008). There was no uniformity in the outcome measures. Satoh et al. (2008) reported improved gait rhythm indicated by higher knees, smoother turns and reduced number of steps while walking and turning, and improvement in the UPDRS motor scale following internal cue gait training. Baker et al., (2007, 2008) used internal cues whilst also carrying out a dual motor task (carrying a tray while walking). Gait speed and step amplitude improved with attentional cues, despite the dual tasking. However internal cues alone did not improve gait stability while performing the same dual task (Baker et al., 2008). These three studies suggest there is capacity to use internal cueing even whilst stressing attentional and cognitive capacity through multi-tasking. This may be especially true if the secondary task represents a daily functional activity which utilises less attentional resources.

Effect of internal cues and pharmacy

Dopaminergic medication leads to improved bradykinesia and force output in PD resulting in increased stride length and improvement in walking speed (Rochester et al., 2011). Only one study in this review tested the effect of internal cues in *on* and *off* dopaminergic medication in PD (Rochester et al., 2011). As reported earlier, the results established that internal cues improve stride length in both *on* and *off* dopaminergic medication compared with no cues. However, cues are more effective when combined with medication. Internal attentional cues have no effect on gait variability measures such as stride time CV and DLS CV *off* medication, but these cues significantly increased stride time CV when combined with dopaminergic medication.

This has important clinical implications, and indicates that internal cues selectively target dopamine gait dysfunction related to spatial domain and stride length impairment only. PWP are unable to modify gait variability fluctuations which might be controlled by additional neural mechanisms as mentioned earlier, not dependent on dopaminergic pathways and only targeted by external cues (Rochester et al., 2011). More studies should focus on evaluating the effect of internal cues with and without dopaminergic medication to better understand the underlying neural mechanisms.

Effect of internal cues on habituation

Internal cue habituation was not explicitly tested in any of the studies in this review. Werner et al. (2010) reported progressive improvement in stride length and gait velocity over the course of training sessions implying the efficacy of internal cues was maintained throughout the internal cue gait training. Given that internal cueing is a self-generated strategy requiring one’s own attentional skills, hypothetically it would be difficult to ignore them and habituate to them due to consistent vigilance required to incorporate these skills. This becomes more challenging if attention is divided due to environmental distractions or during attentional overload. A second reason for attenuation of internal cue efficacy is the progressive nature of cognitive decline in PD, which leads to executive and attentional impairment. In this instance, despite making significant improvements in gait performance with internal cue training, PWP might revert back to their baseline measurement due to an inability to self-employ the attentional strategy. This highlights that more work is indicated to evaluate cue habituation with internal cues.

Effect of internal cues on dual tasking

Although this review has only focused on the effect of internal cues in a single task condition (walking), it is important to point out a few things related to functional activities in daily life. Most of our daily functional tasks involve performing more than one activity simultaneously; therefore in order to be effective outside a clinical setting, internal cues should be able to improve gait parameters in dual task conditions. Only three studies assessed the effect of internal cues in a dual task situation (Baker et al., 2007, 2008; Lohnes & Earhart., 2011). A range of secondary tasks were used in the dual task paradigm. Lohnes & Earhart. (2011) employed a cognitive word generation task while walking and Baker et al. (2007, 2008) used a secondary motor task of carrying a tray while walking. Lohnes & Earhart. (2011) established no additional benefit of attentional or a combination of attentional and auditory cueing on any of the gait parameters in the dual task condition in PD. The secondary motor task employed by Baker et al. (2007) resulted in improved walking speed and step amplitude with attentional cueing in PWP. This led to a more normalised gait and reduced stride time gait variability with a combination of attentional and auditory cueing in the subsequent study (Baker et al., 2008). The reason for this discrepancy has been attributed to task difficulty rather than task type (O' Shea et al., 2002). The unfamiliar nature of a secondary word generation make people direct their attention to that task, leading to full engagement of cortical resources on the secondary task dividing attention and reducing gait quality (primary task). On the other hand, walking while carrying a tray is both a familiar and functional activity most of us are accustomed to in our daily life. This utilises less cognitive resources to perform, resulting in an attention cue still maintained on the primary task of walking.

Motor learning (retention and carry-over effects)

It is outside the scope of this structured review to explore motor learning deficits in PD, although in relation to the findings from the review, a brief summary is discussed. Research investigating retention of performance in PD suggests PWP have capability to acquire new skills, implying an unaffected procedural or implicit learning (Cohen et al., 2007; Marinelli et al., 2009). Although the effect of retention is inconsistent with some studies concluding PWP are unable to maintain the carry over effects in short and long term (Cohen et al., 2007; Marinelli et al., 2009) whilst others report no such deficit (Pendt et al., 2011). Retention in the context of motor learning is defined as the preservation of movement specific skill over a period of rest (Pendt et al., 2011). Pendt et al. (2011) found delayed response time (time shift deficit) in their PD group in retention of the skill. However this performance decrease was associated with delayed movement initiation at the beginning of a new session secondary to hypokinesia. This finding was not related to poorer retention; this view is also supported by previous studies (Platz et al., 1998).

The study by Ferrazzoli et al. (2017) evaluated reaction times of 103 PWP in *on* state before and after a goal based, aerobic and intensive rehabilitation treatment. They found an improvement in motor functional outcome measures. The authors suggested focused and sustained attention is modified following motor-cognitive neuro-rehabilitation in PD. Only three of 14 studies assessed retention and carry over effects on gait domains post internal cueing (Morris et al., 1996; Lehman et al., 2005; Werner et al., 2010). Two studies found the maintenance of statistically significant improvement in step length for short and long term post internal cue training (Lehman et al., 2005; Werner et al., 2010). Although future research should focus on testing retention and motor learning post internal cue gait training this information is useful for motivating PWP to increase the frequency of practise sessions.

Summary of key findings

Internal attentional cueing uses a cognitive or volitional mechanism of motor control and reroutes the movement through a non-automatic pathway bypassing the basal ganglia (Morris et al., 1996; Lehman et al., 2005; Baker et al., 2007; Werner et al., 2010). The primary motor deficit in PD relates to the impaired capability of generating adequate movement amplitude and force causing hypokinetic movements (Morris et al., 1994 a, 1996). Focussing attention on increasing step length by internal cueing therefore leads to improved stride and walking speed in PD, as shown in previous studies reported earlier.

Musical cues have also proven to be an effective intervention due to their capacity to not only activate the auditory system but also engage cortical and sub-cortical regions of the brain, thereby improving motor output in PD (Rao et al., 1997; Jancke et al., 2000; Lewis et al., 2004; Chen et al., 2006, 2008; Grahan & Rowe 2009; Kung et al., 2013). Self-generated vocal cues are also an established rehabilitation approach in improving speech impairment in PD, with research also reporting improvement in upper extremity tasks such as grasp with these cues (Narayana et al., 2010; Haneishi., 2001; Maitra., 2007). Despite evidence supporting the efficacy of musical cues, research also suggests employing externally generated music cues increase gait variability in PD due to cognitive demand for step synchronisation as mentioned previously.

Internal musical cues on the other hand are deemed comparatively simple due to using attentional resources only in the output or motor control phase, as compared to external musical cues when attention is required for music perception (input signal) and matching footsteps to the beat or motor control (output phase) (Satoh et al., 2008; Harrison et al., 2017, 2019). Matching movement to one’s own voice through a self-generated internal rhythm is more efficient and enables more accurate motor entrainment through ‘vocal motor coupling’ in comparison to matching movement to externally generated cues that rely on ‘auditory motor coupling’ (Harrison et al., 2017, 2019). Externally generated musical cues require consistent vigilance to match the footsteps to an external auditory beat, which likely increases gait variability (Harrison et al., 2018, 2019). In addition to the improvement in motor response, musical cues are also associated with upliftment of mood (Dunbar et al., 2012). PWP display apathy and compromised emotional response (Harrison et al., 2018), these symptoms are attributed to impairment of dopaminergic pathways related to reward and motivation (Vickery et al., 2011). Active music making in the form of self-generated musical cues is reported to increase endorphin release as compared to passive music listening in the normal healthy population (Dunbar et al., 2012).

Improvement of gait performance using cueing strategies is dependent on the cognitive ability of PWP. If cognitively intact they may be able to learn to self-instruct in the use of an internal cueing strategy. On the other hand, PWP with cognitive impairment may use external sensory cues more effectively to improve gait parameters. The findings from this structured review confirmed that internal attentional strategy by creating a mental image of the correct step length helps to improve gait dysfunction in PD by improving stride length and walking speed. This cue however is not effective in improving gait stability due to the possibility of increasing attentional demand in complex unfamiliar situations.

Internal musical cues in the form of mental singing or singing aloud, on the other hand, are not additionally taxing on the brain due to minimal or nil change in gait variability measures as reported by this structured review’s findings. The past musical experience of participants did not impede the capability of employing the musical cues either. An individual might prefer to use one intervention over the other, for example self-generated active singing can pose a logistical challenge for some people not comfortable singing aloud while walking on the street. Instead they may prefer to mentally sing or use an attentional strategy to improve their gait. PWP also often exhibit speech impairment leading to hypo phonia and difficulty with word formation (Harrison et al., 2019). In these situations PWP would likely prefer internal cueing in the form of mental singing in their own head to avoid vocalisation. The majority of the research in external cueing is carried out in clinical settings such as walking on a straight path, which does not represent our daily functional activities. In everyday life people walk in confined spaces such as in the kitchen, laundry or toilet, and have to turn around. The additional cognitive demand for PWP matching footsteps to external cues in this situation can increase attentional demand, cause panic and increase the risk of falls.

The advantage of internal cues is the ability of the person to internally modulate and control the provision of cues depending on the environment, and set up their own tempo to match their fluctuating symptoms through the day. This increases its likely applicability in real world situations in everyday life. The combination of an internal and external cue has been shown to improve gait stability in the past (Baker et al., 2008; Horin et al., 2020b). This combination can be replaced by two internal cueing approaches of attention and musical cues likely yielding a similar benefit. Future research should focus on reviewing the effect of combining these two internal cueing strategies.

However similar to external cueing strategies, internal cueing is not applicable in all stages of PD. Self-employment of internal cues requires cognitive and executive function limiting the applicability and optimal therapeutic window of internal cueing techniques to the early stages of PD when disease severity and cognitive impairment is mild to moderate.

Conclusion

Internal cueing improves both gait speed and step length in PWP but fortunately not at the expense of increasing gait variability. Less conclusive are the effects of internal cueing on rhythmic and stability features of gait. Improvements in stability require a combination of internal and external cues. Although the body of evidence supporting these views is mixed, it does include one Level A evidence study (Nieuwboer et al., 2007) alongside several other experimental studies (Baker et al., 2008; Horin et al., 2020b).

This review also points to the selective response of cueing strategies on gait outcomes. The effect of internal cues is different, for example, than a combination of internal and external cues. Further research is required to tease out this selectivity which informs a tailored treatment approach for each individual.

This research is of significance due to the growing prevalence of PD both nationally and internationally. As a treatment option, internal cueing is cost-effective and a self-managed component of symptom-relief and disease management. Furthermore, the ability to self-generate and control the tempo of cues makes this technique diverse and increases its applicability in a dynamic real world setting. The findings of the structured review are illustrated in Figure 5.

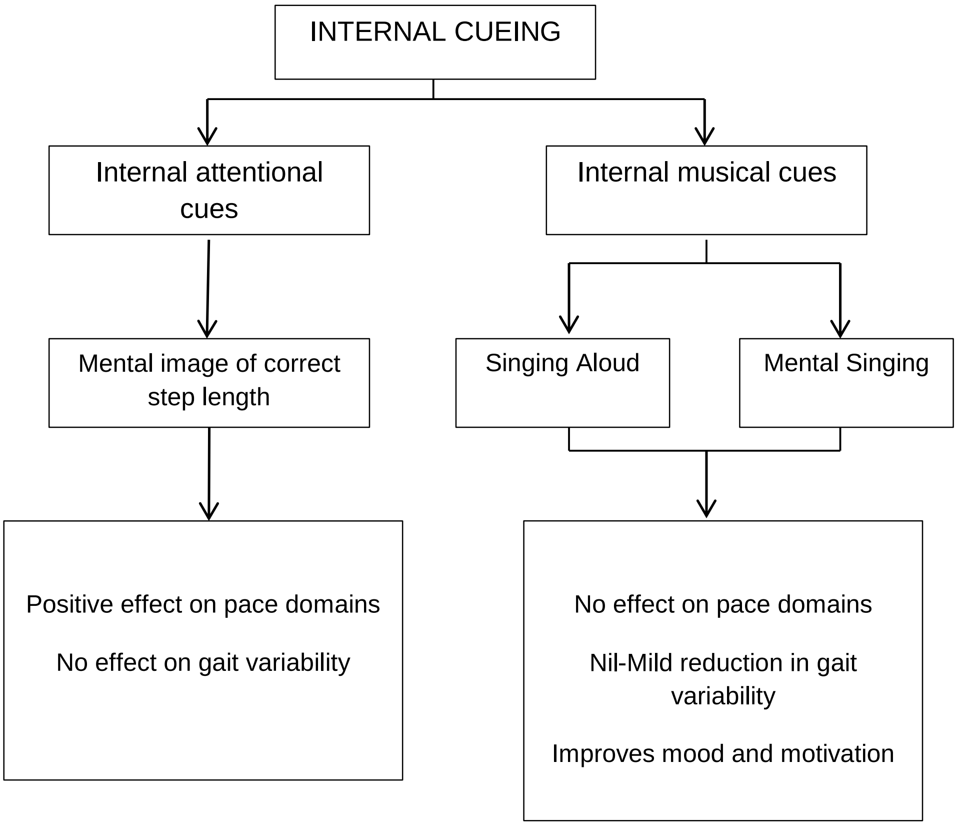


Figure.5. Findings of the structured review

Limitations

Current research

The methodological quality of included studies was evaluated using McMaster critical review form for quantitative studies (Wells et al., 2014). This critical tool was utilised because it is comprehensive in assessing methodological quality of quantitative evidence and also has good inter-rater reliability (Wells et al., 2014). As mentioned earlier, due to the structured nature of this review a score was not given to each study. The tool assisted the author highlight overall quality of the body of quantitative research. The effect of internal cues was not explicitly articulated in the research question in some studies (Morris et al., 1996; Lowry et al., 2010; Werner et al., 2010; Rochester et al., 2011; Horin et al., 2020a). The author read the full text articles to determine the eligibility of the study in the structured review. Only one randomised control trial was included (Lehman et al., 2005), and most studies had a small number of participants. All studies except Rochester et al. (2011) included PD participants with mild to moderate disease severity and non-significant gait deficits which limited generalisation of the review findings to the wider PD population with more advanced disease severity. Spatial and temporal gait parameters as well as symmetry of the gait contributes to overall gait stability resulting in meaningful improvement in mobility and reducing risk of falls (Harrison et al., 2019). Only five studies assessed both pace domains and gait variability characteristics (Lowry et al., 2010; Rochester et al., 2011; Harrison et al., 2018, 2019; Horin et al., 2020 b). Only one study in this review tested the efficacy of internal cues in *on* and *off* dopaminergic medication in PD (Rochester et al., 2011).

Studies were conducted in a laboratory setting while participants walked on a straight pathway, only one study included turns in the gait training protocol (Satoh et al., 2008). Although internal cueing strategies resulted in improved gait performance in a clinical setting when walking a straight path, it is uncertain whether these strategies will be able to assist PWP in daily life to improve their functional activities in a real world setting. Intensive internal cue gait training was provided to all participants challenging the applicability of this intervention in the real world physiotherapy clinical settings with limited staff, time and health resources available. The findings of this structured review are summarised for single task conditions only, with limited evidence for efficacy of internal cueing strategies under dual task conditions. In studies employing internal generation of musical cues in one's own head, researchers only relied on the participants' subjective account and overt lip movements to assess cue performance. The studies did not outline objective measures reporting functional improvement following internal cue gait training. Habituation to internal cues and participants' personal preferences to cue types were also not reported in the studies.

Review process

There are a number of limitations of this structured review process. The use of broader search terms ensured relevant article retrieval, perhaps more exploration of possible synonyms, may have yielded more robust search results. The search yield and articles selection were only analysed by the author and lacks peer review. As already mentioned, the article search and selection was an extensive process due to several barriers; this include a lack of uniformity defining external and internal cues and the interchangeable use of terminology for internal cueing. The results are only reported for the effect of internal cues in PD. More thorough synthesis to compare and present the effect of different cueing interventions within PD participants and between different groups would have provided better insight into underlying mechanisms and strong rationalisation for future research in this area. Data extraction was also challenging due to incorporation of mixed cueing techniques, five studies employed combination of internal and external cueing interventions (Baker et al., 2007, 2008; Rochester et al., 2011; Lohnes & Earhart., 2011; Harrison et al., 2017). For better interpretation of study findings and results, the cueing interventions are explicitly listed and presented in Table 2. The results are presented as *p* value, analysis and synthesis of mean difference and 95% confidence intervals in the extracted data may have aided more robust interpretation of the results.

Clinical Implications, Future Direction and Recommendations

Physiotherapy implications

The practical implication of internal cue gait training is diverse in physiotherapy clinical practise, both for clinicians and PWP. Incorporating service delivery of internal cued gait rehab in physiotherapy clinical practise does not require additional set up of equipment or space for the clinician. As a result the gait training can be provided in an inpatient or outpatient setting, or at home and in the community. As already discussed in the earlier section, internal cues are more beneficial in the early stages of PD. An alternative could be to provide internal cued gait training to PWP in group setting due to mild physical and cognitive deficit. Although future research is required to investigate the economic cost, this will be time effective and less resource intensive which will reduce the burden on the public health system.

Demonstration and use of verbal instructional strategy is a most common tool used by clinicians to communicate with patients. In physiotherapy practise, a clinician may demonstrate to a patient how to stand up from a chair or provide verbal cues to guide a movement for specific muscle activation. A clinician does not need to be formally trained to provide internal cued gait rehab as the instructions to focus on *big steps* or *internally sing* are provided through verbal and physical prompts. The easy applicability to use this approach increases its utility not only in physiotherapy field but also in retirement villages and aged care facilities to improve quality of life in elderly PWP with mild cognitive and gait deficits.

Exorbitant cost of visual cueing modalities is a significant hurdle to its limited use in PD. In New Zealand public health system, Ministry of health (MOH) is able to fund the essential equipment needs of a disabled person through disability support services (for more information on the criteria please refer to MOH website, link provided in Appendix B). This means PWP are eligible to receive standard walking frame to assist with mobility free of cost if they fulfil the eligibility requirements. Although MOH does not provide funding for customised projection light mounted walking frames or walking sticks which provide visual cues and are beneficial in PD. This is done so services are allocated fairly through a consistent, principled and equitable approach across the diverse range of people the Ministry serves. This implies PWP have to rely on privately purchasing these specialised walking aids and cost acts as a barrier. The advantage of internal cues is their self-generation removing cost and making it more feasible approach.

Visual and auditory cues are difficult to incorporate in restricted and compact spaces limiting their use in daily functional tasks in PWP. Examples of such activities include walking between different rooms of the house whilst negotiating furniture and different floor surfaces (rugs, tiles or wooden surface) or stepping backwards to sit in the chair. It is nearly impossible to follow the visual markers or walk to a predetermined beat in the above situations. The beneficial effect of internal cues through self-generation leads to its easy implementation in indoor activities. For instance internal cueing strategy may be incorporated while attending to personal care needs in compact bathroom space when stepping in or out of the shower, to access uneven driveway of the house, external or internal steps, stepping through the front door or walking in the narrow hallway. Internal cueing may also be used for outdoor or community ambulation due to its discrete application; such as while walking on uneven terrain or on footpath, stepping on the kerb or on pedestrian crossing.

Studies in this structured review provided frequent sessions of gait training to PD participants which overall resulted in improved gait speed and step length post training sessions. However, in clinical practice, approximately 10 minutes of gait training is typically provided (Werner et al., 2010). As PWP require more practice for motor learning, education and guidance should include continuing the training at home for improved outcomes and maintain compliance to the programme. The gait profile of PWP is inconsistent and exhibits motor fluctuations during the day. Due to this, PWP with mild cognitive and gait deficit should be educated to use internal cueing strategy during their fluctuating symptoms when gait deviates from normal.

Physiotherapists should focus on providing internal cued gait training to replicate practical environmental and real life situations such as walking and turning, during gait initiation, walking over different surfaces and backward stepping. These skills can then be transferred to daily functional tasks such as standing up from chair to walk or stepping back to sit in the chair.

Future Direction and Recommendations

Maintaining gait stability while walking is an automatic process in the normal healthy population which becomes attention demanding in PWP and worsens with the addition of a secondary task due to attentional overload (Satoh et al., 2008; Harrison et al., 2017). Most of our daily activities involve dual tasking, whether walking and talking or walking and carrying a plate of food or cup of tea. Although few studies assessed the effect of internal cueing on dual task, the paradigms differed between studies with an employment of secondary cognitive or motor dual tasks resulting in a lack of uniformity which might have influenced the research findings. Future research should focus on reviewing the effect of internal cueing strategies on its own and in combination under dual task conditions replicating real life situations. For overall gait stability and meaningful improvement in mobility, gait characteristics including spatial and temporal parameters as well as variability measures should be tested to review efficacy of internal cueing.

The studies did not specifically include PD patients with freezing which is associated with worse internal beat timing (Horin et al., 2020 a) leading to an inability to use internal cues effectively. The increased attentional demand of employing the internal cueing technique implies that PD patients with freezing might not positively respond to this cueing strategy. Future research should focus on the effect of internal cueing on PD patients with freezing. Only one study (Satoh et al., 2008) conducted the research which included a real life setting such as turns. More studies need to conduct research in real life settings to see the effects of internal cueing in daily activities. There are inconsistencies in the research regarding PWP capability of learning new skills and carry over effects. Future research should also focus on testing retention post internal cue gait training to investigate motor learning in PD.

Internal cues through attentional control might only be effective in addressing dopamine gait dysfunction and spatial gait parameters (Rochester et al., 2011). More studies should focus on evaluating the effect of internal cues with and without dopaminergic medication. In the current review, back ground musical training did not impede participants' ability to incorporate musical cues in gait training; future research should include participants with musical experience to determine if a stronger response is elicited with past musical experience and review the suitability of this technique for the appropriate patient group. Internal cue gait rehab might be suitable to deliver in group classes due to the targeted PD population with less cognitive and physical deficits. Future research should focus on comparing the functional outcome and economic cost of delivering internal cued gait training in an individualised and group setting in PD to investigate most effective service delivery method.

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Glossary

List of Abbreviations

A	
AP	Antero posterior
C	
CI	Cognitive impairment
CV	Coefficient of variation
cms/sec	Centimetres/second
cms	Centimetre
D	
DLS	Double limb support duration
DT	Dual task
E	
Ext cue	External cue
F	
fMRI	Functional magnetic resonance imaging
G	
GPi	Internal globas pallidus
GPe	External globas pallidus
H	
H&Y scale	Hoehn & Yahr disease severity classification
HC	Healthy controls
HR	Harmonic ratio
I	
Int cue	Internal cue
M	
Med	Medication
ML	Mediolateral
m/sec	Meter per second
m/min	Meter per minute
m	Meter
min	Minute
ms	Millisecond
mm	Millimeter
P	
PD	Parkinson's disease
PWP	People with Parkinson's
PD-FOG	PD without freezing of gait
PD+FOG	PD with freezing of gait
PIGD	Postural instability gait disturbance
PET	Positron emission topography
PMC	Premotor cortex
p	P value (clinically meaningful improvement)
S	
SMA	Supplementary motor area
SST	Single support time
STMS	Short test of mental status dementia
ST	Single task
Steps/min	Steps per minute
sec	Second
U	
UPDRS	Unified parkinson's disease rating scale
V	
VI	Verbal instruction

Appendix A

External cue device cost

External cue device	Cost price	Website
U Step walker frame with cueing module and laser	NZ $1700	https://mobilitymanawatu.co.nz/u_step.html
Laser cane	NZ $ 402.50	https://mobilitymanawatu.co.nz/u_step.html
Laser guided shoes	£1100	https://walkwithpath.com/product/path-finder/
Metronome auditory cues	Free	https://pianotraders.co.nz/collections/tuners-metronomes

Appendix B

Ministry of health (MOH) Equipment manual

Website page	https://www.health.govt.nz/our-work/disability-services/contracting-and-working-disability-support-services/equipment-and-modification-services
Document	https://www.health.govt.nz/system/files/documents/pages/equipment-manual-nov2014.pdf

Appendix C

Hoehn and Yahr stages

Stage 1.0: Unilateral involvement only.

Stage 1.5: Unilateral and axial involvement.

Stage 2.0: Bilateral involvement without impairment of balance.

Stage 2.5: Mild bilateral involvement with recovery on retropulsion (pull) test.

Stage 3.0: Mild to moderate bilateral involvement, some postural instability but physically independent.

Stage 4.0: Severe disability, still able to walk and to stand unassisted.

Stage 5.0: Wheelchair bound or bedridden unless aided