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Review Article

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Abstract

Regular exercise can improve wellbeing, yet data are scarce on how persons with disabling conditions may benefit from active lifestyles, due to the complexities of exercise prescription in this population. A novel medical concept for exercise prescription called activity pacing is the subject of this review, which identifies the potential for this strategy to optimally integrate existing medical and sports medicine approaches in promoting physical activity in persons with disabling conditions. Activity pacing is a goal-directed behavioural process of empowering people to confidently develop decision-making and planning over how and where to distribute available energy across daily activities. Currently, different conceptual traditions and definitions of pacing exist with important implications for the implementation and subsequent effectiveness of activity pacing. Application of activity pacing has mostly focused on symptom-reduction to improve self-regulatory behaviour, and less on physical activity stimulation for health and wellbeing. Further studies and greater connection between medical and sports science research are needed on how to adapt, tailor and optimise activity pacing to make it successful. The potential of activity pacing to increase physical activity and lessen fatigue could be a powerful tool to help fight the growing incidence of physical inactivity, particularly in persons with disabling conditions.

Keywords: physical activity, fatigue, pacing behaviour, disabling conditions, self-regulation

Introduction

Worldwide public health data clearly demonstrate physical activity levels are low across the general population, but worryingly this is even more prevalent in persons with disabling conditions (World Health Organization, 2002). There are many causal elements behind this observation, but engaging in regular physical activity depends on successfully managing and distributing physical efforts across daily activities. However, this can be particularly challenging to those with disabling conditions due to varying degrees of physical impediments and psychological disturbances such as depression (Kargarfard, Etemadifar, Mehrabi, Maghzi & Hayatbakhsh, 2012; Motl, McAuley & Snook, 2005), in addition to reduced confidence to exercise and self-awareness of one's physical limits (Barnett et al., 2012; Durstine et al., 2000). Worryingly, studies investigating the effects of exercise in people with disabling conditions report a high number of dropouts, and identified that participants struggle to continue engaging in activity post-intervention (Larun, Brurberg, Odgaard-Jensen & Price, 2017; Roehrs & Karst, 2004). This indicates that the way exercise is introduced, delivered and/or undertaken might influence its long-term adoption within a physically active lifestyle.

The importance of habitual physical activity has been extensively documented (Kayes et al., 2011; Lee et al., 2001; Motl, McAuley & Snook, 2005). Persons with disabling conditions such as multiple sclerosis, chronic fatigue syndrome, fibromyalgia, and osteoarthritis often struggle with mobility and consequently sedentary behaviours are common; however, this makes engagement in physical activity of even greater importance. Increasing physical activity is associated with an estimated gain of 4.5 years of life compared with being inactive (Moore et al., 2012), reduced fatigue, and psychological conditions in persons with chronic conditions who often are affected by these consequences of their condition (Motl, McAuley & Snook, 2005; Murphy and Kartz, 2014). Thus strategies to promote physical activity ought to be a primary goal for persons with disabling conditions (Motl, McAuley & Snook, 2005).

Several approaches have been successful in stimulating an active lifestyle in persons with disabling conditions (Alingh et al., 2015; Larun, Brurberg, Odgaard-Jensen & Price, 2017; Murphy, Lyden, Smith, Dong & Koliba, 2010; Nielson & Jensen, 2004; Roehrs & Karst, 2004) but not much is yet known on the overarching principles of how to achieve this for a wide range of persons with disabling conditions. Existing approaches (graded exercise therapy and cognitive-behavioural therapy) to promote physical activity in persons with disabling condition are typically expensive, resource-intensive and not widely accessible (Castell, Kazantzis & Moss-Morris, 2011).

This review overviews literature related to physical activity and condition-induced fatigue in persons with disabling conditions, and explores the potential of better promoting self-regulatory behaviour through activity pacing, a recent medical concept to aid engagement in physical activity and accurately distribute available energy throughout the day (Smits, Pepping & Hettinga, 2014). With appropriate education and experience (Micklewright et al., 2012), this approach may be beneficial to stimulate persons with disabling conditions longitudinal engagement in physical activity.

Physical activity in persons with disabling conditions

An estimated 10.2–46.1% of the world's population have moderate to severe disabilities and experience significant functional difficulties (World Health Organisation, 2004). However, there is a dearth of physical activity data available on persons with disabling conditions (World Health Organisation, 2004; Murray & Lopez, 1997). A disturbing statistic is that physical activity levels among persons with disabling conditions are significantly lower across all age groups compared to non-disabled people (Durstine et al., 2000). Consequently, many persons with disabling conditions do not achieve the recommended amount of physical activity required for maintaining health (Garber et al., 2011; Van den Berg-Emons, Bussmann & Stam, 2010). Therefore, the greater time spent in sedentary behaviour compared with the general population means that this population, often already with limited physical functionality, has a problem exacerbated by an inactive lifestyle (Van den Berg-Emons et al., 2008).

Several studies rightly emphasised that excessive rest and the lack of physical activity found in persons with disabling conditions can result in reduced physical functionality and increased physical deconditioning (Afari & Buchwald, 2003; Bakkum, de Groot Sonja, van der Woude & Janssen, 2013; Boutron et al., 2008; Clark & White, 2005). This consequently perpetuates early-onset fatigue when active and further compounds the impact of physical disability on mobility and participation in activities of daily living, work, and other meaningful activities causing a downwards spiral (Grotle, Hagen, Natvig, Dahl & Kvien, 2008; Sutbeyaz, Sezer, Koseoglu, Ibrahimoglu & Tekin, 2007; Theis, Murphy, Hootman, Helmick & Yelin, 2007; World Health Organisation, 2001).

Knowing that physical activity has health-enhancing impacts such as positive effects on symptoms, quality of life, mobility and participation in daily life (Anderson, Jason, Hlavaty, Porter & Cudia, 2012; Goudsmit, Nijs, Jason & Wallman, 2012; Rimmer & Marques, 2012; Roehrs & Karst, 2004; Van Koulil et al., 2010) inevitably means a physically active lifestyle is strongly recommended for persons with a disabling conditions (National Institute for Health and Clinical Excellence, 2007; Plotnikoff et al., 2013). Consequently, because there is, as of

yet no cure for disabling conditions, the promotion of an active lifestyle has been considered to be an important factor in the treatment of disabling conditions symptoms (National Institute for Health and Clinical Excellence, 2007).

Activity pacing as potential intervention to manage fatigue and promote an active lifestyle in persons with disabling conditions

Too vigorous exercise, or even a 30% increase in activity, has been shown to exacerbate symptoms in persons with disabling conditions (Black, O'Connor & McCully, 2005; Jammes, Steinberg, Mambrini, Brégeon &, Delliaux, 2005). In addition, specific activities, expected to exacerbate symptoms have been shown to be less frequently performed by persons with disabling conditions (Kayes et al., 2011; Vercoulen et al., 1997) indicating an exercise programme based around greater opportunities for self-regulation may aid adherence and minimise condition-induced avoidance of exercises or drop-out. Therefore applying a self-regulatory exercise therapy such as activity pacing to persons with disabling conditions is potentially important, particularly in terms of its long-term adoption within a physically active lifestyle (Nijs, Paul & Wallman, 2008).

Activity pacing is a new therapeutic intervention that has the potential to stimulate an active lifestyle by lowering fatigue and increasing physical activity in persons with a disabling condition. Activity pacing as defined in medical settings, is a strategy to educate and develop individuals' self-regulatory skills to divide one's daily activities into smaller, more manageable portions, in a way that should not exacerbate their symptoms, which then allows gradual progressive increases in activity (Andrews, Stron & Meredith, 2012). The concept of activity pacing postulates that by perceiving an increase in physical activity without exacerbation of symptoms, patients are likely to feel more in control of their fatigue and focus less on fatigue, which can lead to positive effects such as task enjoyment, better fatigue management and physical function (Chalder, Goldsmith, White, Sharpe & Pickles, 2015).

The rationale for activity pacing as an intervention to stimulate engagement in physical activity can also be found elsewhere in literature (Nijs Wallman & Paul, 2011). In rehabilitation practice, several activity engagement strategies have been observed in daily lives of persons with disabling conditions. These include reduced activity levels resulting from and in anticipation of fatigue (Clark & White, 2005; Nijs et al., 2009; Nijs, Wallman & Paul, 2011), activity peaks followed by very long rest periods (van der Werf, Prins, Vercoulen, van der Meer & Bleijenberg, 2000), and the ability to perform short periods of light to moderate activity without exacerbating symptoms (Cook et al., 2005). However, activity pacing as a potential treatment to stimulate engagement in an active lifestyle for persons with disabling conditions has not been fully explored (Amato et al., 2001).

The unpredictable illness trajectory and symptoms characteristic of disabling conditions bring challenges specific to this population and to their engagement in physical activity (Anderson, Jason, Hlavaty, Porter & Cudia, 2012; Crook et al., 2005; Kayes et al., 2011). Consequently, in some persons with disabling conditions, physical activity/exercise may exacerbate symptoms and thus may not be beneficial for such individuals. Also, activity pacing as a treatment option may not be possible to practice in some persons with disabling conditions due to loss of function and/or cognitive dysfunction (Goudsmit, Nijs, Jason & Wallman, 2012; Grotle, Hagen, Natvig, Dahl & Kvien, 2008; Micklewright et al., 2012; Motl, McAuley & Snook, 2005). Thus alternative ways of treating symptoms and improving quality of life in such individuals are needed.

The concept of pacing has long been established in a sporting context (Hettinga et al., 2017), mostly in endurance activities, whereby physical capabilities are managed by an athlete in order to finish a race or event in an optimal performance time, depending on the goal of the athlete. Several researchers (Edwards, Bentley, Mann & Seaholme, 2011; Smits et al., 2014) have examined the balance of performance and recovery periods holistically, and have stressed the importance of self-regulatory skills for effective pace-regulation particularly in longer exercise tasks involving fatigue, both within a race as well as en route towards the long term goal of athletic excellence (Brick, MacIntyre & Campbell, 2016; Elferink-Gemser & Hettinga, 2017). Several different theoretical frameworks on pacing in sports have in some way suggested that competition between psychological, physiological and/or social factors is essential for decision-making regarding the regulation of exercise (Konings & Hettinga, 2018; Marcora, 2008; Renfree, Mytton, Skorski & St Clair Gibson, 2014; Smits et al., 2014; St Clair Gibson, Swart & Tucker, 2017; Venhorst, Micklewright & Noakes, 2017), with fatigue as a crucial factor. Pacing decisions have been suggested to be the outcome of the interplay between the sensation of fatigue and exercise expectations (Lambert, 2005; Noakes, St Clair Gibson & Tucker, 2009). In addition, planning and self-regulation skills have been identified as essential (Elferink-Gemser & Hettinga, 2017).

As early as 1996, Ulmer theorized the existence and functioning of a closed-loop feedback control system for optimal adjustment of effort during exercise to manage physical energy resources in relation to the known demands of the task. A framework for examining extracellular regulation of muscular metabolic rate during exercise was provided, which suggested central regulation occurred by optimising the perception of effort or teleoanticipation along with feedback from peripheral physiological systems (e.g. working muscles) so that tasks could be completed within physiological capacity (Edwards & Polman, 2012; Marino, 2014). Based on previous experiences, the pacing process can be learned and optimised (Foster et al., 2009; Micklewright et al., 2012), and a distinction has been made

between pre-planned deliberate strategic elements that determine optimal pacing (i.e. macro pacing), and more intuitive adaptations that occur while engaging in activities (i.e. meso and micro pacing) (Edwards & Noakes, 2009; Micklewright, Kegerreis, Raglin & Hettinga, 2017). These factors are relevant when exercising in diverse environments where multiple factors of varying importance impact on exercise-related decision-making (Smits et al., 2014).

Though the relevance of understanding the regulation of exercise intensity for a broader audience of exercisers has been highlighted (Smits et al., 2014), the majority of pacing research has tended to be limited to managing and describing competitive performances. However, the principles underlying pacing and the regulation of exercise intensity could also apply in medical and clinical contexts, extending well beyond the maintenance of physical efforts in a single task. By self-managing and spreading physical efforts across multiple daily tasks, it is possible for individuals to have greater confidence to engage in many activities they may not have previously thought possible, which accumulatively represent a more active, fulfilling lifestyle, of greater physical engagement. This can be achieved by employing better strategies to manage fatigue symptoms and distribute the limited available energy resources to prevent overactivity causing periods of subsequent inactivity. The next section overviews the literature regarding activity pacing and its potential to stimulate a physically active lifestyle.

Activity pacing as a concept to influence physical activity behaviour

Within the concept of activity pacing in rehabilitation, a distinction can be made between '*naturalistic pacing*' and '*programmatic pacing*'. The distinction between naturalistic pacing and programmatic pacing is analogous to the distinction between macro pacing, and meso and micro pacing in sport. The main difference between concepts being that in rehabilitation, it is applied to the pacing of activities over a day instead of the pacing of a single race or exercise bout in sports. Naturalistic pacing comprises the level of activity pacing that a person implements in daily life without a specifically instructed activity pacing programme (Nielson, Jensen, Karsdorp & Vlaeyen, 2013). Programmatic pacing involves treatment with pacing instruction to allow individuals to participate in activities in a way that should not exacerbate their symptoms, which then allows planned and calculated increases in activity (Andrews, Strong & Meredith, 2012). While pacing in sport is very much oriented towards the relatively straightforward goal of setting the best performance and using all the available energy as efficiently as possible, activity pacing has added complexities. These complexities are underpinned by the need to engage in physical activity behaviour to improve fitness and mobility, while at the same time preventing too severe fatigue symptoms that will impact on any subsequently planned physical activity. It is therefore more of a lifestyle strategy.

Within the concept of naturalistic activity pacing, there is a lack of clarity in the direction of the relationship between physical activity behaviour and symptom outcome. The conundrum here is do persons engage in more pacing behaviour in daily life due to an increase in perceived symptoms (symptom-contingent) or do persons engage in more pacing behaviour and thereby reduce their perceived symptoms (symptom-reduction) (Antcliff et al., 2015; Nijs et al., 2008). More insight in relations between physical activity, fatigue and naturalistic pacing could provide input to develop strategies and possible interventions to help persons with high fatigue complaints manage their fatigue through 'programmatic' pacing.

In programmatic pacing, patients receive a specific treatment with pacing instructions to learn and stimulate optimal activity pacing behaviour. The specific goal of this training varies depending on the theoretical orientation of the treatment and may include a focus on pain reduction, lessening of fatigue, and/or increased overall activity (Nielson, Jensen & Hill, 2001). It is more of an instructional and educational pacing strategy where individuals may learn to become more naturalistic in their approach to their pacing of life activities.

While several studies support links between programmatic pacing and lower levels of fatigue and disability (Murphy et al., 2008; Nielson and Jensen, 2004; van Koulil et al., 2010; Kos et al., 2015), a number of studies show no association (Murphy et al., 2010; Nijs et al., 2009; White et al., 2011). In a sample of people with chronic fatigue syndrome, programmatic pacing was associated with low fatigue severity, high leisure time physical activity, improved personal activity goal progress and health related quality of life (Marques et al., 2015).

Likewise, in 2010 Murphy, Lyden, Smith, Dong & Koliba reported in their study that programmatic pacing was associated with low fatigue severity. Similarly, van Koulil et al., (2010) found a reduction in fatigue severity and a trend towards improvement in physical function related to concurrent programmatic pacing and exercise training. Additionally, though not statistically significant, participants in a study of programmatic pacing demonstrated increased physical activity and physical functionality (Murphy et al., 2008).

Contrariwise, White et al., (2011) showed that programmatic pacing did not improve fatigue and physical functioning compared to graded exercise therapy and cognitive behavioural therapy. Additionally, Nielson et al., (2013) reported that increased pacing was associated with higher levels of pain and fatigue and suggested that future research should be based on a clear theoretical foundation and consider the context in which the behaviour occurs. These findings may suggest that if programmatic pacing has a role then it may be to develop a more self-directed naturalistic pacing approach to lifestyle management which would aid longitudinal engagement in physical activity.

In a study to measure naturalistic pacing behaviour in 30 women with osteoarthritis (OA), Murphy, Smith & Alexander, (2008) reported that naturalistic activity pacing was related to lower physical activity. Furthermore, when compared with low engagement in activity pacing, high engagement in activity pacing persons had more severe, escalating symptoms. Alternatively, Murphy, Kratz, Williams & Geisser, (2012) in their study on associations between symptoms, coping strategies, and physical activity in adults with OA reported that naturalistic pacing moderated the relationship between fatigue and physical activity. Those with high levels of activity pacing have the smallest association between fatigue and physical activity. Also, with decreasing use of pacing, the association between fatigue and physical activity becomes increasingly negative.

In addition, Murphy and Kartz, (2014) studied naturalistic pacing in 162 older adults with OA and reported that high activity pacing was associated with higher subsequent levels of fatigue and that naturalistic pacing seemed symptom-contingent and not reinforced by symptom reduction. They further stated that naturalistic pacing may be distinct from programmatic pacing in terms of outcomes. Similarly, Andrews et al., (2012) reported that an increase in disability relating to naturalistic pacing may reflect the ineffectiveness of pacing if not used to gradually increase an individual's activity level. They further suggested that people with better psychological functioning who experience more disruption through fatigue in daily life are more inclined to pace their activity.

While not the focus of this review, some interesting works have examined self-paced and imposed-pace exercise in sports. Together, they demonstrate that imposed-paced exercise presents a significantly greater physiological challenge than self-paced exercise (Edwards et al., 2011; Lander, Butterly & Edwards, 2009). However, the ability to dynamically self-pace effort is an important behavioural response to homeostatic challenges. In this way, the individual is able to down regulate effort when necessary and up regulate when feeling strong. Knowing physical limitations is an important part of self-regulated exercise and so developing these skills in programmatic pacing would be an important strategy to aid further independent self-regulation.

From the preceding paragraphs, most of the few studies on activity pacing focused on programmatic pacing with little emphasis on naturalistic pacing (Antcliff et al., 2015; Nielson et al., 2001). Together, these findings demonstrate that despite the frequent use and theoretical benefits of activity pacing, there is a dearth of and conflicting empirical evidence regarding effects of activity pacing (Jones et al., 2015; Nielson et al., 2001), although its application to clinical and rehabilitation contexts appears promising.

Over-activity vs. under-activity

The existence of different concepts and definitions of activity pacing which translate into its implementation may have contributed to the current lack of clarity about the nature and impact of activity pacing (Murphy and Kratz, 2014). In some studies, activity pacing is described as adjusting to one's condition and staying within limited amounts of energy by alternating activities and incorporating rest periods (Murphy et al., 2010; White et al., 2011). In other studies, activity pacing is described as modifying behaviour by going slower, taking breaks, maintaining a steady pace and splitting tasks into manageable pieces, managing symptoms whilst reducing relapses and gradually increasing activity (Antcliff et al., 2015; Kos et al., 2015; Nielson et al., 2013; Nijs et al., 2009; Nijs et al., 2008).

Most interventional designs of activity pacing focused on symptom-reduction and in particular on preventing over-activity. Instructions are based on limiting or avoiding those activities that exacerbate symptoms. While some studies advised patients not to undertake activities that demanded more than 70% of their perceived available energy levels (White et al., 2011), others advised activity duration 25–50% lower than the capacity participants reported (Kos et al., 2015). The evidence that over-activity may perpetuate fatigue and subsequent functional decline may have contributed to this phenomenon of focusing mostly on symptom reduction and preventing symptom exacerbation by curtailing over-activity.

The large focus on preventing over-activity may however represent a gap in literature as underactivity has also been linked to functional impairment (Birkholtz et al., 2004). It is possible that the current inconclusive findings on activity pacing may be accounted for by variation in characteristics such as illness duration, physical behaviour and attitudes towards both naturalistic as well as programmatic activity pacing. Studies that reported poor outcomes may have sampled persons with prior underactive behaviour for whom instructions regarding prevention of over-activity is likely to be non-beneficial (Andrews et al., 2012; Murphy and Kartz, 2014; Murphy, Smith & Alexander, 2008), while positive outcomes may have been obtained in an overactive sample of the population (Kos et al 2015; van Koulil et al., 2010). It can thus be inferred that interventions modelled based on the assumption that over-activity needs to be prevented are less likely to be effective in underactive persons. Equally, with activity pacing related to activity management, it is imperative to consider the physical behaviour and attitudes towards physical activity of persons when delivering an intervention (Murphy et al., 2008). An individually-tailored approach, based on characteristics that are unique to that person, related to the outcome of interest, and derived from an individual assessment (Rimer and Kreuter, 2006), is therefore needed.

Recommendations for future research

There is growing consensus for the need of a clear definition of activity pacing (Antcliff et al., 2012; Birkholtz et al., 2004) based on a clear theoretical concept and considerations of the context in which the behaviour occurs (Nielson et al., 2001). This would allow activity pacing studies to be replicated, providing clarity on optimising the effectiveness of activity pacing interventions in the future.

Given that different activity profiles (underactivity, overactivity and uneven spread of activity) exist between patients, an individualised approach to activity pacing should be considered in future interventional studies. Thus persons with disabling conditions associated with high fatigue may need to be advised differently constructed on their activity profile. This type of tailored-activity pacing techniques appear warranted to manage fatigue and stimulate physically active lifestyle, to improve health and increase participation of patients.

Although studies support the efficiency of self-paced exercise in sports (Edwards and Polman, 2012; Edwards et al., 2011; Lander et al., 2009), little remains known about how persons with disabling conditions naturally pace and plan multiple activities across a day and how this relates to fatigue, quality of life and physical activity in the context of their lifestyle. Further research that investigates the nature of pacing in persons with disabling conditions is warranted. Insight into this will contribute to better understanding and explain the current considerable variation in response to activity pacing. Additionally, this will help tailor, adapt and optimise activity pacing interventions to make this more effective and efficient.

There is also a need for further evidence-based validity studies of current measures of activity pacing. A number of measures of activity pacing are recent and have undergone limited validity testing (Antcliff et al., 2015; McCracken and Samuel, 2007). Given the variance in definition and implementation across studies, there may be a need to develop new measures or refine existing ones. For example, it may be worthwhile to develop a measure that detects risk of overactivity and underactivity as dimensions of pacing behaviour. This may offer valuable insights into how to tailor activity pacing interventions to help persons with disabling conditions remain or become physically active (Plotnikoff et al., 2013).

Conclusion

Physical inactivity and premature, debilitating fatigue sensations are often reported in persons with disabling conditions and are associated with deconditioning and disability. A physically active lifestyle is of utmost importance to improve quality of life and participation in daily life in persons with disabling conditions. Activity pacing could be a novel, useful adaptive strategy to

stimulate a physically active lifestyle in persons with disabling conditions. However, most studies on activity pacing have thus far focused on symptom reduction and curtailing over activity. Empirical work is now required to explore this strategy and this review may be the catalyst to stimulate future work.

Considering that both underactivity and overactivity are linked to disability, it is necessary to adopt an individualised approach to activity pacing intervention to provide extra and optimal guidance and support for those with high fatigue complaints. Given the efficacy of self-pacing in sports, there is a need for further exploratory studies on the use of naturalistic pacing in persons with disabling conditions within the context of daily life. Additionally, encouraging persons with disabling conditions to learn to 'listen' to their symptoms and develop a performance template based on previous experience in pursuit of optimal performance may be an efficient way to manage fatigue and stimulate an active lifestyle. This could further improve the effectiveness of activity pacing intervention.

The current limited evidence on activity pacing calls for closer inspection of the dimensionality of pacing as it is currently operationalized and its relations to physical activity and fatigue in daily life. Future research on activity pacing and physical behaviour will be welcome to fully understand the link between activity pacing and disability. This will play a key role in the management of disabling conditions and fight the growing incidence of physical inactivity in persons with disabling conditions.

Conflict of Interest

Abonie S. Ulric, Edwards M. Andrew and Hettinga J. Florentina declare that they have no conflict of interest.

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