**Introduction**

Manual wheelchair users rely on their upper limbs for all activities of daily living, such as wheelchair propulsion. Yet, shoulder pain is common and severe pain may lead to loss of independence and quality of life (Finley & Rodgers., 2004). Despite mild or moderate shoulder pain many wheelchair users continue to independently propel their wheelchairs and engage in physical activity (Alm et al., 2008; Briley et al., 2020b; Finley & Rodgers., 2004; Samuelsson et al., 2004). Therefore, understanding the interplay between shoulder pain and wheelchair propulsion biomechanics is of clinical importance but remains surprisingly unclear. Previous studies have identified an association between shoulder pain and wheelchair propulsion parameters such as greater peak magnitude, rate of rise and jerk of push rim forces and lower kinetic and scapular kinematic variability (Beirens et al., 2020; Briley et al., 2020b; Dysterheft et al., 2017; Rice et al., 2014). However, these findings are based on cross-sectional studies; as a result, the time-varying relationship between shoulder pain symptoms and wheelchair propulsion biomechanics is currently undetermined. Hence the need for longitudinal investigations.

Several studies have examined longitudinal changes in shoulder pain and explored factors that may be associated with pain (Eriks-Hoogland et al., 2014; Mulroy et al., 2015; Walford et al., 2019). This work has primarily investigated factors such as muscle strength and joint range of motion during early manual wheelchair use.  To date, only Walford et al. (2019) have examined shoulder pain in relation to wheelchair propulsion biomechanics in a large cohort of 102 individuals with paraplegia. They identified that wheelchair users who developed shoulder pain, over 18 or 36 months, displayed greater internal shoulder rotation, lower trunk flexion, and larger contact angle variability at baseline compared to those that remained pain-free (Walford et al., 2019). Previous studies have only explored shoulder pain in relation to baseline factors; thus, only potential predictors of pain have been examined (Mulroy et al., 2015; Walford et al., 2019). Consequently, the association between within-subject changes in shoulder pain and alterations in wheelchair propulsion biomechanics in response to pain has yet to be established.

Recent evidence suggests that wheelchair propulsion biomechanics, during fixed propulsion speeds and average power output, is adaptable to a variety of short-term interventions and acute fatigue (Bossuyt et al., 2020; Leving et al., 2016). To date, the short-term wheelchair propulsion biomechanical modifications that correspond with worsening shoulder pain have not been investigated. From a theoretical perspective the short-term motor response to pain is one of protecting the painful or threatened body part during movement tasks that may provoke pain (Hodges et al., 2015; Merkle et al., 2018). However, the specific biomechanical adaptations to pain vary, based on factors such as pain location and the constraints of the task (van Dieën et al., 2003). As a result, it may be expected that under fixed propulsion conditions worsening shoulder pain may coincide with certain changes in wheelchair propulsion biomechanics that may protect the shoulder. That said, the specific biomechanical alterations are unknown. Subsequently, the purpose of this study was to investigate the longitudinal association between within-subject changes in shoulder pain and alterations in wheelchair propulsion biomechanics in manual wheelchair users. Specific research objectives were to: i) quantify the longitudinal changes in shoulder pain in manual wheelchair users, ii) investigate whether changes in shoulder pain correlated with changes in wheelchair propulsion biomechanics, and iii) to examine whether individuals with increased shoulder pain and those that did not change/reduced pain altered propulsion biomechanics differently. Based on the points raised above, it was hypothesised that individuals with increased shoulder pain would display longitudinal reductions in range of motion of the shoulder during wheelchair propulsion to protect the shoulder.

**Methods**

*Participants*

Eighteen manual wheelchair users (13 men, 5 women; age = 33 ± 11 years; body mass = 72.2 ± 11.8 kg; duration of wheelchair use = 13 ± 11 years) provided written informed consent and participated in this study. All participants met the following inclusion criteria: full-time manual wheelchair user, aged 18-55 years. Participants were a combination of athletic and nonathletic manual wheelchair users following our previous work that reported no biomechanical differences in daily wheelchair propulsion between these populations (Briley et al., 2020a). Participants primarily resided in the local community and were recruited through direct contacts, previous study participation and advertisements. Participants primary impairments were spinal cord injury (SCI) C6 or below, spina bifida, and cerebral palsy. Exclusion criteria were a history of shoulder surgery and major trauma to the upper extremity in the previous year. Ethical approval was obtained through the University’s local ethics committee.

*Shoulder pain*

Shoulder pain over the previous seven days was evaluated using the Performance-Corrected Wheelchair User Shoulder Pain Index (PC-WUSPI) (Curtis et al., 1999). The PC-WUSPI uses a 10 cm visual analogue scale (VAS) to quantify shoulder pain experienced during 15 activities of daily living. Total scores for the PC-WUSPI range from 0 (no pain) to 150 (highest degree of pain). The severity of shoulder pain was classified following PC-WUSPI thresholds described in Briley et al. (2020b). Specifically, a PC-WUSPI score of ≤ 51 was classified as no or mild pain, between 52.5 and 111 moderate pain, and >112.5 severe pain. A modified upper extremity pain questionnaire (PSQ) was used as an auxiliary questionnaire to the PC-WUSPI to report the location (right/left), frequency, and severity of shoulder pain (van Drongelen et al., 2006).

*Physical activity*

Physical activity was quantified via the Leisure Time Physical Activity Questionnaire for people with Spinal Cord Injury (LTPAQ-SCI) (Martin Ginis et al, 2007). The LTPAQ-SCI is a brief (~ 5 minutes) self-administered questionnaire that reports the total duration of mild, moderate, and heavy intensity physical activity over the previous seven days (Martin Ginis et al., 2012). Total physical activity was calculated from the combined duration (number of days x average duration of activity) of all physical activity intensities.

INSERT FIGURE 1 HERE

*Experimental protocol*

Participants completed two laboratory visits (T1 and T2) between 4 and 6 months apart (Figure 1). The study duration is comparable to the time frame used in wheelchair user-specific shoulder pain intervention studies and provided adequate time for changes in shoulder pain to occur (Curtis et al., 1999; Mason et al., 2020; Nawoczenski et al 2006). Physical characteristics (age, body mass, sex, primary impairment, years of wheelchair use) were collected. Wheelchair configuration of participants own daily living wheelchair were assessed. Participant’s wheelchair characteristics were chair mass 12.4 ± 1.4 kg; wheel diameter 0.60 ± 0.01 m; rim diameter 0.55 ± 0.01 m and wheelbase 0.55 ± 0.03 m. No changes in wheelchair configuration occurred between laboratory visits.

Participants were tested in their own daily living wheelchair on a dual roller wheelchair ergometer (Lode Esseda, m988900, Groningen, Netherlands). A five-minute warm-up was performed which involved wheelchair propulsion at a self-selected speed and dynamic stretching. Followed by a three-minute wheelchair propulsion trial at 1.11m.s-1 (Figure 1; Mason et al, 2014). At the end of the trial participants reported their Rating of Perceived Exertion (RPE) using a Borg scale (Borg, 1982), which ranges from 6 (no perceived exertion) to 20 (maximal exertion). A Vicon motion capture system (Vicon, Motion Systems Ltd. Oxford, UK) consisting of 10 cameras (MX T40-S) acquired kinematic data during wheelchair propulsion at 200 Hz. Eighteen retroreflective markers (B&L Engineering, California, USA) were attached to anatomical landmarks of both upper limbs and the torso in accordance with the International Society of Biomechanics (ISB) recommendations (Wu et al., 2005). Scapular orientation during wheelchair propulsion was tracked using Acromion marker clusters (AMC), as described by Warner et al. (2015). Glenohumeral joint centres (GHJC) were determined using the Symmetrical Centre of Rotation Estimation (SCoRE) method from a bilateral circumduction trial (Ehrig et al., 2006).

To monitor shoulder pain and physical activity between laboratory visits, participants completed the PSQ and LTPAQ-SCI at four-week intervals (Figure 1). Both questionnaires were sent to participants via email.  The PSQ was used to quantify shoulder pain between visits rather than the PC-WUSPI as it was easier to administer and complete during the study period. This monitoring provided an opportunity to maintain regular contact with all participants to maximise study adherence and to identify any sudden change in shoulder pain or physical activity status.

*Data analysis*

Biomechanical data processing and analyses were conducted using custom written MATLAB scripts (Matlab R2017a, The Mathworks Inc, Natick MA, USA). To ensure steady-state propulsion, biomechanical parameters were calculated from the final 60 seconds of the propulsion trial. The following spatio-temporal and kinetic variables were calculated from the ergometer data: stroke frequency, contact angle, contact angle coefficient of variation (CV), peak torque, peak torque coefficient of variation (CV) and work done per push (Briley et al., 2020a, Goosey-Tolfrey et al., 2018). A fourth-order, low-pass Butterworth filter with a cut-off frequency of 6-Hz was applied to the marker trajectories (Morrow et al., 2011). Euler angles were calculated for thorax (thorax to global), scapulothoracic (scapula to thorax), and glenohumeral (humerus to scapula) motion (Kontaxis et al., 2009, Wu et al., 2005). Peak angles, range of motion (ROM), and standard deviation were extracted from 20 consecutive propulsion cycles. Peak angles were for thorax flexion, glenohumeral flexion, abduction and internal rotation and scapulothoracic internal rotation, downward rotation, and anterior tilt. These peak angles were selected as they represent directions of motion assumed to impose stress on the subacromial tissue of the shoulder (Mozingo et al., 2020; Zhao et al., 2015). Joint kinematic variability for each joint angle was calculated from the mean of the standard deviation of each joint angle (Srinivasan & Mathiassen, 2012). For participants with unilateral shoulder pain the painful side was analysed and for those with bilateral pain the most painful side at baseline was analysed.

*Statistical analysis*

The Statistical Package for Social Sciences (SPSS Version 23, IBM, New York, USA) was used to perform all statistical analyses. Data normality was assessed by Shapiro-Wilk tests. Paired samples t-tests determined if differences in shoulder pain (PC-WUSPI scores), body mass, and total physical activity between laboratory visits were statistically significant (α=0.05). A one-way independent analysis of variance (ANOVA) was used to assess the effect of impairment type on change in PC-WUSPI scores. Pearson product-moment correlations quantified the relationship between within-subject changes in shoulder pain (PC-WUSPI scores) and within-subject changes in body mass, physical activity, and biomechanical parameters of wheelchair propulsion. Changes in all variables were calculated as the value at T2 subtracted by the value at T1. Correlations determined the relationship between shoulder pain according to PSQ and physical activity reported during each visit at each interval between visits.

Separate two-way mixed-model analysis of variance were used to determine main effects for time (T1, T2), group (increased pain, no change in pain), and a time\*group interaction for each outcome variable. Participants were retrospectively categorised as having increased shoulder pain, no change or reduced shoulder pain symptoms using the minimal detectable change (MDC) for the PC-WUSPI of 5.1 points (Curtis et al., 1995; Curtis et al., 1999). Participants who reported an increase in PC-WUSPI scores between visits ≥ the MDC were classified with increased shoulder pain, a change < the MDC or a decrease > the MDC were classified with no change/reduced shoulder pain. Data normality, homogeneity of variance, and sphericity were assessed by Shapiro-Wilk tests, Levene’s test, and Mauchly’s test of sphericity, respectively. Differences in how each group altered propulsion biomechanics parameters over time were identified as significant (time\*group) interactions from the two-way mixed ANOVA. The alpha level was set at P < 0.05. For parameters that had a significant interaction effect post-hoc t-tests were performed for each participant group to establish where differences occurred. This enabled the change in propulsion biomechanics to be evaluated for each group separately via paired t-tests. Independent t-tests examined group differences at each time point. A Bonferonni correction was applied to correct for multiple testing (α = 0.05/4) and effect sizes (Cohen, 1988) were calculated.

**Results**

*Longitudinal changes in shoulder pain*

Of the 18 participants, 16 (89 %) were categorised as having no/mild shoulder pain and two (11 %) moderate shoulder pain at T1. Between laboratory visits participants mean PC-WUSPI scores increased by 5.4 points (P = 0.03, 95 % CI = 0.74 to 10.1). Nine (50 %) participants were classified as having increased shoulder pain, seven (39 %) no change in shoulder pain and two (11 %) decreased shoulder pain (Figure 2). The mean PSQ trace for the increased pain group demonstrated that these individuals gradually increased pain between visits (Supplemental Figure 1). No significant main effect for impairment type on change in shoulder pain (F (3,14) = 0.264, P = 0.850) was observed. Additionally, both pain groups (increased pain and no change in pain) were composed of a mixture of impairment types.

INSERT FIGURE 2 HERE

No significant difference between laboratory visits was observed in either body mass (T1 = 72.2 ± 11.8 kg vs T2 = 72.5 ± 11.1 kg, P = 0.66) or total physical activity (T1 = 699 ± 453 mins vs T2 = 733 ± 417 mins, P = 0.211).  Change in shoulder pain was not significantly correlated with change in body mass (R = -0.02, P = 0.472) or total physical activity (R = -0.01, R = 0.964). No correlation was observed between shoulder pain (PSQ) and total physical activity at any time point between laboratory visits (Supplemental Table 1).

*Correlations with propulsion biomechanical changes*

Significant correlations were observed between within-subject changes in shoulder pain and alterations in eleven wheelchair propulsion biomechanics parameters (Table 1). Overall, increasing shoulder pain severity was associated with increased contact angle, decreased peak torque, and increased peak torque CV. Increasing shoulder pain was also significantly correlated with increased thorax ROM, decreased glenohumeral flexion ROM, decreased peak glenohumeral abduction, and increased glenohumeral abduction inter-cycle variability. Also, increasing shoulder pain was associated with decreased scapular internal/external rotation ROM, decreased scapular down/upward rotation ROM, decreased peak scapular downward rotation, and increased scapular anterior/posterior tilt variability.

INSERT TABLE 1 HERE

*Comparison between increased pain and no-change/reduced pain*

The group comparison identified significant interaction (time \* group) effects for ten of the eleven propulsion biomechanics parameters (Figure 3). The significant interaction effects indicated that the alteration in the wheelchair propulsion biomechanics parameters over time differed between pain groups (increased pain, no change in pain). Pairwise comparisons revealed that the increased shoulder pain group significantly increased contact angle (P < 0.001; 95 % CI 6.0 to 11.6; ES = 2.39), reduced peak torque (P = 0.001, 95 % CI -1.9 to -0.77; ES = 1.79) and increased peak torque CV (P < 0.001; 95 % CI 3.8 to 8.3; ES = 2.06) between T1 and T2. Whereas no significant change in either contact angle, peak torque, or peak torque CV was observed in the no change in pain group. The increased shoulder pain group exhibited significantly greater thorax flexion/extension ROM (P = 0.004; 95 % CI 0.8 to 2.8; ES = 1.35), and significant decreases in scapular peak downward rotation (P = 0.009; 95 % CI -2.5 to -0.5; ES = 1.15) and down/upward rotation ROM (P = 0.003; 95 % CI -1.6 to -0.4; ES = 1.36) between visits. Due to the use of Bonferonni corrections, the increased shoulder pain group displayed non-significant decreases in glenohumeral peak abduction (P = 0.013; 95 % CI -3.2 to -0.5; ES = 1.06) and scapular int/external rotation ROM (P = 0.023; 95 % CI -2.8 to -0.3; ES = 0.94). In terms of kinematic variability, the increased pain group displayed significantly increased glenohumeral abduction variability (P = 0.006; 95 % CI 0.2 to 1.0; ES = 1.25) and scapular ant/posterior tilt variability (P = 0.011; 95 % CI 0.1 to 0.7; ES = 1.10). In contrast, the no-change group displayed no differences between visits in any kinematic or kinematic variability parameters. Finally, the increased shoulder pain group displayed significantly lower peak torque CV (P = 0.008; 95 % CI -11.5 to -2.1; ES = 1.44) and greater glenohumeral peak abduction angle (P = 0.010; 95 % CI 2.4 to 15.1; ES = 1.37) at T1 compared to the no change in pain group.

INSERT FIGURE 3 HERE

**Discussion**

This study examined the longitudinal association between within-subject changes in shoulder pain and alterations in wheelchair propulsion biomechanics over 4 – 6 months in a sample of manual wheelchair users. Overall increases in shoulder pain symptoms correlated with increased contact angle, thorax ROM and movement variability but reduced motion at the shoulder during wheelchair propulsion. In support of the study hypothesis these biomechanical alterations were displayed by participants who experienced increased shoulder pain, whereas propulsion biomechanics of those with no change/decreased pain remained unaltered.

Individual changes in shoulder pain between laboratory visits varied widely, ranging from -13.5 to + 20.9 PC-WUSPI points. Of the eighteen participants, nine (50 %) reported shoulder pain increases above the MDC for the PC-WUSPI. Despite this, all participants were able to propel their wheelchair at a functional speed and maintained current physical activity levels. Furthermore, no correlation between physical activity levels and shoulder pain existed at any time point between laboratory visits. These findings support previous studies that report a similar frequency of wheelchair use and physical activity in those with and without shoulder pain (Alm et al., 2008; Mulroy et al., 2011).

It was revealed that wheelchair users who increased shoulder pain transitioned towards propelling using larger contact angles, increased thorax ROM and reduced peak torque and peak scapular downward rotation and ROM. A larger contact angle extends the distance forces can be distributed over, thereby reducing the magnitude of peak forces (Boninger et al., 2005). This alteration has been demonstrated as a favourable adaptation in previous motor learning and shoulder pain intervention studies (Boninger et al., 2005; Rice et al., 2009). It is generally accepted that even small but sustained reductions in peak force at a given propulsion velocity may be beneficial in terms of reducing further shoulder pain (Cowen et al., 2008). Similarly, the joint kinematic alterations exhibited by those with increased shoulder pain are likely linked to the increases in push rim contact angle. To produce larger contact angles individuals must increase the ROM at the shoulder and/or the trunk. The current findings indicate that experienced wheelchair users with worsening shoulder pain produce larger contact angles by increasing motion of the thorax but constrain the range and specific orientations of the shoulder that may impose mechanical stress on tissues within the shoulder (Mozingo et al., 2020; Zhao et al., 2015). Generally, these findings support the protective response theory which proposes that during tasks that may provoke painful symptoms the nervous system searches for movement patterns that are less painful by constraining motion at the painful joint/area (Hodges et al., 2011; 2015). Furthermore, this altered movement may mitigate acute shoulder pain symptoms during wheelchair propulsion but also reduce pain over a short-term period (Hodges et al., 2011; Merkle et al., 2018). However, the long-term consequences on shoulder pain symptoms are unclear.

It is important to note that large inter-individual biomechanical differences were present in the increased pain group. It is well recognised that biomechanical alterations to pain are not uniform and that the nervous system possesses a range of options to achieve the movement task outcomes (Hodges et al., 2015; van Dieën et al., 2003). A possible explanation for these biomechanical differences may be the varied nature of impairments possessed by participants in this study. Indeed, further work is needed to understand the adaptations to worsening shoulder pain in wheelchair users with poor trunk control, such as those with higher-level spinal cord injury, as the kinematic alterations observed in the current study may not be possible in these individuals.

Regarding inter-cycle variability, wheelchair users with increased shoulder pain displayed a concomitant increase in peak torque, glenohumeral abduction, and scapular posterior tilt variability over time. Previous studies of movement variability during wheelchair propulsion have employed cross-sectional or prospective designs (Briley et al., 2020b; Rice et al., 2014; Walford et al., 2019). The current study agrees with previous research that has observed increased kinetic and kinematic variability in response to acute or short-term pain (Côté, & Bement., 2010). From a theoretical perspective greater variation in forces and movements allows the repeated stress imposed by wheelchair propulsion to be distributed more widely thereby reducing the risk of overuse injury (Côté, & Bement., 2010; Rice et al., 2014). Therefore, greater kinetic and joint kinematic variability may be another aspect of the wider short-term strategy demonstrated by wheelchair users with increased shoulder pain to protect the shoulder. However, interpretation of these biomechanical alterations should be made carefully as changes in these biomechanical parameters may have contributed to worsening shoulder pain symptoms.

Finally, only two biomechanical parameters differed between wheelchair users with increased pain and those with no change in pain during the first visit. Specifically, those with increased shoulder pain exhibited lower peak torque variability and greater peak glenohumeral abduction at T1 compared to the no-change group. It is postulated that greater peak glenohumeral abduction applies mechanical stress to subacromial tissue of the shoulder and lower peak torque variability imposes a more uniform distribution of stress on the tissue (Hamill et al., 2012; Mozingo et al., 2020). Thus, the combination of both biomechanical differences preceding the shoulder pain increase indicates greater cumulative tissue fatigue which may lead to shoulder pain development (van Drongelen et al., 2005). However, as participants were not pain-free at the start of this study these differences may reflect an adaptive strategy to the development of pain. The lack of biomechanical differences prior to increases in shoulder pain indicates that most changes in wheelchair propulsion biomechanics were not the result of detrimental propulsion biomechanics before the pain increased. Additionally, some participants in the no pain/decreased pain group had greater pain at both T1 and T2 than some participants in the increased pain group at T1 and T2. Therefore, while alterations in biomechanical parameters were related to increases in pain in this study there does not appear to be an association between biomechanical parameters and magnitude/severity of the pain. Future work should focus on understanding the short-term and long-term alterations in wheelchair propulsion biomechanics in response to important constraints including shoulder pain, physical activity, and wheelchair configuration.

Limitations

This study had many unique features, yet it was notable that our study duration, which occurred within 4 - 6 months to suit the participants, was shorter than previous longitudinal studies (Eriks-Hoogland et al., 2014; Mulroy et al., 2015). That said, while it is unclear whether the biomechanical changes in this study were temporary or long-term, having observed biomechanical adaptations in this short timespan was promising. The varied duration between visits was decided due to the practicalities of participants visiting the laboratory. Additionally, it should be noted that sample size was smaller than that of previous longitudinal studies of wheelchair users. Grouping participants according to change in PC-WUSPI scores was carefully considered. Since the focus of the study was primarily on wheelchair users with increased shoulder pain the two individuals with reduced shoulder pain were included in the no change in shoulder pain group. The experimental design used a prescribed speed that participants maintained on an ergometer that reflected daily propulsion (Cowan et al., 2008). Future work could use self-selected speeds and assess over-ground propulsion. As an instrumented wheel was not used in this study it was not possible to quantify alterations to either the push-rim force vector or joint kinetics of the shoulder. As these biomechanical parameters have been linked to both shoulder pathology and pain (Walford et al., 2019) authors should consider utilising instrumented wheels in future longitudinal investigations.

**Conclusions**

The current study revealed that longitudinal increases in shoulder pain were associated with alterations in wheelchair propulsion biomechanics. Wheelchair users with worsening shoulder pain displayed greater contact angle, thorax ROM and movement variability but reduced peak torque, shoulder ROM and peak angles over time. These biomechanical changes are interrelated and suggest that wheelchair users with increased shoulder pain symptoms maintain short-term functional independence by modifying how they propel their wheelchair. Generally, the short-term wheelchair propulsion biomechanical response to increases in shoulder pain symptoms appears to be protective but the long-term consequences remain unclear.

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