Gait & Posture 72 (2019) 234–238

Contents lists available at ScienceDirect

Gait & Posture

journal homepage: www.elsevier.com/locate/gaitpost

Full length article
GCMAS Best Paper 2018

Factors influencing post-surgical variability in StepWatch data in youth with cerebral palsy

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ARTICLE INFO

Keywords:
Activity monitor
Cerebral palsy
Step count variability
Surgical burden
Bootstrapped regression

ABSTRACT

Background: Over the past several years, activity monitors have become very popular in the general population, and due to their low cost and ease of use, are starting to be seen as clinical tools for the assessment of interventions. This presents researchers with the opportunity to better understand how activity, or lack thereof, is related to the recovery of patients. However, even in individuals without disabilities, there is a high degree of variability in activity monitor data which must be better understood in order to produce clinically meaningful interpretation of such data.
Research question: What sources of variability contribute the most to the daily scatter in activity data as measured by StepWatches in youth with Cerebral Palsy (CP)? In particular, do non-clinical factors such as weather and location contribute to this variability significantly?
Methods: This was a retrospective study making use of data from our activity monitoring protocol of youths with CP who obtain single event multi-level surgeries. Before and after these surgeries, 57 such youths aged 4.2–21.3 years were issued StepWatches to monitor daily activity for 8 day periods over 24 months duration. Weather data and walk scores for the patients' home locations were collected from online databases. Steps per hour were predicted from clinical and environmental data using bootstrapped regression to determine the stability of regression coefficients and the percent variability explained by each variable.
Results: Time since surgery, age, season, GMFCS level, and surgical burden were significant variables in the model. Of them, GMFCS level was most important and explained nearly 16% of the variability in the data. Temperature, precipitation, and walk score had small effects on step count variance.
Significance: Understanding sources of variability in step-counts is important if such a measure is to be used as a clinical measure of recovery, and may be important in the consideration of future surgical planning.

1. Introduction

The current ubiquity of wearable accelerometers has presented clinicians with the opportunity to monitor patient outcomes in novel ways. In particular, using such devices enables clinical teams to determine patients' recovery of gait function after surgical interventions based on the number of steps walked. The working hypothesis is that as patients improve post-surgically, they will walk more so that step-counts can serve as a proxy for recovery [1]. Unfortunately, step-count data is quite variable even in the best of cases [1–3], and the authors' personal experiences with the devices leads to the conclusion that such data must be scrutinized more closely to find true trends in the noise.

Some of the observed variability in step-counts is thought to be predictable. For example, in healthy individuals, a greater number of steps are taken during weekdays than on weekends [3]. Likewise, some populations of school aged children walk more during the school year than during the summer break [4]. In children with cerebral palsy (CP), it has been found that GMFCS level is inversely proportional to step-count [5].

However, there exist other possible sources of step-count variability which may be important in explaining individual variability in the youth with CP: surgical burden, where patients live, and the daily weather. In children recovering from single event multilevel surgery (SEMLS), step-counts vary considerably among individuals in the same GMFCS level and age [6]. Location may also play a role in the ability to maintain a healthy step-count. A recent large study has shown that the

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https://doi.org/10.1016/j.gaitpost.2019.06.017

walkability of a neighborhood is predictive of health outcomes in individuals without disabilities [7]. A walk score is an index that measures the ease with which an individual can walk to various amenities, the pedestrian friendliness based on traffic, sidewalks, and population [8]. Therefore, it is possible that in neighborhoods with higher walk scores, there will be an increase in the number of steps, on average, than in neighborhoods with lower walk scores. Similarly, one might presume that patients are likely to walk more in better weather. Weather effects have been shown to be important in physical activity levels of children [9] although this seems to be location or population specific [10]. Using bootstrapped multiple linear regression, this study seeks to determine the factors that influence variability and number of step-counts including clinical factors such as time since surgery, GMFCS level, age, and surgical burden, as well as considerations such as day of the week, season, walk score, and weather.

2. Methods

IRB approval was obtained to study retrospective data of 57 patients who underwent surgery for the treatment of CP (Age 4.2–21.3 years, mean = 12.2, sd = 4.3). As part of the clinical protocol, patients were assigned StepWatch devices just prior to their surgeries and again at three month intervals post surgically out to 24 months. Each monitoring period was eight days in length. Patients were included if they had pre-surgical step monitoring and at least three post-surgical monitoring periods. Based on gait analysis recommendation, patients underwent various surgeries and were assigned to groups based on the surgical burden (Table 1). The final dataset included 2013 patient days of data. Patient address data were used along with the R packages “ggmap” [11] and “rnoaa” [12] to retrieve temperature and precipitation data, and the R package “walkscoreAPI” [13] to retrieve walk scores for patients' neighborhoods. R packages are computer code published on the Comprehensive R Archive Network (CRAN) and which are often described in detail in the R Journal [14].

A bootstrapped multiple linear regression written in R was conducted to determine how various independent variables related to the number of steps taken per hour. While total steps per day was considered as a final outcome variable, since the time per day the StepWatch was worn was known, the normalized step-count (steps per hour) was considered to be a better indicator of functionality because there tended to be less variability in this measure. Both continuous (A_i coefficients) and categorical (B_i coefficients) dependent variables were considered in the regression analysis as per the regression equation:

Table 1
Surgical burden categories listed by procedure.

Surgical Burden	Description
1 – lowest	Unilateral, Soft Tissue Only
2 – low	Bilateral, Soft Tissue Only
3 – moderate	Unilateral, Bony Surgery (1 procedure)
4 – high	Bilateral, Bony Surgery (1 procedure) -OR- Unilateral, Bony Surgery (2-3 procedures)
5 – highest	Bilateral, Bony Surgery (2-3 procedures) -OR- Unilateral, Bony Surgery (≥ 4 procedures)

Table 2
List of independent continuous and categorical variables in the model. The dependent variable, Nstep, was the step-count normalized by time, or steps per hour.

Continuous Variable	Categorical Variable
Days.Since.Surgery	Day (Day of the week)
walkScore	GMFCS (levels I-III)
tmax (Maximum daily temperature)	Season
prcp (Daily precipitation)	SurgicalBurden (5 levels)
Age	

$$\begin{aligned} NStep = {} & A_0 + A_1 Days.\ Since.\ Surgery + A_2 walkScore + A_3 tmax \\ & + A_4 prcp + A_5 Age \\ & + B_1 DayTuesday + B_2 DayWednesday + B_4 DayThursday + B_5 DayFriday \\ & + B_6 DaySaturday + B_7 DaySunday + B_8 GMFCSII + B_9 GMFCSIII \\ & + B_{10} SeasonSpring + B_{11} SeasonSummer + B_{12} SeasonFall \\ & + B_{13} SurgicalBurden2 \\ & + B_{14} SurgicalBurden3 + B_{15} SurgicalBurden4 + B_{16} SurgicalBurden5 \end{aligned} \quad (1)$$

and prior to the analysis, continuous variables were checked via a correlation matrix to ensure that there was low multicollinearity. A summary of variables is shown in Table 2. Since the magnitude of the maximum correlation coefficient in this matrix was small (0.13), and related variables, age and maximum temperature, were conceptually orthogonal, it was deemed that collinearity of independent variables was not a problem, and all listed variables were included in the regression model. The purpose of the regression was to identify 1) the direction of the influence of the variable on the predicted number of steps (positive or negative), 2) the strength of influence of the variable on the number of steps, 3) the percent variability in the model explained by the variables, and 4) the relative significance of each variable.

Bootstrapping was selected as the method of choice to identify important variables since results of backwards and forward regression strategies can give different answers dependent on the order of variable entry [15]. The actual bootstrapping was conducted by sampling the data with replacement and then running the regression 1000 times, and the alpha level for significance was set to 0.05. The naive bootstrapping method used here is relatively common and was described in detail in Efron & Tibshirani [16]. While k-fold cross-validation is sometimes suggested as having a lower bias, it can produce highly variable results [17]. Increasing the number of bootstraps can decrease bias, but this depends on the size of the sample population and its skew. Bootstrapping can also provide less variable, more stable estimates of parameters [17]. The resultant standardized regression coefficients, percent variabilities, and p-values of each independent variable were stored and then graphed. Box and whisker plots are shown for all predicted variables indicating their range of values as well as range of significance.

3. Results

In bootstrapped regression, variables whose standardized regression coefficients changed little regardless of the data used in the model while also having a stable percent variability and p-value are more likely to show a real effect. Fig. 1 shows bootstrap results by way of a) box plots of the standardized beta coefficients, b) box plots of significance values for each variable, and c) box plots of variability explained. Based on this, regression results indicated the existence of a number of significant variables that were stable across the bootstrap. In multiple linear regression involving categorical variables such as day of the week or season, significance is measured with respect to the first category in the variable. For example, all days of the week are compared to Monday,

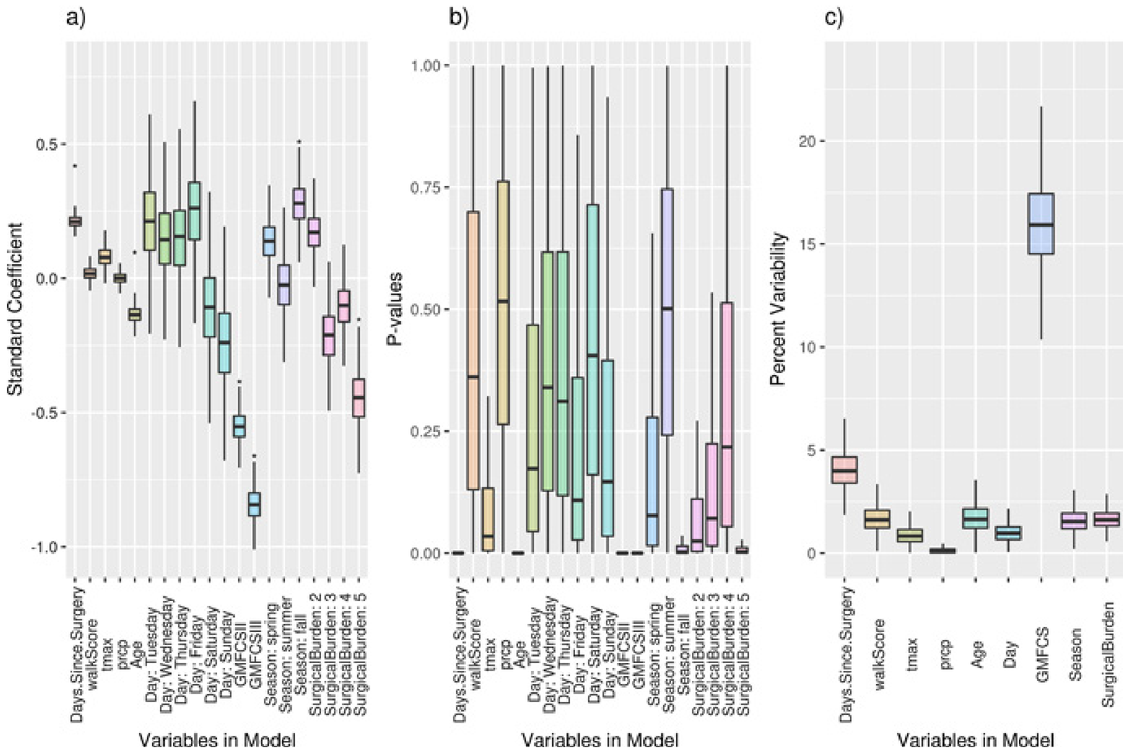

Fig. 1. Results of bootstrap regression in boxplot format (adjusted $R^2 = 0.269$, p = 2.2e-16) to predict steps per hour. a) Standard coefficients (beta) shown with interquartile ranges (box and whiskers) and median as a horizontal line. Mean beta and standard deviation are shown by variable. Where a variable was significant, an asterix (*) is shown above the whisker. b) Ranges of p-values for regression coefficients. Where there is little variability, there is more confidence in the result. c) Ranges of percent variability for each variable as obtained via ANOVA. Colors are redundant with x-axis tick labels are are there more easily distinguish adjacent box plots.

Table 3
Mean (standard deviation) regression coefficients for bootstrapped regression. For continuous variables, the meaning of the coefficients is straight-forward. A ten day increment in Days.Since.Surgery results in an elevation of hourly step-count by 12. For categorical variables, the coefficient indicates the elevation in number of steps if that category is true. For example, if it is Tuesday, subjects took, on average 17.11 steps more than subjects did on Monday, and none of the other day of the week variables contribute. Likewise, subjects with GMFCS level III took 245.74 fewer steps per hour than did GMFCS level I subjects. Significance is listed on the right with codes "." $P < 0.1$, "*" $P < 0.05$, "**" $P < 0.01$, "***" $P < 0.001$.

Variable	Mean (coef)	Std (coef)	P-value	Pr (> |t|)
(Intercept)	351.63	29.95	< 2.0e-16	***
Days.Since.Surgery	0.12	0.01	8.65e-15	***
walkScore	0.15	0.19	0.41	
tmax	1.09	0.54	0.14	
prcp	0.07	0.43	0.55	
Age	−4.46	1.06	0.0011	**
DayTuesday	17.11	10.7	0.25	
DayWednesday	10.75	12.83	0.39	
DayThursday	11.81	13.2	0.36	
DayFriday	18.43	12.32	0.25	
DaySaturday	−9.92	12.48	0.42	
DaySunday	−18.57	13.43	0.27	
GMFCSII	−160.91	16.11	2.55e-27	***
GMFCSIII	−245.74	17.73	1.84e-38	***
SeasonSpring	20.41	11.42	0.19	
SeasonSummer	−2.66	15.32	0.51	
SeasonFall	40.6	12.26	0.018	*
SurgicalBurden2	21.11	10.57	0.14	
SurgicalBurden3	−28.33	12.64	0.14	
SurgicalBurden3	−12.74	11.93	0.29	
SurgicalBurden4	−57.36	13.95	0.012	*

and all seasons are compared to fall, but neither Monday nor fall appear in the equation. Regression coefficients of categorical variables are to be taken as the amount that the step-count increases, on average, if conditions fit that category (see Table 3 caption).

Table 3, which shows the non-standardized regression coefficients, indicates that there were generally more weekday steps than weekend steps, though not significantly so. The standardized regression coefficients shown in Fig. 1a reflect the same trend. Days Tuesday through Friday all have positive betas, but for Saturday and Sunday, these values are negative. About one percent of the variability in the data was accounted for by day of the week. Seasonal results accounted for 1.67% of the variability and reflect that although more steps were taken in the spring (p = 0.19) and fall (p = 0.018) than in the winter (the base value for comparison), there were only slightly more summer steps (p = 0.51) than in the winter (Fig. 2a). Both higher GMFCS levels (Fig. 2b) and surgical burdens predicted significantly fewer steps (Fig. 2c). However, significantly decreased step-counts compared to the first surgical group were observed only for surgical group five. GMFCS level accounted for the majority (15.96%) of the data's variability, while surgical burden accounted for substantially less (1.68%).

Of the continuous variables in the model, the length of time since surgery was significant in predicting the normalized step-count (p = 8.65e-15) as was the subject age (p = 0.0011) (Table 3). These accounted for 4.09% and 1.65% of the variability of the data, respectively (Fig. 1c). While not significant according to our criteria, the maximum daytime temperature was significant at the 0.05 level almost half the time despite the average p-value of 0.15, was positively correlated to normalized step-count, and accounted for 0.88% of the variability (Fig. 1c). Walk score, precipitation, and age were not significant in the analysis, and these accounted for 1.74%, 0.12%, and 0.27% of the variability, respectively (Fig. 1c). Other than days since surgery, the continuous variables did not have substantive effect sizes

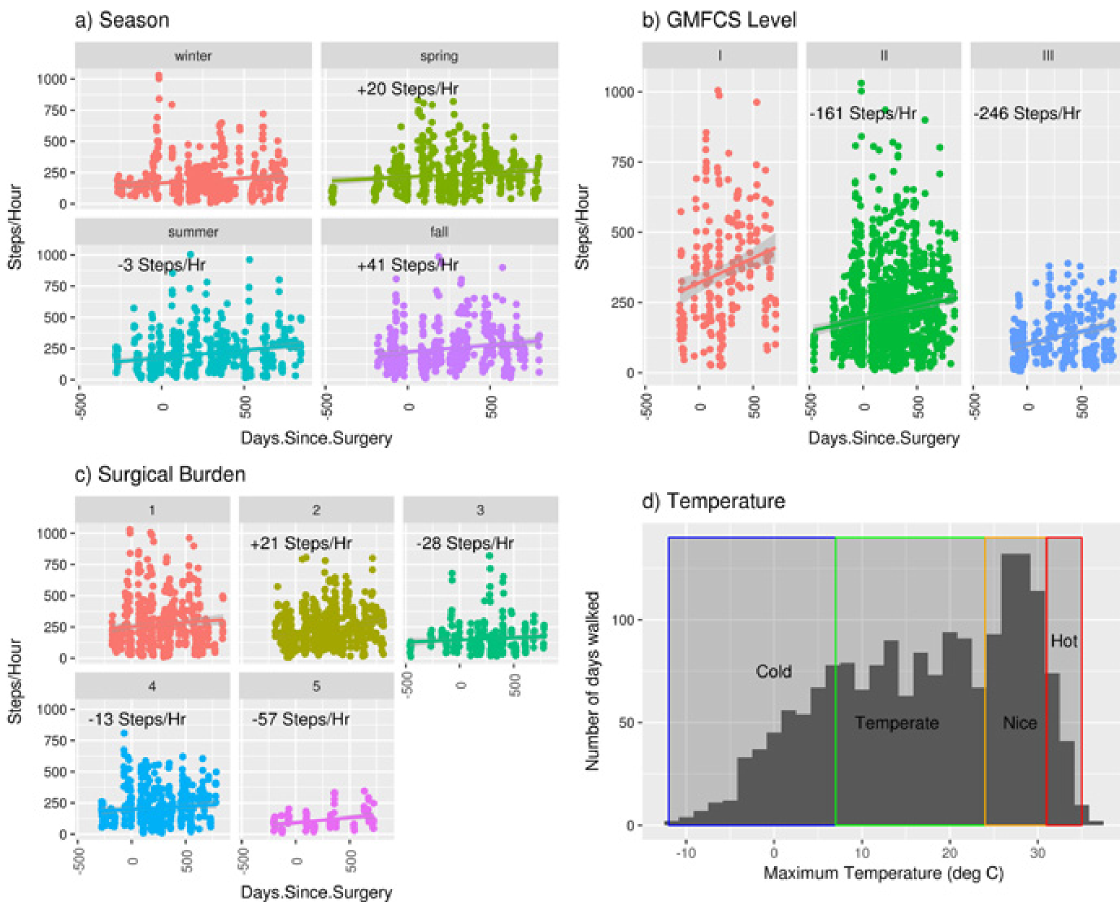

Fig. 2. Representation of how specific variables influence step-counts according to the bootstrap regression results. Categorical results are presented in a)-c). Generally, these graphs show an upward trend with increasing days past surgery. Offsets to regression lines are based on the category within the variable as shown in Table 3. Other variables from the regression have influence so it within a given panel, there is not a pure offset, and the slope changes. Figure d) shows a histogram of maximum daily temperature. Walking frequency increases when the temperature is from 24 to 31 °C ("Nice") weather, and is relatively stable across "Temperate" weather from 7 °C to 24 °C. This frequency analysis was not apparent in the regression, although temperature was weakly but positively correlated with step-count.

as indicated by the standard regression coefficients.

4. Discussion

Activity monitors offer the promise of improving patient outcomes both in offering incentive to users to be more active, but also in helping to identify critical moments in the recovery process for patients [18]. But for this promise to be fulfilled, a better understanding of the sources of variability of these data are needed. In this paper, we have considered some of the more traditional sources of variability: patient functional level, age, time since surgery, and surgical burden. In addition, we also considered temporal variables such as season and day of the week. In these measures, our outcomes are comparable to what has come before [2–6]. In the pediatric population, step-count is known to decrease on weekends and during the summer since children are often less active at home than at school [3,4]. Likewise, patients with higher GMFCS levels tend to have lower step-counts for functional reasons [3–5]. Our prior work has also demonstrated the influence of surgical burden [6]. The age effect shown in our model is more important, according to standard coefficient magnitudes than are the environmental variables. Yet although it is quite significant, the effect size is small: during the two years of the study, participants would lose on average about nine steps per hour due to gains in stature allowing them to cover more distance in fewer steps.

Walk scores, which showed great promise as a metric for health outcomes [7], had less of an impact in this study. While there was a slight effect which bordered on significant at times (Fig. 1b), little of the variability in step-count could be explained by this metric. Additionally, there was a small effect size: a change in walk score of 100 would result in just 15 additional steps per hour which was substantially less than the effect of temperature wherein a modest increase of 14–15 degrees would lead to the same result. The walk score's lack of effect in this study could be explained by a couple of factors. If our sample is primarily walking at school and not at home, then the walk score becomes irrelevant in this group. Also, it is worth considering that Althoff's data was from healthy working adults [7], and as such, may show different trends compared with youth with CP who are recovering from surgery.

Related to walk scores are temperature and weather. If the hypothesis is that people walk more when it is nice out, it is also presumed in this that they are walking outside. As indicated in Fig. 1b, the temperature effect on steps per hour was more significant than precipitation and walk score, although it did not rise to the level of formal significance ($p < 0.05$) (Table 3). The fact that there is some slight increase in walking during the spring and fall compared to winter, despite the fact that school is in session during all three seasons, lends credence to the fact that outside temperature matters. Likewise, the drop in step-counts during the summer could be due to the temperature as well (Table 3, Fig. 2a). However, it may be that patients lives are less dictated by the weather and more by their schedules. For example, total daily precipitation may not play such a large role in that the timing of rain shower is likely to influence whether or not one is outside being active. A more nuanced look at our temperature data shows that there more days walked during "nice" weather, here defined as between 24 and 30 °C (Fig. 2d). This effect is not captured by our modeling of step-count. Although other studies have examined the effect of weather and location on physical activity in children [9,10], this has not been previously done for this population, nor has the specific walkability of a location been related to their physical activity counts. Both Remmers et al. [9] and Harrison et al. [10] showed weather can affect walking totals. In particular, it is suggested that there is an optimal temperature range for peak physical activity [9]. However, Harrison et al. [10] have shown that such is at least population specific in that in cities in different countries, walking activity does not follow the weather in the same way. Some populations are more resilient to poor weather conditions than are others.

Other sources of variability could not be accounted for in the model. We did not have access to data reflecting the pain experienced by the patients, and it has been shown to affect a patient's willingness to participate in rehabilitation activities [19]. Likewise, we did not track physical therapy participation. Such interventions may increase step-counts both via the activity itself, but also due to travel to and from the activity. Finally, as is implied by work showing decreased step-counts during the summer and on weekends, a patient's walking habits may be determined substantially by their schedule either at school or elsewhere. A patient who participates in school clubs or other social activities is likely to have an increased step-count over those who do not participate in such activities. Therefore, a survey of frequency of patients' social involvement may help to better understand some of the random variability in the step-count data not explained by the model thus far.

Our methodology is also worth noting in that we have considered a number of factors simultaneously rather than one at a time. Also, unlike most of the studies noted here which used neither bootstrapping nor cross-validation, we have attempted to determine the stability of our results. StepWatch and other fitness tracker data are particularly noisy, and therefore, slight changes in the composition of the data set can cause wild swings in the unwary researcher's conclusions. Therefore, we deemed that it was better to include rather than ignore all variables that were not multicollinear and for which we could offer up a reasonable hypothesis relating them to the step-count in order to control for their effects or lack thereof. By using a bootstrapping approach to quantify variability of regression coefficients, p-values, and variability, it is possible to see which variables are the most important, and why that may be the case. Ideally, a variable's standard regression coefficient would be stable under bootstrapping so that it changed little, along with its p-value and percent variability in the model. However, bootstrapping may exhibit bias in parameter estimation if the subsample of data is not representative of the population [17]. We have identified several variables that exhibit stability under bootstrapping: time since surgery, GMFCS level, surgical burden, and to some extent season. Although the effect is small, the importance of season in predicting step-count is such that it suggests that season should be considered in surgical planning, even if seasonal effects are only due to attendance in school rather than weather, especially if it is accepted that early improvement in step-count leads to better long term outcomes. This last remains to be seen, and the potential effect size should be studied more closely in a larger cohort of patients to confirm it.

5. Conclusions

Step-counts are now frequently used as a proxy for measuring recovery post-surgically, but as these are highly variable, it can be difficult to directly relate the daily step-count to a patient's recovery progress. This study quantifies a number sources of variability in step-counts by way of bootstrapped regression. Our results have confirmed the importance of GMFCS, season, and surgical burden on walking activity levels and measured how hourly step-counts improve with time since surgery. Because of daily and seasonal variation observed in the data, it is suggested that scheduling and hence, social activities, might be worth considering in future studies.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of Competing Interest

None of the authors have any conflicts of interest to report.

CRediT authorship contribution statement

Timothy A. Niiler: Conceptualization, Data curation, Formal analysis, Methodology, Software, Visualization, Writing - original draft. **Kristen Nicholson:** Conceptualization, Data curation, Investigation, Methodology, Writing - review & editing. **Lydia Fischer:** Data curation, Investigation, Writing - review & editing. **Nancy Lennon:** Conceptualization, Methodology, Supervision, Writing - review & editing.

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