Title: A validation study of the modified Bouchard activity record that extends the concept of uptime to Rett syndrome

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Abstract

AIM To investigate the validity of responses to the Bouchard activity record (BAR) in Rett syndrome using pragmatic gold standard measures of walking status and step counts recorded using the StepWatch™ Activity Monitor (SAM).

METHOD During the waking hours of one day, 43 participants (mean age 21 yr SD 9 yr) wore a SAM whilst a proxy completed the BAR. Responses to the BAR were compared between participants grouped by walking status using the Mann-Whitney two-sample rank-sum test. Relationships were sought between BAR responses and step counts using linear regression.

RESULTS Compared with those who walked independently, in those who needed assistance to walk, the proxy classified more time using the BAR as sitting (median 9h15min [IQR 8h8min – 10h30min] vs. 6h15min [4h15min - 8h30min]; $p<0.001$) and less time standing (1h [38min – 1h30min] vs 2h15min [45min - 3h45min]; $p=0.04$). In those who walked independently, time classified as "uptime" (standing and walking) using the BAR was associated with increased step count ($r^2=0.58$; $p<0.001$).

INTERPRETATION These data support the validity of proxy-reported BAR responses. In those who walked independently, time classified as uptime using the BAR was able to estimate daily step count. This tool offers an inexpensive method for clinicians to gain insight into physical activity levels in this population.

Shortened title:

Physical activity in Rett syndrome

What this paper adds

- Our data provide evidence for the validity of a proxy-reported measure of physical activity in people with a neurodevelopmental disability.
- For those who walked independently, time classified as standing and walking (uptime) was strongly associated with step count over a 24-hour period.
- In Rett syndrome, increasing uptime could be a target for day activity programs to increase daily step count.

Rett syndrome is a neurodevelopmental disorder predominantly occurring in females and is usually associated with a mutation on the methyl-CpG-binding protein 2 (*MECP2*) gene.[1] It has an incidence in Australia of approximately 1 in 9,000 live female births.[2] Rett syndrome is characterised by largely normal early development followed by the loss of communication, hand function, impaired gross motor skills as well as the development of hand stereotypies.[3] The disability is severe with dependence for most activities of daily living.[4] Video data collected in 2004 on girls and women in the Australian Rett syndrome population-based study demonstrated that the majority were able to sit, slightly fewer than half could stand or walk, and a smaller proportion could transfer from sitting to standing independently.[5] Some will experience a decline in motor abilities with increasing age but the loss of walking is not inevitable.[6]

Assessment of physical function in this population often focuses on gross motor skills. In other words, measures are available for 'capacity' rather than 'performance'. One measure of gross motor performance is daily physical activity. Evaluating physical activity provides insight into the extent to which gross motor skills are used during daily life. Physical activity has been defined as "bodily movement generated by skeletal muscles resulting in energy expenditure" and can be broadly grouped as non-purposeful or purposeful movements.[7] Although both types of movement result in energy expenditure, non-purposeful movement (e.g. fidgeting) are not useful for activities of daily living whereas purposeful movements are those aimed at accomplishing a goal. Activities such as standing, walking and running when grouped together have been described as "uptime"[8] and in contrast to sedentary behaviours of lying and sitting, they provide opportunity for physical activity. Walking is a form of purposeful movement and is the most common type of physical activity.[9] There is evidence regarding the physical and mental health benefits of physical activity such as walking for populations of all ages.[10] Those with a disability would likely demonstrate similar health related benefits from participating in physical activity to persons in the general population, and benefits related to the development and maintenance of motor skills.[11,12]

We recently investigated the accuracy of physical activity monitors in girls and women with Rett syndrome. Compared to the Actigraph™ GTX3 (ActiGraph, Pensacola, FL) and ActivPAL™ (PAL Technologies, Glasgow,UK), the StepWatch™ Activity Monitor (SAM; OrthoCare Innovations, WA, USA) was best able to quantify the number of steps taken,[13] regardless of the level of gross motor skills.[14] A subjective method that provides useful information regarding physical activity would provide a cheaper and more convenient alternative to the SAM but the usefulness of proxy-report measures of physical activity in this population is not known.

The aim of this study was to determine whether a proxy-report measure of physical activity, a modified Bouchard Activity Record, could provide valid information in girls and women with Rett syndrome. We explored responses of a proxy-report diary card in relation to two pragmatic gold standard measures, namely walking status and step counts measured using the SAM.

Methods

Participants

Participants were recruited from the population-based Australian Rett Syndrome Database (ARSD).[15,16] A diagnosis of Rett syndrome was confirmed by the presence of a pathogenic *MECP2* mutation or by fulfilling the most recent diagnostic criteria.[3] To be eligible to participate in this study, video data collected in 2012 needed to be available to assess gross

motor skills[17] and the girl or woman with Rett syndrome needed to be able to walk either independently or with assistance. During the course of 2012, families of participants were provided with a diary card and the SAM device to be completed by a proxy. As part of a larger study, each participant wore the SAM over a seven-day period in their usual environment and the parent/guardian completed the diary card over any single 24-hour period within the SAM data collection period when they were with their daughter all day. The Human Research Ethics Committee at Curtin University approved this study (HR 139/2011).

Equipment

The Bouchard activity record (BAR) is a whole day diary card where every 15-minute epoch is assigned a number between one and nine, each representing either sedentary behaviour or a level of intensity of physical activity.[18] For the current study, the number of categories was reduced from nine to five levels to be more relevant to the severe disability associated with Rett syndrome. That is, the original categories of leisure activities, light and moderate manual work were grouped into the slow walking category, and the intense sports and manual work category were grouped into the vigorous walking category. The modified scale comprised the following items: lying; sitting for mealtime or other activities; light activities in standing; walking at a slow intensity; and walking at a vigorous intensity. "Walking at a slow intensity" was defined as walking gently such as when walking around the house. "Walking at a vigorous intensity" was defined as walking that was fast or more vigorous than gentle walking". The parent was asked to record the dominant category of activity for each 15-minute epoch on a day their daughter was at home, and to report immediately following each epoch or within 30 minutes.

The SAM is a small sensor that attaches to the ankle using a Velcro strap. It responds to acceleration, position and timing of movement, and measures the number of steps taken.[19] The SAM has demonstrated superior accuracy compared with other physical activity devices in free-living conditions across a range of walking speeds in the general population[20] and in Rett syndrome.[13]

Data management and analyses

The SAM data were grouped and summed across the same 15-minute epochs reported using the BAR. Each of the SAM data epochs was then labelled using the modified categories selected on the BAR. Epochs of BAR data were excluded if one dominant activity had not been classified, if the activity was water-based and the SAM had therefore been removed for that activity, or if an activity such as horse riding was described for which the intensity could not be accurately assigned. An additional BAR variable, labelled "uptime" was derived by merging the BAR categories for light activities in standing, slow and vigorous walking.

The distribution of data was examined for normality using frequency histograms and the Shapiro Wilks test. Data for participant age were normally distributed and are reported and mean and standard deviation values, whereas BAR physical activity data were non-normally distributed and are reported as median [IQR]. The start time and duration of monitoring for the BAR and SAM records were compared with a paired t test. The number of epochs for each BAR category for those who walked independently and needed assistance to walk was compared using a Mann-Whitney two-sample rank-sum test. The relationship between the number of step counts (measured using the SAM) and time classified in weight bearing activities (using the BAR) was assessed using linear regression. Data were analysed using STATA® (version 13, StataCorp, College Station, Texas). A $p<0.05$ was considered statistically significant.

Results

In December 2012, families of 361 females with confirmed Rett syndrome and born since 1976 had provided information to the ARSD, among whom 297 females were alive. Of those who were alive, data on walking status were available for 262 (88.2%) females. Of those, 151 (57.6%) had some walking skills [106 (70.2%) walked independently and 45 (29.8%) walked with assistance] and 114 (43.5%) were wheelchair dependent. The sample for the current study comprised 43 girls and women at a mean age of 21 yr SD 9 yr, 13 younger than 12 years. Thirty one (72%) participants were able to walk independently and 12 (28%) needed assistance to walk, proportions that were similar to the overall population who were alive. A total of 38 (88%) had a pathogenic mutation with most of the common mutation categories represented: for those who walked independently (C-terminal deletion [n=2], p.Arg104Trp [n=4], p.Arg133Cys [n=3], p.Arg168* [n=1], p.Arg270* [n=2], p.Arg294* [n=3], p.Arg306Cys [n=2], p.Thr158Met [n=2], large deletion [n=2], other [n=7]) and for those who needed assistance to walk (C-terminal deletion [n=1], p.Arg104Trp [n=1], p.Arg270* [n=2], p.Arg294* [n=1], p.Arg306Cys [n=2], p.Thr158Met [n=1], other [n=2]).

The SAM began recording steps (mean 8:21am SD 1h 10min) at a similar time to when the proxy began filling in the BAR (mean 8:15am SD 1h12min; p=0.92). The total time the SAM was worn each day (mean 11h 28min SD 1h 24min) was similar to the time over which the proxy filled in the BAR (mean 11h 41min SD 1h 30min; p=0.35). There was no difference in the total SAM wear time between those who needed assistance to walk (mean 11h 32min SD 1h 49min) compared to those who could walk independently (mean 11h 29min SD 1h 31min; p=0.94). The mean number of steps recorded by the SAM was 6,628 (SD 5,481) steps and was higher for those who could walk independently (mean 8,472 SD 5,392) compared with those who needed assistance to walk (mean 1,868 SD 1,074; p<0.001).

A total of 119 of the 3,966 epochs of BAR data (3%) from the 43 records were excluded from the analyses, mostly due to water-based activities or the proxy filling in more than one BAR activity category per epoch of time. The categories of lying and sitting were most frequently selected followed by light activities in standing and slow walking activities (Table 2). Vigorous walking was selected for 7 (16%) girls and women, two of whom walked vigorously for one epoch, one each for two and three epochs, three walked vigorously for five epochs in the day. Compared with those who could walk independently, those who needed assistance to walk spent more time sitting (p<0.001) and less time standing (p=0.04) than those who walked independently, but both groups spent similar time lying (p=0.23), slow walking (p=1.00) or vigorous walking (p=0.90) as classified using the BAR (Table 1).

Frequency histograms of step counts by BAR category indicated that the category of sitting was most frequently described. Visual inspection of Figure 1 shows that as the intensity of weight bearing activity selected using the BAR increased (i.e. standing and slow walking), the proportion of epochs during which a step count of zero was recorded appeared also to reduce (Figure 1). The median [IQR] steps for the 22 epochs classified as vigorous walking was 202 [12-476].

Total uptime derived using BAR data (summed standing and walking time) was greater for those who walked independently than those who needed assistance to walk (4h 30 min [2h 30min – 5h 30min] vs 2h 30min [2h-15min – 3 h], p=0.02) (Table 2). For all participants, total uptime was associated with step counts measured with the SAM (coefficient 32, 95% CI 23.4, 39.8). This relationship was only significant amongst those who could walk

independently (coefficient 29, 95% CI 19.1, 37.4) and not in those who required assistance to walk (coefficient 0.2, 95% CI -14.8, 15.2). For those who walked independently and as indicated by the coefficient, each additional minute of uptime classified using the BAR was associated with an additional 29 steps measured using the SAM (Table 2).

Discussion

To our knowledge, this is the first study to investigate the capacity of a proxy-report measure of physical activity in girls and women with Rett syndrome to yield useful data. Our findings indicated; (i) evidence of the validity of proxy-reported BAR responses and, (ii) for those who walked independently, the measure of 'uptime' derived from BAR responses was able to estimate daily step count.

This study examined the relationships between the modified BAR and two pragmatic gold standard objective measures: the participant's walking status and step counts measured using the previously validated SAM.[13,14] The BAR is a subjective measure of physical activity,[18] and has been validated in healthy populations of young adults aged 18 to 23 years[21] and adolescents aged 12 to 16 years.[22] We collapsed some of the categories to fit the range of activities relevant to the severe disability of Rett syndrome and our findings were able to provide data to validate the BAR when completed by a proxy for this group. In the first instance, we demonstrated its construct validity by the comparing BAR responses between two known groups; those who walked independently and those who walked with assistance. As would be expected, we observed that those who were able to walk independently spent less time sitting and more time standing compared to those who needed assistance to walk. Uptime comprised the sum of standing and walking time, as classified by the BAR, and was strongly associated with step count as measured by the SAM; a finding which supports its concurrent validity. Our sample also included 13 girls who were younger than 12 years of age. To our knowledge, this is the first study to investigate the usefulness of the BAR in any population with a neurological disability and also in any child younger than 12 years of age.

Graphical display of the distribution of step count in all five BAR categories indicated a high frequency of epochs with low numbers of step counts which is consistent with the severe disability seen in girls and women with Rett syndrome. Perhaps surprisingly, time classified using the BAR as slow and vigorous walking did not differ between participants grouped according to whether or not they required assistance to walk. Although it is possible that there were no differences in the time spent engaging in slow or vigorous walking, it is more likely that the BAR lacked the resolution and specificity to discern between standing and the different levels of walking activities. Firstly, parents categorised each epoch by the dominant activity rather than describing all activities. Secondly, balance is impaired in Rett syndrome and when upright, taking steps may be more likely than standing still,[5] yet the parent could perceive the dominant activity as standing. And finally, perceptions of walking intensity may differ between proxies. When we derived the blunter measure of uptime by merging the BAR categories of standing and walking, we found differences between participants grouped according to whether or not they required assistance to walk. Our composite variable of uptime measured by proxy-report is similar to the original objective measurement of uptime which has been measured in the general population of children and those with cerebral palsy using an 'Uptimer', a device that measures the position of body segments.[8,23] Uptime in 529 typically developing children aged 8 to 15 years was on average five hours across a 24 hour day, the sample representing children who attended public and private schools in both urban and rural locations.[8] In 300 children with cerebral palsy, mean uptime was lower than in the general population and varied with clinical severity ranging from one to five hours per day.[23]

Broadly similar to the cerebral palsy sample, the girls and women with Rett syndrome in our sample spent less time weight bearing than the general population and weight bearing time varied by walking status. Whilst we were unable to distinguish between the intensities of different weight bearing activities, our findings extended the concept of uptime from cerebral palsy to another diagnostic group and suggest that the BAR is an inexpensive and easy to use tool to quantify this concept.

Our data revealed that in those who could walk independently, for each additional minute of weight bearing time, an additional 29 steps were taken. In a healthy population, it is not surprising that weight bearing time leads to walking.[8] In contrast, skills in Rett syndrome are usually impaired by truncal ataxia, dyspraxia and altered muscle tone,[24] and therefore transitional skills are particularly difficult, such as the attainment of standing from sitting and the initiation of walking.[5] Consistently, our findings illustrated that even those who could walk independently, spent a sizeable proportion of their day in sedentary activities but that the opportunity to weight bear was likely to lead to walking. The determinants of physical activity could also be influenced by physical health status including comorbidities such as epilepsy[25] and opportunities for participation. Therefore given that the child is well, our finding has implications as to how parents and carers can provide opportunities to influence their child's physical activities. They would be more inclined to facilitate the initiation of standing as it would lead to more walking providing the health benefits that come with increasing physical activity and simultaneously reduced sedentary behaviour.[11] Weight bearing time could also be integrated into activity programs by therapists in an effort to promote a more active lifestyle in Rett syndrome.

The participants in this study were recruited from the ARSD, a national database of girls and women with Rett syndrome and comprised variability by age, gross motor skills and mutation category. The diary record of physical activity has important advantages as well as limitations. Each BAR epoch is categorised with a broad descriptor, the structure allowing more accuracy than estimates over longer time frames and validation has been demonstrated in adolescents and young adults in the general population.[18,22] However, the procedure relies on approximation, and some data were excluded due to human error in reporting and during epochs when the SAM could not be worn during water based activities. This proportion of missing data was however small and would have had little effect on the results. Using the BAR could also have been burdensome to parents although its simplicity and use over one waking day was acceptable to our cohort. We used pragmatic gold standard measures of walking status and the SAM, but we acknowledge that the ideal gold standard measure would be direct observation.

The concept of uptime has been explored in typically developing children in the general population and in children with cerebral palsy. Our paper extends this concept to another diagnostic group with a neurodevelopmental disability and demonstrated measurement of uptime in Rett syndrome using a proxy-report measure. This inexpensive and simple subjective measure has capacity to provide useful information on uptime and estimate the daily step count in those who could walk independently. Use of the modified BAR in Rett syndrome provided insights into physical activity and sedentary behaviour levels and further studies are justified to examine how its use could facilitate the planning of suitable day activities and programs in response to measurable goals.

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Table 1: Median [IQR] duration of time selected in each Bouchard Activity Record category for the total sample and with participants grouped according to assistance needed to walk

Activity category	Total sample (n=43)	Able to walk independently (n=31)	Needed assistance to walk (n=12)	*p* value[a]
Lying	4h 45min [3h 45min – 6h 30 min]	4h 45min [3h 45min - 6h 45 min]	3h 52min [3h – 5h 45min]	0.23
Sitting	8h 15min [4h 30 min -9h]	6h 15min [4h 15min - 8h 30min]	9h 15min [8h 8min – 10h 30min]	<0.001
Standing	1h 30min [45min – 2h 45min]	2h 15min [45min - 3h 45min]	1h [38min – 1h 30min]	0.04
Slow walking	1h 15min [30min – 2h]	1h 15 [30min – 2h 15min]	1h 24min [45min – 2h]	1.00
Vigorous walking	0min [0min – 15min]	0min [0min -15min]	0min [0min - 15min]	0.90
Uptime[b]	3h 15 min [2h 15 min – 5 h 30 min]	4h 30 min [2h 30min – 5h 30min]	2h 30min [2h - 15min – 3 h]	0.02

[a] Mann-Whitney two-sample rank-sum test used; [b] Uptime is a composite of time classified as standing, slow walking and vigorous walking

Table 2: Summary of linear regression findings for estimating total daily step count (measured using SAM) using weight bearing time, derived from BAR data.

	R-squared	Coefficient[a]	95% confidence intervals	*p* value
Total sample (n=43)	0.60	32	23.4, 39.8	<0.001
Able to walk independently (n=31)	0.58	29	19.1, 37.4	<0.001
Needed assistance to walk (n=12)	0.0001	0.2	-14.8, 15.2	0.98

[a]coefficient = change in the daily step count (measured using the StepWatch™ Activity Monitor; SAM) for each minute spent as uptime (standing or walking) classified using the Bouchard activity record (BAR).

Figure 1: Distribution of step counts measured with the StepWatch™ Activity Monitor in the Bouchard activity record (BAR) categories of lying, sitting, standing and slow walking. Vigorous walking was classified in seven subjects during 22 epochs and is not shown.