**­Title:** Physical activity and sedentary behavior in children and adolescents with healed Legg-Calve-Perthes Disease, Slipped Capital Femoral Epiphysis (SCFE), and Developmental Dysplasia of the Hip (DDH).

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**Project Summary:**

Once children or adolescents with Legg-Calve-Perthes (Perthes) disease, Slipped Capital Femoral Epiphysis (SCFE), or Developmental Dysplasia of the Hip (DDH) have healed, they are no longer restricted in their participation in physical activity. Generally, these patients are expected to return to normal activity levels after recovery; however, whether or not this actually occurs is unknown. There is currently no data on the amount of physical activity or sedentary behavior that these patients engage in or if their activity levels are adequate. This has impeded the ability for orthopedic surgeons to provide recommendations that prevent poor health outcomes associated with physical inactivity and sedentary behavior without compromising hip joint function or accelerating the development of osteoarthritis. This cross-sectional observational study looks to understand activity levels within this cohort, focusing on the extent to which healed pediatric hip patients meet current guidelines for physical activity and sedentary behavior, as indicated by the 24-Hour Canadian Movement guidelines. We anticipate that hip patients will have both inadequate physical activity levels and increased levels of sedentary behaviour. The study will further examine the factors that might be associated with activity levels and guideline adherence. Our hope is that this study will inform a larger multisite project, creating an international database of physical activity levels and sedentary behavior in pediatric hip patients. From this, larger prospective cohort and interventional studies would address the needs of this population by providing standardized, disease-specific recommendations on the types, frequency, and duration of physical activity hip patients should engage in once they have healed.

**BACKGROUND**

Legg-Calve-Perthes (Perthes) Disease, Slipped Capital Femoral Epiphysis (SCFE), and Developmental Dysplasia of the Hip (DDH) are three of the most common hip conditions affecting children and adolescents. Developmental Dysplasia of the Hip (DDH) is the most prevalent of the three conditions, with 1-3% of all newborns diagnosed at birth (Mulpuri et al., 2016). DDH represents a range of disorders, from mildly dysplastic to fully dislocated hips, where there is a complete loss of contact between the femoral head and acetabulum (Aronsson et al., 1994). Hip dysplasia is a leading cause of early onset hip osteoarthritis in people under 60 years (Jacobsen and Sonne-Holm, 2005; Pun, 2016).

Perthes is a disease of an unknown etiology where there is a disruption of blood supply to the hip joint that results in the collapse and flattening of the femoral head (Kim, 2011). Incidence of Perthes in the general population is quite rare (0.2 to 19.1 per 100,000) (Perry et al., 2012). Long-term prognosis is generally good, (Canavese & Dimeglio, 2008), nevertheless, long-term studies have found that adults who had Perthes during childhood or adolescence are more likely to develop osteoarthritis later on in life (Heesakkers et al., 2015).

Slipped capital femoral epiphysis (SCFE) is another common pediatric hip condition that is characterized by the slippage of the femoral head off the neck of the femur at the physeal plate. SCFE is relatively uncommon, with an incidence between 0.71 and 10.8 per 100,000 children (Novais et al., 2012); however, it is the most common hip disorder in adolescents. Early diagnosis is essential for a good outcome, but early onset osteoarthritis is a potential consequence of disease (Bitersohl et al., 2015).

In order to protect the hip joint, patients with SCFE, Perthes, and DDH are restricted in their sports and physical activity until they have healed. In SCFE cases, activity is restricted until the physis has closed. For Perthes and DDH patients, healing is contingent on the containment of the femoral epiphysis within the acetabulum and the shape of the femoral head. After healing, children can generally return to normal activity; however, many patients are left with residual deformity, experience hip symptoms and functional limitations, and may go on to develop hip osteoarthritis.

Physical activity (PA) is any bodily movement produced by skeletal muscles that results in

energy expenditure (Caspersen et al., 1885). PA plays an important role in bone and joint health, body

composition, cardiovascular health, and mental health (Janz et al;, 2010; Specker et al., 2015; Janssen &

Leblanc, 2010). The positive effects of PA are especially important during youth, as it has been shown to

reduce risks of future chronic diseases in adulthood (Raitakari et al., 1994; Twisk et al., 1997). PA habits

at this time may also be carried on into adulthood, making it important that health-promoting behaviors

are established (Telama et al., 2005). Physical inactivity, on the other hand, is associated with many chronic diseases including obesity, diabetes, cardiovascular disease, bone and joint diseases, cancer, and mental illnesses such as anxiety and depression (Booth, Roberts, & Laye, 2012).

While physical inactivity is associated with poorer health outcomes, sedentary behavior is also as important to consider. Increased sedentariness has been linked to metabolic syndrome, hypertension, and lowered self-esteem and pro-social behavior scores (Tremblay et al., 2010). In fact, sedentary screen time has been associated with risk of being overweight and poorer metabolic health, regardless of how long one exercises (Engberg et al., 2019; Owen et al., 2010). Thus, the benefits of PA may only exist if sedentary behavior is also limited.

The CSEP *Canadian 24-Hour Movement Guidelines for Children and Youth* were developed with an understanding of the health benefits related to maximizing PA and minimizing sedentary behavior. The guidelines suggest that children and adolescents should accumulate at least 60 minutes of daily moderate to vigorous physical activity (MVPA) and no more than 2 hours of recreational screen time per day (CSEP, 2016). In more recent studies, meeting these requirements has been operationalized as having average at least 60 min of MVPA or less than 2 hours of recreational screen time per day. Although the benefits of increased PA and reduced sedentary behavior in children and youth have been well established, adherence to the guidelines is low. Data from the Canadian Health Measures survey of children and youth aged 5-17 indicated that only 39% of children accumulated at least 60 minutes of MVPA on average and that just over half (53%) met the recommendation for 2 hours or less of average recreational screen time (Statistics Canada).

In pediatric hip patients, the same health benefits of PA exist; however, PA has the added benefits of improving hip physical function, reducing pain, and reducing the risk of future disability (Ayers, Franklin, & Ring, 2013; Fransen et al., 2014). In addition to the health benefits, the resulting consequences of inactivity may be more severe in hip patients. For example, weight gain adds stress to the affected hip joint and can worsen hip symptoms and function (Cooper et al., 1998). This can lead to a vicious cycle where hip symptoms/osteoarthritis further prevent an individual from being active, resulting in weight gain that leads to the further worsening of hip symptoms (Issa & Griffin, 2012).

Studies of other pediatric conditions, including juvenile arthritis and pediatric asthma have shown that these patients engage in even less PA compared to their healthy peers (Henderson et al., 1995; Bourdier et al., 2019; Glazebrook et al., 2006; Lang et al., 2004; Lelieveld et al., 2008). These studies examined many factors that predict PA, including parent/child perceptions of the disease and activity limitations, time of disease onset, disease outcome, and disease severity (Pianosi et al., 2004; Glazebrook et al., 2006; Takken et al., 2003). From these studies, as well as the known impacts of hip disease, one can infer that the pediatric hip population may similarly have reduced PA due to real and/or perceived limitations stemming from their disease (Riner & Sellhorst, 2013; Walker et al., 2015). Despite this possibility, only one study has examined activity pediatric hip patients; however, it only focused on Perthes disorder and did not use any objective measures of physical activity (Hailer et al., 2014).

This study will fill the current gaps in the literature by examining physical activity and sedentary behavior in three of the most common pediatric hip disorders. Since the orthopedic surgeon only provides care/guidance up to the end of adolescence, our target population will include children and adolescents only. We will examine the proportion of pediatric patients that are currently meeting the Canadian physical activity and sedentary behavior guidelines. We also hope to further understand the factors that affect physical activity and sedentary levels in order to make suggestions for surgeons when guiding hip patients in the future.

**Research Objectives & Hypotheses**

**Primary objective:** Understand levels of physical activity and sedentary behavior in healed pediatric hip patients.

Hypothesis 1: We hypothesize that the proportion of hip patients meeting the CSEP guidelines for average daily MVPA will be lower than nationally reported values (39%). We also hypothesize that the proportion of hip patients meeting the CSEP guidelines for average daily recreational screen time will be no different than or less than nationally reported values (53%).

We believe that our population of hip patients will have a lower proportion meeting the MVPA guidelines, as previous studies on other pediatric disorders have shown that patients have lower levels of physical activity than their normal peers. Additionally, the impact hip disease may have on activity (Riner & Sellhorst, 2013) and previous clinical experience further lead us to believe that this population may be less active than their unaffected peers.

With regards to sedentary behavior, there have been no studies in this population. Studies in other pediatric disorders have been inconsistent, with most finding no difference between their patients and unaffected peers.

Hypothesis 2: Time since healed, disease laterality, disease severity at presentation, radiographic outcome, parental support of PA, and HOOS scores, will be significant predictors of activity levels.

These factors have been chosen in accordance with previous studies assessing disease-specific predictors of physical activity in other pediatric disease populations.

**Secondary Objective:** Evaluate the validity of Physical Activity Questionnaires (PAQ-C and PAQ-A) in children and adolescents with healed hip disorders.

Hypothesis 1: There will be moderate to strong positive correlation between PAQ scores and accelerometry-derived physical activity metrics (rho>0.3).

A previous study by Voss et al. (2017) showed that the PAQ-C and PAQ-A were valid for use pediatric hip patients diagnosed with congenital heart defects. In this study, they found that the strength of correlation between PAQ score and total PA was 0.52 while the correlation between PAQ score and average MVPA was 0.48 (Voss et al., 2017)

**Methodology**

**Study Design:** This will be a cross-sectional, observational, correlational study design.We will be observing and describing sedentary behavior and physical activity levels in a population of healed pediatric hip patients at one time point. We will determine the proportion of patients meeting the CSEP MVPA and sedentary behavior guidelines. We will also be examining the relationships between activity levels (PA and sedentary behavior), which are our criterion variables, with multiple predictor variables.

**Inclusion Criteria:** Patients of the BC Children’s Hospital Orthopaedic clinic between the ages of 6 and 18 who have history of DDH, SCFE or Perthes that has since healed, which is operationalized as one year since successful intervention. A parent/guardian must be able to provide informed consent for their own participation and participation of their child. The child must be able to provide informed assent.

**Exclusion criteria:** Participants will be excluded if the patient has a comorbidity that limits their ability to be physically active. These comorbidities include cerebral palsy, muscular dystrophy, spinal deformity, lower limb deformity, osteogenesis imperfecta, or any lower limb surgery within the past year. Participants will be excluded if they cannot commit to returning the device, questionnaires, and activity log within 2 weeks of the appointment.Participants will also be excluded if they plan to travel during the time of accelerometry wear, as travelling may cause atypical levels of physical activity.

**Recruitment:** Participants will be recruited during their yearly follow-up visits at the Orthopedic clinic at BC Children’s Hospital (BCCH). It does not matter when after recovery the patient is recruited, as long as they have reached the healed stage. We will use a form of criterion sampling, where patients will be pre-identified from the orthopaedic department clinic lists based on the inclusion criteria. The pre-identified patient and their families will be approached in the clinic by a research assistant and invited verbally and with a formal invitation letter. Since all pediatric hip patients are followed-up with yearly until skeletal maturity or the age of 18, we do not foresee any recruitment bias where patients with certain disorders or more severe disorders are more likely to be recruited.

**Assent and consent:** We will obtain written informed parent/guardian consent and written informed participant assent to study participation and access to patient medical records. There will be two versions of the assent form: one for children and one for adolescents. Participants will be reminded that consent/assent is continuous and that they can withdraw at any point.

**Anthropometry:** The researcher will measure stature (cm) and body weight (kg) in order to calculate Body Mass Index (BMI). BMI will be calculated with the formula kg/m2 and will be used to determine Z scores that classify the participants ordinally as underweight (<-2SD) normal weight (between -1 and +1 SD), overweight (>+1SD), or obese (>+2 SD) in accordance with age and sex-specific tables (WHO).

**Chart Review**: We will categorically classify the laterality of disease (unilateral vs. bilateral) and determine the time since the patient has reached the healed stage (rounded to nearest year). An orthopaedic surgeon will examine past radiographs to classify the disease as mild, moderate, or severe at initial presentation using the following disease-specific grading scales:

Developmental Dysplasia of the Hip: IHDI Grade (Narayanan et al., 2015)

Legg-Calve-Perthes: Catterall classification (Catterall, 1971)

Slipped Capital Femoral Epiphysis: Southwick Slip Angle (Southwick, 1967)

The orthopaedic surgeon will also determine radiographic outcome using the following measures:

Developmental Dysplasia of the Hip: Tonnis, Severin classification

Legg-Calve-Perthes: Stulberg, SDI, Moses

Slipped Capital Femoral Epiphysis: AP alpha angle

**Subjective Hip Outcome:** Patients will fill out the International Perthes Study Group (IPSG) Modified Hip Osteoarthritis Outcome Score (HOOS), which is a 16-item questionnaire that asks about symptoms, pain, quality of life, function during everyday life, and function during sports and recreation (IPSG). This version of the HOOS has been previously modified for use in pediatric hip patients. The HOOS is scored on an interval scale ranging from 0-100, with higher scores indicating better hip function. In a systematic review by Thorborg et al. (2010), the HOOS was found to have adequate measurement qualities for test–retest reliability, floor and ceiling effects, construct validity, and responsiveness.

**Physical Activity and Sedentary Behavior:** The Physical Activity Questionnaire (PAQ-C PAQ-A)**:** The Physical Activity Questionnaire for Children (PAQ-C) and adolescents (PAQ-A) is a 7-day recall questionnaire that has been commonly used to self-report general physical activity levels (Kowalski, 2004). In 2011, an expert panel identified the PAQ as one of the most suitable self-report instruments for measuring physical activity in youth because of its validity, reliability, and ease of administration (Biddle et al, 2011).

Participants 13 years old or younger will be given the PAQ-C while children 14 years old or older will be given the PAQ-A. All patient participants 8 years old or younger will complete the PAQ-C by parent proxy. The PAQ is scored on a continuous interval scale of 1 to 5 (1=low physical activity, 5=high physical activity). The first question of the PAQ, which asks participants to select the types of frequencies of the sports and recreational activities that they engage in, is not scored.

In order to measure sedentary behavior, patients will be asked to self-report their average daily recreational screen time. Patients will also be given a series of 6 screen-based activities (e.g. watching TV/movies, playing electronic games) and will be asked to select approximately how much time they spend per day doing each activity (survey adapted from Harris lab).

Parents will also be asked to report their child’s average PA and sedentary behaviors as *x* minutes of MVPA per day and *x* minutes of recreational screen time per day.

**Accelerometry:** Each participant will be outfitted with a triaxial accelerometer (GT3X+; ActiGraph LLC, Pensacola FL). The ActiGraph monitor has repeatedly demonstrated good validity in children and adolescents (de Vries et al., 2006). The ActiGraph GT3X has also demonstrated acceptable reliability and validity for use in a patient population of children with cerebral palsy (O’Neil et al., 2014), reliability for measuring PA in free-living adults (Aadland et al., 2015), and is also the most commonly used accelerometer in published studies (Wijndaele et al 2015).

The device will be worn over the right hip from the time the individual wakes up until the time they go to bed with the exception of water-based activities such as bathing or swimming. There is evidence to show that placement of the device on the hip performs better than wrist placement when classifying activities into intensity categories (Ellis et al., 2016; Rosenberger et al., 2013).

The participant will wear the device for 7 consecutive days. A study by Trost et al. (2000) indicated that a 7-day monitoring period was enough to produce acceptable estimates of daily MVPA participation and account for differences in weekday and weekend physical activity. Although it may seem logical to collect more days, protocol adherence decreases with more days of wear.

It is up to the discretion of the parent whether or not the child will wear the device during contact sports. We will recommend that the participant consult with their coach if they have concerns; however, there is no evidence to show that the device poses any significant physical harm.

**Activity Log:** Patients will be given a one-page activity log. The log will ask participants to approximate the time the device was put on and taken off each day. It also asks for reasons why the device was not worn, for how long it was not worn, and to document reasons why a day might not have been “typical” such as due to illness.

**Data Collection and Management**: All patient participants will be coded with a study identification number. Personal identifiers will be stored separately from the data. Documents containing patient information will be stored either in a locked drawer in the research lab or will be encrypted and stored on a BCCH computer. Data will only be accessed by trained members of the research team.

**Procedure:** Once written consent is received from the parent/guardian and patient assent is received, the researcher will take the participant’s anthropometric measurements. The participants will then be fitted with an accelerometer and given verbal device instructions. The participants will be instructed on how to fill out the questionnaires (parent and patient questionnaires) and will be sent off with a page of instructions for accelerometer wear and return, the activity log, and a copy of the both the parent and the patient questionnaires. The parent/guardian will also be asked if they would prefer to return the materials after in-person or via mail). Those who choose to return the study materials by mail will be provided with a pre-paid courier package with the clinic address.

The patient will then wear the device and fill out the activity log for 7 consecutive days in their normal environment. Each patient will receive two elastic belts so that they can be rotated and washed if necessary. During the first day of wear, a research assistant will call the parent to ensure that the protocol is being followed and to answer any additional questions. Reminders will be sent out in the days following via text. At the end of the 7 days, the parent and the patient will complete their respective questionnaires at home. The parent questionnaire asks about parental support, their report of child’s activity, and additional sociodemographic factors. The patient questionnaire consists of the HOOS and PAQ questionnaire, as well as self-reported screen time and basic questions about schooling. Once the questionnaires are complete, the device and all documents will be returned to the research lab. If the device is not received within 2 weeks of the clinic visit, the research assistant will call the parent to follow-up. If there are any incomplete responses to the questionnaire, the research assistant will contact the parent via phone or email to ask if the question was intentionally left blank or accidentally missed. In the case of the latter, we will obtain the missing response from the participant.

Participants will be compensated with a $50.00 CAD to Indigo or Sport Chek for partaking in the study. Both stores have online websites and are able to ship across Canada. The parent will also be emailed the patient’s individual report card, which will provide individual accelerometry data (average MVPA) as well as information about national averages and recommended activity.

The questionnaires will be scored and data will be inputted onto the computer by trained members of the research team. The accelerometer will be wiped down with alcohol wipes and the elastic belts will be washing thoroughly after each use.

**Data Analysis:** The ActiLife 6 software (ActiGraph LLC, Pensacola FL) will be used for accelerometer initialisation (30 Hz), file download (15s epoch), processing, and data analysis. A day is considered valid if the device was worn for a minimum of 600 minutes—this criteria is recommended and has been previously shown to maximize reliability (Penpraze et al., 2006).

All participants who have at least 4 days of active days of wear (including one weekend day) will be included in the analyses. A study by Rich et al., (2013) demonstrated that a reliability coefficient of 0.93 can be achieved if the device is worn for at least 4 days. The reliability decreases slightly (0.90) when the minimum wear time decreases to 3 days (Rich et al., 2013). So, if there are few participants who meet this 4 day criteria, we will compare to less stringent criteria (3 valid days)(Voss et al., 2017). It is important to collect both weekdays and weekend days, as studies of children and adolescents show significant differences in weekend vs. weekday MVPA, with children exhibiting higher levels of MVPA on weekends and adolescents exhibiting lower MVPA on weekends (Trost et al., 2000).

We will compare the wear time with the activity log. If the time does not match up (>2 hour difference in wear time) or if the patient documented a day as “atypical,” the day will be excluded from analysis.

We will use Evenson cut-points to define PA intensity: MVPA (>2296 CPM), sedentary (<100 CPM)(Evenson et al., 2008). Although there are many cut points designed specifically for use in youth, a study by Trost et al. (2011) determined that only the Evenson cut-points performed among all age levels and provided acceptable classification accuracy for all levels of PA intensity. Physical activity levels will be operationalized as the total number of MVPA minutes from all valid days divided by the number of valid days. Sedentary levels will be operationalized as the average between the parent report and patient’s self-reported daily recreational screen time. Adherence to PA guidelines will be operationalized as having an average daily MVPA greater than or equal to 60 minutes. Adherence to sedentary behavior guidelines is defined as having both the parent and child report average daily screen time as less than or equal to 120 minutes.

**Statistical Analysis:** Pearson correlation will be used to assess the association between PAQ score and MVPA. Descriptive statistics will be calculated for applicable variables, including levels of MVPA, hours of screen time, PAQ, and HOOS scores. Using results from question 1 of the PAQ, we will also identify the most common sports/activities among our study sample. We will also determine the overall and age (child and adolescent) specific proportion of patients who meet the Canadian 24-Hour Movement guidelines for physical activity and/or sedentary behavior and conduct hypothesis testing to compare to previously reported Canadian population proportions (Statistics Canada).

In order to analyze predictors of physical activity levels, we will also conduct a 2-step analysis. First, we will do a univariate analysis with each predictor variable in order to identify important factors that are contribute the MVPA levels. We will then take the important factors and apply them to a multivariate analysis. We will control for age, sex, BMI, and household socioeconomic status.

**Significance**

This project will provide critical data on the amount of PA and sedentary that healed hip patients currently engage in. In most of orthopaedic research, a return to health is primarily regarded as the absence of symptoms (Ayers, Franklin, & Ring, 2013). However, it is important to take into account other factors (such as PA) that have an effect on overall health but specifically on hip health, even though the effects are not evident until long after these patients are out of pediatric care. Previous studies that have examined PA levels in this population have focused on how the patients’ activity levels change pre and post-intervention, with the purpose of the studies to use PA as an evaluation tool for a certain surgical procedure rather than as important health outcome in and of itself. This study will hopefully inform current orthopaedic practice on the importance of understanding PA in their patients.

While the vast majority of studies have demonstrated improved PA in hip patients post-surgical intervention, no study has actually examined the adequacy of these levels or the factors that contribute to these levels. Only one study so far has examined PA levels as a health outcome; however, it only focused on only PA in Perthes and did not use objective PA measures (Hailer et al., 2014).

Our understanding of PA and sedentary behavior is particularly important in pediatric hip patients because they are at greater risk of a more physically inactive and sedentary lifestyle. In addition to the increased risk, the consequences are more severe. Besides the risk of chronic diseases, inactivity and increased sedentariness can worsen hip joint function and aid in the progression or development of hip osteoarthritis, which these patients are already at risk for as a result of their disease. Osteoarthritis or any hip symptoms in adulthood would add significant financial and social burden to these patients.

Thus, it is vital that we understand how pediatric hip patients partake in PA and sedentary behavior and the factors that affect it. This knowledge would inform orthopaedic practice about the need to understand physical activity in their patients and promote greater activity and less sedentariness.

Determining the validity of the PAQ is also important. Although the PAQ-A and PAQ-C have

been validated in children and adolescents, we cannot assume that the PAQ is valid in our population of

interest. This study will determine if the PAQ can be administered in place of accelerometry as an

assessment of PA in this population. This is beneficial because self-report is more cost-efficient, easier to

administer, and has a lower participant burden compared to objective measures of PA (Warren et al.,

2010).

Since the pediatric hip patient population is quite small in British Columbia, we hope to

collaborate with other centers to create a multisite registry of PA and sedentary behavior. From this,

future interventional studies that aim to break the cycle of hip osteoarthritis, allowing patients to improve hip joint function while also achieving all the other benefits associated with PA and reduced

sedentariness. Ultimately, this research will lead to standardized guidelines for surgeons and other

healthcare providers to recommend PA in a way that optimizes overall patient health while also

minimizing risks for hip-related problems in the future. Recommendations may involve the modification

of activity, exercise intensity, and exercise volume, taking into account the associated predictors. These

guidelines have the potential to impact pediatric hip patients around the world.