Master's thesis

NTNU
Norwegian University of Science and Technology
Faculty of Medicine and Health Sciences
Department of Neuromedicine and Movement Science

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Habitual Ankle-Foot-Orthosis use and user perception of AFO helpfulness in ambulant children with cerebral palsy

Master's thesis in Human Movement Science
Supervisor: Karin Roeleveld
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Abstract

Background: Children with cerebral palsy (CP) experience impaired gait function and increased energy cost (EC) of walking compared to typically developing (TD), which leads to limitations in activity and participation. Ankle-foot-orthoses (AFOs) are commonly used orthotic devices in children with CP, to improve gait function. And although there has been reported evidence for AFOs e.g. improving gait efficiency, evidence levels are consistently reported to be low, and little discussion has been made around the translation of this effect into daily life functioning. Clinical experience has shown us that AFOs may actually be limiting in some activities. How AFOs are habitually used in daily life and activity in children with CP and what the user perspective is has not been investigated thoroughly, and may uncover challenges that need to be addressed to provide children with CP improved treatment.

Aim(s): The main aim of this study was to investigate when, in what activities and possible reasons why or why not AFOs are used in the daily life of children with CP, and what the user perspective is related to perceived effect of the AFOs. A secondary aim was to investigate if improvements in the EC of walking with AFO was linked to more positive user perceptions and a higher degree of habitual AFO use.

Method: 8 children (mean age 11,4) with spastic unilateral CP who used clinically prescribed AFOs were recruited through Trøndelag Ortopediske Verksted. They went through a semi-structured interview asking about when, in what activities and why or why not they used their AFOs at home, at school, and in leisure time, as well as the perceived helpfulness of the AFOs in keeping up with peers, feeling of stability, degree of fatigue at the end of the day and walking in uneven terrain. Three accelerometers (back, thigh and AFO) were worn for 7 consecutive days and analyzed with activity recognition software to find total weekly hours of AFO use and what activities AFOs were used in. To estimate the EC of walking, they performed two walking tests with a portable calorimeter, in randomized order with 1) Shoes only, compared to 2) Shoes with AFO.

Results: AFOs were almost always or often used at school in n=5, in leisure time in n=4 and none of the participants ever used their AFO at home. The connection between most AFOs and shoes were brought up multiple times as reasons for not using AFOs at school and at home. Other recurring themes were not having the need, pain, discomfort or the AFO being limiting in higher activities. Mean total weekly hours of AFO use was 14,6 (range 0-34,9). AFOs were used 25,2% (mean, range 0-52%) of the total time spent in walking activities, and 21,1% (mean, range 0-46%) of the total time spent in standing activities. AFOs were perceived as helpful in feeling of stability in n=4, where the rest felt no difference or were unsure. Fatigue was perceived as reduced in n=3 with AFOs, but increased in n=2, due to pain or altering of gait pattern. Keeping up with peers was perceived as being harder with AFO in n=3, and easier in n=2. Walking uphill was reported to be harder with AFO than without in n=3, and easier in n=1. One participant had a clinically significant decrease (-1,7 J/kg/m) in the EC of walking when walking with AFO compared to shoes only; 5 participants had non-significant decreases and two participants had non-significant increases in EC.

Conclusion: AFO use was variable within the group, but were mainly used in school, in walking and standing activities, and adherence was lower than previously reported findings. Subjective perception of AFO helpfulness was also highly variable, but especially in higher activities, such as running and sports, the AFOs were mostly perceived as limiting the children. The connection between most AFOs and shoes were brought up multiple times; changing the AFO between shoes was an irritant and limiting factor, while having an extra AFO attached to other shoes was a facilitating factor for more use. The only participant with a clinically significant improvement in EC with AFO had the most hours of AFO use and reported perceiving the AFO as being more helpful than the other participants.

Sammendrag

Bakgrunn: Barn med cerebral parese (CP) opplever nedsatt gangfunksjon og kan ha økt energikostnad under gange, sammenlignet med funksjonsfriske barn. Dette fører til begrensninger i aktivitet og deltakelse. Ankel-fot-ortoser (AFO) er et vanlig brukt ortopedisk hjelpemiddel hos barn med CP, for å forbedre gangfunksjon. Selv om det finnes bevis for at AFO'er kan forbedre f.eks. gangøkonomi har evidensnivåene konsekvent blitt rapportert som lave, og det har vært lite diskusjon rundt den antatte overføringen av denne effekten til funksjon og fungering i dagliglivet. Klinisk erfaring har vist oss at AFO'er faktisk kan virke begrensende i noen aktiviteter. Hvordan AFO'er vanligvis blir brukt i daglig liv og aktivitet hos barn med CP og hva brukerperspektivet er har ikke blitt nøye undersøkt, og kan bidra til å avdekke utfordringer som bør adresseres for å forbedre behandlingstilbudene til barn med CP.

Formål: Hovedmålet med denne studien var å undersøke når, i hvilke aktiviteter og mulige grunner for hvorfor eller hvorfor ikke AFO'er blir brukt i daglig liv til barn med CP, og hva brukerperspektivet er relatert til subjektiv oppfattelse av AFO'enes effekt. Et sekundærmål var å undersøke om forbedringer i energikostnad under gange med AFO var koblet til mer positive brukerperspektiv og høyere grad av AFO-bruk i dagliglivet.

Metode: 8 barn (gjennomsnittsalder 11,4) med spastisk unilateral CP som brukte klinisk foreskrevne AFO'er ble rekruttert gjennom Trøndelag Ortopediske Verksted. De gjennomgikk et semi-strukturert intervju der det ble spurt om når, i hvilke aktiviteter og hvorfor eller hvorfor ikke deltakerne brukte AFO'ene sine hjemme, på skolen og på fritiden, samt hvordan de subjektivt opplevde om AFO var fordelaktig i situasjonene å holde følge med venner, følelse av stabilitet, grad av fatigue på slutten av en dag og i gange i ulendt terreng. Tre akselerometre (rygg, lår og AFO) ble brukt i 7 dager i strekk for å måle aktivitet, og ble analysert med aktivitetsgjenkjennende software for å finne totale antall timer med AFO-bruk og i hvilke aktiviteter AFO'ene ble brukt i. For å beregne energikostnaden under gange gjennomførte deltakerne en 5-minutters gangtest og en 3-minutters gangtest med et bærbart kalorimeter, i randomisert rekkefølge av 1) Bare sko, sammenlignet med 2) Sko med AFO.

Resultat: AFO ble brukt nesten alltid eller ofte på skolen i n=5, på fritiden i n=4, og ingen av deltakerne brukte AFO hjemme. Koblingen mellom de fleste AFO'er og sko ble brakt opp flere ganger som grunn for hvorfor AFO ikke ble brukt på skolen og hjemme. Andre gjentagende temaer var at de ikke følte behov, på grunn av smerte eller ubehag, eller at AFO'ene var begrensende i høyere aktiviteter. Gjennomsnittlig totalt timeantall gjennom uken med AFO-bruk var 14,6 (range 0-34,9 timer). AFO'er ble brukt i gjennomsnittlig 25,2% (range 0-52%) av den totale tiden brukt i gangaktiviteter, og i 21,1% (range 0-46%) av den totale tiden brukt i stående aktiviteter. AFO ble opplevd som fordelaktige i følelse av stabilitet i n=4, der resten ikke følte noen forskjell eller var usikre. Fatigue ble opplevd å bli redusert i n=3 med AFO-bruk, men økt i n=2 på grunn av smerter eller endring av gangmønster. Å holde følge med venner ble opplevd som vanskeligere med AFO i n=3, og lettere med AFO i n=2. Gange i oppoverbakke ble rapportert å være vanskeligere med AFO enn uten i n=3, og lettere i n=1. Bare en av deltakerne hadde en klinisk signifikant reduksjon (-1,7 J/kg/m) i energikostnad under gange med AFO sammenlignet med bare sko; 5 deltakere hadde en ikke-signifikant senkning og to deltakere hadde en ikke-signifikant økning.

Konklusjon: AFO-bruk var veldig variabel innad i gruppen, men ble brukt mest på skolen, under gange og stående, og adherence var lavere enn tidligere funn. Subjektiv opplevelse av AFO'enes fordelaktighet var også variabel, men spesielt i høyere aktiviteter som under løping og sport ble AFO'ene opplevd som begrensende hos barna. Koblingen mellom AFO og sko ble nevnt flere ganger; å bytte AFO mellom sko var brysomt og en begrensende faktor, mens det å ha en ekstra AFO i andre sko var en fasiliterende faktor for høyere bruk. Den eneste deltageren med en klinisk signifikant forbedring av energikostnad under gange brukte AFO'en i flest timer og rapporterte at AFO'en var mer fordelaktig enn de andre deltagerne.

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Abbreviations

CP = Cerebral palsy

GMFCS = Gross Motor Function Classification System

TD = Typically developing

EC = Energy cost

ICF = International Classification of Functioning, Disability and Health

AFO = Ankle-foot-orthosis

GRF = Ground reaction force

ROM = Range of motion

REK = Regional Etisk Komitè

TOV = Trøndelag Ortopediske Verksted

NTNU = Norges teknisk-naturvitenskapelige universitet

SIAS = Spina Iliaca Anterior Superior

1 Introduction

1.1 Background

Cerebral palsy (CP) is a neurological disorder affecting the development of movement, balance and muscle coordination caused by malformation or damage to the brain, either before or during birth or up to 2 years of age (Morris, 2002). CP is a highly heterogeneous condition, where the degree and location of primary injury results in a variety of clinical symptoms and gait abnormalities. Both primary and secondary impairments – such as muscle weakness, abnormal tone, contractures and bony deformity - cause different degrees of activity limitations in children with CP (Morris, 2002). The Gross Motor Function Classification System (GMFCS) is a five-level approach to determine the current gross motor function level and functional limitations put on a child with CP (Palisano, 2007). A classification of level 1 represents children who can walk, run, jump, and use stairs without support, although with limitations in balance, coordination and velocity (Palisano, 2007). Level II children might experience more limitations walking across larger distances, and use railing or other support devices like wheelchairs and crutches in a higher degree. They might also have a bigger need for adaptations to facilitate for participation and activity (Palisano, 2007). In comparison, the highest level (V) represents children with no postural control, that are transported in a manual wheelchair (Palisano, 2007). Other classifications include the type and topography of CP, where spastic, dyskinetic and ataxic subgroups describe the main motor manifestations, where spastic type is further classified as unilateral and bilateral (Rosenbaum, 2009). Spastic type is reported in around 80% of CP cases in Norway, of which 33% is unilateral (Andersen, 2007).

Compared to typically developing (TD) children, it has been reported that children with CP can have a three-fold increase in the energy cost (EC) of walking (Norman et al., 2004) due to deviations in muscle activity and gait kinematics (Balaban et al., 2007), and fatigue and musculoskeletal pain are common areas of complaint (Dickinson et al., 2007; Ramstad et al., 2011). Impaired mobility is found to be one of the most important factors in reducing participation (Bjornson et al., 2016; Bjornson, 2014), and energy efficiency in gait correlates with activity limitations for ambulatory children with CP (Kerr et al., 2008). It has also been found that only 25% of children with unilateral CP meet the public health recommendations for physical activity (PA) during a week (Mitchell et al., 2015). An effective and efficient gait is therefore an important treatment goal (Aboutorabi et al., 2017; Figueiredo, 2008). Participation (involvement in daily life situations) and activity (the execution of a task) are crucial domains in the model of the International Classification of Functioning, Disability and Health (ICF) (WHO, 2002). The ICF is a framework that can be used to research and define the impact on an individual caused by impairments at the body structure level and the reciprocal links to activity limitations at the personal level and participation restrictions at the societal level (WHO, 2002). Many treatment interventions in children with CP are aimed at the body structure and body function level, such as surgery, botulinum toxin injections, physiotherapy and applying orthoses (Gage, 1991), to facilitate for positive changes in the other domains.

Ankle-Foot-Orthoses (AFOs) play an important role in the management of ambulatory children with CP, and about 60% of children with CP in Norway are fitted with them (Jahnsen, 2019). AFOs are prescribed to either affect body structure (prevent or correct deformity, compensate for muscle weakness), assist body function (improve gait efficiency) or, many times, both in conjunction (Morris, 2002; Wingstrand et al., 2014). An AFO is primarily constructed in thermoplastics or carbon fibre, either as a prefabricated orthosis or from a positive model of the intended user. They usually extend distally under the toes and proximally to the tibia plateau. It thus encompasses the ankle, foot and leg segment, and can either limit or assist movements and range of motion (ROM) in the ankle and foot joints (Lin, 2000; Morris, 2002). An example is the incorporation of a spring-adjusted ankle joint, which can allow for both physiological plantarflexion at initial contact and for assisting dorsiflexion in swing phase and controlled ankle-rockers through the gait cycle (FIOR&GENTZ, 2018). AFOs may also indirectly affect more proximal joints, i.e. the knee and hip, by manipulating the line of action of the ground reaction force (GRF) (Bowers, 2009). Pictures of regular AFOs prescribed to children with CP are shown in figure 3.1. The main justification for AFO prescription can be summarized as "the collective improvement of biomechanical variables to increase the ease of taking an individual step" (Bjornson et al., 2016). That is, to improve or optimize gait and self-sufficient mobility, with the expectation that this improves physical functioning and increases participation and activity (Bjornson, 2014).

There has been reported evidence that AFOs can decrease the EC of walking for children with CP (Aboutorabi et al., 2017; Balaban et al., 2007), improve spatiotemporal gait parameters such as increasing velocity, cadence, stride and step length (Aboutorabi et al., 2017; Bowers, 2009; Kerkum et al., 2016), and affect ankle kinetics and kinematics such as improving power generation at push-off and increasing ankle dorsiflexion at initial contact (Balaban et al., 2007; Bowers, 2009). There has sadly also been reported generally low levels of quality in these studies, as many have methodological flaws, including failing to provide important details of both participants, control conditions and the AFOs – the intervention itself – which makes it difficult to extrapolate results, compile data into larger meta analyses and inform evidence-based prescription guidelines (Figueiredo, 2008; Firouzeh et al., 2021; Harlaar et al., 2010; Morris, 2007). Therefore, the prescription and fitting process of an AFO relies in big part on the clinical experience of the orthotist and the physicians. In the city of Trondheim, Norway, a recurring prescription goal is conservation of energy along with correction or prevention of deformities. As earlier research is primarily focused on outcome measures related to the Body structure dimension according to the ICF (Firouzeh et al., 2021), we only really rely on a body of low-level evidence related to *capacity,* defined as what a person can do in a standardized, controlled environment (Bjornson et al., 2016; WHO, 2002). Clinical experience also tells us that the fitting and optimization of AFOs in the clinics are performed on level floors, at self-selected speeds, thereby replicating the environment of the laboratories. Further, few individual prescription goals are evaluated with validated outcome measures in the clinics. There are few studies on *performance*, how children with CP use their AFOs in their daily life situations (Bjornson et al., 2016; Firouzeh et al., 2021; WHO, 2002), and even fewer studies on user or relative's perspective and experience with AFO use (Firouzeh et al., 2021). We therefore do not have a good understanding of how hypothesized benefits and effects of AFOs on gait-related outcomes are thought to translate into improving everyday function, increasing time in other activities and participation.

Working as a clinical orthotist providing children with disabilities with orthoses, we often come across reports of children not using their AFOs as prescribed, and especially in young children we observe that the AFOs may be limiting in floor play and some transitional movements, which has also been reported in a study on parent's perception (Näslund et al., 2003), one of few qualitative studies on AFOs in children with CP.

Gaining insight into user perspective and patterns of daily AFO use might uncover challenges, limitations, and how other motor function skills such as transfers, running, the execution of sports, dancing, and moving on uneven terrain are affected according to the users themselves. After all, children do not live permanently on level floors, and general PA have been found to be associated with a range of benefits in both psychological and physiological health in children and adolescents with CP (Johnson, 2009), and should therefore be facilitated. Understanding the children's and their families' motivation and adherence to the AFO as a treatment intervention is also crucial, as no matter the reasoning behind the prescription of an AFO, the intervention has failed if the AFO is not used (Eddison et al., 2020). To start reaching a better understanding of how AFO use affects day-to-day life and activity of children with CP, we need to investigate possible limitations or challenges through finding out when, in what activities, and why or why not they use their clinically prescribed AFOs and what the user perspective is.

1.2 Research aims

The primary objective of this study is therefore to investigate how children with CP with clinically prescribed AFOs use their orthoses at home, at school and in leisure time; in what activities and why or why not are they used in the different settings and situations? How do the children themselves perceive the helpfulness of the AFO in keeping up with peers, degree of fatigue at the end of the day, walking in uneven terrain and feeling of stability?

A secondary aim is to investigate if improvements in the EC of walking, when walking with AFO compared to shoes only, is linked to more positive user perceptions and a higher degree of habitual AFO use.

2 Method

This study was approved by the Regional Committee for Medical and Health Research Ethics (REK #28777).

2.1 Participants

Participants were recruited through Trøndelag Ortopediske Verksted (TOV), where they had received clinically prescribed AFOs that they were accustomed to. To be eligible for inclusion, they had to be between the ages of 5-17, diagnosed with unilateral spastic CP with a corresponding GMFCS classification of level I or II. They also had to be able to receive and understand verbal instructions and to walk consistently for at least five minutes without assistive walking aids. The recruitment process lasted from december 2019 to november 2020.

2.2 Protocol and procedure

Organizationally, this study was part of a larger protocol contributing to three master thesis projects at the Norwegian University of Science and Technology (NTNU). Only the protocol relevant to this study will be described.

Written consent was obtained from both the participants and their parents or guardians. They were informed that they could withdraw from the study at any time without giving a reason. The participants went through a testing protocol lasting 2-3 hours on the same day, located at the gait laboratory at St. Olavs hospital in Trondheim, Norway. The protocol included:

1. *Semi-structured interview*
2. *Anthropometric measurements and AFO description*
3. *Accelerometer placement for activity measurements*
4. *Walking tests to estimate the energy cost of walking*

Before the testing started, the participants were thoroughly explained the protocol, using pictures to illustrate and show the equipment to be used.

2.2.1 Semi-structured interview

The interview questions are attached as Appendix 1. The questions were chosen to get information on habitual AFO use in three different life settings: at home, at school and in leisure time outside of the home, and if there were specific activities they always or never wore their AFO. Answers were directly categorised according to how often AFOs were used and any commentary regarding *why* or *why not* AFOs were used. We also wanted information on the children's subjective experience and the perceived helpfulness of the AFOs in different settings: keeping up with peers, walking in uneven terrain (uphill/downhill), degree of fatigue at the end of the day and feeling of stability. Therefore, the children were asked if the different tasks were better with AFO or worse with AFO. We did not find validated questionnaires available that included all the aspects we wanted. The interview was semi-structured to give room for explanations or possible changes to the words to allow full understanding for the participants, many of them being young children.

The interview was performed as the first part of the testing protocol. It was completed with one or both parents/guardians in a private room without disturbance. The participants were asked to try their best at answering for themselves, but if they were stuck or didn't understand the questions, parents/guardians were allowed to contribute. One researcher completed the interview while scribbling small notes or keywords to give as much attention to the participants as possible. Another researcher transcribed the answers in real-time on a laptop computer. All questions were included, and no new ones were added, except that every interview ended with the open question, "Is there anything else you wish I would have asked?" to give room for additional information.

2.2.2 Anthropometrics and AFO description

A stadiometer was used to measure height, and a digital scale was used to measure weight. To measure any presence of gastrocnemius equinus, the Silfverskiöld test was performed on every participant, using a goniometer to measure degrees of dorsiflexion with bent knee and straight knee, on both the sound side and afflicted side. One researcher performed the test with two hands as described elsewhere (Goss et al., 2020), while another measured with a goniometer. The presence of any knee flexion contracture was also tested, as well as measuring leg length discrepancy (LLD) using a measuring tape from spina iliaca anterior superior (SIAS) to the medial malleolus plateau on both sound side and afflicted side.

The type of AFO, material and movements prevented or assisted in the AFO were noted down for every individual after visual inspection.

2.2.3 Activity measurements

The accelerometer as a measure of habitual physical activity is reported to be feasible in children with CP (Gorter, 2012) and valid in reflecting time in sitting, lying, standing and walking (Mitchell et al., 2013).

Three or four accelerometers of the type Axivity AX3 sensor (Axivity, Newcastle, United Kingdom) were used per participant. Configuration of the accelerometers was done in version 1.0.0.28 of the Open Movement GUI Application. The accelerometers were set to sample at a frequency of 200 hertz and a range of ±8 times gravity.

The sensors were placed on the mid-thigh (anterior) on the unaffected leg, on the lower back (at or close to the L3 vertebra) and on the lateral proximal part of the AFO where they would be least in the way, but close to the mid leg. Some of the participants had a second AFO, for example attached to shoes used inside at school, and in these cases a fourth sensor was placed on this second AFO. The accelerometers were attached to a piece of soft and pliable fixation tape on the skin, and then sealed by transparent adhesive film. The accelerometers were oriented so that the x-axis corresponded to the mediolateral axis of rotation, the y-axis corresponded to the anteroposterior axis of rotation, and the z-axis corresponded to the longitudinal axis of rotation.

The AX3 sensors were worn for 7 consecutive days starting with the test day. Parents or guardians were shown how to attach and remove the sensors and given extra rolls of tape in case they should detach. Additionally, the participants were asked to write an activity diary for the week, where they would note down specific activities they had been doing, and if they had sick days home from school or similar situations not reflecting everyday life, which could be used to control the quality of the data output later. It was emphasized that the participants should go about their everyday life as usual, and to not worry about wearing the AFO more than usual just because of the sensor. At the end of the test period, the sensors were sent back to the researchers in a pre-paid envelope.

2.2.4 Walking tests to estimate energy cost

The energy cost of walking was estimated using a portable indirect calorimeter, Metamax II (CORTEX Biophysik GmbH, Leipzig, Germany). Air inhaled through a Cortex face mask with a disposable flow turbine and a small mixing chamber provided respiratory values for every 10 seconds. Before each test, the equipment was calibrated using ambulant air, a reference gas (15% O2 and 5% CO2) and a 3-liter cylindrical pump that calibrated the flow turbine (Hans-Rudolph, Shawnee, KS). Time and heart rate during the walking test were collected using Polar M400 (Polar Electro Oy, Finland) with an attached chest strap with heart rate monitor. The distance walked over the tests was measured using a standardized measuring wheel with a 1-meter circumference.

The participants completed two walking tests, walking back and forth along a 35-meter hallway wearing the Metamax and heart rate monitor. The test was started with a 5-minute walking test (5MWT) to ensure steady-state measurements, followed by one 3-minute walking test (3MWT). Between the tests, pauses were held at a minimum and only used for changing between the randomized conditions and checking the quality of the data. As per pre-defined randomization the conditions were 1) Shoes only, 2) Shoes + AFO. An extra condition was included in the larger protocol this study is a part of (Shoes + adjusted AFO), meaning the participants completed one 5MWT and two 3MWTs. To control for level of fatigue, the order of testing of the different conditions will be presented.

Before starting the tests, it was emphasized that the participant should walk at their self-selected speed and without talking to secure the accuracy of the measurements. Participants were asked at regular intervals during the test if they felt OK and were instructed to respond with a “thumbs up” or “thumbs down”, where in the latter case the test would be immediately stopped and the equipment removed.

Two testers walked behind the participant, one keeping track of the time and equipment, the other tracking distance with the measuring wheel. One additional tester was located near the computer to secure the data quality and executing quick condition changes between the tests.

2.3 Data analysis

2.3.1 Semi-structured interview

To synthesize the answers into a meaningful overview, the transcribed answers and the interviewer's keywords and notes were cross-checked and read thoroughly as a quality control before categorization. As well as categorizing degree of use, recurring themes were gathered, especially in the context of facilitators or barriers for use in different settings and activities.

AFO use at school, at home, and in leisure time were categorized into four brackets of degree of use: "Almost always"," Often", "Sometimes" and "Hardly ever". Subjective perception of AFO use were categorized according to if the situation or setting was «Better with AFO» or «Worse with AFO».

2.3.2 Energy cost of walking

Respiratory exchange ratio (RER), calculated as VCO2/VO2, and mixed venous oxygen saturation (sVO2) were extracted from MetaSoft (Cortex Biophysic, 2005) and exported to Excel 2016 version (Microsoft, Inc., Redmond, WA, USA). Data from the walking tests were plotted for visual inspection, and the most stable 60 seconds with less than 10% variation in VO2 and ventilation, and less than 5% variation in RER were defined as steady-state (Thomas et al., 2009).

Energy cost expressed as J/kg/m was calculated using energy consumption (ECS) divided by speed (m/min) (De Groot et al., 2010):

EC (J/kg/m) = ECS / Walking speed

ECS expressed as J/kg/min was calculated using relative VO2 and RER:

ECS (J/kg/min) = ((4.96 * RER) + 16.04) * *VO2 / kg*

VO2 was calculated using the collected weight of the participants:

*Relative VO2 (mL/kg/min) = (VO2 / weight) * 1000*

It has been reported that children with CP have an energy cost of walking corresponding to 6.84 J/kg/m (standard deviation (SD) 2.0 J/kg/m) and that the smallest detectable difference of this measure is 0.464 J/kg/m (or 6.8%) (Brehm et al., 2006). Considering this, a 10% improvement in the EC of walking was defined to be clinically significant.

2.3.3 Activity measurements

Raw accelerometer data was transferred from the sensors to a computer using the Axivity AX3 sensor software Omgui. The total three or four sensor output data channels were plotted together in Matlab. This resulted in timelines of accelerometer activity per day over the entire week, with a cut-off point before 07:00 and after 23:00. An example of a weekly overview is attached as Appendix 2. The time periods when AFO channels were active and corresponding to movements with the thigh and L3 sensors were extracted from this overview manually, to provide time periods with AFO on for further processing. This was also done manually to ensure the AFOs were corresponding to movement with the other sensors, and not, for example, being carried by others. An example of sensor activities not corresponding and therefore not classified as in use by the participant is shown in figure 2.1.

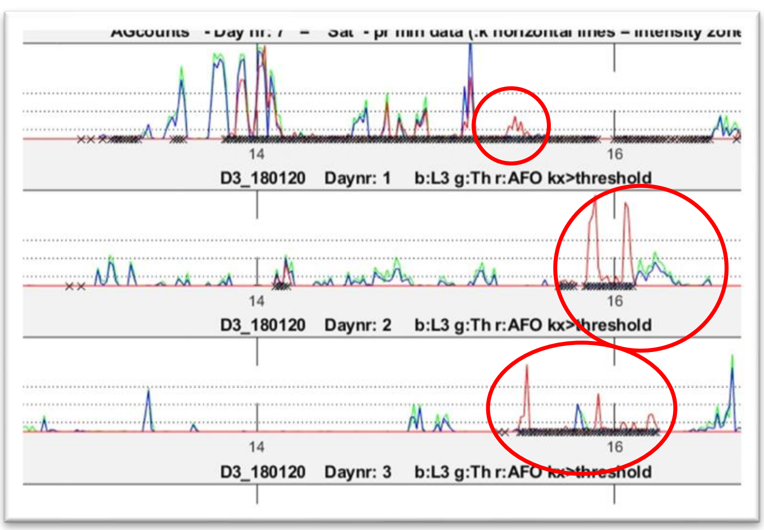

Figure 2.1. Examples of ankle-foot-orthosis (AFO) sensor activity not corresponding to third lumbar (L3) and thigh sensor activity, leading to them not being classified as a time period of AFO usage. L3 sensor is marked by blue line, thigh sensor by green line and AFO sensor by red line.

Raw data from the L3 and thigh sensors were processed by NTNU's Department of Computer Science using a human activity recognition model developed for children. For every 3-second periods, data was categorized as walking, standing, sitting, lying, cycling (sitting), cycling (standing), running, ascending or descending stairs. The data received from NTNU was presented in a Windows Excel file, and further processed using a Matlab script. To simplify, activity types were merged by combining «cycling (sitting)» into sitting activities, «shuffling» and «cycling (standing)» into standing activities, and «stairs (up)» and «stairs (down)» into walking activities. To control the quality of the activity recognition analysis, we checked that recognition of walking activity was true for a period of time we knew the participants had a longer walking period - during the gait test in the laboratory on the test day. This was done by manually examining overviews of time spent in the different activity categories per hour, per day, over the entire week. An example of this overview is attached as Appendix 3. This was also done to examine other sources of error (i.e. loss of sensors). If potential errors were discovered, such as long periods or days without other activities than laying and standing, meaning a sensor might have fallen off, the activity diary was checked to exclude other reasons, like sick days home from school. The amount of time spent in the 5 different activity categories was then investigated further through a Matlab script to find total hours of AFO use per week, as well as hours and % of time spent in different activities with/without AFOs.

2.3.4 Links between improvements in EC, user perception and AFO use

To look for any possible trends in links between improvements in EC, positive user perceptions and habitual AFO use, the participants were divided into two groups according to if the AFO reduced or increased the EC of walking with AFO compared to shoes only, and were presented in descending order of the magnitude of difference in EC between the two conditions. Participants were further categorized into low, medium and high degree of AFO use according to the activity measurements results in total hours of AFO use in a week. Low degree of use was set at 0-10 weekly hours, medium at 10-20 weekly hours and high degree of use at 20+ weekly hours, which was based on the results in this group. User perception was denoted with the number of positive (better with AFO), negative (worse with AFO) and neutral (unsure/no difference) answers on the semi-structured interview related to perceived helpfulness of the AFO.

3 Results

3.1 Participants

Of 12 patients at TOV asked to participate, 2 declined without giving a reason and 1 was scheduled for an operation with a following period of serial casting, leaving n=8. Participant characteristics are shown in table 3.1.

The included participants consisted of three boys and five girls with a mean age of 11,4. 4 were classified as GMFCS level I, and 1 as GMFCS level II. GMFCS classification were missing from available epicrises in 3 of the participants. Five participants used the prefabricated ToeOFF® (AllardAFO, 2019) orthosis or variants of this, all of them with individual adjustments. One participant had a custom-made carbon fibre orthosis with a Becker TripleAction® ankle joint (Becker Orthopedic, Detroit, Michigan) that allowed for more degrees of ROM both in plantarflexion and dorsiflexion. The remaining 2 participants used a custom-made orthosis in 2-3mm copolymer with a Tamarack Flexure® ankle joint (Tamarack Habilitation Technologies, Inc, Minnesota, Usa) allowing free dorsiflexion with a plantarflexion stop at neutral (90˚ in the ankle). Similar orthoses are shown in figure 3.1a-c.

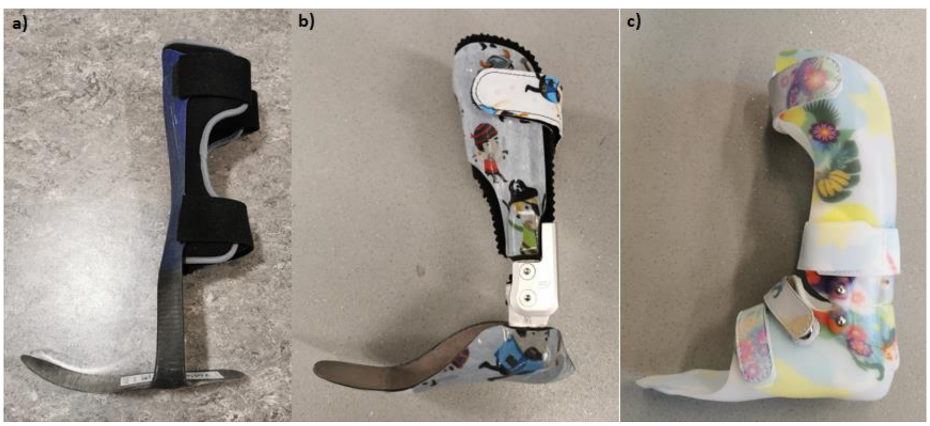

Figure 3.1. Pictures of similar ankle-foot-orthoses as used by the participants. a) ToeOff® Blue Rocker. b) Custom carbon fibre AFO with spring-adjusted joint. c) Custom copolymer polypropylene AFO with free dorsiflexion-plantarflexion stop joint.

Table 3.1. Participant characteristics

ID	Age	Gender	GMFCS	Afflicted side	Height	Weight	LLD *(cm)*	**ROM ankle joint*** straight knee/bent knee *Left*	*Right*	AFO
D1	7	M	I	R	117	18,2	-0,8	15/31	10/20	KiddieGait®◊ with custom-made footbed with 4mm thickness, +5mm wedge under heel of orthosis
D2	9	M	I	R	131,5	30,2	-0,7	10/22	0/5	Custom-made carbon fibre AFO with semi-flexible footplate, anterior calf support, TripleAction® Becker joint
D3	17	M	÷	R	168,8	57	-2	6/17	0/8	ToeOFF® with custom-made footbed with 8mm thickness under the heel
D4	12	F	÷	L	158,8	50	-1,1	÷	5/17	Custom-made ToeOFF® with custom-made footbed with 6mm thickness under the heel
D5	15	F	II	L	÷	÷	-1,5	-7	-1/14	BlueRocker®◊ + 15mm sole lift on shoes
D6	9	F	÷	L	141,9	33,6	-0,4	-1/12	10/18	Custom-made 3mm copolymer polypropylene AFO with Tamarack joint, free dorsiflexion, plantarflexion stop at 90˚ in ankle
D7	13	F	I	R	155,5	34	-1,1	÷	÷	Custom-made 3mm copolymer polypropylene AFO with Tamarack joint, free dorsiflexion, plantarflexion stop at 90˚ in ankle
D8	9	F	I	L	135,5	28,8	-3	÷	÷	KiddieGait®◊ with custom-made footbed with 5mm thickness under heel, 10mm sole lift on shoes

Abbreviations: ID=identification number, GMFCS=Gross Motor Function Classification System, LLD=leg length discrepancy, ROM=range of motion, AFO=ankle-foot-orthosis, M=male, F=female.

*ROM of the ankle joint is described in degrees from neutral toward dorsiflexion, where a negative number indicates degrees from neutral towards plantarflexion. ◊Variants of the ToeOFF® model. ÷Missing data.

3.2 Semi-structured interview

3.2.1 Reported AFO use

The results from the interview regarding reported AFO use at home, at school and in leisure time are presented in table 3.2.

Table 3.2. Self-reported AFO use at home, at school and in leisure time.

		Almost always	Often	Some times	Hardly ever
AFO use at home	D1				**X**
	D2				**X**
	D3				**X**
	D4				**X**
	D5				**X**
	D6				**X**
	D7				**X**
	D8				**X**
AFO use at school	D1	**X**			
	D2	**X**			
	D3		**X**		
	D4				**X**
	D5	**X**			
	D6			**X**	
	D7			**X**	
	D8		**X**		
AFO use in leisure time	D1	**X**			
	D2		**X**		
	D3		**X**		
	D4				**X**
	D5	**X**			
	D6				**X**
	D7				**X**
	D8				**X**

Abbreviations: AFO=ankle-foot-orthosis

Reported AFO use at home

All the participants answered that they hardly ever used their AFOs at home. A recurring theme when trying to map why they did not use their AFO at home, 6 out of 8 participants brought up not wearing shoes inside as one of the primary reasons. Other recurring themes were that being at home was «free time», that they mostly just sit around relaxing, or that they did not feel the need. One participant reported that the AFO was uncomfortable.

> *…Often when I'm at home, I just sit around, relaxing. [The AFO is] mostly used when I'm outside, wearing shoes…*

Reported AFO use at school

AFOs were almost always used at school with 3 of the participants, and often with 2 of them. Only one participant hardly ever used their AFO in school, but the settings of use were very variable within the group. A few of them used their AFO both inside, outside and in gymnastics class, while others did not use the AFO for higher activities or indoors. The topic of shoes also seemed to be a recurring theme for why or why not AFOs were used at school.

> *...I use the AFO both inside at school, in the recesses and in gymnastics class. I use them in inside shoes because we are supposed to wear inside shoes at school…*

Some participants had a second AFO that they wore with inside shoes, which seemed to facilitate for a higher degree of use, as moving the AFO from one pair of shoes to another was brought up as an irritant or hard task to do by themselves. In the three participants using their AFO some times (2) or hardly ever (1), both pain from pressure or the AFO causing limitations in movement were some recurring themes, and one participant mentioned that they didn't want their peers to see and ask questions.

> *…Sometimes I have to take it off, because it can be a little painful. I don't use it when I'm going to run, because it feels strange……*

Reported AFO use in leisure time

Four participants used their AFO almost always (2) or often (2) in leisure time outside of the home, but in most cases it was only reported in «outside wear» in terms of walking or playing. Most participants reported using their AFO for *walking*, moving from point A to point B, but a recurring theme was that AFOs were not used in specific activities with higher demands, like football, handball, dancing, running or similar. The remaining four participants reported hardly ever using their AFOs in the context of specific leisure time activities, because the AFO caused limitations in movements.

…I use the AFO always outside and at shooting practice, but never at football practice. I haven't tried yet with the new brace, but the old one was too stiff in the ankle when I tried to kick the ball…

…The AFO is limiting me in running. At handball practice I just use my footbed [custom made insole], *and then I feel more at level with the others…*

3.2.2 User perception

Results from the interview regarding user perceptions are shown in table 3.3.

Keeping up with peers

When asking if keeping up with peers was easier with or easier without AFO, only two participants reported that the AFO was helpful, in terms of making it easier to walk fast or to run. Three participants felt no difference between the two conditions, and three answered that the AFO made it more difficult to keep up with peers, stating for example changing between shoes as an irritant when trying to get fast into the playground during school breaks; that they feel faster without the orthosis; and, for one participant, that walking was easier with the orthosis, but running was harder.

Feeling of stability

When asking about feeling of stability, four participants did not feel a difference or were unsure. The remaining four felt more stable using their AFO. In some of the cases, where the child was unsure or felt no difference, parents unprovoked commented that they felt the AFO made the children more stable, that they were stumbling less, and seeing the difference in joint alignment.

Table 3.3. User perception of AFO helpfulness in keeping up with peers, feeling of stability, degree of fatigue at the end of the day and walking uphill/downhill.

		Better with AFO	**Worse with AFO**	**Don't know**	**No difference**
Keeping up with peers	D1	**X**			
	D2	**X**			
	D3				**X**
	D4				**X**
	D5				**X**
	D6		**X**		
	D7		**X**		
	D8		**X**		
Feeling of stability	D1	**X**			
	D2	**X**			
	D3				**X**
	D4				**X**
	D5			**X**	
	D6	**X**			
	D7	**X**			
	D8			**X**	
Degree of fatigue at end of day	D1	**X**			
	D2				**X**
	D3			**X**	
	D4				**X**
	D5	**X**			
	D6		**X**		
	D7		**X**		
	D8	**X**			
Walking uphill/downhill	D1	**X**			
	D2			**X**	
	D3			**X**	
	D4				**X**
	D5			**X**	
	D6		**X**		
	D7		**X**		
	D8		**X**		

Abbreviations: AFO=ankle-foot-orthosis

Degree of fatigue at the end of the day

When asking about degree of fatigue at the end of the day, three participants felt less fatigue with AFO, where one of them attributed this to less musculoskeletal pain. Two participants felt more fatigue with AFO, one of them attributing this to more pain with the brace. This was the question sparking most comments from the parents, reporting that this was a difficult question because more activity and walking is usually related to more AFO use, and more activity is related to more fatigue.

…[Guardian] *They are always out walking more when they are using the AFO, so maybe they are more tired because of the level of activity…*

…I feel like you [adressing child] *complain less about pain after days with more AFO use, and that fatigue and pain often go together…*

Walking in uneven terrain

When asking about walking in uneven terrain with an emphasis on uphill/downhill walking, four participants had difficulty answering, feeling that it was just the same, or had not tried without their AFO in a while, making it difficult to compare. One participant felt that it was easier with the AFO both uphill and downhill, while the remaining 3 felt it was more difficult, specifically walking uphill.

Every interview was ended with asking what the participants felt were the best thing and the worst thing with using an AFO, as well as asking if the families felt we should have asked something else or wanted to add anything. Comments in this section was generally low, but recurring themes were difficulties with pain from pressure and chafing from the orthoses. Clamminess and increased sweating were irritants, as well as difficulties with shoes:

…[Guardian] *Well, now they're getting big and more independent, but the thing with moving the AFO from one pair of shoes to another, inside, outside… The wear and tear on shoes is also a thing… It's just that extra bit of hassle, trying to facilitate for getting them out in the recesses at the same time as their classmates…*

3.3 Activity measurements

The results of the activity measurements are presented in table 3.4, 3.5 and 3.6. Data was missing from the L3 sensor the last two days of wear in participant D7, but the AFO sensor was never active except for the test day. As this represents one realistic pattern of use in the group, the data was included, but activity recognition of the last two days were not included in the statistics. The last two days of activity measurements for D5 were also not included due to possible errors or losing a sensor.

Total hours of AFO use during a week ranged from 0 to 34,9, with a mean of 14,6 hours. AFOs were used in the highest degree in walking and standing activities, with a mean 25,2% of total walking activity time and mean 21,1% of total standing activity time. While wearing AFO, 36% (mean) of the time was spent in standing activities, 25% (mean) in sitting activities and 21,7% in walking activities. In total weekly activity, with or without AFOs, 52% of the time was spent in laying, 20,6% in standing, and 8,4% in walking.

Table 3.4. Total hours with AFO on in different activities

	Hours with AFO on in different activities					
	laying	sitting	standing	walking	running	Total
D1	4,5	11,7	11,1	7,1	0,6	34,9
D2	1,9	4,6	11,4	6,0	0,3	24,1
D3	0,1	5,8	3,9	2,8	1,4	14,0
D4	0,0	0,0	0,6	1,3	0,0	1,9
D5*	0,2	8,5	6,1	2,3	0,0	17,1
D6	0,3	5,7	12,2	3,3	0,0	21,6
D7*	0,0	0,0	0,0	0,0	0,0	0,0
D8	0,0	0,9	1,7	0,4	0,0	3,1
Mean	*0,9*	*4,7*	*5,9*	*2,9*	*0,3*	*14,6*

*Last two days of activity measurements not included in analyses due to missing data.
Abbreviations: AFO=ankle-foot-orthosis

Table 3.5. Total hours in different activities and percentage of time spent in different activities

	Hours in different activities					% of time in different activities				
	laying	sitting	standing	walking	running	laying	sitting	standing	walking	running
D1	86,9	29,2	24,3	13,6	1,0	56	19	16	9	1
D2	77,8	22,9	37,3	13,5	0,4	51	15	25	9	0
D3	90,2	16,0	20,0	17,0	11,2	58	10	13	11	7
D4	77,1	23,1	36,4	13,8	0,3	51	15	24	9	0
D5*	58,4	28,9	16,0	4,6	0,0	54	27	15	4	0
D6	69,9	24,5	44,6	11,6	0,1	46	16	30	8	0
D7*	44,0	23,2	18,4	7,4	0,1	47	25	20	8	0
D8	81,2	25,0	34,5	13,9	0,1	52	16	22	9	0
Mean	*73,2*	*24,1*	*28,9*	*11,9*	*1,6*	*51,9*	*17,9*	*20,6*	*8,4*	*1*

*Last two days of activity measurements not included in analyses due to missing data.
Abbreviations: AFO=ankle-foot-orthosis

Table 3.6. Percentage of total time in each activity with AFO on, and percentage of time spent in each activity while wearing AFO.

	For each activity, %of total time AFO on					While wearing AFO, % of time in different activities				
	laying	sitting	standing	walking	running	laying	sitting	standing	walking	running
D1	5	40	46	52	60	13	33	32	20	2
D2	2	20	31	44	75	8	19	47	25	1
D3	0	36	20	16	13	1	41	28	20	10
D4	0	0	2	9	0	0	0	32	68	0
D5*	0	29	38	50	-	1	50	36	13	0
D6	0	23	27	28	0	1	27	57	15	0
D7*	0	0	0	0	0	0	0	0	0	0
D8	0	4	5	3	0	0	30	57	13	0
Mean	*0,9*	*19,0*	*21,1*	*25,2*	*18,5*	*3*	*25*	*36,1*	*21,7*	*1,6*

*Last two days of activity measurements not included in analyses due to missing data.
Abbreviations: AFO=ankle-foot-orthosis

3.4 Energy cost of walking

The results of the walking tests are presented in figure 3.1. Walking energy cost data from one participant (D5) was excluded because the equipment failed during testing.

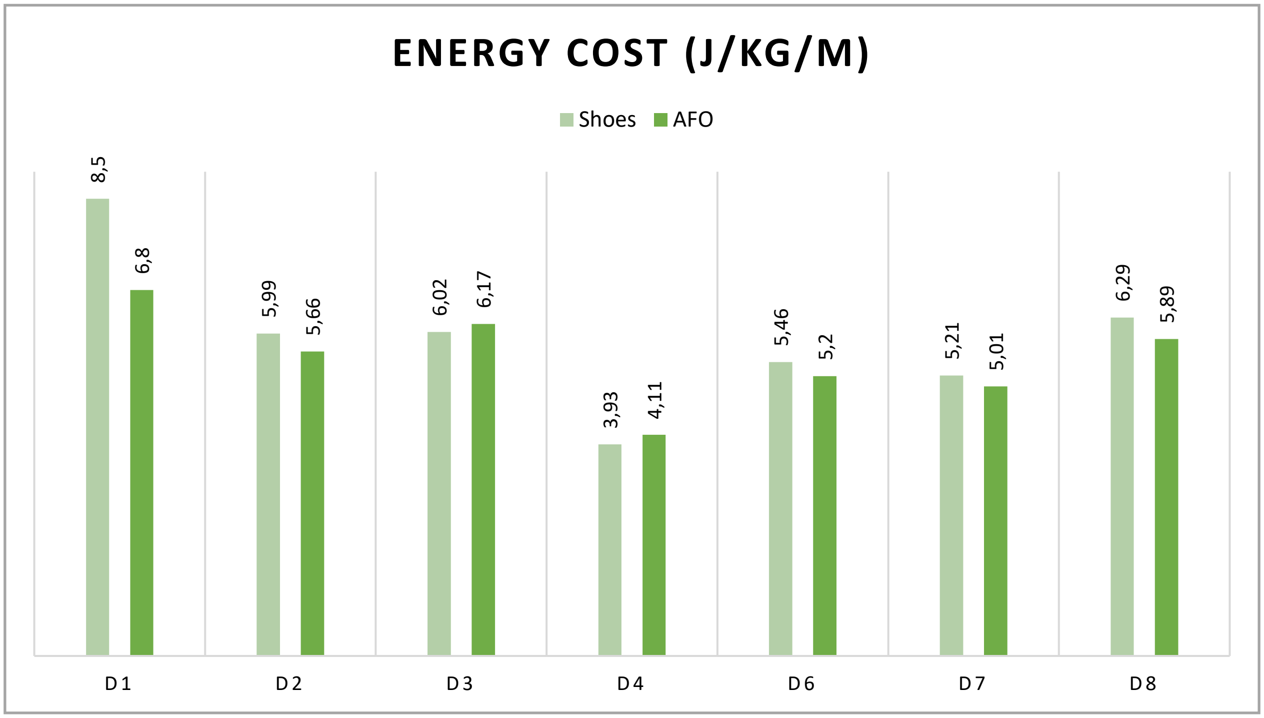

Figure 3.1. The energy cost of walking, in joules per kilogram per meter, comparing walking with shoes only to walking with shoes and ankle-foot-orthoses.

Table 3.4. Order of completion of randomized conditions during walk tests.

	D1	D2	D3	D4	D6	D7	D8
Shoes only	3	2	1	3	3	3	1
Shoes + AFO	1	1	3	2	2	1	2

Abbreviations: AFO=ankle-foot-orthosis

Only one participant had a clinically significant improvement in the energy cost of walking when wearing their AFO, when using a pre-defined threshold of 10% improvement. Four participants had non-significant decreases in EC when walking with AFO, while the remaining two participants had a non-significant increase in EC when walking with AFO.

Gait speed (figure 3.2), calculated as total distance travelled over 3 minutes in both conditions, showed decreases in gait speed with AFO compared to shoes only in five participants, and increases in the remaining two.

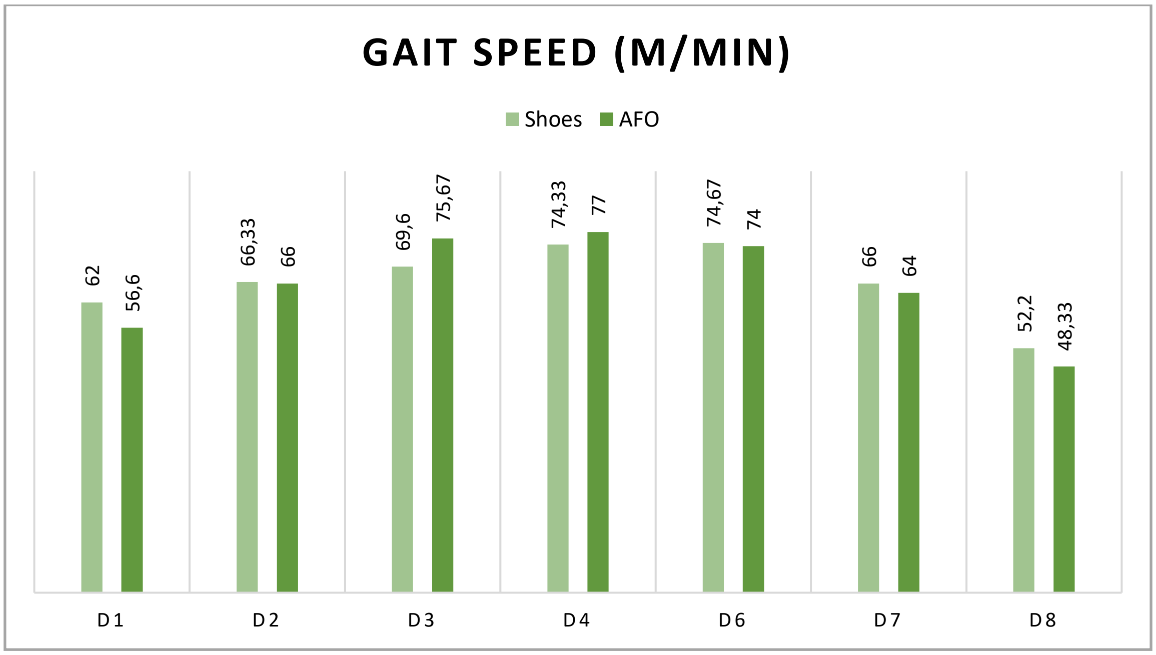

Figure 3.2. Gait speed in meters per minute, comparing walking with shoes only to walking with shoes and ankle-foot-orthoses.

3.5 Improvements in gait efficiency, user perception and habitual AFO use

Table 3.5 shows the characteristics of the participants after being divided into two groups: 1) decreased EC of walking and 2) increased EC of walking with AFO compared to shoes only, where the difference between the two conditions are plotted in descending order. Because the equipment failed during the walking tests in participants D5, that data was excluded. D1 was the only participant with a clinically significant decrease in the EC of walking when using their AFO, had the highest total hours of AFO use and gave the highest number (4/4) of positive answers on the semi-structured interview related to perceived helpfulness of the AFO. No other plausible trends were discovered.

Table 3.5. Participant characteristics in terms of level of AFO use and number of positive (better with AFO), negative (worse with AFO) or neutral (unsure or no difference) answers on subjective perception of AFO helpfulness in keeping up with peers, feeling of stability, degree of fatigue at the end of the day and walking in uneven terrain; divided into group 1) Decrease in EC when walking with AFO, and 2) Increase in EC when walking with AFO, compared to shoes only.

	Decreased EC with AFO					**Increased EC with AFO**	
Participants	D1	D8	D2	D6	D7	D4	D3
Difference in EC (J/kg/m)	-1,7†	-0,4	-0,33	-0,26	-0,2	+0,18	+0,15
AFO use level (hours)	High (34,9)	Low (3,1)	High (24,1)	High (21,6)	Low (0,0)	Low (1,9)	Medium (14,0)
User perception◊	4☺	2☹ 1☺ 1😐	2☺ 2😐	3☹ 1☺	3☹ 1☺	4😐	4 😐

Abbreviations: EC=energy cost, AFO=ankle-foot-orthosis.

*Difference in the EC of walking was calculated by subtracting the lowest number from the highest number, where negative sign describes a decrease, and a positive sign describes an increase in EC when walking with AFO compared to shoes only.

†Clinically significant decrease.

◊User perception describes number of positive ☺, negative ☹ or neutral 😐 answers.

4 Discussion

The primary research aim of this study was to investigate when, how much and in what activities children with CP use or do not use their prescribed AFOs, and what the user perception was. A secondary aim was to investigate if improvements in gait efficiency was linked to more positive user perceptions and a higher degree of habitual AFO use.

The main results show that use of AFOs in daily life was highly variable within the group, ranging from 0 to 34,9 weekly hours of wear, with a mean of 14,6 hours. Longer mean wear times than this has been reported in another study relying on user feedback, where five participants reported wearing their AFO for a minimum of 18 hours a week (6-8 hours per day, 3-7 days a week) (Eddison et al., 2020). In the same study they reported that AFOs were mainly being worn at school (Eddison et al., 2020), which was replicated in this study where 5/8 used their AFO almost always or often at school, 4/8 used their AFO almost always or often in leisure time and none of the participants used their AFO at home. As reported in the interview, AFOs were primarily used for walking and moving from one place to another, as was confirmed by activity measurements showing 25,2% of total walking activity time was spent with AFO on. Whereas in higher activity demands, such as running, dancing and in sports, AFOs were consistently reported to be limiting in performing the tasks. In total weekly hours of running activities, a mean 18,5% of the time AFOs were used, but there were outliers in this statistic, with a range of 0-75%. Participant D2 used the AFO 75% of the time spent in running activity, but total hours of running time were less than half an hour during the entire week, so this statistic is not representative. AFOs limiting young children in sit-to-stand transfers and floor play were also brought up in Näslund et al. (2003), a study on parent's perception of AFO use in their children. In addition, in the interview regarding perceived helpfulness of the AFO in different settings it was revealed that 3/8 participants reported keeping up with peers and walking uphill as being harder with AFO than without, where 2 participants experienced the AFO as helpful in keeping up, and only 1 in uphill walking, further highlighting an important issue uncovered in this study: the clinically prescribed AFOs in this study sample were mostly reported to be limiting the children in sport and physical recreation, which are important activities for social participation (Clutterbuck et al., 2020). This sample was small and may not be representative, and the children themselves did not seem to be bothered by this restraint, simply choosing to not use their AFO in their sport of choice. But further research into different AFOs effect on high level motor skills is warranted. Looking at the characteristics table (table 3.1), 5 of the participants used the original ToeOff® (AllardAFO, 2019), with small adjustments made by the orthotist. The dynamics of the carbon fibre in both the foot plate and the lateral stirrup allow for a few degrees of ROM in walking (AllardAFO, 2019), but might be too stiff to make running, uphill walking and larger positional changes easy. Nonetheless, the two participants (D6, D7) with free dorsiflexion joints in their AFOs still reported uphill walking, dancing and running to be harder with their AFOs and displayed the most negative subjective perception of the group. Many AFOs are bulky and may not be possible to fit into a variety of sport-specific shoes, such as football shoes. Adjustment of the footwear to accommodate AFOs, or development of sport specific AFO designs might be an area of future product development.

Feeling of stability was reported as better with AFO in 4 participants, where the rest were unsure or felt no difference. Degree of fatigue at the end of the day was reported as better with AFO in 3, versus worse with AFO in 2. Where two participants reported more fatigue at the end of the day with AFO, this was related to more pain or irritation with the AFO altering their gait pattern. These two participants (D6, D7) were the only two using custom-made orthoses in thermoplastic, which have a more circular, enclosing construction both around the calf and the foot, thereby providing a higher degree of control, but possibly also with bigger risks of chafing and clamminess, as well as being bulkier in shoes and under clothes. In terms of cosmetic appearance, only one participant reported not wanting their peers to see their AFO and ask questions in this study, and no-one brought up cosmetic issues, while all the participants in Eddison, Healy et al. (2020) reported not liking wearing their AFO-footwear combination because they did not like the way they looked.

When stating reasons for not using AFOs in different settings the connection to shoes, the AFO causing limitations in movements, pain, discomfort or sweating were the recurring themes. Similar themes were found in Näslund et al. (2003), where problems with finding shoes that fit, and the burden of putting on and taking off orthoses were also brought up. Shoes, either as a prerequisite for using their AFO, or problems with changing between shoes were brought up by many of the participants themselves. Six of the participants were using AFOs that must be worn with shoes. Participant D6 and D7 used AFOs that can be worn without shoes, but the plastic can be slippery on floors and may be more optimally aligned in shoes, because the heel height in combination with a given plantarflexion stop results in changes in inclination of the tibia, which have been shown to influence kinematics at the ankle, knee and hip (Jagadamma, 2015; Norman et al., 2004).

In this study on children with unilateral CP at level I-II on the GMFCS, we did not find consistent improvements in the EC of walking when walking with AFO compared to shoes only. This is supported by Brehm et al. (2008), who did not find decreases in the EC of walking in children with unilateral CP, when compared to barefoot condition. Only one participant (D1) had what we can call a clinically significant decrease in the EC of walking with AFO compared to shoes only. Together with 5 other participants, a decrease in the EC of walking with AFO also coincided with a decrease in gait speed, as well as an increase in EC coincided with an increase in gait speed for the last 2 participants. The relationship between speed and EC is not directly linearly related (Weyand, 2013), so it is unlikely that the decrease in gait speed in D1 is the total cause of the significant lowering of EC in participant D1. The relationship between EC and gait speed needs further research to enable evaluation of improvements in EC when allowing self-selected walking speeds. D1 also completed the walking tests in such an order (shoes + AFO first, shoes only third) that fatigue could have contributed to the result, but this did not seem to affect D7, who had the same order of randomization.

Due to a small sample size and qualitative method choices, statistical analyses were not performed. What we can see when looking at all the data, are *trends:* The only participant with a clinically significant decrease in the EC of walking when walking with AFO was the same participant that reported the most satisfaction with AFO in terms of subjective experience of AFO helpfulness and reported using the AFO almost always at school and in leisure time. They also had the most total hours of AFO use in a week according to the activity measurements and used their AFO in 58% of the total time spent in walking activities. Participant D6 and D7 reported using their AFO some times in school, and never in leisure time and at home, and had the most negative perception of AFO helpfulness in the group. D7 never used their AFO during the week, except during data collection, while D6 still used their AFO in 28% of total time in walking activities and 27% of standing activities. They both had non-significant decreases in the EC of walking when walking with AFO, even though they both performed the walking test with shoes only last. No other plausible trends were discovered, except that high level users and number of positive and negative answers were clustered slightly in the group with decreased EC, and low-medium level users with only neutral answers were clustered in the group with increased EC. But in this small sample, where only 7 of the 8 original participants were included, and only 2 participants were in the increased EC group, no conclusions can be made.

One participant (D4) reported feeling no difference on all aspects of the subjective experience part of the interview, had the lowest energy cost of walking in both conditions of the group, and was in the higher spectrum of the group in terms of hours of overall physical activity. D4 reported wearing the AFO only sometimes in leisure time, and almost never at school or at home. A comment they made during the interview might explain this pattern, as they reported that their treating physician told them to just wear the AFO during long walks or trips, and to not use it during training sessions or everyday activities. This was probably due to having minimal impairments. They described that the main prescription goal was reducing musculoskeletal pain in the evenings, which was caused by higher activity and after longer walks. This information is interesting and important in terms of analysis, because it explains why the participant had low levels of use. It also highlights the variability within the group and the «problem» with heterogeneity in CP in research. It shows that it might be important to evaluate and report individual prescription goals in future research, as well as during the process of designing studies. With recruitment in mind, maybe a more homogenous sample can be reached by including participants within the same GMFCS classification level and with similar prescription goals. Or else, controlling for GMFCS level, main gait deviation or similar factors in analyses when including mixed classifications and levels should be included, as outliers can have a large influence on results. Choosing outcome measures that are directly related to the participating individuals' prescription goals may also ensure that the results are valuable, clinically relevant, and relevant to the user and participant as well. User perception and user participation in formulating research questions that are important to them might also uncover new themes that may have a larger clinical value, and fit into the framework of the ICF in terms of looking at a person through all the environmental and personal factors affecting them, and not purely in medical terms. After all, the effects on one specific outcome measure, like energy expenditure, is not paramount to the overall effects of AFOs on children with CP. No reduction in oxygen cost does not mean that the AFOs are not doing for the child what they were meant to do. In fact, energy efficiency in gait has been shown to correlate with activity limitations, but is not reflective or participation restriction or general health (Kerr et al., 2008).

This study has multiple limitations. The sample size was small, owing both to the limitations in recruitment possibilities within the city of Trondheim, and missed opportunities for recruitment throughout the year due to the emerging Covid-19 pandemic in the spring of 2020. Small sample sizes seem to be a recurring limitation in studies on AFO use in children with CP (Balaban et al., 2007; Bjornson et al., 2016; Figueiredo, 2008) and might highlight the need for well-designed multi-center studies and cooperation across both national and international borders. With the qualitative aspect of this study in focus, the limitation of few participants is less hard-hitting, and makes the application of thorough within-subjects analyses easier. Other studies have also gained new insight by applying within subject comparisons, like in Bjornson et al., who found no significant group differences between walking with shoes only and walking with AFOs. But when using within subject comparison, two of the participants significantly increased their total steps per day, %walking per day and peak intensity over the other participants when wearing AFOs, coinciding with being the only two subjects having undergone tuning processes for AFO optimization (Bjornson et al., 2016). The reporting of individual details such as ROM in the lower extremities and the construction details of the AFOs is easier with smaller study populations, and is important to ensure confidence in a study's findings, the generalizability of the results and give readers the possibility to assess the quality of the intervention (Ridgewell et al., 2010). Presenting the results individually also limits effects from outliers when relying on central tendencies.

A bigger limitation of this study is the use of a non-validated questionnaire/set of interview questions, especially considering the interview subjects were young children. In hindsight, the important aspect of musculoskeletal pain should also have been incorporated as a question, and pain was brought up, unprovoked, by some of the parents when talking about fatigue at the end of the day. Also, the interview seemed to be difficult for the children in terms of remembering and comparing situations with and without AFOs, especially regarding subjective perception of AFO helpfulness. In most cases, this was due to having no experiences of it, for example of walking uphill/downhill without AFO, because they always wore their AFO outside in walking. Maybe the subjective experience of walking in rough terrain, degree of fatigue, keeping up with peers and added questions about degree of musculoskeletal pain would have been easier to record in a diary during the 7 days of activity measurements, with questionnaires they could complete at the end of the day with their parents/guardians.

During the interview process, without asking explicitly for it, there seemed to be a recurring theme of the children not having a real understanding for why they used AFOs. When asking why or why not they used their AFOs in the different settings and activities, hoping for specific reasons related to function, comfort or similar, the answer was more often related to «*Dad says I have to wear it*». Most AFOs are prescribed not only to improve gait function, but to prevent long-term deformities (CPUP, 2020; Wingstrand et al., 2014). This is an important aspect that is probably not regarded by the children themselves, and an aspect that must be weighted when designing studies just looking at habitual use and user perspective in children.
AFOs are usually prescribed by the treating paediatrician with specific goals, but communication with the children themselves is important to evaluate their effect and map motivation for adherence, remind them what the reasoning and goal is, and evaluate together what can be done to achieve them. As this study has discovered some recurring limitations for AFO use, especially in higher activities and walking in rough terrain, it shows the importance of asking follow-up questions to map any problem areas and limitations the AFOs put on them.

4.1 Conclusions

AFO use was variable within the group, but were mainly used in school, in walking and standing activities, and mean adherence was lower than previously reported findings. Subjective perception of AFO helpfulness was also highly variable, but especially in higher activities, such as running and sports, the AFOs were mostly perceived as limiting the children. The connection between most AFOs and shoes were brought up multiple times; changing the AFO between shoes was an irritant and limiting factor, while having an extra AFO attached to other shoes was a facilitating factor for more use. The only participant with a clinically significant improvement in

EC with AFO on had the most hours of AFO use and reported perceiving the AFO as being more helpful than the other participants.

4.2 Future directions

Future studies should aim to investigate associations between improvements in gait and their direct effect on day-to-day life of children with CP, for example through intervention studies where they have longer periods with and without AFOs. It should also be investigated how AFOs affect children in activities with higher demands such as speed, transitional movements and ROM, to inform prescription and maybe even advise product design. Longitudinal studies on AFOs effect on the prevention of deformities could provide a better base of evidence for prescription, and support reasoning for maintaining adherence when talking to the users themselves and their families.

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Appendix 1

Se for deg en vanlig uke den siste tiden:

<u>Hjemme</u>

- Hvor ofte bruker du AFO når du er hjemme?
  - Nesten alltid ☐
  - Ofte ☐
  - Av og til ☐
  - Nesten aldri ☐
- Hvorfor bruker du ikke AFO hjemme (er det spesielle situasjoner/forhold)?:
- Hvis sjelden/aldri, hvorfor ikke? Svarene kan også rangeres hvis det er flere årsaker:
  - Komfort ☐
  - Utseende ☐
  - Ikke riktig funksjon ☐
  - Trenger ikke ☐
  - Annet:

<u>På skolen</u>

- Hvor ofte bruker du AFO på skolen?
  - Nesten alltid ☐
  - Ofte ☐
  - Av og til ☐
  - Nesten aldri ☐

- Er det spesifikke aktiviteter du bruker den/de, eller ikke bruker den? (For eksempel friminutt, kroppsøving)
  - Bruker:
  - Bruker ikke:

- Hvorfor/hvorfor ikke?

<u>På fritiden (ikke hjemme)</u>

- Hvor ofte bruker du AFO i fritiden?
  - Nesten alltid ☐
  - Ofte ☐
  - Av og til ☐
  - Nesten aldri ☐

- Er det spesielle fritidsaktiviteter du alltid/ofte bruker AFO og hvorfor?
- Er det spesielle fritidsaktiviteter du sjelden/aldri bruker AFO?
  - Hvorfor/hvorfor ikke?
    - Komfort ☐
    - Utseende ☐
    - Ikke riktig funksjon ☐
    - Trenger ikke ☐
    - Annet

<u>Subjektiv opplevelse av AFO-bruk:</u>

Jeg vil nå spørre deg litt mer om hvordan du selv opplever å bruke AFO'en din.

<u>Sirkle rundt riktig svar</u>

- Føler du at AFO hjelper deg å holde **mer eller mindre** følge med vennene dine?
  - Ja, ofte ☐
  - Av og til ☐
  - Nei, sjelden ☐
  - Annet:

- Føler du deg **mer eller mindre** sliten på slutten av en dag når du har brukt AFO enn når du ikke har brukt AFO?
  - Ja, ofte ☐
  - Av og til ☐
  - Nei, sjelden ☐
  - Annet:

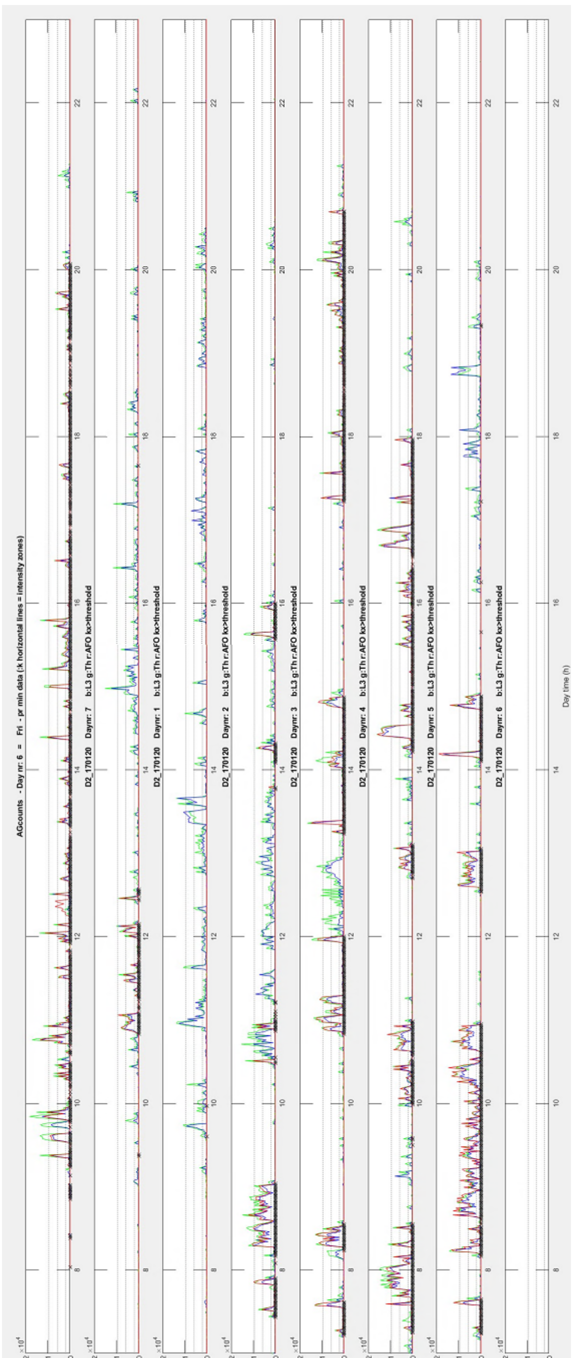
- Føler du at AFO'en din gjør deg **mer eller mindre** stabil?
  - Ja, ofte ☐
  - Av og til ☐
  - Nei, sjelden ☐
  - Annet:

- Føler du at det er **mer eller mindre** vanskelig å bevege deg på ulendt terreng (opp/nedoverbakke) med AFO?
  - Ja, ofte ☐
  - Av og til ☐
  - Nei, sjelden ☐
  - Annet:

- Hva synes du er det verste med å bruke AFO?
- Hva synes du er det beste med å bruke AFO?

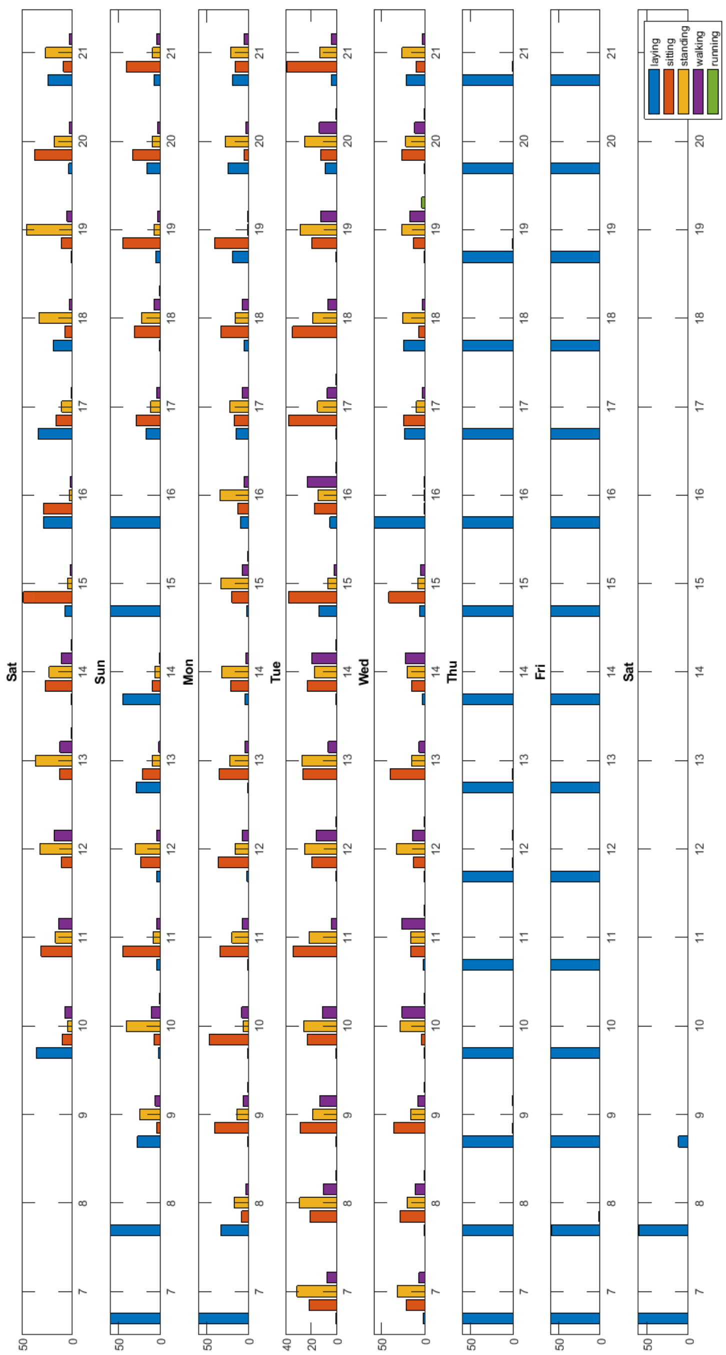
5 Appendix 2

Example of weekly overview of accelerometer activity data, where periods of AFO use were manually extracted.



6 Appendix 3

Example of weekly overview of activity categories per hour. Loss of sensors last two days.



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