

Title: Exploratory investigation into energy expenditure using tuned versus non-tuned ankle foot orthoses- footwear combinations in children with cerebral palsy

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Abstract

Study Design: Within-subjects design.

Background: Children with cerebral palsy (CP) commonly expend two to three times as much energy to walk as typically developing children. Research shows that the effects of non-tuned ankle-foot orthoses (AFOs) on energy expenditure is inconclusive. Tuning of an ankle-foot orthosis-footwear combination (AFO-FC) has demonstrated an improvement in the kinetics and kinematics of pathological gait, particularly knee flexion during stance phase, which are key determinants of an energy efficient gait.

Objective: To compare the submaximal energy expenditure via indirect calorimetry, speed and distance walked, of tuned and non-tuned AFO-FCs and barefoot gait, in children with cerebral palsy (CP).

Methods: Performance assessment of four children aged between 7-10 years with a diagnosis of CP (one hemiplegic and three diplegic participants, two female, two male, with a Gross Motor Function Classification System (GMFCS) of 2) at a Gait Analysis Laboratory.

Results: There was a reduction in gross submaximal energy expenditure and energy efficiency index (EEI) based on O₂, in three out of the four participants tested when wearing tuned compared to a non-tuned AFO-FC, the reduction ranged from 9.2% to 33.7%. Speed and distance covered also showed improvement in the tuned condition.

Conclusions: Tuning the AFO-FC of children with cerebral palsy has the potential to decrease energy expenditure and increase speed and distance compared to providing a non-tuned AFO-FC.

Clinical Relevance: There is a lack of research on the effect of using a tuned compared to a non-tuned AFO-FC on energy expenditure. This paper provides a comparison of energy expenditure in children with CP, during Barefoot, Non-tuned and Tuned AFO-FC walking, intending to inform clinical practice.

Keywords: Cerebral Palsy, Orthotic Devices. Energy expenditure, ankle-foot orthosis, AFO tuning.

Introduction

Cerebral palsy (CP) has often been considered the prototype childhood ‘neurodisability’(1). It is a group of disorders with widely varying type, timing, location and extent of injury to the brain and has been defined as:

“A group of permanent disorders of the development of movement and posture, causing activity limitation that is attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems”. (1)

50%-80% of individuals with CP will achieve some ability to walk(2,3). Pathological gait associated with CP is widely documented in the current literature(3–10). Patients with pathological gait have abnormal lower limb kinematics, particularly at the shank segment. Attempting to normalise the shank kinematics offers a higher chance of optimum thigh and trunk kinematics and knee and hip kinetics(11,12). Measuring the energy expenditure during walking provides a way to quantify the physiological strain resulting from pathological gait(13).

In pathological gait the ground reaction force (GRF) is further away from the joint centres when compared with normal gait, resulting in an increased joint moment which requires an increase in muscle activation to control the joint, thus, resulting in an increase in energy expenditure and a less efficient gait. Therefore, it is widely accepted that pathological gait requires more energy expenditure than normal gait(14–21). Children with CP commonly expend two to three times as much energy to walk as typically developing children(22), thereby predisposing children with CP to early fatigue in carrying out activities of daily

living. There is a debate as to how pathological changes in gait effect energy expenditure, Saunders, Inman and Eberhart(23) propose that a set of kinematic features help to reduce the displacement of the body's centre of mass (COM). This theory assumes that the vertical and horizontal displacements of the COM require increased energy. However, recent studies indicate that three of the determinants listed (stance phase knee flexion and fore-aft axes) may contribute very little to reducing the vertical displacement of the COM may contribute very little to reducing the vertical displacement of the COM(24). Thus the "six determinants" are perhaps better described as "six kinematic features of gait". Conversely, the inverted pendulum theory(25) proposes that it requires less energy for the stance limb to act like a pendulum with the COM following an arc profile. The pendulum theory also presents a dilemma in that if pendulums can swing freely, why is there an energy cost to walking?(24).The mechanical explanation of the features of pathological gait remains unresolved(24).

In rehabilitation, interventions which improve physical mobility by reducing energy expenditure are important treatment modalities to maintain or enhance independent functioning(15). Ankle-foot orthoses (AFOs) are routinely prescribed to children with CP, in an attempt to improve their gait and function(26). Equivocal findings exist in the literature as to whether the intervention of an AFO can reduce the metabolic cost of walking in CP patients(22,27–33).

Current evidence seems to indicate that there is a reduction in oxygen consumption, with the introduction of an AFO, when walking speed is standardised. A noticeable issue with these studies is that they tend to compare different AFOs with each other, on the same subject. Furthermore, there is no detail regarding the clinical justification for the AFO design and a

dearth of information regarding the design of the AFOs used(34); thereby, contravening the best practice reporting guidelines for research on AFO interventions in children with CP(35). As such, the confidence in the findings are diminished(34), but critically, concerning this research, the AFOs studied were not tuned.

AFO-FC Tuning (biomechanical optimisation)

Ankle-foot orthosis – footwear combination (AFO-FC) tuning can be defined as the process whereby fine adjustments are made to the design of the AFO-FC to optimise its performance during a particular activity. The term biomechanical optimisation is used to encompass the whole process of designing, aligning and tuning the AFO-FC(36).

Tuning of an AFO-FC has demonstrated improvements in the kinetics and kinematics of pathological gait (12,37,38), in particular, knee flexion during mid-stance and knee extension at terminal stance; which are widely accepted to be key factors in an energy efficient gait(23). Detailed information on the tuning process has previously been described(11). Two essential components are the angle of the ankle in the AFO (AAAFO); which can be defined as the angle of the foot relative to the shank in the sagittal plane in the AFO. It is measured as the angle between the line of the lateral border of the foot (base of the 5th metatarsal head to the base of the heel) and the line of the shank and should represent the passive length of the gastrocnemius. Secondly, the shank to vertical angle (SVA) which can be defined as the angle of the shank relative to the vertical, measured in the sagittal plane. The SVA is described as inclined if the shank is inclined forward from the vertical and reclined if it is reclined backwards from the vertical(11).

Although AFO-FC tuning is considered an essential aspect of AFO prescription(39), previous research(40) reported it is not yet standard practice. There is currently no research which has looked at the effect of using a tuned (as defined in this study), compared to a non-tuned AFO-FC, on energy expenditure. The study aimed to compare energy expenditure in children with CP, in three conditions: 1) Barefoot, 2) Non-tuned AFO-FC and 3) Tuned AFO-FC, to better inform clinical practice. The barefoot condition was deemed necessary, to provide a baseline measure of the participants' natural gait with no intervention.

Methods

Participants

Due to CP being a heterogeneous disorder with a wide-ranging topographical presentation single participant research has been advocated, with a focus on functional change at the individual level(41). Therefore, a case series approach was utilised.

Four children aged between 7-10 years with a diagnosis of CP and a Gross Motor Function Classification System (GMFCS) of 2, as determined by an experienced paediatric physiotherapist, took part in this study. All participants were recruited from the author's paediatric orthotic clinic. All participants were long-term solid AFO users (meaning the user had worn AFOs for five years or more). (See table 1 for participant anthropometrics).

Ethical Approval

After necessary approvals from the University Ethics Committee, this study also received ethical approval from the **NRES Ethics Committee West Midlands(Ref: 12/WM/0378)**. The parent/guardian and the child provided written informed consent and verbal assent prior to inclusion in the study, respectively.

Materials

AFO-FC

Each participant was assessed by an experienced orthotist and prescribed with a bespoke solid AFO. The AFOs (see figure 1) used in this study were deemed appropriate for each participant on an individual basis and ensured there was no visible movement of the AFO regarding deformation during stance phase. (See table 1 for AFO design details).

The trim-lines at the ankle finished anterior to the malleolus. The height of each AFO finished 30mm below the fibula head. All the footplates were full length. The AAAFO was determined by an examination of the passive length of gastrocnemius with the knee extended, using a goniometer. If the AAAFO was set in plantar flexion, the AFO had the addition of a shank angle to bench (SAB) build up (see figure1) to ensure the resulting AFO captured the length of gastrocnemius but had a bench alignment of 90°. All participants were issued with the same over splint footwear, in either black or white (Blacky style; Salts Healthcare), which had a heel-to-sole differential of 8mm before any adaptations were added.

Equipment/instrumentation

The area was a dedicated thermostatically controlled gait laboratory with a figure of 8 track to ensure walking was continuous with no abrupt turns. The walkway measured 30.5 metres in total (see figure 2). Its design also precluded bias to the same leg on corners by balancing the number of left and right turns. Two sets of timing gates were set up on the walkway to measure the participant's speed and distance.

A portable gas analyser system (3B Metamax® cortex, Germany) was used to measure oxygen uptake. The 3B MetaMax® is a portable cardiopulmonary exercise system (CPX) for pulmonary gas exchange measurements. During a CPX test with 3B MetaMax®, the participant wears a small facemask, breathing out through a volume transducer fixed to the facemask, which measures volume continuously and simultaneously determines expired CO₂ and O₂ concentration and thus energy expenditure can be estimated from this. From these recordings the calorie uptake can also be determined via the ratio of VCO₂ to VO₂, defined as the respiratory exchange ratio [RER]). The equipment was calibrated before each participant was tested.

Testing procedure

Testing took place over two days, three weeks apart at the gait laboratory: Room temperature and time of testing were kept constant on both testing days to control for time-of-day effects. Day one consisted of barefoot and non-tuned AFO-FC trials and day two consisted of tuned trials. Participants were issued with their non-tuned AFO-FC three weeks prior to visit one, to allow acclimatisation.

Visit 1: Barefoot and non-tuned AFO-FC

Participants were restricted from eating for two hours before the start of the trial, sipping water was permitted. Each participant was fitted with the gas analyser and a heart rate monitor (see figure 3).

Once fitted with the equipment, each participant was given a 20 minute habituation period to allow familiarisation with the testing area, equipment and procedure. 30 minutes rest period,

where the participant remained seated, followed to enable heart rate to return to approximately pre-exercise levels. The order of testing for each condition was randomised.

Testing commenced with each participant sitting for two minutes prior to walking, to establish baseline heart rate and oxygen consumption data. Walking was at a self-selected speed for 3 x 4-minute trials, resting (supported sitting) for eight minutes in-between trials. Each trial commenced once the participant's heart rate was 100 beats per minute or less.

The Metamax equipment was held by the researcher who walked beside the participant to ensure the extra weight of the equipment didn't impede the participant's gait; as such equipment can potentially distort performance(42). It was deemed essential to allow the participant to walk at a self-selected speed to ensure speed and distance could be compared between conditions. There was a 60 minute rest period between conditions, with the second condition (barefoot or non-tuned AFO-FC) carried out on the same day, following the same protocol.

At the end of the testing period on visit one; each participant had their AFO-FC tuned by an experienced orthotist, using 2D video vector analysis, to establish the optimum SVA. The tuning process followed Owen's(43) algorithm. Non-tuned in this study means the AFO-FC was not set to an optimum SVA and the footwear was not adapted to optimise entry and exit in gait. However, it was deemed unethical to supply the participants with an AFO, which did not have the correct AAAFO to represent the length of gastrocnemius, as doing so may have caused the participant pain and put them at risk of a pressure sore.

Once the SVA was determined, the footwear was sent for permanent modification (see figure 4) and returned to the participants within five working days. Following this, a period of three weeks was allowed, to enable acclimatisation to the tuned AFO-FC, before testing.

Visit 2: Tuned AFO-FC trials

Participants followed the same protocol as visit one, this time wearing a tuned AFO-FC.

Data recording methods

Data recording using the Metamax cortex, timing gates and heart rate monitor was commenced simultaneously, recording throughout in parallel.

All data were taken as an average of the three trials in each condition, from minute 5-6 of each trial (minute 3-4 of the walking trial), ensuring cardiovascular steady state had occurred (See figure 5). Steady state occurs when the body has adjusted to the workload and oxygen uptake plateaus. It has been previously reported that steady state whilst walking, usually occurs between minutes two and four(44).

Calculating energy cost measures

The VO_2 was collected breath-by-breath and then averaged over 15 second epochs. The data was then averaged for the 4 x 6 minutes trials, equalling 24 data points in total.

- Gross submaximal energy expenditure (VO_2 mL/minute/metre)

VO_2 (Volume of oxygen uptake per mL/min/m). Gross energy expenditure is the total energy required for an activity, whilst net energy expenditure subtracts resting energy from the total energy produced. Net energy expenditure reduces the effects of the variables that may change

over time such as altered cardiac or pulmonary function(45). As testing for all conditions in this study was only three weeks apart, this was not a relevant factor to consider. Therefore, Gross rather than net energy expenditure was calculated for this study.

- EEI based on O₂

The EEI based on O₂ indicates the amount of energy required to walk a specified distance and reflects energy economy(17,46–50) and is measured by O₂ uptake per kilogram of body weight per minute (VO₂mL/kg/min), divided by walking speed (m/sec

- Walking speed

Walking speed = metres per minute

$$\frac{\text{Distance (metres)}}{\text{Time}}$$

- Calorie uptake

Measured in kilocalories (kcal)

Average volume of oxygen in litres (VO₂L) during walking minute 3-4

Respiratory quotient (RQ)

RQ value corresponds to a caloric value for each litre (L) of O₂ produced.

RQ = CO₂ eliminated / O₂ consumed

VO₂L x Calorific equivalent of O₂ = kcal per minute

Statistical methods

Because of the study design, sample size and the heterogeneity of CP, descriptive statistics were employed instead of inferential statistics.

Results

Gross submaximal energy expenditure

The results indicate a reduction in gross VO_2 in three of the four participants tested when using a tuned AFO-FC compared to non-tuned and barefoot gait. The reductions ranged from 10.2% - 33.7%. Results for participant five show that gross VO_2 increased by 14% in the tuned condition compared to barefoot walking, and there was very little difference between the tuned and non-tuned conditions (3%). (See figure 6).

EEI (O_2)

The results also indicate that the EEI (O_2) was lowest for the same three out of the four subjects in the tuned condition compared to non-tuned and barefoot gait, with one subject (subject five) showing no difference between tuned and non-tuned conditions. The reductions ranged from 3.2% - 31%.

Distance, speed and calories (Kcal) used

All of the participants covered the most distance in the tuned condition compared to non-tuned, the speed of all participants was also highest in the tuned condition when compared to non-tuned, and ranged from an increase of 1.5% to 12.4%. The number of calories (Kcal) increased in the tuned condition when speed also increased, for subjects four and five. However, for subjects one and three, the number of calories (Kcal) they used reduced in the tuned condition compared to non-tuned.

Discussion

This study is the first to measure the metabolic responses during walking at a self-selected speed, in children wearing a tuned or non-tuned AFO-FC.

The results reveal that gross submaximal energy expenditure, ($\text{VO}_2\text{mL}/\text{min}/\text{m}$, which will be referred to as gross VO_2), when walking, was lower with a tuned AFO-FC for three out of the four participants tested when compared with the other two conditions(see table 2). The decrease in gross VO_2 ranged from 10.2% - 33.7%, a change in gross VO_2 which meets or exceeds 10% is considered clinically relevant(51). EEI (O_2) was also lowest for the same three out of the four subjects in the tuned condition, with one subject showing no difference between the tuned and the non-tuned condition (see table 2). The findings of this study are in line with the mean economical EEI(O_2) for children with CP, reported by Rose et al.(17), which is 2.9 times higher than that of healthy children. This is important because interventions that decrease the O_2 cost of walking could potentially benefit activities of daily living in children with disabilities(27).

The high O_2 cost of walking in CP is associated with excessive co-activation in the lower limb(27,52). It has been reported that an AFO can provide increased stability during stance, decreasing the co-activation in the lower limb, and reducing the O_2 cost of walking(27). The results demonstrated in this study may indicate that the tuned AFO-FC improved the positioning of the ankle during stance, resulting in a reduction of gross VO_2 . Previous studies(27,28) which showed no reduction in O_2 uptake, at self-selected speeds, are in contrast to the results of this study. However, the AFO-FCs used in this study were all biomechanically optimised, and this may explain why energy expenditure was reduced at self-selected speeds.

All participants covered the most distance in the tuned condition compared to non-tuned (increase range 1.2% - 16.6%). In three of the four participants tested, wearing a non-tuned AFO-FC decreased the distance they covered compared to barefoot walking (range 0.9%-8.1%, see table 2). Thus, possibly indicating that the intervention of a non-tuned AFO-FC, rather than improve, actually hindered their walking ability.

Similar results are shown in the speed of each subject, with all participants increasing their speed in the tuned condition when compared to non-tuned; with the increase ranging from 1.5% to 12.4%. Interestingly, although speed increased, three participants also reduced their gross VO_2 . The increase in the gross VO_2 in the non-tuned condition (compared to tuned) may be due to the reduction in their speed, which increases the mechanical power required to maintain the body in motion(17). Three of the four subjects had a speed of 49-58 m/min, which is significantly below that of the speed of healthy children(42).

Not surprisingly, the number of calories (Kcal) used increased in the tuned condition when speed also increased for subjects four and five. However, for subjects one and three the number of calories (Kcal) used reduced in the tuned condition compared to non-tuned even though their speed increased, which suggests the tuned AFO-FC provided a more efficient gait pattern (see table 2).

Thus, in summary, participant one performed better against all measures, in the tuned AFO-FC when compared to non-tuned, with the non-tuned condition resulting in deterioration against all measures compared to the barefoot condition. Similarly, participant three's results indicate the same improvements in the tuned condition, compared to non-tuned. Participant four demonstrated an improvement in all conditions except calories (Kcal) used, in the tuned

condition. However, the increase in calories (Kcal) is not unexpected since speed and distance both increased. Participant five's results indicate an improvement in speed and distance in the tuned condition compared to non-tuned, but an increase in gross VO_2 and calories (Kcal) and no difference in EEI (O_2).

The increase in gross VO_2 for participant five is not in line with the results from the other participants, although the increase versus the non-tuned condition is only 3%, the increase against the barefoot condition is 17%. The increase in speed and distance may have contributed to the increase in gross VO_2 , although the reason why this occurred in this participant and not the other three is unknown.

Limitations of the study

The authors recognise that the sample size used in this study is small. However, CP is an extremely heterogeneous disorder, and as such, the aim was to look at the effects of the intervention on the individual participant, in contrast to the vast majority of studies in the available literature, which emphasise group and mean differences(41).

The AFOs in the non-tuned condition had the correct AAAFO as dictated during the patient assessment; this is an essential aspect of AFO-FC tuning. It is hypothesised that setting the AFOs to an incorrect AAAFO, as is common in clinical practice(40), would have further increased energy expenditure but this was deemed unethical. A treadmill was considered to ensure constant velocity; however, treadmills are impractical in clinical applications involving participants with CP as participants with disabilities have difficulty adjusting to walking on a treadmill(14,15). Furthermore, if data are to be used to aid clinical decision making, it is preferable for it to be collected on level ground(53). Additionally, unregulated

walking reduces so-called velocity artefacts that result from artificially imposed conditions(29). The literature also notes that in both disabled and able-bodied individuals, the most efficient rate of ambulation is very close to the individual's freely chosen velocity(29) and enforcing the participants' speed may result in modifications to the gait pattern.

The researcher held the portable gas analyser system, whilst the participant walked ahead, it is possible that this affected the participants' self-selected walking speed. However, the researcher was an able-bodied adult, therefore; it is unlikely that they would not be able to maintain the walking speed of a disabled child.

A further limitation is that no data was collected with footwear alone, to use as a comparison between barefoot and the AFO conditions. However, this study aimed to compare the effects of tuned and non-tuned conditions and the footwear during these conditions remained the same. The footwear used were over splint orthopaedic footwear which are designed to be worn with an AFO. Thus, another shoe would have had to be used, which would mean the results would not have been comparable to the AFO conditions, as it is widely documented that footwear is a crucial aspect of the AFO prescription.

7.8 Conclusion

The results of this study indicate that tuning an AFO-FC can potentially reduce energy expenditure and increase speed and distance covered during walking at a self-selected walking speed, in children with spastic cerebral palsy. The first aim of any clinical intervention is to do no harm; however, this study indicates that the introduction of a non-tuned AFO-FC has the potential to increase energy expenditure and reduce speed and distance in children with CP.

Further research is required on a larger sample to validate these findings and learn more about which patients benefit the most from AFO-FC tuning, and why.

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Table and Figure Legends

Table 1: Participant anthropometric data and AFO design (AAAFO = angle of the ankle in the AFO, SVA = shank to vertical angle)

Table 2: Energy expenditure average of all three trials per subject, measured in Volume of oxygen in millilitres per minute per metre walked. Energy expenditure index (EEI) based on VO_2 , speed, and distance on per subject per condition.

Figure 1 and 1A: Example of an AFO with a SAB build up

Figure 2: Diagram of the layout of the walkway

Figure 3: Participant wearing a gas analyser and heart rate monitor

Figure 4: Permanently adapted footwear

Figure 5: A graph showing the cardiorespiratory steady state was achieved during minute 5-6. The walking trial began at minute 2.

Figure 6: Comparison of Gross VO_2 in barefoot, non-tuned AFO-FC and Tuned AFO-FC

Supplier List

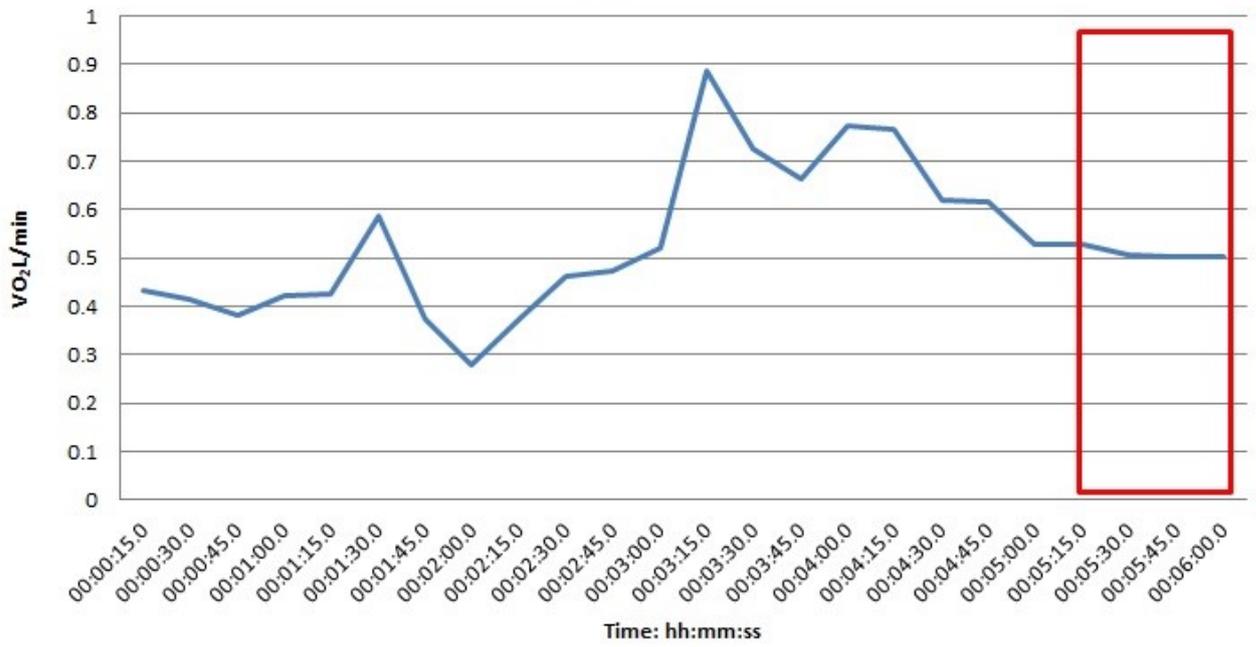
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UK
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Cortex Biophysik GmbH
Walter-Köhn-Straße 2d, 04356 Leipzig, Germany
Germany

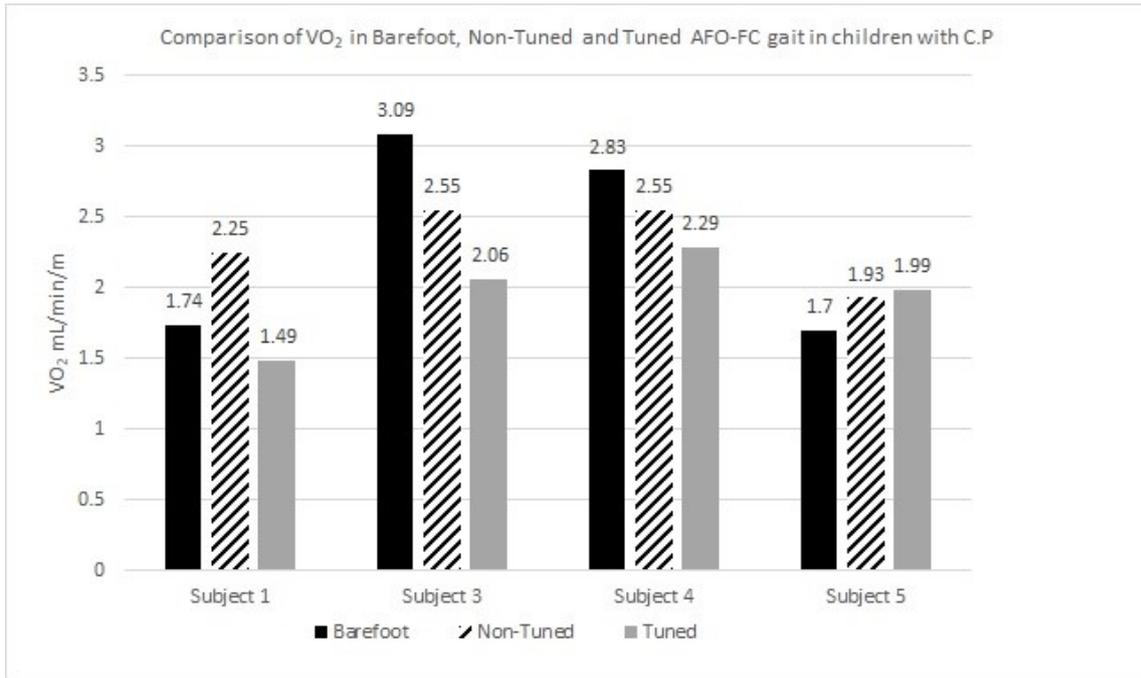
| Subject ID | CP Classification | Sex | Body Mass (kg) | Age (Years) | Stature (cm) | Passive length of gastrocnemius with knee extended | Bare foot gait classification | AAAFO | AFO | Foot Plate |
|------------|---------------------------------------------------------|-----|----------------|-------------|--------------|----------------------------------------------------|-------------------------------|----------------------------|---------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| 1 | Spastic hemiplegic right side affected | F | 23.6 | 8 | 122 | 5° dorsiflexed | Group II (Winters (4)) | 90° | Right solid AFO | Full length with M-L flanges distal to 5 th MTPJ to control fore foot abduction. Flexible at the MTPJs to facilitate 3 rd rocker. Flexible sole rounded profile. |
| 3 | Spastic diplegic | F | 27.7 | 7 | 131 | 90° | Group IV (Winters (4)) | 90° | Bilateral Solid AFO | Full length, carbon fibre stiffener. M-L flanges distal to MTPJs to block 3 rd rocker and limit knee flexion Sole unit stiff with a point loading rocker. |
| 4 | Spastic diplegic with left side predominantly affected | M | 31 | 10 | 140 | 8° plantar flexed | Group IV (Winters (4)) | 8° Plantar Flexion SAB 90° | Left Solid AFO | Full length, M-L flanges distal to MTPJs to block 3 rd rocker and limit knee flexion. Sole unit stiffened with a rounded forefoot rocker. |
| 5 | Spastic diplegic with right side predominantly affected | M | 25.8 | 9 | 131 | 90° | Group II (Winters (4)) | 90° | Right solid AFO | Full length, M-L flanges proximal to MTPJs flexible to facilitate 3 rd rocker Flexible sole, rounded profile. |

| Subject | Condition | Speed (metres per minute) | Distance (metres) | Energy expenditure (kcal per hour) | EI (O ₂) | Mean average VO ₂ mL/min/m |
|---------|------------------|---------------------------|-------------------|------------------------------------|----------------------|---------------------------------------|
| 1 | Bare foot | 71.25 | 285 | 145.6 | 0.3 | 1.74 |
| | Non-tuned AFO-FC | 65.5 | 262 | 172.2 | 0.38 | 2.25 |
| | Tuned AFO-FC | 69.5 | 278 | 126.3 | 0.29 | 1.49 |
| 3 | Bare foot | 46.27 | 185.1 | 171.1 | 0.4 | 3.09 |
| | Non-tuned AFO-FC | 44.29 | 177.2 | 131.4 | 0.36 | 2.55 |
| | Tuned AFO-FC | 52.39 | 210.2 | 126.8 | 0.29 | 2.06 |
| 4 | Bare foot | 36.4 | 145.6 | 118.8 | 0.36 | 2.83 |
| | Non-tuned AFO-FC | 45.6 | 182.7 | 133.8 | 0.32 | 2.55 |
| | Tuned AFO-FC | 49.91 | 199.7 | 139 | 0.31 | 2.29 |
| 5 | Bare foot | 56.6 | 226.6 | 112 | 0.27 | 1.7 |
| | Non-tuned AFO-FC | 56.13 | 224.5 | 127.6 | 0.3 | 1.93 |
| | Tuned AFO-FC | 57.17 | 227.3 | 143.7 | 0.3 | 1.99 |



Participant 5: Non-Tuned AFO-FC Trial 2 VO₂L/Time





Path = 30.5m

