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Abstract
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Document type : Article de périodique (Journal article)

Référence bibliographique
DOI : 10.1111/j.1365-2516.2011.02711.x
ORIGINAL ARTICLE Musculoskeletal

Functional impact of custom-made foot orthoses in patients with haemophilic ankle arthropathy

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Summary. Although foot orthoses are often prescribed to patients with haemophilia (PWH) and ankle arthropathy, the efficacy and biomechanical effects of such devices are not fully understood. We experimentally investigated the effects of orthopedic insoles (OI) and shoes (OS) in PWH presenting ankle arthropathy, with specific attention being paid to pain, spatiotemporal parameters, kinematics and kinetics of lower limb joints, as well mechanical and energetic variables. Using three-dimensional gait analysis (3DGA), synchronous kinematics, kinetics, spatiotemporal, mechanics, and metabolic gait parameters were measured in 16 PWH with ankle arthropathy. The revised Foot Function Index (FFI-R) and 3DGA were determined in patients wearing neutral running shoes at two time points (T0 and T1), with OI (n = 11) or OS (n=5) being subsequently prescribed. Patients, while wearing their orthoses, were re-evaluated using 3DGA, FFI-R, and satisfaction questionnaires (T2). OI and OS provided significant pain relief and comfort improvement in more than half of the patients, with minimal side effects. OI had limited impact on gait pattern, whereas OS significantly improved the propulsive function of the ankle. Biomechanical changes induced by OI and OS were independent of their ability to improve comfort, while being insufficient to influence knee and hip kinematics and kinetics, or mechanical and energetic variables. These findings suggest that OI and OS may have beneficial effects on ankle joints in PWH. Self-reported clinical tools such as FFI-R and satisfaction questionnaires are sufficiently sensitive for assessing the efficacy of foot orthoses in PWH.

Keywords: ankle arthropathy, gait analysis, haemophilia, insoles, orthopaedic shoes, orthoses

Introduction

The ankle is a common site for spontaneous bleeding in patients with haemophilia (PWH). Recurrent haemarthroses may progressively damage the cartilage, bone and soft tissues, resulting in irreversible chronic arthropathy [1,2]. Foot deformities are common in patients with haemophilic ankle arthropathy, including valgus/varus malalignment of the hindfoot and flat/cavus foot, due to growth disturbance of the distal tibia, talar deformity and subtalar involvement [1–3]. These deformities may become irreversible in the long-term without appropriate treatment [2], and patients with foot deformities often experience discomfort while walking or standing for long periods [4].

Many conservative therapeutic treatment modalities have been described in PWH with ankle arthropathy, including foot orthoses [2,5,6]. Orthopaedic insoles (OI) and shoes (OS) are designed to prevent or correct deformities as well as improve foot comfort by spreading pressure, unburden painful areas, absorb shock and reduce joint loading [6,7]. They aim to control the amount, rate, and temporal sequence of subtalar joint movements [7].

Orthopaedic insoles are defined as contoured, removable in-shoe devices that are moulded from an impression of the foot [8]. They are made from different soft compressible or semi-rigid materials and incorporate several corrections, including medial or lateral supports, which provide additional stability under the longitudinal arch and metatarsal area (Fig. 1a, b). Orthopaedic insoles are moulded shoes (Fig. 1c, d) that stabilize the foot while standing or walking, and display shock-
absorbing properties that transfer weight-bearing stresses, with a sole form that facilitates the rolling movement of the step [5]. Contrary to OI that promote sagittal plane motion, OS restrict the ankle movements. Although the effectiveness of foot orthoses has been demonstrated in rheumatoid arthritis (RA) [9–13], it is still unclear whether this can be extrapolated to PWH. In a few studies, OI were shown to reduce ankle pain and bleeding [14,15], while improving stability in PWH [16].

The biomechanical impact of OI and OS is not fully understood, as published reports mainly focused on the effects of orthoses at the foot level rather than on the lower limbs and global gait pattern. To address these limitations, we experimentally investigated the effects of OI and OS in PWH presenting ankle arthropathy, with specific attention given to pain and gait variables.

Material and methods

Subjects

Sixteen male patients diagnosed with haemophilia-related bilateral ankle arthropathy were followed-up between March 2008 and December 2010, at the Haemophilia comprehensive centre of the Cliniques Universitaires Saint-Luc, Brussels, Belgium. Their characteristics are presented in Table 1. Of these, 13 patients had severe (one-stage FVIII or IX assay <1 IU dL\(^{-1}\)) and three moderate haemophilia (one-stage FVIII or IX assay between 2–5 IU dL\(^{-1}\)), with 15 patients suffering from haemophilia A and one from haemophilia B. All patients were capable of independent gait, without needing assistance. Exclusion criteria included prior use of OI and OS and co-morbidities with an impact on walking, such as neurological and cardiopulmonary diseases. The median Pettersson radiological score [17] of the ankle was 8 (P25: 7; P75: 9). The study was approved by the institutional ethics committee, and all participants provided informed consent.

Protocol

Patients wearing neutral running shoes (Kalenji success Decathlon\(^{®}\); Villeneuve d’Ascq, France) were assessed using three-dimensional gait analysis (3DGA) at base-line (T0) and after a mean follow-up of 17 ± 5 weeks (T1). Subjects were required to complete the region-specific Foot Function Index-Revised short-form (FFI-R). The FFI-R is the revised version [18] of an anatomically specific outcome instrument with established validity, test–retest reliability, internal consistency and responsiveness [18–21]. The FFI-R assesses self-reported foot function in terms of the following aspects: pain (e.g. ‘During the past week, how severe was your foot pain when you first stood without shoes’), stiffness (e.g. ‘During the past week, how severe was your foot stiffness before you get up in the morning’), difficulty (e.g. ‘how much difficulty did your foot problems cause you climbing stairs’), activity limitation (e.g. ‘how much of the time did you limit your outdoor activities because of foot problem’) and psychosocial issues (e.g. ‘how much of the time did you feel awful because of foot problem’). The maximum possible score is 100, indicating maximum alteration. Psychometric properties of the FFI-R have been tested using Rasch analysis established on a sample of 92 patients, of whom 69% (63/92) reported having degenerative arthritis [18]. Given its robust psychometric properties and sensitivity in PWH [14,22], the FFI-R was selected to assess the subjects in this study.

Following T1, patients were referred to a multidisciplinary podiatry clinic consultation. Eleven patients were prescribed OI and five OS, with the delivery time being 3 weeks for OI and 6–8 weeks for OS. After

![Fig. 1. Orthopaedic insoles were proposed as first-intention option to patients with partially correctable rear-foot pronation (Fig. 1a, b). Orthopaedic shoes were moulded on the patient’s foot (Fig. 1c) under semi-weight-bearing conditions, being made of a double layer of leather (Fig. 1d).](image-url)
40 ± 18 weeks (T2), 3DGA was performed, with the subjects wearing their OS or OI fitted inside the neutral running shoes. As some foot orthoses required some modification before the patient felt comfortable, this partially accounted for the longer time period between T1 and T2 than between T0 and T1. Subjects were asked to complete a third FFI-R questionnaire, in addition to a satisfaction questionnaire based on the MOS questionnaire [23], to assess the orthose impact over the T1 – T2 period.

Prior to 3DGA assessment, patients had not experienced acute joint or muscle bleeding during the previous 30 days. Subjects who occasionally used anti-inflammatory drugs (NSAID) were instructed to stop taking them for at least 72 h, whereas daily NSAID users were told not to interrupt the treatment.

**Foot orthoses**

All orthoses were manufactured by the same laboratory. Orthopaedic insoles were fabricated from a plaster model of the patient’s foot taken from a foam box impression. Although most OI were made using sheets of high-density polyethylene foam over the casts (Fig. 1b), two patients received leather-lined cork OI. For metatarsal relief, extra-density padding was incorporated into the OI proximally to the metatarsal to improve longitudinal arch weight-bearing.

OS manufacturing necessitated a custom molding of the patient’s foot (Fig. 1c) under semi-weight-bearing conditions. The insoles of the shoes were made from two different podofoam® XA 1000 and XA 600 layers (Podofrance, Noisy-le-grand, France), with a temporary shoe in thermoformable polymer being made using a vacuum process on the pre-existing mould. The definitive shoe (Fig. 1d) was composed of a double leather layer, while the sole was made of soft rubber with a rocker shape to facilitate the rolling of the step. Features such as shoe depth and model, heel height, sole stiffness, and fastening apparatus (laces, Velcro, or buckle) were taken into consideration.

**Gait analysis**

The basic principles of 3DGA are summarized as follows, although a description of its technical aspects may be found in two previous publications [24,25].

In short, 3DGA involved synchronous spatiotemporal, kinematic, kinetics, mechanics and metabolic measurements. Walking trials were conducted while the patients walked at a self-selected speed on a treadmill that was mounted on 3D strain-gauge force transducers (Fig. 2), and for each subject, the same speed was imposed at T0, T1 and T2. Segmental kinematics was measured with six infrared cameras, which recorded the 3D co-ordinates of reflective markers positioned on specific anatomical landmarks and then computed angular displacements. A force platform located under the treadmill simultaneously measured the ground reaction forces generated by the body in three directions. The net moments of force and power that were generated or absorbed at the major lower limb joint muscles in the sagittal plane were estimated, with spatiotemporal parameters being assessed using 3D position data. The internal work ($W_{\text{int}}$), i.e. positive work performed by the muscles to move the limbs in relation with the body’s centre of mass (CoM), was calculated based on kinematics data [26]. The external work ($W_{\text{ext}}$), i.e. work performed to lift and accelerate the centre of mass in relation to the surroundings, was computed [26], the total work ($W_{\text{tot}}$) being defined as the sum of $W_{\text{ext}}$ and $W_{\text{int}}$. ‘Recovery’ was quantified as the percentage of mechanical energy saved via a pendulum-like exchange between gravitational potential energy and kinetic energy of the CoM [26].

The oxygen consumption rate was measured using an ergospirometer. The mass-specific net cost of transport ($C_{\text{net}}$) was calculated by dividing net energy consumption by walking speed. The efficiency of positive work production by the muscles during walking was calculated as the ratio of $W_{\text{tot}}$ to $C_{\text{net}}$ [27].
Statistical analyses

Statistical analyses were performed using SAS 9.2 (SAS Institute Inc., Cary, NC, USA, 2002–2008). As subjects were measured repeatedly over time, a mixed model for repeated measures data with a compound symmetry covariance pattern was used for each outcome variable, which took into account the correlation between observations relating to the same patient [28]. The outcome variable at the first time point (T0) was used as a continuous baseline variable and the time variable (T1 and T2, pre- and post-treatment) as a binary variable. The mixed model was adjusted for two binary covariates: treatment (OS vs. OI) and satisfaction (satisfied vs. non-satisfied), with interactions between time and treatment, and time and satisfaction also being taken into account. Regression residuals were visually examined by ACL and SL for linearity of the model, and for independence, homoscedasticity and normality of the errors.

Results

Satisfaction questionnaire

In total, ten patients (63%) were satisfied with their foot orthoses, all of them reporting a substantial reduction in pain, with four noting improved proprioception (Table 2). Three patients reported a substantial reduction in pain killer/NSAID consumption, three exhibited a subjectively improved walking endurance, and one observed a decrease in swelling towards the end of the day. Four patients (25%) did not show any improvement while wearing the orthosis, but they did not report any adverse effects. Two patients abandoned the orthoses because of increased ankle pain due to Achilles tendonitis (diagnosed by clinical examination).

Mixed models

Orthoses impact regardless of the OI or OS type. Overall, the mechanical and energetic variables were not influenced by the orthoses, except for the ‘recovery’ index, which was significantly increased by 2.2% (P = 0.037). Regarding kinematic and kinetic variables, wearing orthoses was associated with a 3.1° decrease in external rotation of the foot progression (angle between the long axis of the foot and the direction of travel as seen from above) (19.7° vs. 16.6°, P < 0.001) (Fig. 3i), in addition to a 2.0° increase in knee flexion amplitude during stance phase (9.9° vs. 11.9°, P < 0.001) (Fig. 3f) and 0.32 W kg⁻¹ increase in peak concentric power during push-off phase (1.86 vs. 2.18 W kg⁻¹, P = 0.004) (Fig. 4f).

Impact of orthoses, with the distinction between OI and OS. For several gait variables, the mixed model was able to highlight differences between OI and OS. OI had no influence on gait variables other than that reported when not distinguishing between OI and OS types, whereas OS had a significant influence on spatiotemporal parameters, kinematics and kinetics. Using OS led to a decrease in cadence of 4.5 step min⁻¹ (110.0 vs. 105.5 step min⁻¹, P < 0.001) and was associated with a 2-cm increase in step length (0.66 vs. 0.68 m, P = 0.012) (Table 3), while stance phase duration remained unchanged. Total hip ROM and knee ROM in swing phase measured by 3DGA increased by 3.1° (40.4° vs. 43.5°, P = 0.011) and 4.4° (54.3° vs. 58.7°, P < 0.001), respectively (Fig. 3a, d). Peak plantar-flexion moment of the ankle increased by 0.30 N m kg⁻¹ (0.85 vs. 1.15 N m kg⁻¹, P < 0.001) (Fig. 4a).

Impact of orthoses, when distinguishing between satisfied and non-satisfied patients. Total FFI-R score was found to significantly decrease by nine points in patients who reported being satisfied with the orthoses (33 vs. 24 points, P = 0.007), but no significant change was observed in those being non-satisfied (Fig. 5). This improved total FFI-R score may largely be accounted for by a 13-points decrease in the ‘pain’ subscale among satisfied patients (30 vs. 17 points, P = 0.006).

Aside from the significantly decreased cadence of 3.6 step min⁻¹ (108.3 vs. 104.7 step min⁻¹, P < 0.001) that was reported only in satisfied patients, the biomechanical impact of the orthoses was similar between satisfied and non-satisfied patients.

Discussion

Foot orthoses are widely prescribed for patients with RA [9–13], midfoot OA [29], or plantar heel pain [30], but few studies evaluate their effects in PWH. Our study was designed to evaluate the impact of OI and OS on gait parameters, patient satisfaction and functional improvement in PWH with foot involvement.

Impact of orthoses on self-reported satisfaction and function

The use of orthoses was reported to be satisfactory by more than half of the patients, resulting in a significant reduction in the pain subscale score and total FFI-R score (Fig. 5). South et al. [15] observed that combined physiotherapy and podiatry resulted in excellent patient satisfaction scores and significant pain reduction. Slattery and Tinley [14] also reported a significant pain reduction in 16 PWH using OI over a 6-week period without rehabilitation.

Currently, there is no clear understanding of how foot orthoses affect foot pain and improve comfort. Several theoretical explanations have been proposed,
notably that orthoses resist or facilitate the motion of arthritic joints [29], reduce plantar fascia strain by minimizing arch deformation [31], decrease joint loading by acting as a cushioning interface between the ground and foot [32], or alter proprioception involved in muscle activity regulation [33,34]. Given that high (pes cavus) and flat-arched (pes planus) foot types are common in PWH, these deformities may result in increased load and pressure on the foot structure, which are then transferred to proximal joints, such as the knees, hips and lower back [35]. Foot orthoses may also be instrumental in reducing plantar pressure [9,13,30].

Impact of orthoses on gait pattern

The use orthoses was associated with a decreased external rotation of the foot progression (Fig. 3l). The foot progression angle may provide some indication as to the torsional abnormality of the foot and whole limb. A reduction of the angle may be due to the correction of excessive rear-foot pronation combined...
with fore-foot abduction as a result of using OI or OS, which is in line with most published studies evaluating the effects of OI on rear-foot alignment in the frontal plane [7,37–39].

Orthopaedic insoles had no impact on ankle kine
matics and kinetics. Although significant, the changes associated with OS in hip and knee kinematics (Fig. 3a, d) were probably not clinically important. On the contrary, OS improved the propulsion of the ankle (Fig. 4a, d). At the push-off phase, the peak ankle moment improved with OS (Fig. 4a), whereas peak ankle power improved significantly only when OS and OI data were considered together (Fig. 4f). This was likely due to low statistical power, given the small number of patients (n = 5) treated with OS. As ankle ROM at the push-off phase did not improve with OS, the increased ankle power was probably attributed to increased moments. As the lever arm remained unchanged between neutral shoes (T1) and OS conditions (T2), increased joint moment could only be attributed to increased forces that developed at the ankle level. Increases in ankle moment and knee flexion in the stance phase (Fig. 3d) suggest that patients with OS experience improved weight acceptance, probably due to improved comfort and reduced ankle pain. These results are in contradiction with those of Chen et al. [4] who reported that the peak ankle plantar-flexion moment tended to be smaller in flat foot patients walking with OI.

Regarding spatiotemporal parameters, previous publications reported that foot orthoses increased the step length in RA patients [40]. In our study, OS was shown to increase step length, while decreasing cadence, which is thought to be related to increased total hip ROM (Fig. 3a) and improved propulsion phase of the ankle.

Otman et al. [41] reported an 8% decrease in oxygen consumption in flat feet patients when walking with OI. Furthermore, Kavlak et al. [40] showed that using OI for 3 months lowered energy expenditure during gait in RA patients. However, in our series, we did not detect any difference between pre- and postorthoses conditions in terms of metabolic cost, mechanical work and gait efficiency. The recovery index, a measure of the muscular work undertaken during the pendulum exchange between potential and kinetic energy, was the only mechanical variable, which improved slightly when using orthoses.

The principal limitation of this study was the sample size. Haemophilia is a rare disease and our exclusion criteria were very strict (e.g. prior use of OI and OS). It

Table 2. Results of the satisfaction questionnaire and adverse effects of custom-made foot orthoses.

<table>
<thead>
<tr>
<th>Questions</th>
<th>Number of patients (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total responses to satisfaction questionnaire</td>
<td>16(100)</td>
</tr>
<tr>
<td>Are you satisfied with the comfort of your orthoses?</td>
<td></td>
</tr>
<tr>
<td>Yes, improvement</td>
<td>10(63)</td>
</tr>
<tr>
<td>No change</td>
<td>4(25)</td>
</tr>
<tr>
<td>No, deterioration</td>
<td>2(13)</td>
</tr>
<tr>
<td>What are the disadvantages of your orthoses?</td>
<td></td>
</tr>
<tr>
<td>Pain reduction</td>
<td>10(63)</td>
</tr>
<tr>
<td>Improved proprioception</td>
<td>4(25)</td>
</tr>
<tr>
<td>Diminution of pain killer/NSAID</td>
<td>3(19)</td>
</tr>
<tr>
<td>Improved walking endurance</td>
<td>3(19)</td>
</tr>
<tr>
<td>Decrease of swelling</td>
<td>1(6)</td>
</tr>
<tr>
<td>What are the disadvantages of your orthoses?</td>
<td></td>
</tr>
<tr>
<td>Tendonitis</td>
<td>2(13)</td>
</tr>
</tbody>
</table>

NSAID, non-steroidal anti-inflammatory drug.
was therefore difficult to recruit more patients in the trial. As our sample was small, it should be highlighted that the absence of significance could be due to the low statistical power. This also could explain that when a variable is applicable for both limbs independently (e.g. peak plantar-flexion moment of the ankle), significant results could be separately observed in OS, as the presence of two limbs for each patient multiplied the sample size by two. On the contrary, when considering unique variables among the patients (e.g. recovery index), changes could only be observed when both OI and OS were considered together (n = 16).

**Indications, perspectives and side effects**

There is no consensus with respect to the best type of foot orthoses for managing foot pain in PWH. Empirically, OI were proposed as the first option for patients with moderate ankle arthropathy or partially correctable rear-foot (Fig. 1a, b), whereas OS were prescribed to patients with more severe pain or poor ROM. With respect to gait kinematics, the rocker sole of OS provides several advantages, being thus used when there is only minimal motion at the forefoot joint or hindfoot joint [42,43]. It facilitates controlling joint motion by rocking the foot from heel strike through to toe-off. Our statistical analysis revealed enhanced functional impairment (lower FFI-R score) and gait disturbances (increased metabolic cost, and decreased hip and knee ROM and plantar-flexion moment of the ankle) in OS patients compared with OI patients. Given that our results show that foot orthoses are beneficial for some, but not all PWH, it is important to understand which factors are associated with potential foot orthosis benefits, before drawing definitive conclusions on this modality. Previous studies reported significantly reduced ankle bleeding in PWH when using OI [14,15]. In contrast, Jorge Filho et al. [16] noted significantly increased traumatic bleedings when using OI over 6 months, owing to that fact that patients felt so safe when wearing OI that they indulged increasingly in activities likely to cause sprains and bleeding. In addition, although this study provided useful insights into the immediate effects of OS and OI,
long-term effects were not assessed. Therefore, further studies are required to determine the impact of foot orthoses on joint bleeding, along with their long-term effects.

In our series, two patients suffered from Achilles tendonitis due to OI. Although foot orthoses are considered to have minimal side effects, our two observed cases of Achilles tendonitis support the idea that correction in patients with a long history of foot deformation must be progressive.

Conclusion

Our study results suggest that foot orthoses may have beneficial effects on ankle joints in PWH. OI and OS provided significant pain relief and improved comfort in more than half of patients, with minimal side effects. Although OI had a limited impact on gait pattern as evaluated using 3DGA, OS significantly improved ankle propulsion. The biomechanical changes caused by OI and OS were independent of their ability to improve comfort, as they were too limited to influence knee and hip kinematics and kinetics, or mechanical and energetic variables. Overall, 3DGA was not discriminative enough to determine the potential benefits of foot orthoses in PWH. Self-reported scales, such as FFI-R and satisfaction questionnaires, proved sufficiently sensitive to assess the efficacy of foot orthoses in PWH. Foot orthoses will likely make a substantial difference in terms of comfort and function for patients with limited access to replacement therapy.

Acknowledgements

The authors acknowledge Mr Christian Pintaux, orthotist, (Orthopedie Van Haesendonck) for his technical help. This work was supported in part by a grant from the Fondation Salus Sanguinis.

Author contributions

Sébastien Lobet performed the research. Sébastien Lobet and Christine Detrembleur designed the research study. Sébastien Lobet and Anne-Catherine Lantin analysed the data. Sébastien Lobet and Cedric Hermans wrote the manuscript.

Disclosures

The authors stated that they had no interests which might be perceived as posing a conflict or bias.

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