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Plantar pressure patterns in women affected by Ehlers–Danlos syndrome while standing and walking

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ABSTRACT

This study aims to quantitatively characterize plantar pressure distribution in women affected by Ehlers–Danlos syndrome of the hypermobile type (EDS-HT) to verify the existence of peculiar patterns possibly related to postural anomalies or physical and functional lower limb impairments typical of this disease.

A sample of 26 women affected by EDS-HT (mean age 36.8, SD 12.0) was tested using a pressure platform in two conditions, namely static standing and walking. Raw data were processed to assess contact area and mean and peak pressure distribution in rearfoot, midfoot and forefoot. Collected data were then compared with those obtained from an equally numbered control group of unaffected women matched for age and anthropometric features. The results show that, in both tested conditions, women with EDS-HT exhibited significantly smaller forefoot contact areas and higher peak and mean pressure than the control group. No differences in the analyzed parameters were found between right and left limb. The findings of the present study suggest that individuals with EDS-HT are characterized by specific plantar pressure patterns that are likely to be caused by the morphologic and functional foot modification associated with the syndrome. The use of electronic pedobarography may provide physicians and rehabilitation therapists with information useful in monitoring the disease's progression and the effectiveness of orthotic treatments.

1. Introduction

The Ehlers–Danlos syndrome (EDS) comprises a number of heritable disorders of the connective tissue characterized by peculiar symptoms such as hyperextensible skin, dystrophic scarring, easy bruising, joint hypermobility (JHM) and tissue fragility, often accompanied by a wide range of visceral, pelvic neurologic and cognitive dysfunctions (Sacheti et al., 1997; Castori et al., 2012).

The consequences of such a condition on musculoskeletal (MS) system functionality are quite relevant, especially due to ligamentous and capsular laxity. The main orthopedic issues commonly observed in individuals with EDS are dislocations

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and joint instability, spinal abnormalities, asymmetry of the thoracic cage and osteoarthritis (Beighton & Horan, 1969). Moreover, the majority of individuals affected by EDS report chronic pain in joint locations such as elbow, shoulder and knees and in the lower extremities (ankles, feet and toes, Sacheti et al., 1997). In particular, Beighton and Horan (1969) and Tompkins and Bellacosa (1997) reported that besides the general MS affections caused by EDS, foot appearance and functionality appear severely altered by the pathology. In particular, individuals with EDS exhibit foot anomalies such as asymptomatic pes planus, hallux valgus, claw toes, and "moccasin feet" (i.e. foot skin loosening that looks like the individual is wearing an oversize ankle sock).

The combination of such alterations with discomfort/pain and JHM in the lower limbs may be considered responsible for reducing the general mobility of individuals with EDS as well as their capability to keep a simple upright quiet posture and their willingness to be involved in physical activity, as reported by several recent studies (Berglund, Nordström, Hagberg, & Mattiasson, 2005; Rombaut, Malfait, Cools, De Paepe, & Calders, 2010). As the efficiency of simple motor tasks such as standing and walking are greatly influenced by the stress patterns that set in at the foot–ground interface, it appear important to verify whether individuals with EDS are characterized by a specific plantar pressure distribution that may possibly reflect on gait and postural anomalies.

Nevertheless, to the authors' knowledge no quantitative studies (e.g. using devices such as pressure sensitive platforms, mats or insoles) have been carried out to investigate the main foot–ground interface parameters such as contact areas and peak or mean contact pressure in EDS. This appears to be a serious limitation in view of a global evaluation of mobility issues in individuals affected by EDS, since pedobarography represents a reliable clinical tool useful in assessing the effectiveness of surgical procedures and orthotic devices, as well in acquiring a better comprehension of foot functionality, especially in pathological subjects (Lord, Reynolds, & Hughes, 1986; Hughes, 1993).

On the basis of the aforementioned considerations, this study proposes an analysis of foot–ground contact features in a sample of individuals with EDS for the purpose of evaluating contact areas and plantar pressure distribution in terms of peak and mean values acting on the different plantar sub-regions, namely forefoot, midfoot and rearfoot.

We hypothesize that considering the modifications induced by the disease in foot morphology and functionality, individuals with EDS present peculiar plantar pressure patterns. Such data will then be discussed in the light of known physical impairments related to foot–ground contact, especially in terms of standing posture and gait (Galli et al., 2011a; Galli et al., 2011b; Rombaut et al., 2011a), as well as outlining a possible role of pedobarography as a useful tool in monitoring the evolution of the syndrome and evaluating the effectiveness of rehabilitative treatments.

2. Methods

2.1. Participants

In November 2012, 26 females with Hypermobility Type (HT) EDS in the age range 17–62 were examined at the Movement Analysis Lab of the IRCCS "San Raffaele Pisana" (Italy). Diagnosis was established based on both the Villefranche and Brighton criteria (Beighton, De Paepe, Steinmann, Tsipouras, & Wentrup, 1998; Grahame, Bird, & Child, 2000) and patients were considered affected if meeting at least one of the two sets of diagnostic criteria (Celletti, Castori, Grammatico, & Camerota, 2012). As EDS is a diagnosis of exclusion, the absence of features suggestive of other heritable connective tissue disorders was assessed in a clinical genetics outpatient clinic.

An equally numbered age-matched control group was established by recruiting healthy individuals on a voluntary basis after a public announcement. The main anthropometric features of the two groups are reported in Table 1.

The study was approved by the Ethics Committee of the Institute and written informed consent was obtained from all the participants after a detailed explanation of the purposes of the study and a description of the experimental methodology.

2.2. Data acquisition and post-processing

Plantar pressure measurements were performed by means of a pressure platform (FDM-S, Zebris Medical GmbH, Germany) composed of 2560 capacitive sensing elements arranged in a 64×40 matrix, and connected via USB interface to a personal computer.

Firstly, static plantar pressure distribution was acquired under quiet upright stance conditions. All participants were asked to stand as still as possible on the platform for 10 s, while their feet were freely placed in a self-selected comfortable position.

Table 1
Anthropometric features of the individuals recruited for the study. Values are expressed as mean ± SD.

	Ehler–Danlos	Control group	*p*-Value
Participants (#)	26	26	–
Age (years)	36.8 ± 12.0	37.2 ± 12.4	0.985
Height (cm)	164.3 ± 5.7	161.7 ± 4.0	0.069
Weight (kg)	58.0 ± 8.1	57.9 ± 7.8	0.960
BMI ($kg\,m^{-2}$)	21.5 ± 3.1	22.1 ± 2.9	0.138

As the acquisition frequency was set at 50 Hz, a total of 500 temporal frames were acquired for each test. The corresponding text matrices containing the contact pressure value for each element of the platform's sensitive grid were exported as an ASCII file and post-processed with a custom-made routine (developed in the Matlab® environment) able to calculate the following parameters on a frame-by-frame basis:

- Overall, forefoot, midfoot and rearfoot contact areas (expressed in mm^2) calculated according to the method described by Cavanagh and Rodgers (1987)
- Peak of the plantar pressure for each sub-region (i.e. the highest pressure value detected by a single sensor in a certain sub-region, expressed in kPa)
- Mean value of the plantar pressure for each sub-region (i.e. the mean value calculated taking into consideration all the sensors belonging to a certain sub-region, expressed in kPa)

For each of the aforementioned parameters, the average value of all the frames acquired was selected as representative of the whole trial and used for comparisons between the two groups.

Dynamic foot–ground contact parameters were acquired by asking participants to walk at a self-selected speed over a 4 m long walkway in which the pressure platform was embedded. The trial was considered concluded when at least three steps for each limb were correctly acquired (i.e. the platform was struck with a single foot). In this case, the sampling frequency was set to 100 Hz. The raw data were again exported as ASCII files and processed as previously described. The average value of the foot–ground contact parameters calculated for the three steps related to each limb was considered representative of the whole trial.

2.3. Statistical analysis

Differences in the foot–ground interaction parameters introduced by the EDS were assessed using one-way multivariate analyses of variance (MANOVA). The independent variable (IV) was individuals' status (EDS or control group), and the ten dependent variables (DV) were total contact area, sub-region (rearfoot, midfoot and forefoot) contact area, peak and mean contact pressure. The level of significance was set at $p = 0.05$ and the effect of size was assessed using the eta squared coefficient (η^2). Follow-up analyses were conducted using one-way ANOVAs for each dependent variable by setting the level of significance at $p = 0.005$ (0.05/10) after a Bonferroni adjustment for multiple comparisons.

3. Results

The acquired data were preliminarily checked to verify the existence of possible bilateral asymmetry in foot parameters for the 114 (52×2) analyzed limbs. A one-way ANOVA, performed using the limb (left/right) as IV and the foot–ground contact parameters as DV, revealed no significant bilateral differences for any of the DVs. For this reason, the data related to both limbs were grouped and the results described below can be considered representative of both feet.

The results obtained from experimental tests are summarized in Fig. 1. In the case of static standing (Table 2), MANOVA revealed a significant effect of individuals' status ($F(10,93) = 4.96$, $p < 0.001$, Wilks $\lambda = 0.65$, $\eta^2 = 0.35$) on foot–ground contact parameters. In particular, individuals with EDS were characterized by a smaller (−11%) forefoot area with respect to control group ($F(1,103) = 14.31$, $p < 0.001$, $\eta^2 = 0.12$), larger peak (+40%, $F(1,103) = 19.87$, $p < 0.001$, $\eta^2 = 0.16$) and mean (+19% $F(1,103) = 10.13$, $p = 0.002$, $\eta^2 = 0.09$) contact pressure.

Even in the case of walking tests (Table 3) a significant effect of status was detected ($F(10,93) = 6.26$, $p < 0.001$, Wilks $\lambda = 0.60$, $\eta^2 = 0.40$). Similarly to what was observed for upright posture, the differences between EDS and control group basically involved the forefoot. In fact, EDS subjects exhibited smaller forefoot contact areas (−5% ($F(1,103) = 10.15$, $p = 0.002$, $\eta^2 = 0.09$)) as well as larger peak (+24% ($F(1,103) = 10.15$, $p = 0.002$, $\eta^2 = 0.09$)) and mean contact pressure (+13% ($F(1,103) = 20.73$, $p < 0.001$, $\eta^2 = 0.17$)).

4. Discussion

Previous studies carried out on individuals with EDS highlighted that their balance and gait abilities appear impaired in some way. As the foot represents the interface between the body and the outside world, and thus the site where all the forces exchanged take place, it is reasonable to suppose that EDS may negatively influence such motor task performances even by altering the physiological plantar pressure patterns. In this regard, our initial hypothesis was supported by the results obtained since there were significant differences between individuals with EDS and healthy controls that basically involve the forefoot both in static and dynamic conditions. In fact, in individuals with EDS this plantar sub-region is smaller and characterized by higher mean and peak pressure.

Unfortunately, to the authors' knowledge, this is the first quantitative study on plantar pressures in EDS patients, and thus no information is available to compare our results, even though foot morphology and functional anomalies have been extensively reported in previous studies (Beighton & Horan, 1969; Tompkins & Bellacosa, 1997; Berglund, Nordström, Hagberg, & Mattiasson, 2005). Nevertheless, it is to be noted that the main distinctive feature of EDS-HT,

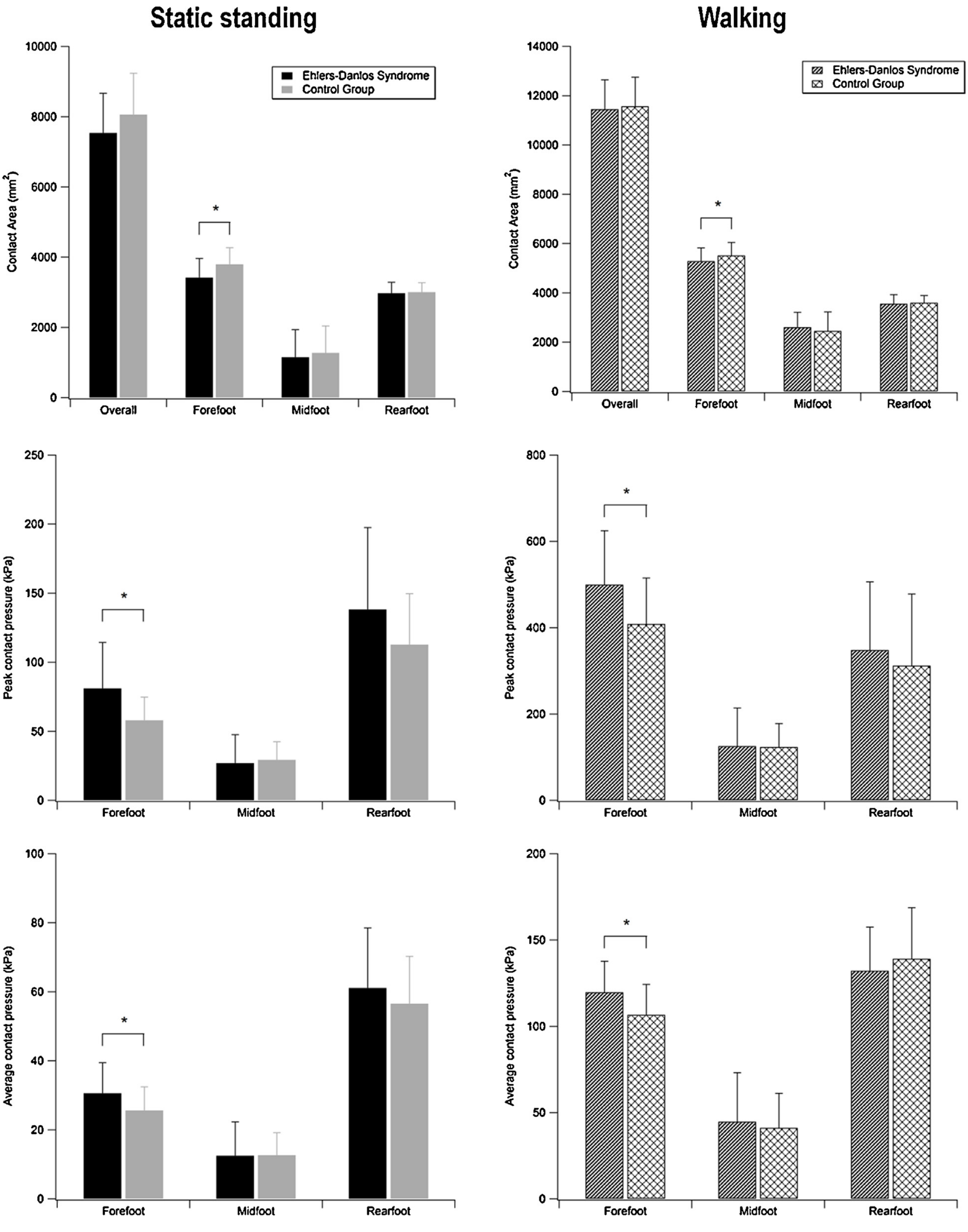

Fig. 1. Top to bottom: overall and sub-region contact area, peak and mean contact pressure for static standing (left) and walking (right) in individuals with EDS and control group. *Denotes significant differences between EDS and control group ($p < 0.005$ after Bonferroni correction).

Table 2
Foot–ground contact areas and pressures for static standing. Values are expressed as mean ± SD.

Static standing		Ehlers–Danlos	Control group	*p*-Value
Contact area (mm^2)	Overall	7533.15 ± 1131.65	8061.40 ± 1176.22	0.022
	Rearfoot	2969.05 ± 310.90	2993.57 ± 274.64	0.671
	Midfoot	1147.25 ± 786.59	1272.06 ± 767.23	0.415
	Forefoot	3416.84 ± 544.89	3795.77 ± 474.11	<0.001[a]
Peak contact pressure (kPa)	Rearfoot	138.13 ± 59.41	112.64 ± 36.87	0.010
	Midfoot	26.88 ± 20.69	29.26 ± 13.10	0.485
	Forefoot	81.00 ± 33.33	57.92 ± 16.80	<0.001[a]
Mean contact pressure (kPa)	Rearfoot	61.06 ± 17.43	56.54 ± 13.75	0.145
	Midfoot	12.48 ± 9.81	12.60 ± 6.52	0.945
	Forefoot	30.58 ± 8.90	25.62 ± 6.86	0.002[a]

[a] Denotes a significant effect of EDS (follow-up after MANOVA, $p < 0.005$ after Bonferroni correction)

Table 3
Foot–ground contact areas and pressures for walking. Values are expressed as mean ± SD.

Walking		Ehlers–Danlos	Control group	*p*-Value
Contact area (mm^2)	Overall	11459.00 ± 1092.29	11590.46 ± 1119.20	0.542
	Rearfoot	3578.36 ± 332.31	3597.76 ± 276.84	0.747
	Midfoot	2611.50 ± 545.06	2450.43 ± 722.30	0.202
	Forefoot	5269.14 ± 427.97	5542.36 ± 446.18	0.002[a]
Peak contact pressure (kPa)	Rearfoot	342.42 ± 108.64	308.62 ± 94.7	0.094
	Midfoot	127.18 ± 66.33	120.77 ± 34.89	0.539
	Forefoot	497.04 ± 124.66	401.59 ± 125.14	<0.001[a]
Mean contact pressure (kPa)	Rearfoot	133.00 ± 20.29	137.24 ± 28.08	0.381
	Midfoot	46.69 ± 21.86	39.85 ± 13.74	0.059
	Forefoot	119.89 ± 14.86	106.11 ± 15.98	<0.001[a]

[a] Denotes a significant effect of EDS (follow-up after MANOVA, $p < 0.005$ after Bonferroni correction)

namely joint hypermobility, together with other functional impairments, chronic pain and foot alterations are often encountered in other kinds of rheumatoid diseases such as, for example, rheumatoid arthritis (RA) (Rombaut et al., 2011b) so that in the past RA was even incorrectly diagnosed instead of EDS (Bridges, Smith, & Reid, 1992). Given such clinical similarities, it is possible to compare, at least indirectly, the results of the present study with previous investigations on plantar pressures in patients affected by RA. It is interesting to observe that either higher contact pressures or force-time integrals in the forefoot (Bowen et al., 2011; Lord, Reynolds, & Hughes, 1986; Otter, Bowen, & Young, 2004) similar to what was found in the present study, were observed in patients with RA when compared to healthy controls and such stress concentration was also found to be significantly correlated with perceived pain (van der Leeden, Steultjens, Dekker, Prins, & Dekker, 2006). The peak pressure values in the forefoot (but also in midfoot and rearfoot) calculated here for the EDS patients are in good agreement with those reported by van der Leeden et al. (2006) even from a quantitative point of view. Consequently, it has been hypothesized that such anomalous plantar pressure patterns may be associated with impaired foot function that reduces the ability to perform simple daily activities such as standing or walking.

On the other hand, smaller forefoot areas and the corresponding high plantar pressures may represent a consequence of the individual's sensation of instability due to poor performance of the postural control system, a fact that has been reported in previous studies on postural sway of patients with EDS-HT. Thus it is likely that such instability leads patients with EDS to contract the toes in an attempt to obtain a hold on the ground to improve stability.

It is also to be noted that anomalous plantar pressures may represent per se a co-factor able to further disturb postural control performance, as plantar mechanoreceptors are continuously hyperactivated, especially when dynamic motor tasks are performed, and thus the quality of the sensory information may be worsened. This hypothesis has been proposed to partly explain the poor balance abilities in all those conditions (such as obesity, for example) that are characterized by the presence of excessive plantar pressure peaks (Hue et al., 2007).

Moreover, anomalous plantar pressure patterns may contribute to foot discomfort and pain that are likely to limit the willingness of individuals with EDS to maintain the standing posture or walk for prolonged times. Of course, the reduction in physical activity contributes to creating a vicious circle that relates to poor balance, fear of falling, discomfort, pain and muscular weakness, with the final result that the quality of life in individuals with EDS is significantly affected.

4.1. Limitations of the research

Some limitations of the study are to be acknowledged: first of all, both standing and walking tests were performed barefoot and this condition, although essential to ensure direct comparison with similar studies, is not fully representative of actual everyday life conditions. In particular, a specific selection of footwear, made on the basis of perceived discomfort and pain, may possibly attenuate some of the plantar stress concentrations observed in the present investigation. Secondly, given the cross-sectional nature of the present study, patients with a large difference in age (17–62 years old) were grouped, and thus it was not possible to explore possible effects of the progression of the disease on foot–ground contact.

4.2. Future developments of the research

Considering the lack of quantitative data regarding foot–ground interaction in EDS patients, having available a larger database of plantar pressure data at various stages of the pathology is certainly a priority, so the authors are currently recruiting more individuals with EDS to test by means of pedobarography. Moreover, as previously recalled, since the data presented here refer to a specific laboratory condition, it would be important to integrate them with those obtainable with insole sensors, which are more suitable in testing actual daily conditions. Finally, projects are ongoing to assess the effectiveness of simple orthotic treatments specifically designed to reduce the pressure peaks on forefoot, such as metatarsal pads, which have been effective in reducing both the pressure peak and whose effect is well correlated with subjective pain perception (Kang, Chen, Chen, & His, 2006).

5. Conclusions

This study proposed the application of electronic pedobarography techniques to investigate foot–ground contact in individuals affected by EDS-HT, with the main purpose of assessing whether such a disease is associated with peculiar plantar pressure patterns. The findings from this research suggest that a stress concentration in the forefoot exists, and this fact may act as a co-factor able to perturb standing and walking and cause the discomfort and pain frequently reported by individuals with EDS. Such information may be useful in planning orthotic or rehabilitative approaches as well as physical training protocols. Above all, pedobarography should be considered part of the diagnostic process for EDS-HT, especially in terms of periodic monitoring of deterioration, whenever present, of the foot–ground interaction.

Conflicts of interest

The authors report no conflicts of interest

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