Introduction
Systemic sclerosis (SSc), often referred as scleroderma, is an autoimmune rheumatic disease that is characterized by three principal features: vasculopathy, immune activation with the production of autoantibodies and alterations in immune cells, and fibroblast dysfunction with excessive matrix deposition leading to fibrosis of the skin and internal organs (1-2).

In SSc, hand problems are well known and have always been investigated; however, studies and detailed information on feet involvement are lacking.

Skin fibrosis and a hypoxic environment compromise the mechanical and biological properties of the skin in patients with SSc, causing instability in the collagen structure (3). Moreover, vasculopathy, skin fibrosis, and calcinosis are known factors contributing toward the development of foot ulceration, relatively common in people with SSc, and impaired healing (4-6). Raynaud’s phenomenon contributes to peripheral ischemia and may lead to apical digital ulceration, subcutaneous calcinosis, skin thickening, plantar hyperkeratosis, and foot ulcers (3, 7-8). In addition to these factors, intrinsic foot mechanics may play a further role. Abnormal plantar foot pressure is widely noted as a risk factor for ulceration in people with diabetes, and this additive relationship has also been established in diseases closer in nature to SSc, such as rheumatoid arthritis (8-11). Foot ulcers in SSc can last for a long time and can lead to lower limb amputation (7, 12).
In the feet, musculoskeletal system involvement is frequent and often disabling; this includes tendonitis, arthritis, arthropathy, and flexion contractures. Radiological lesions include joint space narrowing, bone demineralization, joint margin erosions, and degenerative changes (12).

Studies regarding podiatric problems related to SSC are lacking, and there are no data specifically evaluating biomechanical changes of the foot. However, foot problems occur in majority of patients (12). Thus, patients with SSC require information about foot health, including the impact of their condition on their feet. Occasionally, they may need specialist treatment targeting foot problems specific to this disease.

The aim of the present pilot study was to evaluate podiatric problems in an Italian cohort of patients with SSC by assessing their received podiatric services, foot pain and disabilities, and biomechanical foot deformities.

**Material and Methods**

From the Division of Rheumatology, Department of Clinical and Experimental Medicine of Florence University, 25 consecutive female patients with SSC [age: 66.68±11.54 (range 37–84)] years, with a disease duration of 18.28±16.62 months (range 1–41 months), diagnosed according to the criteria of the American Rheumatism Association Diagnostic and Therapeutic Criteria Committee were enrolled. Of these patients, 17 suffered from limited cutaneous SSC and 8 from diffuse cutaneous SSC (13). Data were collected over a 3-month period. Patients signed a written informed consent form, and procedures were conducted in accordance with the 1975/83 Declaration of Helsinki. The study was approved by local Ethical Committee.

The inclusion criterion was a current diagnosis of SSC by a rheumatologist (1).

Exclusion criteria were the presence of leg and/or foot ulcers because clinimetric assessment could be influenced by the presence of pain and disability due to ulcers.

**Assessments**

All assessments were performed by the same podiatrist.

All patients enrolled were assessed by the following questionnaires:

**Standards of care for people with musculoskeletal foot health problems: Audit tools**

The original questionnaire, ideated and drawn in Great Britain, consisted of 29 questions to which patients could answer yes or no (12). In order to evaluate foot health services in an Italian context, we chose 13 questions for evaluating the manner in which the visit was performed. Explanations about behavior and education of patients were provided by the podiatrist (Table 1).

**Foot Function Index (FFI)**

The FFI is a specific tool widely used for the assessment of foot pain and disability. It is composed of 23 questions divided into three sections that quantify, by a visual analogue scale (VAS 0–10), the disease impact on foot health by measuring the intensity of pain, disability, and limitations of activities. In the first section, assessing the pain, the scale ranges from no pain (0) to the worst imaginable pain (10), while, in the second section, evaluating disability, the scale ranges from no difficulty (0) to impossible (10); then, in the third section, assessing the limitation of activities, the scale ranges from never (0) to always (10) (14).

**Podiatric evaluation**

All patients who underwent a podiatric evaluation were divided for non-weight-bearing foot joint and weight-bearing foot joint assessments.

**Non-weight-bearing foot joint assessment according to the standard method described by Root (15-18), with the subtalar joint in the neutral position assessing the following:**

- Subtalar joint, to evaluate the deformity of the rearfoot: rearfoot varus (when the bisector of the calcaneus remains in an inverted position with respect to the bisector of the leg distal third), valgus (when the bisector of the calcaneus remains in an eversion position with respect to the bisector of the leg distal third), or straight (when the bisector of the calcaneus is in line with the bisector of the leg distal third)
- Midtarsal joint, to evaluate the deformity of the forefoot: forefoot varus (when the plantar plan of the metatarsal heads remains in an inversion position with respect to the bisector of the calcaneus), valgus (when the plantar plan of the metatarsal heads remains in an eversion position with respect to the bisector of the calcaneus), or straight (when the bisector of the calcaneus is in a perpendicular position with respect to the plantar plan of the metatarsal heads)
- Ankle joint, to assess dorsiflexion (more than 10°, with knee flexion): ankle equinus (when the dorsiflexion of the ankle is less than 10°), ankle talus (when the plantarflexion of the ankle is less than 20°), or ankle physiological (when the dorsiflexion of the ankle is at least 10° and the plantarflexion of the ankle is at least 20°)

The first ray, to assess dorsiflexed or plantarflexed position: first ray plantarflexed (when the plantar plan of the first metatarsal heads is in a lower position with respect to the second metatarsal heads), first ray elevated (when the plantar plan of the first metatarsal heads is in the highest position with respect to the second metatarsal heads), or first ray straight (when the plantar plan of the first and second metatarsal heads is the same height)

**Weight-bearing foot joint assessment (the knee fixed with no possibility of movement)**

**Foot Posture Index (FPI)**

The FPI is a diagnostic and validated tool quantifying the grade of foot pronation, supination, or neutral position. The podiatrist assigns a score related to a series of observations proportional to the posture of the foot that may vary from the neutral position, which is assigned a value of 0, to the pronated position, which is assigned a positive value (+1, +2), and to the supinated position, which is assigned a negative value (−1, −2) (high pronation, +2; pronation, +1; normal, 0; supination, −1; high supination, −2). High positive aggregate values indicate a pronated posture, and high negative aggregate values indicate a supinated overall foot posture; a value of zero indicates a neutral posture.

The six clinical criteria employed in the FPI are talar head palpation supina curvature, infralateral malleolar curvature, calcaneal frontal plane position, prominence in the region of the joint talonavicular, congruence of the medial longitudinal arch, and abduction/adduction of the forefoot on the rear foot (19-20).

**Gait assessment**

The complete gait cycle corresponds to the time that elapses from the heel strike of one foot to the contact of the heel of the same foot in the following step. Gait evaluation was performed by subdividing the stance phase into three periods to facilitate the clinical observation of foot movement and position: contact (27%), midstance (40%), and propulsion (33%).

**Disability and general health evaluation**

The Health Assessment Questionnaire (HAQ) is a self-report questionnaire organized into 20 items divided into eight categories: dressing and grooming, standing, eating, walking, per-
sonal hygiene, reaching, gripping, and other activities. Each item is rated from 0 (no difficulty) to 3 (unable to do) (21).

The Medical Outcomes Survey Short Form 36 (SF-36) is a self-report questionnaire evaluating health related to quality of life (HRQoL) by 36 items organized into eight domains that are combined into a summary physical index (SPI) and a summary mental index (SMI). For all, the scales are scored from 0 to 100, with higher scores corresponding to a better HRQoL and lower scores to a worse HRQoL (22).

Statistical analysis
Descriptive statistical analysis was performed by calculating the mean±standard deviation (M±SD) and ranges for continuous data and number and percentages calculated for binomial data.

Results
The results of the questionnaires administered are the following:

Standards of care for people with musculoskeletal foot health problems: Audit tools
7 (28%) out of the 25 patients with SSc reported a specific podiatric assessment and treatment. The answers of these 7 subjects to the 13 selected questions of Standards of Care for People with Musculoskeletal Foot Health problems are shown in Table 1.

Foot Function Index
Values of pain (4.7±5.1), disability (5.1±3.2), and activity limitations (3.2±3.1) are reported in Table 2.

Podiatry assessment
Non-weight-bearing foot joint assessment
The non-weight-bearing foot joint assessment is presented as percentages in Table 3.

Weight-bearing foot joint assessment
Foot Posture Index
The weight-bearing foot joint assessment made by the FPI showed that 38% of patients have a normal foot, 20% have a pronated foot, 34% have a highly pronated overall foot posture, 4% have a supinated foot, and another 4% have a highly supinated overall foot posture.

Gait assessment
The observation of gait cycle shows the following data: 64% of patients have inverted contact of the calcaneus and the remaining 36% have a contact of the calcaneus in eversion.

In the midstance, 78% of the patients have the foot in pronation and 22% in supination. In the last period of the gait, propulsion, 12% present a takeoff of the foot in supination while 88% in pronation.

HAQ and SF-36:
Global disability was assessed by the HAQ and the results, expressed as M±SD, are 1.13±0.80.

HRQoL was assessed by SF-36: SPI and SMI scales, assessing physical and mental QoL, have scores (M±SD) of 32.38±10.65 and 38.67±11.40, respectively.

Discussion
In the literature, only a few studies about podiatric problems and SSc exist, and detailed information is lacking about feet involvement despite the fact that foot manifestations are frequent and often disabling in patients with SSc (3, 7).

To the best of our knowledge, our study is the first work that evaluates podiatric services received, biomechanical foot deformity, HRQoL, and global disability in an Italian cohort of patients with SSc.

Our results, in an Italian casuistry of 25 patients with SSc, suggest that most of the patients had not received adequate information about foot health in SSc, and preventative information was scarce. In fact, no patient received a foot health assessment within the first 6 months of disease diagnosis and no patient received information about SSc consequences on foot health.

Only 1 patient (4%) received an assessment of the foot every year; only 1 patient (4%) received specific information about the disease for self-management in foot care; and 5 patients (20%) received information about the benefits of using adapted footwear and insoles.

These data are similar to those of an English study of 91 patients with SSc in Leeds (UK) (12). Only 37 of 91 (40.7%) with podiatric problems were addressed to the care of a podiatrist, 36 of 91 (39.6%) reported that they had received some information about foot health, regardless of whether or not they had previous podiatric problems. Only 27 (29.7%) received specific information on foot health in relation to SSc. 23 (25.3%) participants were given no information about the potential effect of SSc on their feet, or about how to access care in case they presented with problems. Hence, these studies show that information about foot problems and podiatry assessment and treatment are inadequate in patients with SSc, particularly in Italy.

The impact of SSC on foot health has been poorly studied. However, the few published studies show that podiatric problems are common and potentially disabling for patients with SSc (3, 7). This is confirmed in our study by FFI results that indicate an average intensity of pain of 50% and disability of approximately 50%, while the foot activities are limited to approximately 30%. To put this in perspective, our patients with SSc have less pain, disability, and activity limitations compared to patients with rheumatoid arthritis (RA), which when assessed by the FFI, had an average value of 6.7 (24).

Regarding foot examinations, we have no reference data for comparisons. Biomechanical deformities in healthy subjects are poorly evaluated in the literature; only a population of athletes has been evaluated. Thus, the most common biomechanical deformities found in these subjects are rearfoot varus, forefoot valgus, forefoot varus, ankle equinus, and first ray plantarflexed (25).

In our patients with SSc, podiatric assessment was performed using the Root method (15), a very useful tool in clinical practice. Despite the scarcity of studies on the reliability of this method, there are many studies on Kirby’s method that are based on the Root evaluation (16-18).

In the present study, biomechanical evaluation of the subtalar joint shows that majority of the patients had a rearfoot varus deformity (64%) and that no patient presented a rearfoot valgus deformity. In the biomechanical evaluation of the midtarsal joint, the absence of a deformity is prevalent, while 42% of patients with SSc had a forefoot varus deformity and 6% had a forefoot valgus deformity.

The ankle joint was normal in majority of the patients, while 34% of them had a limitation of dorsiflexion only with the knee extended. This is not due to a structured deformity, but to a series of causes: a shortening of the gastrocnemius muscle or a contracture or spasticity.

Only 2% presented an equinus ankle deformity with limitation of ankle movement, both with the knee extended and with the knee flexed. In addition, patients showed deformities of the first ray both in the dorsiflexed and plantarflexed positions.

These deformities can be severe in patients with SSc as they may cause serious conse-
<table>
<thead>
<tr>
<th>Question</th>
<th>YES Percentage (number)</th>
<th>NO Percentage (number)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Does the health professional explain what is being done and the equipment they are using all the way through your foot health appointment?</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>2. Have you been given information in your foot health clinic that promotes good musculoskeletal foot health?</td>
<td>28% (2)</td>
<td>72% (5)</td>
</tr>
<tr>
<td>What information was included?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>The benefits of exercise and remaining active</td>
<td>28% (2)</td>
<td>72% (5)</td>
</tr>
<tr>
<td>The benefits of managing and controlling your weight and a balanced diet</td>
<td>42% (3)</td>
<td>58% (4)</td>
</tr>
<tr>
<td>The benefits of foot hygiene and how to maintain general foot health</td>
<td>28% (2)</td>
<td>72% (5)</td>
</tr>
<tr>
<td>The benefits of suitable footwear and insoles</td>
<td>72% (5)</td>
<td>28% (2)</td>
</tr>
<tr>
<td>An explanation of how your foot problem affects your foot and how you walk</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>How to prevent injuries, including occupational i.e., through work and sports and leisure</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>3. Have you been given information to help you manage your own foot problems?</td>
<td>28% (1)</td>
<td>72% (6)</td>
</tr>
<tr>
<td>If yes, what information have you been given to help you care for your feet at home?</td>
<td>28% (1)</td>
<td>72% (6)</td>
</tr>
<tr>
<td>Information to help you identify factors that lead to foot problems</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>Information to help you identify the signs and symptoms of foot problems</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>Information to help you prevent problems such as corns and calluses from forming</td>
<td>28% (2)</td>
<td>72% (5)</td>
</tr>
<tr>
<td>Information on how to manage pain in your feet at home</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>Information on how to access the equipment you need to keep as mobile as possible</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>Information on how to recognize the signs and symptoms for when you need to seek help from a health professional</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>The consequences of your condition on your feet and on your overall health</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>4. Does your health professional explain how your lifestyle may affect the health of your feet and the symptoms you experience?</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>5. When you first saw a health professional about your feet, did they</td>
<td></td>
<td></td>
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<tr>
<td>Ask you questions about your general health</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>Do a complete examination of your feet</td>
<td>42% (3)</td>
<td>58% (4)</td>
</tr>
<tr>
<td>Ask you questions about your lifestyle, work, and leisure activities</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>Ask you questions about pain in your feet</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>Take X-rays or other types of scans</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>Carefully watch you while you walk</td>
<td>42% (3)</td>
<td>58% (4)</td>
</tr>
<tr>
<td>6. Were you given written information about your diagnosis or symptoms?</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>7. Were you given a choice of treatment options?</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>8. Do you feel that your foot health has been monitored well?</td>
<td>42% (3)</td>
<td>58% (4)</td>
</tr>
<tr>
<td>9. Were you asked if you understood and accepted the results of your last appointment and that you understood what would happen next?</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>10. Have you ever been recommended for surgery?</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>11. Have you received self-management information specific to your condition?</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
<tr>
<td>12. Did you receive a foot health assessment within 6 months of the diagnosis of your connective tissue disorder (scleroderma)?</td>
<td>0% (0)</td>
<td>100% (7)</td>
</tr>
<tr>
<td>13. Have you received a foot assessment every year?</td>
<td>14% (1)</td>
<td>86% (6)</td>
</tr>
</tbody>
</table>

SSc: systemic sclerosis
The presence of biomechanical foot deformities imposes a compensation in a pronated overall foot posture; this posture of the foot occurs in both static and dynamic positions. This pronation is especially pathological if it occurs during the propulsion period of the gait cycle, and it is responsible for joint hypermobility and instability of the distal bony segments. This situation leads to postural changes in the entire body and subluxations in the metatarsal joint and phalanges.

A biomechanical intervention with plantar orthotic therapy has the aim to reach the neutrality of target joints and could reduce the painful symptoms and prevent future complications. Plantar orthotic therapy, by increasing the patient autonomy, can reduce pain and disability and improve the QoL.

According to the literature, SSc is a disease causing disability and with a negative impact on the QoL, with consequences on daily activities, on a patient’s employment, and on social relationships. The results of our study are in agreement with the literature, as shown by the SF-36 and HAQ (27-29).

The impact of SSc on foot health is poorly assessed. However, the few published studies show that podiatric problems are potentially disabling for patients with SSc (3, 7). This is confirmed in our study by the values of pain, disability, and activity limitations assessed by the FFI.

Foot health could influence and/or cause a reduced QoL and disability in patients with SSc.

The aims of the present study was to investigate the status of received podiatric services, foot pain, disabilities, and biomechanical foot deformities.

Large surveys of foot problems in the general population are still lacking; therefore, we could not compare the frequency of foot problems in patients with SSc with that in healthy subjects. We suggest that future studies on this subject should search for correlations between foot problems, quality of life, and disability in patients with SSc.

Our study is not without its limitations; further research on biomechanical podiatric evaluation with a control group and a larger sample is needed. However, in this study, we demonstrated the near absence of a podiatric evaluation for patients with SSc. Moreover, information on foot health due to the disease and preventative information are insufficient, and a multicenter study is needed to strengthen these podiatric results.

In conclusion, communication between the rheumatologist and podiatrist should be strengthened in order to address foot problems adequately and not to underestimate them. Moreover, SSc should be treated by a multidisciplinary team, and current practice should include a podiatrist in order to assess foot problems and then create a therapeutic plan. Care of the foot is an important part in the management of patients with SSc. An improved information system would facilitate the self-management of low-risk problems and would be useful in identifying high-risk problems.

Ethics Committee Approval: Ethics committee approval for this study has received from the ethics committee of AOU Careggi.

Informed Consent: Informed consent has obtained from the patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.S.M.; Design - S.S.M.; Supervision - S.S.M.; Data Collection and/or Processing - G.R., B.C., K.E.A.; Analysis and/or Interpretation - A.D.R.; Writing Manuscript - G.R., K.E.A.; Critical Review - B.C., A.D.R.

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References


