Surgical Management and Outcomes of Patients with Idiopathic Peroneal Spastic Flatfoot: A Retrospective Case Series

Vahdet Uçan, MD*  
Mehmet Demirel, MD*  
Orkhan Aliyev, MD*  
Fatih Yıldız, PhD*  
Gökçer Uzer, PhD*

*Department of Orthopaedics and Traumatology, Bezmialem Vakif University, Istanbul, Turkey.

Corresponding author: Vahdet Uçan, MD, Department of Orthopaedics and Traumatology, Bezmialem Vakif University, Fatih, Istanbul, Turkey 34100. (E-mail: vahdetucan@gmail.com)

Background: Although tarsal coalition represents the most common cause of peroneal spastic flatfoot, its existence cannot be verified in several cases. In some patients with rigid flatfoot, no cause can be detected after clinical, laboratory, and radiologic examination, and the condition is called idiopathic peroneal spastic flatfoot (IPSF). This study aimed to present our experience with surgical management and outcomes in patients with IPSF.

Methods: Seven patients with an IPSF, who were operated on between 2016 and 2019, and followed for at least 12 months were included, whereas those with known etiologies, such as tarsal coalition or other etiologies (traumatic, etc.) were excluded. All patients were followed up for 3 months with botulinum toxin injection and cast immobilization as a routine protocol,
and clinical improvement was not achieved. The Evans procedure and grafting with tricortical iliac crest bone graft in 5 patients and subtalar arthrodesis in 2 patients were performed. The American Orthopaedic Foot and Ankle Society ankle-hindfoot scale scores (AOFAS) and Foot & Ankle Disability Index (FADI) scores were obtained pre- and postoperatively from all patients.

**Results:** In physical examination, all feet manifested by rigid pes planus with varying degrees of hindfoot valgus and limited subtalar motion. Overall, the mean AOFAS and FADI scores significantly increased from 42 (range = 20-76) and 45 (range, 19-68) preoperatively ($p = 0.018$) to 85 (range = 67-97) and 84 (range, 67-99) ($p = 0.043$) at the final follow-up, respectively. No major intra- or post-operative complications were observed in any of the patients. All CT and MRI scans revealed no evidence of tarsal coalitions in any of the feet. All radiological workups failed to demonstrate secondary signs of fibrous or cartilaginous coalitions.

**Conclusion:** Operative treatment seems to be a good option in the treatment of patients with IPSF who do not benefit from conservative treatment. In the future, it is recommended to investigate the ideal treatment options for this group of patients.

Peroneal spastic flatfoot is characterized by a rigid and frequently painful planovalgus deformity with peroneal muscle spasms in children or adolescents. Although tarsal coalition, a bony or fibrocartilaginous intertarsal bar, represents the most common cause of peroneal spastic flatfoot, its existence cannot be verified in several cases. Other rare causes implicated in the

How to cite this article: JAPMA 112 (4): e1-e23; doi: http://doi.org/10.7547/21-210.
etiology of this disorder include osteomyelitis, tuberculosis, trauma, osteoarthritis, rheumatoid arthritis, reflex sympathetic dystrophy, nonspecific tarsal synovitis, osteoid osteoma, and neoplasms \(^2\)\(^-\)\(^6\). In some patients, however, no cause can be detected after clinical, laboratory, and radiologic examination, and the condition is called idiopathic peroneal spastic flatfoot (IPSF) \(^7\).

For the treatment of spastic peroneal flatfoot with an identifiable etiology, several conservative and surgical management strategies are available in the literature \(^8\). Nonetheless, to the best of our knowledge, there is a paucity of published data on the management of IPSF without known etiology (ie. coalition or other diagnosable entities). Accordingly, it may be difficult to determine the optimum treatment for this distinct group of patients.

This study aimed to present our experience with surgical management and outcomes in patients who were referred to our clinic for treatment of IPSF.

**Methods**

Patients who underwent surgical treatment due to spastic peroneal flatfoot between January 2016 and May 2019 at a single tertiary referral center were retrospectively reviewed. Inclusion criteria for the study were: (1) patients with the diagnosis of IPSF whose complete clinical, laboratory, and radiological examination failed to detect a cause, (2) a minimum follow-up of 24 months, and (3) being willing to participate in the study. Exclusion criteria were: (1) patients

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How to cite this article: JAPMA 112 (4): e1-e23; doi: http://doi.org/10.7547/21-210.
with identifiable etiologies for their foot deformities (ie. coalition or other diagnosable entities), (2) lost to follow-up, (3) inadequate medical and/or radiological records, and (4) being unwilling to participate in the study.

A total of 15 children had been operated for the treatment of peroneal spastic flatfoot. Eight children were excluded (one was lost to follow-up, one had inadequate radiographic imaging, and six had the diagnosis of a tarsal coalition), and the remaining seven patients who met the inclusion criteria were enrolled in the study and invited to a final follow-up appointment. Parents were informed that medical records could be used for scientific purposes only; thus, written informed consent was obtained at the final visit. Approval from the institutional review board of Bezmialem Vakif University (N27042020-02) was obtained before the acquisition of written consent and enrollment of participants.

The diagnosis of IPSF was established based on the history, clinical examination, and radiological assessment. Demographic and clinical data were collected from the hospital electronic database including age at the time of surgery, gender, involvement side, duration of preoperative symptoms, and follow-up duration. BMI (kg/m2) on admission was calculated, and children were categorized by actual weight status as obesity (>+2 SD) overweight (>+1 SD), normal (<−2 to +1 SD), and underweight (<−2 SD) in accordance with WHO standards. Intra-and post-operative complications were also recorded.

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Clinical Outcome Measures

The flatfeet were clinically examined preoperatively by the senior author as to whether there is rigidity, fixed hindfoot valgus, and restriction in subtalar motion, in order to distinguish IPSF from the flexible pes planus. The American Orthopaedics Foot and Ankle Society (AOFAS)-Ankle Hindfoot Scale and Foot & Ankle Disability Index (FADI) score were performed immediately preoperatively and at the final follow-up.

Radiological Outcome Measures

All radiographic examinations were done on the weight-bearing anteroposterior (AP) and lateral radiographs of the foot to quantify alignment by a single orthopedic surgeon with a special interest in foot and ankle surgery. On the AP radiographs, the talocalcaneal angle (Kite’s angle) and talonavicular coverage angle were measured preoperatively and at the final follow-up (Fig. 1). On the lateral radiographs, the talus-first metatarsal angle (Meary’s angle), angle of the longitudinal arch (calcaneal–fifth metatarsal angle), calcaneal inclination angle (calcaneal pitch), and lateral talocalcaneal angle was measured preoperatively and at the final follow-up (Fig. 2).

All patients underwent computed tomography (CT) and magnetic resonance imaging (MRI) before surgery. All CT and MRI scans were carefully reviewed in the coronal, axial, and
sagittal planes to rule out a possible fibrous, cartilaginous, or bony coalition across the three facets of the subtalar joint and calcaneonavicular area.

Furthermore, all the feet were examined with particular attention through radiographs, MRI and CT scans with respect to secondary signs of fibrous or cartilaginous coalitions, including irregularity of the cortical surface of the tarsal navicular, proximity of the navicular and calcaneus, flattening of the navicular, and hypoplasia of the talar head.\textsuperscript{11,12}

Treatment strategy and operative technique

Based on the senior author’s preference and experience, all patients initially underwent non-operative treatment including botulinum toxin injection, cast immobilization for four weeks, followed by physical therapy. When non-operative treatment for at least three months failed to relieve symptoms that interfere with daily activities or sleep, surgery was performed.

All operations were performed by a single experienced foot and ankle surgeon. All patients were initially examined under general anesthesia in terms of hindfoot flexibility. In five patients whose heels could be easily reduced from their valgus position, the Evans calcaneal osteotomy with autologous tricortical iliac bone graft was performed for lateral column lengthening. When performing the Evans procedure, an osteotomy 1 to 2 cm proximal to the calcaneocuboid joint was first performed and then distracted using a laminar spreader. Once the graft was inserted, the osteotomy site was fixed with two K-wires. In the other two patients, whose subtalar joint
motion was limited with osteoarthritic changes, and heel valgus could not be reduced under general anesthesia, subtalar arthrodesis with autologous cancellous iliac bone graft was performed. The arthrodesis site was fixed with 4.5 mm cancellous screws. Additionally, peroneal tendon lengthening was performed in all cases. Gastrocnemius recession was performed in one patient undergoing the Evans procedure and one patient undergoing subtalar arthrodesis. The operated foot was placed in a below-knee non-weight-bearing cast for a minimum of six weeks postoperatively in all patients.

Statistical Analysis

All statistical analyses were done using SPSS software (version 25, Armonk, NY: IBM Corp.). A P-value <0.05 was regarded significant. Normality tests were conducted using the Shapiro–Wilk test and histogram graphics. Data are presented as “minimum”, “maximum”, and “arithmetic mean”. For differences from pre- to final follow-up, a paired sample T test was used.

Results

Seven feet of seven patients were analyzed whose clinical, radiologic, laboratory, and rheumatologic workup failed to detect an underlying cause for the peroneal spastic flatfoot.

All patients were male. The mean age at the time of the surgery was 17 (range = 16–19) years. The mean follow-up was 50 (range = 32–77) months. The mean BMI was 25.9 kg/m² (range = 22.5 to 28.4). 5 children were considered as overweight and 2 children as normal.

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Clinical Outcomes

In physical examination, all feet manifested by rigid pes planus with varying degrees of hindfoot valgus and limited subtalar motion. Rigid flatfeet resulted in considerable gait impairment in all cases. The talar head was prominent along with moderate to obvious forefoot abduction in all feet. The pain was localized to the hindfoot in all patients.

Overall, the mean AOFAS and FADI scores significantly increased from 42 (range = 20-76) and 45 (range, 19-68) preoperatively (p = 0.018) to 85 (range = 67-97) and 84 (range, 67-99) (p = 0.043) at the final follow-up, respectively. In patients undergoing the Evans calcaneal osteotomy, the mean AOFAS and FADI scores improved from 43 (range = 20-76) and 48 (range = 19-67) preoperatively to 87 (range = 86-90) and 84 (range = 67-98) at the final follow-up, respectively. Similarly, in two patients undergoing subtalar arthrodesis, the preoperative AOFAS ameliorated from 31 and 48 to 89 and 67 postoperatively, respectively; the preoperative FADI scores improved from 38 and 39 to 99 and 72 postoperatively, respectively (Tables 1 and 3).

No major intra- or post-operative complications were observed in any of the patients. One patient who underwent subtalar arthrodesis developed a superficial wound infection that resolved on treatment with an antibiotic (Table 1).
Radiological outcomes

In all cases, the radiographic solid union was achieved in either osteotomy (Fig. 3) or arthrodesis site (Fig. 4) within six months after surgery. Overall, all radiographic measurements showed statistically significant improvement postoperatively compared to preoperative measurements (p < 0.05).

On the AP radiographic measurements, the talocalcaneal angle (Kite’s angle) and talonavicular coverage angle decreased from 28° (range = 15°-38°) and 13° (range = 1°-31°) preoperatively to 16° (range = 12°-21°) and 1.7° (range = 0°-14°) at the final follow-up, respectively. On the lateral radiographic measurements, the talus-first metatarsal angle increased from -4.71° (range = -8°-0°) to 1.15° (range = -4°-6°) postoperatively. Angle of the longitudinal arch decreased from 163° (range = 153°-173°) preoperatively to 157° (range = 147°-166°) postoperatively. Calcaneal inclination angle increased from 11.5° (range = 8°-15°) preoperatively to 18.6° (range = 13°-26°) at the final follow-up. Lateral talocalcaneal angle decreased from 33° (range = 28°-37°) preoperatively to 22.7° (range = 12°-31°) at the final follow-up (Tables 2 and 3).

Discussion

The foot deformity observed in our patients has been referred to as peroneal spastic flatfoot in the literature, and such patients typically manifest with rigid pes planus and varying degrees of

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hindfoot valgus. While the tarsal coalition has been implicated as the major cause of peroneal spastic flatfoot, osteomyelitis, tuberculosis, trauma, osteoarthritis, rheumatoid arthritis, nonspecific tarsal synovitis, reflex sympathetic dystrophy, osteoid osteoma, and neoplasms have been identified as other causes. When no apparent cause can be identified, the condition has been called IPSF. That is, IPSF is mainly a diagnosis of exclusion, and thus a coalition and all other apparent etiologies must be carefully reviewed and ruled out. In our case series, the diagnosis of IPSF was established as clinical, radiologic, laboratory, and rheumatologic workup failed to detect an underlying cause for the peroneal spastic flatfoot.

To the best of our knowledge, the focus of the existing literature has mostly concentrated on peroneal spastic flatfoot secondary to tarsal coalitions; however, little has been published regarding the disorder as a distinct clinical entity. Accordingly, IPSF is poorly known in orthopedic surgery, and its clinical features, pathophysiology, and proper management have not been explained sufficiently. A few previous studies including patients with IPSF are relatively old and not designed according to the current standards. In one of those studies, Rankin and Baker, in 1974, reviewed 24 adult patients with a rigid flatfoot, 7 of whom had no known etiology for peroneal spastic flatfoot and no evidence of tarsal coalition following a limited analysis of only plain radiographs. Dwyer, in 1976, highlighted a minor group of patients with peroneal spastic flatfeet characterized by subtaloid stiffness without apparent structural radiographic changes in their foot. The author attributed this type of

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peroneal spasm to either general foot strain or a form of joint irritation. As seen above, some of the available information on IPSF is only based on simple radiographic analyses without considering rheumatologic, neoplastic, or infectious disorders.

More recently, Luhmann et al. 12 (2000) identified 9 adolescent patients with 13 painful idiopathic rigid flatfeet without known etiology (ie. coalition or other apparent entities that may result in peroneal spastic flatfoot), 8 of whom were greater than the 95th percentile in weight for their age. Differently from the previous studies, they excluded all possible causes for peroneal spastic flatfoot in their patients based on comprehensive radiologic (including MRI and CT scan), neurologic, and rheumatologic workup. The authors hypothesized that the mechanism for IPSF in the obese skeletally immature patient is a mechanical overload of the ligamentous and bony construct of the longitudinal and transverse arches. Similar to Luhmann et al., we established the diagnosis of IPSF after a thorough clinical, radiologic, laboratory, and rheumatologic workup in young male patients. In our case series, most patients (70%) were also overweight, thus supporting Luhmann et al.’s hypothesis.

Due to the rarity of data on IPSF, to our knowledge, there are no guidelines in the literature for optimal management of such patients. Based on the treatment approach for patients with spastic flatfoot in the presence of tarsal coalition, our first-line treatment was non-operative, including botulinum toxin injection and cast immobilization. As non-operative treatment failed to relieve symptoms and improve function, we operated on all patients.
Similar to our treatment approach, Luhmann et al. \(^{15}\) initially applied non-operative treatment for all their patients. After the failure of non-operative treatment, they performed peroneal lengthening in 4 feet, a calcaneal lengthening in one foot, and subtalar arthrodesis in two feet. We decided which surgical technique to use depending on the assessment of subtalar mobility under general anesthesia in addition to the presence of subtalar osteoarthritic changes.

In our patients, either the Evans osteotomy or subtalar arthrodesis were performed. In the literature, these two techniques are the most recommended treatment options for spastic peroneal flatfoot with tarsal coalition \(^{16,17}\). While the Evans calcaneal lengthening osteotomy was performed in 5 feet whose heels could be reduced from their valgus position, subtalar arthrodesis was performed for the other two feet whose subtalar joint motion was limited with osteoarthritic changes. In all patients, peroneal lengthening was performed to improve foot alignment and to prevent a recurrence of peroneal spasms. Besides, complications related to lateral column lengthening (the Evans procedure) have been reported in the literature \(^{17-20}\), comprising persistent pain, undercorrection, overcorrection, graft slippage/collapse, or calcaneocuboid subluxation. Postoperatively, we observed no such postoperative complications in our case series and obtained significant improvements in radiographic parameters including Meary's angle, calcaneal inclination angle, lateral and anteroposterior talocalcaneal angle (kite angle), and talus coverage angle indicating restcration of flatfoot deformity. Our results indicated that both surgical methods, the Evans procedure or subtalar arthrodesis, can be

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effective at improving function and quality of life in patients with IPSF unresponsive to non-operative treatment based on the significant improvement in AOFAS and FADI scores if the proper method is used.

When interpreting the findings in this study, some limitations and strengths should be considered. The major limitations were its retrospective nature and limited sample size. Despite these limitations, our study is one of two studies that present the surgical management and outcomes of IPSF patients in whom other possible causes of peroneal spastic flatfoot such as rheumatic disorders, infection, or neoplasm were ruled out after a thorough history, clinical examination, and laboratory work-up. These results can be considered as a guide for further studies. Finally, all diagnoses and operations were performed by a single experienced foot and ankle surgeon.

Conclusions
IPSF is mainly a diagnosis of exclusion, and thus a coalition and all other apparent etiologies must be carefully reviewed and ruled out. When conservative treatment fails, surgical intervention can be recommended. The surgical procedure of choice should be individualized for each patient. The Evans procedure or subtalar arthrodesis seems to be effective at improving function and quality of life as well as reconstructing the rigid flatfoot in patients with

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IPSF at mid-term follow-up, with the significant improvement in clinical and radiographical results.

Financial Disclosure: None reported.

Conflict of Interest: None reported.

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3. Cowell HR. Diagnosis and management of peroneal spastic flatfoot. *Instr Course Lect* 1975;24: 94.


How to cite this article: JAPMA 112 (4): e1-e23; doi: http://doi.org/10.7547/21-210.


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This Original Article is a preprint. It has been reviewed, accepted for publication, and approved by the author but has not been copyedited, proofread, or typeset.

Table 1. Demographic characteristics and clinical outcomes of study participants

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of patients</td>
<td>7</td>
</tr>
<tr>
<td>Gender</td>
<td>7 males</td>
</tr>
<tr>
<td>Age (year), mean (min to max)</td>
<td>17 (16 – 19)</td>
</tr>
<tr>
<td>Number of the feet operated for symptomatic rigid flatfoot</td>
<td>7</td>
</tr>
<tr>
<td>Follow-up (month), mean (min-max)</td>
<td>50 (32 – 77)</td>
</tr>
</tbody>
</table>

**Clinical findings**

- **Heel alignment**
  - Heel valgus →

- **Medial longitudinal arch**
  - Low in all feet

- **Subtalar motion**
  - Restriction in all the cases.

**AOFAS score, mean (min-max)**

<table>
<thead>
<tr>
<th></th>
<th>Preoperative</th>
<th>Final Follow-up (p = 0.018)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>42 (20 – 76)</td>
<td>85 (67 – 97)</td>
</tr>
</tbody>
</table>

**FADI score, mean (min-max)**

<table>
<thead>
<tr>
<th></th>
<th>Preoperative</th>
<th>Final Follow-up (p = 0.043)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>45 (19 – 68)</td>
<td>84 (67 – 99)</td>
</tr>
</tbody>
</table>

**Complications**

- Major complication → None
- Minor complication → 1 patient
  - Superficial wound infection

*AOFAS = The American Orthopaedic Foot and Ankle Society; FADI = Foot and Ankle Disability Index Score. The p value was set at p<0.05*
Table 2. Patients’ radiographic measurements

<table>
<thead>
<tr>
<th>Radiographic parameter</th>
<th>Preoperative</th>
<th>Final Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>AP X-ray examination</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Talocalcaneal angle</td>
<td>28° (15°-38°)</td>
<td>16° (12°-21°)</td>
</tr>
<tr>
<td>Talonavicular coverage angle</td>
<td>13° (1°-31°)</td>
<td>1.7° (0°-14°)</td>
</tr>
<tr>
<td><strong>Lateral X-ray examination</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Talus-first metatarsal angle</td>
<td>4.71° (-8°-0°)</td>
<td>1.15° (-4° - 6°)</td>
</tr>
<tr>
<td>Angle of the longitudinal arch</td>
<td>163° (153°-173°)</td>
<td>157° (147° – 166°)</td>
</tr>
<tr>
<td>Calcaneal inclination angle</td>
<td>11.5° (8°-15°)</td>
<td>18.6° (13° - 26°)</td>
</tr>
<tr>
<td>Lateral talocalcaneal angle</td>
<td>33° (28°-37°)</td>
<td>22.7° (12°-31°)</td>
</tr>
</tbody>
</table>

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Table 3. Clinical and radiographical outcomes of all study participants

| Patient no. | Age | Operated Side | Operation                  | AOFAS score Pre- and post-operative | FADI score Pre- and post-operative | Anteroposterior talocalcaneal angle | Talonavicular coverage angle | Talus-first metatarsal angle | Angle of the longitudinal arch | Calcaneal inclination angle | Lateral talocalcaneal angle |
|------------|-----|---------------|----------------------------|-------------------------------------|-----------------------------------|-------------------------------------|-------------------------------|-----------------------------|-----------------------------|-----------------------------|-----------------------------|---------------------------|
| 1          | 18  | Right         | Subtalar arthrodesis       | 31→89                               | 38→99                             | 22→15                               | 8→4                           | 4→4                         | 153→159                    | 19→21                       | 30→16                      |
| 2          | 17  | Right         | Subtalar arthrodesis       | 48→67                               | 39→19                             | 37→18                               | 4→0                           | 4→3                         | 160→166                    | 13→13                       | 37→12                      |
| 3          | 19  | Left          | Evans calcaneal osteotomy  | 20→86                               | 19→93                             | 38→21                               | 26→14                         | 5→0                         | 173→154                    | 10→20                       | 31→30                      |
| 4          | 16  | Right         | Evans calcaneal osteotomy  | 31→86                               | 39→67                             | 34→16                               | 31→0                          | 8→6                         | 165→160                    | 10→13                       | 30→23                      |
| 5          | 16  | Right         | Evans calcaneal osteotomy  | 20→89                               | 68→98                             | 30→16                               | 19→5                          | 7→1                         | 165→147                    | 8→22                        | 37→31                      |
| 6          | 16  | Right         | Evans calcaneal osteotomy  | 76→90                               | 60→67                             | 21→17                               | 4→0                           | 5→0                         | 165→154                    | 12→26                       | 37→26                      |
| 7          | 18  | Left          | Evans calcaneal osteotomy  | 20→67                               | 52→95                             | 15→12                               | 1→1                           | 0→2                         | 164→160                    | 13→15                       | 28→21                      |
Figure 1. Measurement of anteroposterior foot radiographic parameters. The talocalcaneal angle is the angle between the longitudinal axes of the talus (blue line) and calcaneus (yellow line). The talonavicular coverage angle refers the angle between the articular surfaces of the talar head (red line) and proximal navicular (white line).
Figure 2. Measurement of lateral foot radiographic parameters. The talus-first metatarsal angle is the angle between a line drawn from the center of the longitudinal axis of the talus (blue line) and the first metatarsal (red line) (a). The angle of the longitudinal arch is formed between a line along the inferior edge of the 5th metatarsal (white line) and the inclination axis of the calcaneus (yellow line) (a). Calcaneal inclination angle is formed between the calcaneal inclination axis (yellow line) and the horizontal surface of the foot (white line) (b). Lateral talocalcaneal angle is the angle between the calcaneal inclination axis (yellow line) and the mid-talar axis (blue line) (b).
Figure 3. Preoperative (a and b), early (c and d), and late (e and f) postoperative radiographs of a patient with idiopathic peroneal spastic flatfoot undergoing the Evans calcaneal osteotomy. Note that improvement in Meary’s angle before and after surgery.
Figure 4. Preoperative (a) and (b) postoperative lateral standing radiographs of a patient with idiopathic peroneal spastic flatfoot undergoing subtalar arthrodesis.