# $\langle$ Clinical Research angle

# Idiopathic Toe Walking Tests and Family Predisposition

Abstract: The aim of this study is to provide clinical examination methods that were designed specifically to assess the level of severity among children with idiopathic toe walking (ITW). The idiopathic toe-walking pattern of 836 children was recorded and analyzed during 4 years. Questionnaires and clinical measurements were evaluated, along with differential tests, assessing the occurrence and severity of toe walking. Questions about family *bistory and onset of toe walking were* evaluated along with special tests and measurements assessing the occurrence and severity of toe walking. The different measurements apply during this study, ankle dorsiflexion, lumbar lordosis angle, as well as the clinical spin test, walking after spin test, and heel walking test revealed in all cases that children with a positive family predisposition were significantly more affected than children with negative family predisposition. It is concluded that children with ITW and a positive family predisposition were more intensively affected during all performed clinical tests than children with no family predisposition. The tests used during this study have not being used by any other researches, even though they showed significant differences between the children with ITW and children with a normal gait pattern.

Levels of Evidence: Diagnostic, Level II: development of diagnostic test with consecutive patients and control patients

Keywords: pediatric podiatry; age-related problems; toe walking; forefoot, toe, midfoot, hereditary/ genetic disorders; general disorders

diopathic toe walking (ITW) was described for first time in 1967 as a congenitally short tendocalcaneous.<sup>1</sup> Nowadays it is common to find it as idiopathic toe walking or habitual toe walking. ITW

important to describe first the normal ankle kinetic during the normal gait pattern."

In order to describe the ITW

[idiopathic toe walking] pattern, it is

can only be diagnosed in the absence of any medical condition known to cause toe walking, such as cerebral palsy, muscular dystrophy, spina bifida, or autistic spectrum disorders.<sup>2</sup>

Typically, ITW gait pattern in individuals is characterized for walking since the beginning on their first steps on the toes. Nevertheless, they can support their entire foot on the floor on request or when concentrating on their gait.<sup>2-4</sup> Some literature claims that idiopathic toe walkers could only normalize the David Pomarino, MScB, Juliana Ramírez Llamas, MSc (Sports Performance), and Andrea Pomarino, MD

characteristics on the 17% of the trials, but during the other 70% of the trials children with ITW can normalize either the parameters during the stance phase or during the swing phase; however, they were not able to normalize the pattern during both phases.<sup>5</sup>

In some literature, it is found that some children with ITW have  $0^{\circ}$  of dorsiflexion

at the ankle joint; while other idiopathic toe walkers present a decreased or normal dorsiflexion between 5° and 20°.<sup>6</sup>

Furthermore, clinical characteristics such as a decrease on range of motion (ROM) in the upper ankle joint for habitual toe walkers is still under discussion and several studies have mentioned a severe decrease of ROM for this condition.<sup>7,8</sup> This is often the only clinical characteristic, which is used to describe toe walking and suggests the severity of this walking pattern.

DOI: 10.1177/1938640016630056. From the Research Department at Praxis Pomarino, Hamburg, Germany. Address correspondence to Juliana Ramírez Llamas, Research Department at Praxis Pomarino, Rahlstedter Bahnhofstrasse 9, 22143 Hamburg, Germany; e-mail: julir83\_ramirez@yahoo.com. For reprints and permissions queries, please visit SAGE's Web site at http://www.sagepub.com/journalsPermissions.nav. Copyright © 2016 The Author(s)

In order to describe the ITW pattern, it is important to describe first the normal ankle kinetic during the normal gait pattern. According to Perry,<sup>9</sup> the ankle kinetic is divided into 3 rockets. The first rocket, start with at heel strike followed by an ankle plantiflexion. During this instant, there is an eccentric contraction of the tibialis anterior. At the second rocket, the ankle has some degree of dorsiflexion and there is an eccentric contraction of the gastrocnemius. At the third rocket, the ankle plantiflexes and there is concentric contraction of the gastrocnemius and soleus muscles. During the third rocket, the push-off action takes place.

Some studies have showed that during the ITW pattern, there is an absence of the first rocket since there is a landing on the sole or the forefoot instead of the heel; the second rocket is inverted, and during the swing phase the ankle plantiflexion seems right after the foot contacts the ground, followed by a reverse movement of the ankle in relation with the tibia, with an increased plantiflexion.<sup>5</sup> During electromyography, there is a premature activation of the gastrocnemius muscle throughout the swing phase, and it is manifested in a premature ankle plantiflexion.<sup>10,11</sup>

The main objective of this study is to present clinical measurements as well as test procedures of how to evaluate children with ITW. In addition, this study aims to determine whether or not there is a difference in severity between children with positive family predisposition and negative family predisposition.

### **Methods**

#### **Proceeding and Participants**

Between June 2006 and June 2010, 923 children from 2 to 13 years of age were referred for toe-walking treatment. All of them were referred exclusively by local pediatricians with the diagnosis of ITW. In the inclusive criteria for this study, it was considered that the diagnosis was based on the fact that the children were walking on the toes permanently or habitually for more than 6 months and no neurological or orthopedic disease signs were found at examination by the referring pediatricians. A total of 87 children were taken out of the study since the children did not walk permanently on the toes or they walked on the toes for less than 6 months or the parents gave incongruent answers between the first and second questionnaires.

Informed consent was obtained from all the parents, who had to complete a standardized anamnesis. Hereafter, the children were examined by 2 investigators and children underwent a self-constructed standardized initial physical examination.

Regular follow-up examinations with a standardized physical examination form were carried out every 2 to 3 months to monitor and estimate the therapy phase. The therapy used to treat these children was the pyramidal insoles<sup>12</sup>; in some cases, in combination with physical therapy. After the first follow-up examination after 2 months, the parents had to answer the parental questionnaire once again to check the reliability of the answers.

All data of hetero-anamnesis and initial respectively follow-up examinations were stored in the patient files and an intern database. The anonymized data of in total 836 toe walkers was evaluated.

To validate the results of the initial physical examination of the idiopathic toe walkers a randomly assigned control group of 55 children with normal gait (NG) was recruited. The normal walkers were healthy and were reviewed by 2 investigators in order to reject any orthopedic or neurologic gait abnormalities. The statistics of the 836 Toe walkers (TW) and 55 normal walkers are shown in Table 1. The age distributions ITW-total and NG-total are homogeneous.

In order to collect the tests data, 5 different investigators performed the initial and follow-up examinations. To increase the interrater reliability of the results, all the involved investigators were introduced and supervised by the first author to standardized physical examination by special training courses and seminars. Therefore, all measurements were conducted in accordance to the standardized examination guidance. Afterward, retests with healthy volunteers and ITW were carried out and indicated an inter-rater and retest reliability of more than 80%.

The children with ITW were also divided into 2 categories, children with positive family predisposition (PF) and children with negative family predisposition (NF). A total of 484 children had a negative family predisposition, while 352 had a positive family predisposition.

#### Examination

A questionnaire was given to the parents to find out about family predisposition (Questionnaire 1 in Appendix A).

After the hetero-anamnesis, 2 investigators started to examine every child on basis of the standardized physical examination guidelines. This physical examination will be described in the following section.

From past experience, it was observe than many children who were brought for examination would hide the ITW pattern during the consultation with the doctors. Therefore, the following tests were developed in order to provoke this idiopathic pattern without the children's awareness. The spin, and after spin walking tests, induce the children to walk on the toes. The heel walking test was designed to induce the compensations that the children with ITW have to do in order to achieve a heel walking pattern. The ankle ROM and the lordosis measurements are tests than children are not able to manipulate during the medical examination.

#### **Initial Examination**

*1. Performance of Spin Test.* The child was exposed to a challenging situation by spinning fast on the same place. A maximum of 10 spins should be performed and the number of the spin at which the child began toe walking was noted (1-10, 99 = no findings). The test is positive when the child stands on the forefoot after the third spin. The

#### Table 1.

Clinical Tests and Measurements of TW and NG.<sup>a</sup>

Test or Measurement	PF-TW (% ± 95% Cl 95) <sup>b</sup> N = 352 Subjects	NF-TW (% ± 95% CI) N = 484 Subjects	NG (% ± 95% CI) N = 55 Subjects	χ²( <i>P</i> ) <i>P</i> 1°/ <i>P</i> 2 <sup>d</sup> / <i>P</i> 3 <sup>e</sup>
Spin test	239 (67.9 ± 4.9)	284 (58.7 ± 4.4)	2 (3.6 ± 4.9)	.007/<.001/<.001
Walking test	291 (82.7 ± 4.0)	366 (75.6 ± 3.8)	1 (1.9 ± 3.5)	.017/<.001/<.001
Heel walking	177 (50.3 ± 5.2)	295 (61.0 ± 4.3)	53 (96.4 ± 4.9)	.002/<.001/<.001
ROM of ankle joint				
Knee extended $\geq 15^{\circ}$	68 (19.3 ± 4.1)	161 (26.4 ± 4.2)	47 (85.5 ± 9.3)	.017/<.001/<.001
Knee bent ≥20°	78 (22.2 ± 4.3)	142 (29.3 ± 4.1)	45 (81.8 ± 10.2)	.021/<.001/<.001
Lumbar lordosis				
Angle $\ge 35^{\circ}$	185 (52.6 ± 5.2)	234 (48.3 ± 4.5)	7 (12.7 ± 8.8)	.235/<.001/<.001
Angle ≥40°	138 (39,2 ± 5.1)	154 (31.8 ± 4.1)	5 (9.1 ± 7.6)	.028/<.001/<.001
Angle $\geq$ 45°	50 (14.2 ± 3.6)	42 (8.7 ± 2.5)	2 (3.6 ± 4.9)	.014/.029/.296

Abbreviations: TW, toe walking; NG, normal gait; CI, confidence interval; PF, positive family disposition; NF, negative family disposition; ROM, range of motion.

<sup>a</sup>For the spin and walking after spin tests, it is given the number of subjects that were positive for the test. For the heel walking test, the number given is the number of subjects who were able to achieve the test allowing compensations. For the ROM at the ankle joint is given the number of subjects who achieve more than 15° or more than 20°, respectively. For the lordosis measurement there are given the number of subjects with 35°, 40°, and 45° for the different categories, respectively.

<sup>b</sup>Number (%  $\pm$  95% Cl). <sup>c</sup>*P*1 = *P*(PF-TW, NF-TW).

 ${}^{d}P2 = P(PF-TW, NG).$  ${}^{e}P3 = P(NF-TW, NG).$ 

severity of the test is given by the number of steps that the child achieves without showing the tip toe walking pattern. The test was positive when the tip toe walking pattern appeared. The earlier on the number of steps the tip toe pattern appears, the more affected is the child.

2. Performance of Walking After Spin Test. Right after the spin test the child was requested to walk 10 steps. The step in which the child started toe walking was noted (1-10, 99 = no findings). The test was considered positive for TW when the tiptoe walking pattern appeared. The severity of the test is given by the number of steps that the child achieves without showing the tiptoe walking pattern. The earlier the tiptoe pattern appears, the more affected is the child. 3. Performance of Heel Walking Test. The child was requested to heel walk. Heel walking test is considered possible, if the child manages to perform at least 4 steps on its heels. Evasive movements such as flexion and/or external rotation in the hip were allowed. Predefined input: [yes, no]. The compensations or adjustments made by the child were also recorded. The severity of the test was determined by the compensations or adjustments that the child needs to achieve in order to perform the heel walking pattern. Forward inclination of the trunk in combination with the inability to dorsiflex the ankle are the most common compensations. The test was positive for toe walking when the child was unable to heel walk or the child heel walked showing trunk, knee, and ankle compensations.

4. Range of Motion for the Ankle Joint in Dorsiflexion. Measurements of passive ankle dorsiflexion at initial examination were made 2 times by using a goniometer. First time, in supine position (SP) with the knee joint extended, and the second time in prone position (PP) with the knee bent at 90°. Maximum ankle dorsiflexion was measured with the heel in neutral valgus and rounded up to the nearest 5° interval. The motion restriction on the ankle is one main indicator for children with toe walking. The greater the restriction, the more affected the child will be by ITW.

5. Angle Degree of the Lumbar

*Lordosis.* The goniometer was placed at the point of the greatest convexity at the lumbar spine when the child was in upright and neutral zero position. Lumbar lordosis angle was measured and rounded up to the nearest 5° interval. Children with idiopathic toe walking develop a prominent lumbar lordosis. The higher the degrees of lordosis, the more affected will be the child by this medical condition.

The measurements and tests 1 to 5 were carried out to serve in our study as an indicator of the severity of toe walking. To our knowledge the tests 1, 2, and 3 have not been applied to ITW and were created by the first author.

For statistical analysis, SPSS version 18 was used: chi-square and/or Fisher-Yates test,  $\chi^2(P)$ ; Kolmogorow-Smirnow test, KS(*P*); Wilcoxon-Mann-Whitney test, WM(*P*).

## Results

#### **Initial Examination**

Table 1 and Appendix B show the results of the 5 initial examination tests. A total of 352 participants were enrolled in the PF-TW group, 484 in the NF-TW group, and 55 in the NG group.

#### **Spin Test**

The spin test revealed that PF-TW have been more severely affected by toe walking than NF-TW.

#### **Walking After Spin Test**

The walking test revealed that PF-TW have been more severely affected by toe walking than NF-TW.

#### **Heel Walking Test**

The heel walking test revealed that PF-TW have been more severely affected by toe walking than NF-TW, since less PF-TW were able to perform the heel walking test.

## Range of Motion for the Ankle Joint at Dorsiflexion

During the measuring of the ankle ROM it was performed with the knee extended (ROM  $\ge$  15° at the ankle) and with the kneed at 90° of flexion (ROM  $\ge$ 20° at the ankle). In both measurements PF-TW were significantly more severely affected by toe walking than NF-TW.

#### Lumbar Lordosis Angle

In addition to the other tests, the lumbar lordosis angle was also measured. The lumbar lordosis angle measurements show that PF-TW were more severely affected NF-TW by having a larger angle.

## Discussion

Several characteristics of idiopathic toewalking are described in the current literature. ROM at the ankle joint, electromyography studies, cinematic profiles of gait analysis were evaluated to describe and differentiate this idiopathic condition among children.<sup>13-18</sup>

To our knowledge our clinical tests and measurements spin (1), walking after spin (2), heel walking tests (3), and lumbar lordosis angle measurement (5) are described for the first time and are the result of an intensive clinical investigation process due to a sizeable examination group. Among these features, we expanded our examination process with clinical tests to provide a possible assessment of the severity of toe walking.

Spin and walking after spin tests (1, 2)were performed to evaluate equilibrium reaction and to access the toe-walking reactions. Furthermore, to provoke the tiptoe walking pattern, toe-walking children can put their foot flat on the ground on request. During walking, the children with ITW are able to normalize the 2 phases of the walking pattern in only 17% of the trials. Throughout the 70% of the trial children with ITW pattern are able to normalize either the stance phase or the swing phase; however, it is possible to notice the tiptoe walking pattern in 1 of the 2 gait phases.5

In the "spin test (1)" and the "walking after spin test (2)," children with positive family predisposition and negative family predisposition exhibit the ITW pattern by the third spin and/or by the first step, respectively. For both tests the percentage of children with positive family predisposition was higher than the percentage of children with negative family predisposition, showing than the children with a positive family predisposition were more severely affected than children with a negative family predisposition.

The heel walking test (3) was designed to measure whether the child's tibialis anterior has the strength to elevate the forefoot during the heel strike in children with ITW. During the test performance, it is essential to observe the compensations movements like trunk forward flexion and/or hyperextension of the knees indicating that children are using the hip muscles instead of the tibialis anterior in order to achieve the forefoot elevation or dorsiflexion.

During the "heel walking test (3)," only 50.3% of the children with a positive family predisposition were able to complete the task. In contrast 61.0% children with a negative family predisposition were able to take at least 4 steps on the heels. Up to now we have not found any other literature that uses the spinning (1), walking after spin (2), and heel walking tests (3).

The ankle measurement test has being described by other authors in several studies as one of the major distinctions between children with ITW and children with normal ankle conditions. In children with habitual toe walking, contractures on the gastrocnemius muscle are frequently found; therefore, there is a reduction or limitation on the ankle dorsiflexion angle.

In other studies, it was found that children who used to be idiopathic toe walkers have a 3 times greater chance of having a limitation in dorsiflexion than children with a normal gait pattern.<sup>8</sup> Therefore, it is concluded that the measurement of the ankle dorsiflexion angle is one of the most important variables to determine the severity of ITW.

All differences between a positive family predisposition and children with a negative family predisposition during the Spin test (1), walking after spin test (2), heel walking test (3) and ROM of ankle measurement (4) were significant. This confirms that children with a positive family predisposition were more severely affected by toe-walking than children with no family predisposition. Comparing the children with a normal gait pattern and children with ITW, with or without family predisposition, there were significant statistical differences showing that exists a difference in the test results between children with ITW and children with a normal gait pattern for the spinning, walking after spin, heel walking, and dorsiflexion tests.

Lumbar lordosis angle test was used since in early studies it was observed that children with ITW present increase in the lordosis angle, compared to other children. It was observed that about 49% of the children that were referred to us with a habitual toe walking pattern presented a lordosis of 35° or higher, and about 11% of the children presented a hyperlordosis of 45°.<sup>19</sup>

For all the clinical tests, the results show that children with a positive family predisposition are more affected than children with no family predisposition as it was previously showed in Table 1. Therefore, we can conclude that family predisposition could be an important indicator at the time to classify ITW.

For this study, there were a large number of children with ITW who participated in the different clinical tests. To our knowledge, this is the first time that those tests are used in order to measure the severity of ITW. In the performance of the tests it is found that there are significant differences between the children that are affected by ITW and the children with a normal gait pattern showing that the tests are objective and reliable.

## Conclusion

In conclusion, it was determined that among children with ITW with a positive family predisposition, would be more likely to be affected than children with ITW that do not have any family predisposition. In addition to the family predisposition, this research shows a clinical way to evaluate the severity of ITW through observation and specific measurements. The different tests results show clear differences between children with habitual toe walking and children with a normal gait pattern. The development of the test will help doctors to standardize this idiopathic condition and at the same time to evaluated the evolution, progression or improvement of the different therapeutic techniques that are available for this condition like physical therapy, castings, botolinum toxin type A (BTX), surgeries, and other conservative therapies. Moreover, these tests will help measure the progression or improvement of ITW in children who are or who are not being treated.

## **Appendix A**

#### **Questionnaire 1**

Please answer the next questions

- 1 "Are there other TW in the family?" with predefined answers: yes\_\_\_\_ no\_\_\_\_
- 2 If response to 1 is "yes": "Which relatives?" with predefined answers: Father\_\_\_\_ Mother\_\_\_\_ Brother\_\_\_\_ Sister\_\_\_
  - Relatives of father\_\_\_\_
  - Relatives of mother\_\_\_
- 3 "When did your child start toewalking?" with predefined answers:
  Since onset of walking\_\_\_\_\_
  Months after onset of walking\_\_\_\_
  - Years after onset of walking\_
  - Only recently\_\_\_\_

n/a

# **Appendix B**

## Results

*Initial Examination.* In Table 1, the results of the initial examination (1 to 5) of 352 PF-TW, 484 NF-TW, and 55 NG are presented.

*Spin test.* The test was positive when the child started to toe stand maximum after the third spin. A total of 239 (67.9%  $\pm$  4.9%) PF-TW, 284 (58.7%  $\pm$  4.4%) NF-TW, and 2(3.6%  $\pm$  4.9%) NG started toe walking latest by the third spin. The differences between PF-TW and NF-TW (9.2%, *P*1 = .007), between PF-TW and NG (*P*2 < .001), as well as between NF-TW and NG (*P*3 < .001) were significant. The spin test revealed that PF-TW have been more severely affected by toe walking than NF-TW.

*Walking test.* The test was positive when the child started to toe stand maximum after the third step. A total of 291 (82.7% ± 4.0%) PF-TW, 366 (75.6% ± 3.8%) NF-TW, and 1(1.9% ± 3.5%) NG started toe walking latest by the third step. The differences between PF-TW and NF-TW (7.1%, *P*1 = .017), between PF-TW and NG (*P*2 < .001), as well as between NF-TW and NG (*P*3 < .001) were significant. The walking test revealed that PF-TW have been more severely affected by toe walking than NF-TW.

*Heel walking test.* The children were asked to complete at least 4 steps on the heels. A total of 177 (50.3%  $\pm$  5.2%) PF-TW, 295 (61.0%  $\pm$  4.3%) NF-TW, and 53(96.4%  $\pm$  4.9%) NG were able to perform at least 4 steps on the heels. The differences between PF-TW and NF-TW (10.7%, *P*1 = .002), between PF-TW and NG (*P*2 < .001), as well as between NF-TW and NG (*P*3 < .001) were significant.

The heel walking test revealed that PF-TW have been more severely affected by toe walking than NF-TW.

Range of motion for the ankle joint at dorsiflexion. The test was performed to see if the ankle joint had a range of  $\geq 15^{\circ}$  performed with the knee extended. Then a ROM of  $\geq 20^{\circ}$  at the ankle joint was evaluated with the knee at 90° of flexion.

*Knee extended*: 68 (19.3% ± 4.1%) PF-TW, 161 (26.4% ± 4.2%) NF-TW, and 47(85.5% ± 9.3%) NG had a ROM of ≥15°. The differences between PF-TW and NF-TW (7.1%, P1 = .017), between PF-TW and NG (*P*2 < .001) as well as between NF-TW and NG (*P*3 < .001) were significant.

Knee bent: 78 (22.2%  $\pm$  4.3%) PF-TW, 142 (29.3%  $\pm$  4.1%) NF-TW, and 45(81.8%  $\pm$  10.2%) NG had a ROM of  $\geq$ 20°. The differences between PF-TW and NF-TW (7.1%, P1 = .021), between PF-TW and NG (P2 < .001), as well as between NF-TW and NG (P3 < .001) were significant.

In both measurements, PF-TW had less ROM at the ankle joint.

*Lumbar lordosis angle.* In addition to the other tests, the lumber lordosis angle was measured.

Angle ≥35°: 185 (52.6% ± 5.2%) PF-TW, 234 (48.3% ± 4.5%) NF-TW, and 7 (12.7% ± 8.8%) NG had an angle of ≥35°. The difference between PF-TW and NF-TW (4.3%, P1 = .235) was not significant, but between PF-TW and NG (P2 < .001) as well as between NF-TW and NG (P3 < .001) the differences were significant.

*Angle* ≥40°: 138 (39.2% ± 5.1%) PF-TW, 154 (31.8% ± 4.1%) NF-TW, and 5 (9.1% ± 7.6%) NG showed an angle of ≥40°. The differences between PF-TW and NF-TW (7.4%, *P*1 = 0.028), between PF-TW and NG (*P*2 < .001), as well as between NF-TW and NG (*P*2 < .001) were significant.

Angle  $\geq$ 45°: 50 (14.2% ± 3.6%) PF-TW, 42 (8.7% ± 2.5%) NF-TW, and 2 (3.6% ± 4.9%) NG had a measurement of ≥45°. The differences between PF-TW and NF-TW (5.5%, P1 = .014) as well as between PF-TW and NG (10.6%, P2 = .029) were significant. Between NF-TW and NG, the difference was not significant (P3 = .296).

The lumbar lordosis angle measurements show that PF-TW had a significantly larger lumber lordosis angle than NF-TW and NG.[AS]

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