〈 Clinical Research 〉

Outcomes of Noninvasively Treated Idiopathic Toe Walkers

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Abstract: *Idiopathic toe walking (ITW) causes a common problem in pediatric orthopaedics. In the literature, numerous treatment options have been reported, but consensus about the management of ITW is still missing. The aim of the current study was to evaluate conservative treatment with pyramidal insoles. A total of 193 patients underwent conservative treatment between January 2010 and June 2013. Mean age at the beginning of the treatment was 7.75 ± 0.23 years (range 2.0-17.0 years). For all patients, demographic data, comorbid diseases, passive range of motion (ROM), persistent toe walking, and performed operations were retrospectively evaluated. Following operative treatment was defined failure. Eight (4.15%) patients underwent Achilles tendon lengthening operation after mean therapy time of 2.72 years (range 0.1-7.0 years), 174 cases were treated successfully (90.16%). In 50 cases, toe walking suspended completely after mean therapy time of 2.83 years. In cases of failure, patients were older at diagnosis and at the beginning of the treatment. Mean passive ROM increased over the time. In cases of failure, ROM decreased from the first to the second examination. Conservative treatment of ITW using pyramidal insoles can be effective. Ankle dorsiflexion significantly improved in the patients who were successfully treated. Therapy should start early. A decrease of ROM under therapy should lead to critical revisal of individual therapy.*

Levels of Evidence: *Therapeutic, level IV: Case series*

Keywords: pediatric podiatry; age-related problems; orthotic therapy; diagnostic and therapeutic techniques; physical therapy; toe walking; forefoot; toe; midfoot; tendo-Achilles lengthening (TAL); heel; rearfoot; ankle

Toe walking in children from the age of 2 years, for the duration of over half a year, and for more than 50% of the day is known as idiopathic toe walking (ITW).[1-7] ITW appears in approximately 5% of children after their second birthday and causes a common problem in pediatric orthopaedics.[1,8,9] A spontaneous remission occurs in approximately 70% of these cases within half a year after the ITW started.[1,4,8-10] The role of gender and genetics is still unclear.[1,7,9,11-15] In ITW, the cause for the changed gait pattern are not found to be only neurological or myogenic.[1,9,11-13,16] Differential diagnosis like infantile cerebral palsy, muscular dystrophy, spinal amyotrophy, and hereditary motor sensory neuropathy as well as rare metabolic disorders of the musculature must be considered.[1,7,9,11-13,17] ITW is also reported in patients with neuropsychiatric disorders, autistic spectrum disorders, and in children with language or cognitive developmental delay.[4,16,18-21] Untreated ITW may lead to structural changes as limiting mobility in the upper ankle, broadening of the front foot, and the development of a contracted foot.[7,8,12,13,22-26] Further health problems may arise due to the change in body statics, problems including

> "The effectiveness of treatment options is not clear and consensus about the management of ITW [idiopathic toe walking] is still missing."

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Table 1.

Idiopathic Toe Walking (ITW) Treatment Options in the Literature.

Authors	Year	Journal	No. of Cases	Treatment Options and Results
McMulkin et al	2006	*J Pediatr Orthop*	14	*Operative treatment* (gastrocnemius or Achilles tendon lengthening) → Dorsiflexion in stance significantly improved
Kogan et al	2001	*J Pediatr Orthop*	10	*Operative treatment* (percutaneous Achilles tendon lengthening) → No recurrence of the ITW
Hemo et al	2006	*J Pediatr Orthop*	15	*Operative treatment* (Achilles tendon lengthening) → Normalization of ankle kinematics, but plantarflexion power did not show normal levels (1 year postoperative)
Hirsch and Wagner	2004	*Acta Paediatr*	14	CONSERVATIVE treatment (all patients had physiotherapy, 5 had additional orthoses or plaster) → 8 of 14 walked without a tiptoe pattern at follow-up, 1 of 14 was permanently relieved of tiptoe walking
Griffin et al	1977	*J Bone Joint Surg*	6	*Conservative treatment* (casts) → All had normal electromyographic gait patterns during heel-toe gait
Fox et al	2006	*Acta Orthop Belg*	44	*Conservative treatment* (below knee walking casts and Achilles tendon stretching exercises) → 29 of 44 (66%) completely stopped toe-walking, dorsiflexion significantly improved
Engström et al	2010	*J Pediatr Orthop*	15	*Conservative treatment* (botulinum toxin A injection and stretching program) → 9 of 14 showed an improvement
Engström et al	2013	*J Bone Joint Surg Am*	47	*Conservative treatment* (below-the-knee casts versus botulinum toxin A injection and below-the-knee casts) → No difference in any outcome parameter
Current study	2017		193	*Conservative treatment* (all patients received pyramidal insoles, 95.5% had additional physiotherapy, 60% had additional orthoses → 95.8% successful treatment

backache stemming from intensified lumbar lordosis and knee pain due to additional burden on the anterior knee joint compartment.[1,2,4,17,26,27] In the literature, numerous treatment options have been reported, including physical therapy, insoles, and orthopaedic footwear, castings, orthoses, instillation of botulinum toxin type A (Btx A), and soft-tissue surgery. The studies show heterogeneous study population and inconclusive results (Table 1).[11,12,23,27-31] The effectiveness of treatment options is not clear and consensus about the management of ITW is still missing. Because of missing knowledge about "natural history" of ITW, some authors proclaim treatment only in cases of parents' worry.[10] A classification of ITW may be helpful in developing an exact therapy and prognosis. The classification according to Pomarino into 3 categories may be used to do this.[1,2,6,32] In rare cases, a surgical extension of the Achilles

Figure 1.

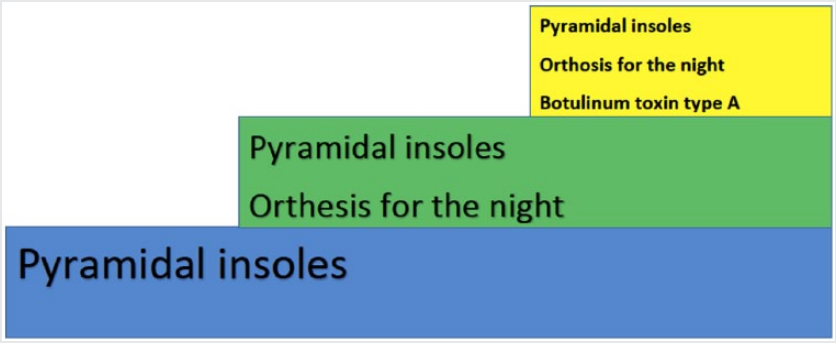
Pyramidal insoles according to Pomarino.

Figure 2.

Close-up view of the pyramidal insoles according to Pomarino.

Figure 3.

Conservative treatment according to Pomarino in 3 steps.



tendon is necessary.[3,11,23,24,28,30] In most of the cases, the range of motion (ROM) increased after an operative Achilles lengthening.[3,11,23,24,28,30] The aim of the current study was to evaluate conservative treatment with pyramidal insoles in a high number of cases.

Material and Methods

The study has been previously approved by the local Ethics Committee (EK No. 2399-2014). Initially, data were collected from 193 idiopathic toe walkers who underwent conservative treatment between January 2010 and June 2013 in a pediatric orthopaedic unit belonging to a university hospital.

In the setting demographic data, comorbidities, treatment, and passive ROM with extended knee are collected consecutively every 6 months (visits 1, 2, and 3). Complications (eg, persisting toe walking) are reported. Following operative Achilles lengthening is defined as failure.

For this study, cases with the International Statistical Classification of Diseases and Related Health Problems code (ICD-10 code) R26.8 were identified from the local hospital information systems OrDIS 14 (AGFA Health Care, Bonn, Germany) and TurboMed (CompuGroup Medical, Koblenz, Germany). All patients were noninvasively treated with pyramidal insoles according to Pomarino,[1,31] which were individually manufactured by an orthopaedic technician. The insoles are all custom made. Structure and height of the pyramids are created referring to anatomic landmarks from an individual static footprint (ossa metatarsalia II-IV, tuberosity of the calcaneus and the central part of the plantar aponeurosis). The insoles are not yet approved by the Food and Drug Administration for any purpose. They are not labeled for the use under discussion in the United States. Using supportive elements in pyramid shape under the second, third, and fourth metatarsal bone, patients are forced to actively step onto their entire foot, due to corrective pressure applied by the cushion (Figures 1-3).[1,2]

The pyramidal insoles were used in most cases in combination with physical therapy.

For all patients, demographic data, comorbid diseases, persisting toe walking, and performed operations were retrospectively evaluated from the local hospital information system (electronic medical records). Additional, 2 different study groups of successful cases and failed cases were formed. Demographic data, comorbid diseases, and ROM were analyzed for both groups.

Statistical Methods

In the first step, data were collected in Microsoft Excel 1997-2003 (Microsoft Corporation, Redmond, WA, USA). Statistical examination was performed using SPSS Version 24.0 (IBM Corp, Armonk, NY, USA) for Windows (Microsoft Corporation, Redmond, WA, USA). For the collected data, a descriptive statistical evaluation was performed, estimating mean value including 95% confidence interval, standard error, median, upper and lower extremity, minimum and maximum, and range. Second, an explorative statistical analysis was performed using Kolmogorov-Smirnov test and Shapiro-Wilk test (depending on the sample size) to evaluate the effects of demographic data, comorbid diseases and ROM on the outcome.

Results

A total of 193 idiopathic toe walkers were included in the analyses. Of these,

Table 2.

Presence of Comorbidities (Number).

Comorbidity	Whole Study Population (N = 193)	Successful Cases (n = 174)	Failure Cases (n = 8)
Cerebral palsy	19	19	0
Developmental speech disorders	10	10	0
Autism spectrum disorders	5	5	0
Hip dysplasia	8	8	0
Vertebral blockade			
Thoracic spine	11	11	0
Lumbar spine	5	5	0
Blockade of the sacroiliac joint	27	27	1
Prematurity	7	7	0

Table 3.

Mean Dorsiflexion for the Right Ankle Joint Over Time.

Dorsiflexion (deg)	Visit 1, n (%)	Visit 2, n (%)	Visit 3, n (%)
0	67 (34.7)	39 (20.2)	23 (11.9)
5	44 (22.8)	29 (15.0)	16 (8.3)
10	39 (20.2)	38 (19.7)	38 (19.7)
15	16 (8.3)	22 (11.4)	27 (14.0)
20	15 (7.8)	25 (13.0)	20 (10.4)
≥25	7 (3.5)	5 (2.5)	5 (2.6)
Total	188 (97.41)	158 (81.87)	129 (66.84)

52 were female (26.9%), 141 were male (73.1%) (female:male ratio is calculated as 1: 2.7). Mean age at diagnosis was 6.10 ± 0.21 years (range 1.0-15.0 years). Mean age at the beginning of the treatment was 7.75 ± 0.23 years (range 2.0-17.0 years). All patients were treated with pyramidal insoles. A total of 185 patients underwent additional physical therapy (95.9%). Overall, 60.1% had additional orthosis for the night. When the treatment started, the toe walking was existent for mean 4.30 ± 0.23 years (range 0-14.0 years). Mean treatment time was 1.94 ± 0.1 years (range 0-6.0 years). Comorbidities are presented in Table 2. Mean ROM over the time is presented in Tables 3 and 4 and in Figures 4 and 5.

Of 193 patients, 8 underwent Achilles tendon lengthening operation (4.15%) after mean therapy time of 2.72 years (range 0.1-7.0 years), 174 cases were treated successfully (90.16%). In 50 cases, toe walking suspended completely after mean therapy time of 2.83 years. In 124 patients, toe walking was still occasionally present, and therapy was still ongoing. In this group, mean therapy time was 4.07 years, 11 patients were lost to follow-up. Operative treatment was defined failure. Demographic data and ROM for successful treatment and in case of failure are presented in Table 5. In cases of failure, patients were older at diagnosis and at the beginning of the treatment (mean 7.25 years at diagnosis vs 6.0 years and 8.88 years at the beginning of the treatment vs 7.7 years).

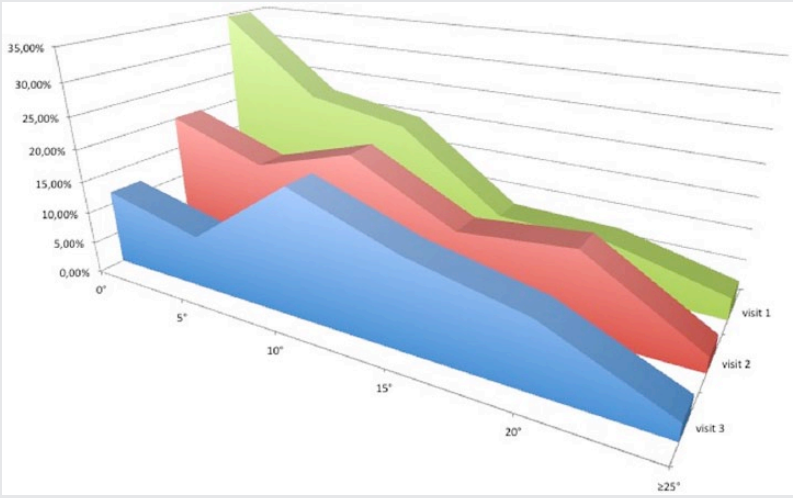
Table 4.

Mean Dorsiflexion for the Left Ankle Joint Over Time.

Dorsiflexion (deg)	Visit 1, n (%)	Visit 2, n (%)	Visit 3, n (%)
0	74 (38.3)	40 (20.7)	25 (13.0)
5	44 (22.8)	27 (14.0)	17 (8.8)
10	38 (19.7)	40 (20.7)	41 (21.2)
15	13 (6.7)	27 (14.0)	22 (11.4)
20	13 (6.7)	23 (11.9)	20 (10.4)
≥25	7 (3.6)	3 (1.5)	5 (2.6)
Total	189 (97.93)	160 (82.90)	130 (67.36)

Figure 4.

Mean dorsiflexion (degrees) for the right ankle joint over time (visits 1-3).



In the therapy failure group, mean dorsiflexion for right and left ankle were less at presentation compared with the successful group. Mean passive ROM increased over time. In cases of failure, ROM decreased from the first to the second examination (Table 5, Figures 6 and 7). Only 1 patient with a comorbidity resulted in a failure.

Discussion

This current study is, to our knowledge, the largest reported in the literature that deals with conservative treatment of ITW. The study results indicate that conservative treatment can be effective. An improvement in all parameters was shown.

ITW is a common problem in pediatric orthopaedics.[1,8,9] The majority of the patients in this study were male (73% male, ratio female:male is 1:2.7). Comparable groups with a majority of male patients were described in most cases in the literature.[3,12,13,16,22,30,33] Contrarian results were described by Griffin et al[29] and Kornbrust.[34] But they dealt with smaller study groups.

The role of genetics in ITW is still unclear. Fox et al[12] described a family history of ITW in 43.3% of their male study population and in 14.3% of their female patients (mean 34.1%). Katz and Mubarak[35] presented a study with hereditary tendo Achilles contracture patients who showed an autosomal dominant condition with variable expression. They described it as a benign condition, and operative treatment was not necessary in these cases.[35] On the basis of this study, we conclude that genetics may also play a role in ITW patients. Other study groups are more critical about this. Sala et al[9] suggest that ITW may be a manifestation of a global neurodevelopmental condition.

Numerous treatment options have been reported in the literature.[11,12,23,27-31] Studies analyzing effects of physical therapy, insoles and orthopaedic footwear, castings, orthoses, instillation of Btx A and soft-tissue surgery lead to inconclusive results.[11,12,23,27-31] Fox et al reported in a prospective study with 44 patients that following below-the-knee walking casts, 66% showed an improved dorsiflexion and improved gait.[12] In an electromyography study, Griffin et al[29] showed successful ITW treatment with plaster casts. After plaster casts, the ROM increased and muscle synergy pattern normalized.

Figure 5.

Mean dorsiflexion (degrees) for the left ankle joint over time (visits 1-3).

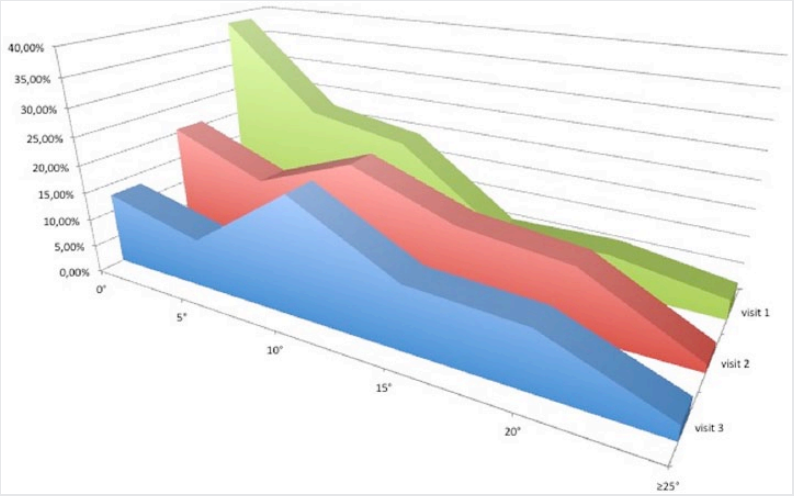


Table 5.

Demographic Data and Range of Motion for Successful Treatment and in Case of Failure.

	Successful Therapy Group	Failed Therapy Group
Mean age at diagnosis (years)	6.05	7.25
Mean age at the beginning of the therapy (years)	7.70	8.88
Ratio male:female	2.7:1	3:1
Mean dorsiflexion (deg) right ankle		
At visit 1	7.60	4.29
At visit 2	9.93	0.71
At visit 3	11.35	2.14
Mean dorsiflexion (°) left ankle		
At visit 1	6.59	2.86
At visit 2	9.77	0
At visit 3	10.97	2.14

Kornbrust[34] analyzed 95 patients who were treated conservatively with insoles (type of proprioceptive insoles). A total of 81.4% of these cases were treated successfully. Nevertheless, Hirsch and Wagner[11] came to the conclusion that nonsurgical treatment of ITW does not have a long-lasting effect and that the natural history of ITW leads to a spontaneous remission.

Randomized controlled studies analyzed the effects of Btx A injections. Engström et al[31] showed in a consecutive series of 15 children that a single Btx A injection combined with an exercise program may improve walking pattern in gait analysis. Engström et al[30] showed in a further study that adding Btx A injection prior to below-the-knee casts does not improve the result.

Dietz and Khunsree[10] conclude that ITW poses a cosmetic problem and should be treated only if the family wishes so. Others believe that untreated ITW will lead to structural changes with limitation of the mobility.[4,8,12,13,22-25,30] Descriptions speak of a shortened Achilles tendon, which limits movement in the ankle joint[1,4,8,12,13,22-24,30] as well as the development of a pes cavus or contracted foot.[1,4] Engelbert et al[36] describe how the probability to develop a severely limited range of movement in the upper ankle joint increases by 3 times in patients suffering from toe walking. Eastwood et al[3] have compared patients who received treatment for their recurrent toe walking with ones who do not.[3] The results find that toe walking continues to exist in patients who were left untreated for recurrent toe walking, 50% of cases will see an increase in toe walking. The changes in structure, which have been described above, can make treatment more difficult and help explain why efforts to treat the toe walking in patients who were diagnosed to have toe walking at an older age often fail. According to current studies treatment for toe walking tends to fail if patients were older at the time of diagnosis and went into treatment late (7.25 years at diagnosis vs 6.0 years and 8.88 years at the beginning of treatment as opposed to 7.7 years). In light of this evidence, it seems reasonable to suggest that treatment should be started as early as possible in order to maximize success.

In the presented study, decreased ROM led to an Achilles tendon lengthening operation in eight cases. The authors therefore conclude that ITW causes more

Figure 6.

Range of motion (ROM) for successful treatment and in case of failure (right foot).

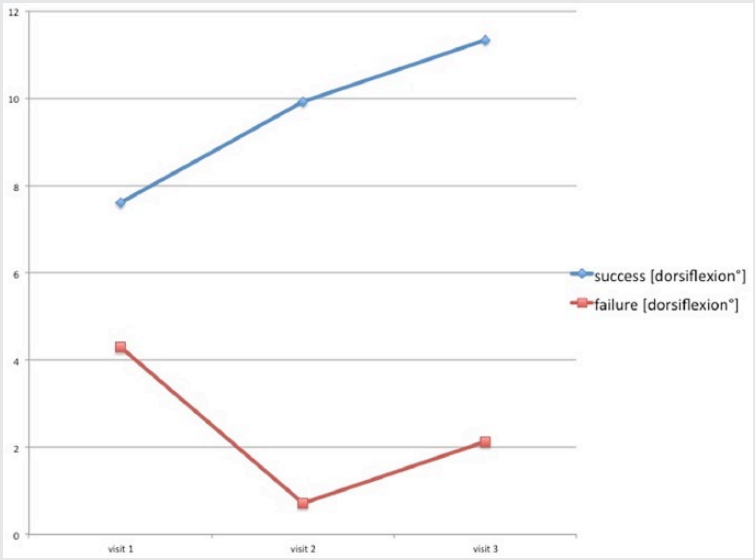
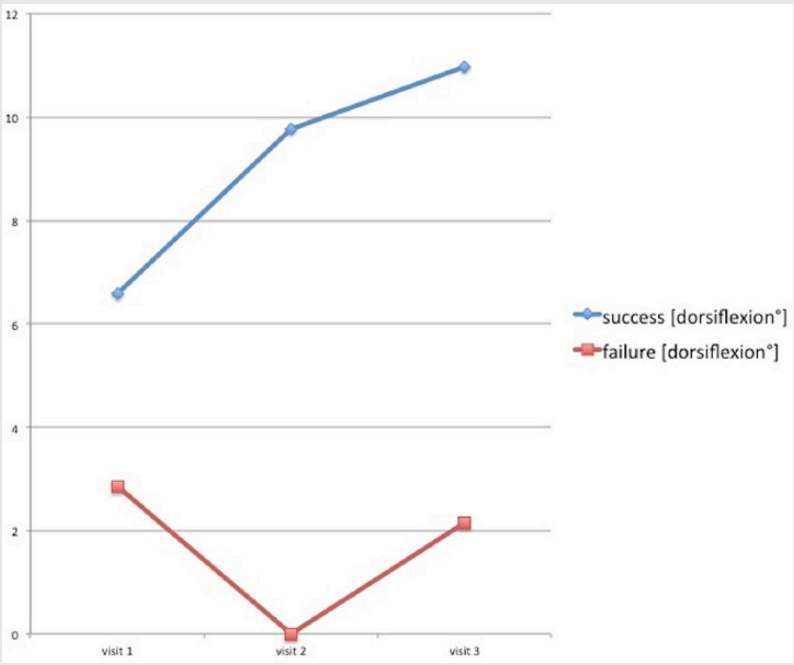


Figure 7.

Range of motion (ROM) for successful treatment and in case of failure (left foot).



than a cosmetic problem and should be treated if diagnosed. Surgery (eg, Achilles tendon or gastrocnemius lengthening operation) is described to be a successful treatment in ITW.[24,27] McMulkin et al[27] showed a normalization of kinematic variables in gait analysis after operative treatment.

A limitation of the study is the retrospective design that cannot control bias. Furthermore, the study population was inhomogeneous. A total of 95.9% had additional physical therapy, and 60.1% had additional orthoses for the night. A mean gap of 1.65 years was reported between the mean age at diagnosis and the beginning of the treatment with insoles. It resulted due to missing absorption of costs by the health insurance, absence of patient's compliance or due to missing prescription. This is a further limitation of this retrospective study design and should be considered in further study protocols with prospective design. Whether the effect of pyramidal insoles can be improved with physical therapy or additional orthoses is unknown and should also be studied in the future.

A prospective randomized controlled trial comparing outcomes of observation and treatment with insoles and treatment with additional physical therapy and with night orthoses would better control for biases. Addition, different types of insoles should be compared in further studies.

Declaration of Conflicting Interests

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Ethical Approval

The study was approved by the local Ethics Committee (EK No. 2399-2014).

Informed Consent

Not applicable.

Trial Registration

Not applicable. FAS

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