

## Systematic Review

# Idiopathic toe walking: What's New? An Integrative Review

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## Abstract

**Objective:** Consolidate the current knowledge on idiopathic toe walking, provide a critical overview, and identify areas for potential future research.

**Methods:** An electronic search was conducted in the following databases up to June 2023: MEDLINE, EBSCO, Embase, CINAHL Plus, and PubMed. The PICO framework was employed to define search terms.

**Results:** Despite considerable methodological heterogeneity among studies, a stronger inclination was found to investigate etiological and prognostic factors. There is a trend for a higher prevalence in boys with a positive family history. Novel classifications have been proposed to improve differential diagnosis. Among conservative treatment options, there is stronger evidence for gait improvement using serial casting followed by orthoses. Severe cases may benefit from surgical zone III gastrocnemius lengthening.

**Conclusion:** Further studies with standardized methodologies are required to clarify questions about this condition's etiology, classification, and treatment. Nonetheless, there is a higher level of evidence supporting conservative treatments with serial casting and orthoses and zone III gastrocnemius lengthening for severe cases in the second decade of life.

**Level of evidence II; Diagnostic studies.**

**Keywords:** Walking; Equinus Deformity; Foot; Treatment.

## Introduction

Idiopathic toe walking (ITW) is a gait deviation characterized by the persistence of a forefoot strike pattern at initial contact throughout most of the gait cycles beyond the age of three years without an apparent cause<sup>(1,2)</sup>. The diagnosis is one of exclusion, necessitating the elimination of neurological, neuromuscular, and primary orthopedic abnormalities; therefore, assessing strength, reflexes, and selectivity must be normal<sup>(3,4)</sup>.

Genetic syndromes like McArdle syndrome, muscular dystrophies, autism spectrum disorder (ASD), and peripheral neuropathies—such as Charcot-Marie-Tooth disease and cerebral palsy—are highlighted among the differential diag-

noses. Children with mild spastic diplegic cerebral palsy have a considerable resemblance to ITW<sup>(5)</sup>. However, in ITW, there is adequate knee extension at initial contact and the end of the swing in the gait cycle. These characteristics indicate good selective motor control, a primary distinctive factor of cerebral palsy<sup>(5,6)</sup>.

The natural history of ITW remains uncertain, and further studies with higher levels of evidence are required to support decisions on when and how to intervene<sup>(7)</sup>. Divergence among authors is substantial, with some mentioning spontaneous resolution for most children<sup>(8,9)</sup>. In contrast, others suggest the persistence of toe walking in approximately 20% to 50% of untreated cases, varying only the severity<sup>(10)</sup>.

Study performed at the Hospital Israelita Albert Einstein, São Paulo, SP, Brazil.

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The prevalence also lacks consensus, with some citing ITW in approximately 5% of children<sup>(8,11)</sup>, while others point to as much as 24% in the preschool population<sup>(5)</sup>.

The first publication on the subject was made by Hall et al. in 1967<sup>(10)</sup>, describing it as a congenital contracture of the Achilles tendon, commonly referred to as congenital short Achilles tendon, a theory that prevailed until the early 21st century, influencing generations of orthopedic surgeons. However, the subject gained new approaches and nomenclatures over the last two decades, and different classifications were proposed. Currently, the term Idiopathic Toe Walking is the most widely accepted and used.

Several etiological hypotheses have gained visibility, such as the theory of sensory processing dysfunction (SPD), with studies suggesting a possible association with vestibular dysfunctions<sup>(12)</sup>. Additionally, genetic etiology has been considered, with the potential of an autosomal dominant variable expression characteristic<sup>(13)</sup>. A positive family history is frequently observed, ranging from 30%<sup>(14)</sup> to 88%<sup>(13)</sup>.

The main consequences of persistent toe walking include the development of plano-valgus feet, increased external tibial torsion, pain, fatigue, imbalance, and psychological impact<sup>(15)</sup>.

Various severity classification methods have been proposed. In 2007, Alvarez et al. proposed categorizing it into mild, moderate, and severe based on the presence or absence of three primary criteria identified in three-dimensional gait analysis. These criteria include the presence of the first ankle rocker, the presence of an early third rocker, and the predominance of the first wave of ankle plantar flexor moment<sup>(16)</sup> (Table 1) (Figure 1).

In 2021, Westberry et al. proposed modifying the Alvarez classification, replacing the kinematic assessment of the first ankle rocker with the sagittal ankle kinetics<sup>(17)</sup>. They introduced a dorsiflexor moment within the initial 10% of the gait cycle as the defining criterion for the first ankle rocker (Figure 2).

Three-dimensional gait analysis adds diagnostic and prognostic value to ITW. Efforts are being made to correlate the most effective treatments for each severity level. However, criticism exists regarding this analysis method, as the environment in which the gait is performed may lead the child to walk differently from their habitual gait. Studies

are investigating alternative methods, such as using inertial sensor garments (wearables)<sup>(18)</sup>, though this technique is still under investigation.

The most common treatment methods described in the literature include physical therapy, sensory therapy, serial casting, botulinum toxin A (BoNT-A) injections, orthotic use (2-10 weeks), pyramid insoles, and surgical gastrocnemius lengthening with or without the use of casting and orthotics.

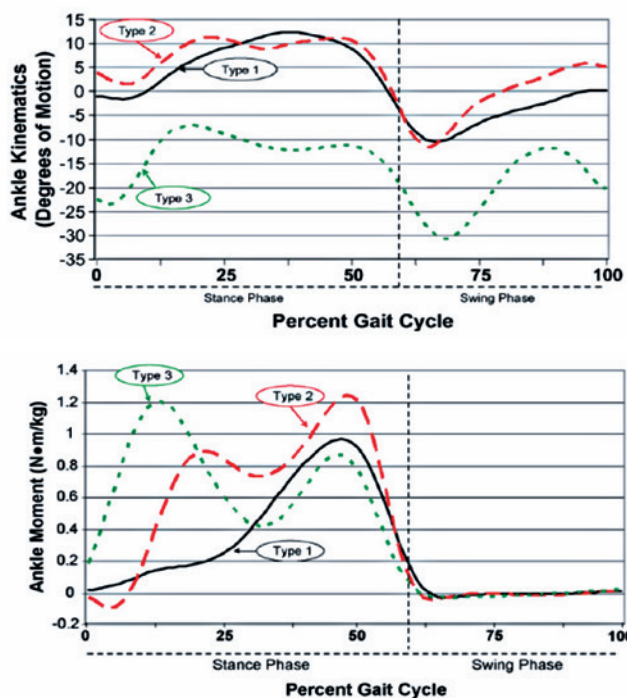
The objective of this study is to consolidate the current knowledge on ITW, provide a critical overview, and identify areas for potential future research.

### Methods

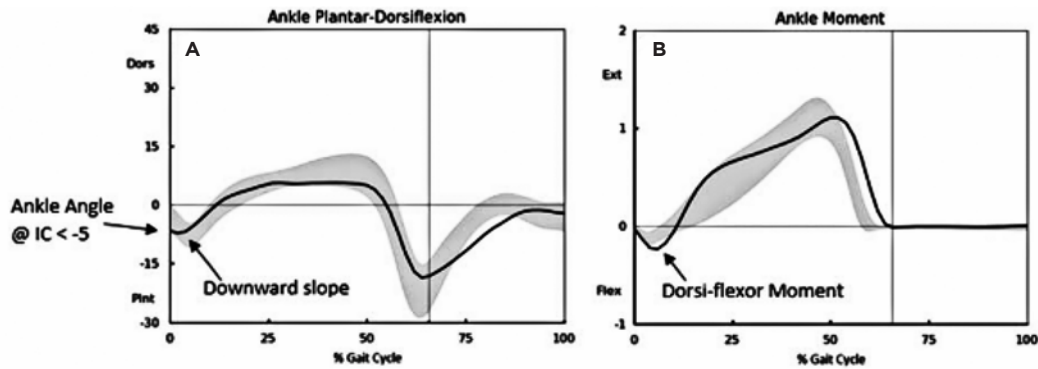
This literature review was conducted following the PICO (Population, Intervention, Comparison, and Outcome) model as the search framework for terms. The following databases were searched from September 2022 to June 2023: MEDLINE, EBSCO, Embase, CINAHL Plus, and PubMed. The keywords were idiopathic toe walking, tiptoe walking, tiptoe gait, gait, and toe walking gait. Inclusion criteria comprised studies involving children diagnosed with ITW discussing diagnosis, risk factors, classifications, treatment, and prognosis. The data analyzed were from published articles in peer-reviewed journals in English. Case reports and studies exclusively focused on patients with cerebral palsy or primary Achilles

**Table 1.** Criteria for ITW classification. (Adapted from Alvarez et al., 2007)<sup>(16)</sup>

Idiopathic Toe Walking: Primary Criteria and Definitions			
	First ankle rocker	Early third rocker	Predominance of the first wave of ankle plantar flexor moment
<b>Type 1</b>	Yes	No	No
<b>Type 2</b>	Yes/No	Yes/No	No
<b>Type 3</b>	No	Yes	Yes



**Figure 1.** Representation of ankle kinematics and kinetics graphs illustrating the assessed parameters for Alvarez classification. (Reproduced from Alvarez et al., 2007)<sup>(16)</sup>



	Toe walking severity group	First Rocker Qualifiers			Primary criteria and definitions		
		Ankle Angle at Initial Contact > -5	Down-going ankle excursion	Presence of dorsi-flexor moment	Presence of first ankle rocker	Presence of early third ankle rocker	Predominant first ankle moment
Alvarez	Type2	No	Yes		No	No	No
Modified Alvarez	Type1			Yes	Yes	No	No

**Figure 2.** Representation of the proposed modification by Westberry et al. to the Alvarez first ankle rocker classification, based on the presence of dorsiflexor moment within the initial 10% of the gait cycle in the sagittal plane ankle kinetics. (Replicated from Westberry et al. 2021)<sup>(17)</sup>

tendon contractures were excluded. The selection process involved reading abstracts, and if they did not meet the exclusion criteria, full articles were assessed.

**Results**  
**Age**

Kelly et al. (1997)<sup>(1)</sup> and Sutherland et al. (1980)<sup>(2)</sup> consider ITW to be the persistence of toe walking beyond the age of three. Sobel et al. (1997)<sup>(14)</sup>, when evaluating children who predominantly walked on their toes since the initiation of walking, observed that 35% of children continued this pattern until age two and 7% between nine and 15 years. Engström and Tedroff (2018)<sup>(8)</sup> noted that 79% of children with ITW ceased toe walking naturally by the age of ten.

Despite the evident spontaneous improvement in most children, studies by Sobel et al. (1997)<sup>(14)</sup> and Engelbert et al. (2011)<sup>(5)</sup> demonstrate that children with persistent toe walking without undergoing treatment have higher rates of shortening and contracture of the calf muscles (Table 2).

**Etiology and risk factors**

Regarding etiology and potential risk factors, there is ongoing research into family history. In Sobel's et al. study (1997)<sup>(14)</sup>, a positive family history was present in 30% of cases, while Fox et al. (2006)<sup>(19)</sup> reported 43.3%, Hirsh and Wagner (2007)<sup>(20)</sup> reported 57%, and Katz and Mubarak (1984)<sup>(13)</sup> reported 88%. It is important to note that Katz and Mubarak (1984)<sup>(13)</sup> identified an autosomal dominant condition when investigating Achilles tendon contracture. Although these are

different conditions, it is not a factor that can be completely ignored (Table 3).

Pomarino et al. conducted two studies<sup>(21,22)</sup> on genetic influence on the development of this condition. They observed a higher predominance in male children, raising the question of whether this pathology could have an X-linked recessive autosomal cause associated with 30% to 42% paternal family history presence.

Furthermore, some studies have proposed an association of ITW with SPD<sup>(12,23)</sup>. It is known that patients diagnosed with ASD might have a higher prevalence of ITW-up to 20.1%- with a high rate of tight heel cords (12%)<sup>(24,25)</sup>. Since patients with ASD frequently present higher rates of SPD, it may be reasonable to consider this as a possible target in the treatment of ITW.

New studies are also investigating neurological changes between children with and without ITW. Donne et al. (2022)<sup>(26)</sup> found differences in the neural pathway activation in their study, as the toe walking group showed lower activation in the left frontal lobe region, which indicates a somatosensory difference between groups. However, more studies are needed to determine the true impact of these differences.

**Classification of ITW**

One of the first reference points to define treatment approaches in different clinical conditions is the severity classification. The most widely used classification for ITW is Alvarez's (2007)<sup>(16)</sup>, based on the presence or absence of the three ankle rocker mechanisms. This classification has

been considered highly sensitive according to the Cochrane systematic review by Caserta et al. (2019)<sup>(27)</sup>.

Other aspects have formed the basis for additional classifications. For instance, O’Sullivan et al. proposal (2018)<sup>(28)</sup> divides children into typical and non-typical-ITW based on differences in gait analysis. Additionally, there are classifications based on anatomical features, as described by Pomarino et al. (2017)<sup>(29)</sup>, highlighting differences in calf shape, forefoot, and Achilles tendon, among others.

Bauer et al. (2022)<sup>(30)</sup> introduced a descriptive classification for children with toe walking. In a straightforward manner, they divided it into toe walking during developmental stages, ITW with autism, congenital contracture, habitual persistent ITW (without contracture), and finally, persistent ITW with contracture (Figure 3).

### Treatment

The literature shows different management options for ITW, including conservative methods such as physiotherapy, orthotics, serial casting, and foot orthoses. Invasive approaches, such as surgical procedures (percutaneous or open), are also considered (Table 4).

### Conservative therapies (physiotherapy, serial casting, orthotics, and botulinum toxin injection)

The effectiveness of stretching and physiotherapy sessions as a curative approach to equinus gait was evaluated, and their efficacy was compared to more invasive techniques. When used in isolation, these methods did not show effectiveness<sup>(31)</sup>.

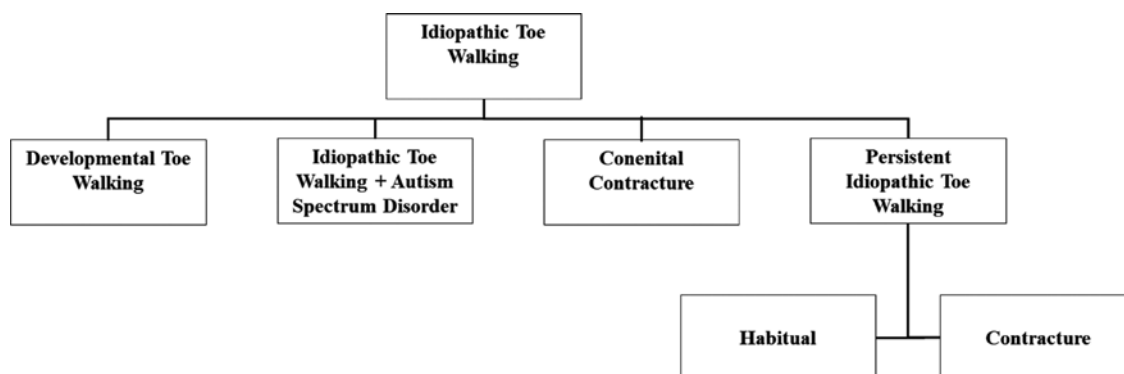


Figure 3. Idiopathic Toe Walking classification, by Bauer et al. (Replicated from Bauer et al., 2022)<sup>(20)</sup>

Table 2. Studies evaluating spontaneous resolution of ITW

Article	Authors (Publication Year)	Journal, Publication year (impact factor 22/23)	Number of ITW in the sample/ Number of the population in the study	Mean age at the beginning of the study (years)	Conclusions of the study
Idiopathic Toe-Walking: Prevalence and Natural History from Birth to Ten Years of Age	Engström P, Tedroff K (2018) <sup>(8)</sup>	The Journal of Bone and Joint Surgery (5.3)	63/1401	5.5	<ul style="list-style-type: none"> <li>- 79% of children with ITW ceased toe walking by age 10.</li> <li>- Among 4 cases of Achilles tendon contractures (defined as &lt;10° of ankle dorsiflexion), two maintained 5°, one worsened from 10° to 5°, and one progressed from 10°/15° to 0° bilaterally.</li> </ul>
Effect of Persistent Toe Walking on Ankle Equinus	Sobel E, Caselli M. A, Velez Z (1997) <sup>(14)</sup>	Journal of the American Podiatric Medical Association (0.7)	60/-	3.5	<ul style="list-style-type: none"> <li>- There was a male predominance in ITW cases (non-significant p-value).</li> <li>- ITW begins right from the onset of a child’s walking, which differs from cerebral palsy cases.</li> <li>- Approximately 30% of cases have a positive family history.</li> <li>- The mean dorsiflexion angle measures 6.2°.</li> <li>- 35% of children exhibited ITW at two years, and 7% between 9 and 15 years.</li> <li>- The presence of ITW decreases with age, accompanied by a decrease in dorsiflexion angle.</li> </ul>

**Table 3.** Studies demonstrating a possible genetic association in children with ITW

Authors (Publication year)	Journal (Impact Factor 22/23)	Percentage of children with positive family history
Katz and Mubarak (1984) <sup>(13)</sup>	Journal of Pediatric Orthopaedics (1.7)	(88%)
Sobel et al. (1997) <sup>(14)</sup>	Journal of the American Podiatric Medical Association (0.678)	(30%)
Fox et al. (2006) <sup>(19)</sup>	Acta Orthopædica Belgica (0.35)	(43.3%)
Hirsh and Wagner (2004) <sup>(20)</sup>	International Journal of Pediatrics (5)	(57%)
Pomarino et al. (2012) <sup>(21)</sup>	Foot and Ankle Specialist (0.562)	(30%-42%)

**Table 4.** Studies that investigated treatment options for ITW

Article (Publication Year)	Authors	Journal (Impact Factor)	Number of ITW sample	Mean age at the beginning of intervention (years)	Mean time of follow-up (years)	Conclusions of the study
Outcomes of Noninvasively Treated Idiopathic Toe Walkers (2017)	Radke K, Karch N, Goede F, Vaske B, Von Lewinsky G, Noll Y, Thren A <sup>(11)</sup>	Foot & Ankle Specialist (0.562)	101	7.75 (2-17)	1.94	Pyramidal insoles: - 95.5% were used with physiotherapy. - 60% were used in conjunction with orthotics. Resulted in a 95.8% resolution of ITW
Surgical Outcomes for Severe Idiopathic Toe Walkers (2021)	Westberry D. E, Carpenter A. M, Brandt A, Barre A, Hilton S. B, Sarawat P, Davids J. R <sup>(17)</sup>	Journal of Pediatric Orthopaedics (1.7)	26	9 (6.7-16.8)	3.6	Surgery: - 100% improvement in ITW for those operated on in zone III and 88% for those in zone II.
Serial casting in the treatment of idiopathic toe-walkers and review of the literature (2006)	Fox A, Deakin S, Petigrew G, Paton R <sup>(19)</sup>	Acta Orthopaedica Belgica (0.35)	44	6.08 (2-14.3)	1.1	Serial casting + stretching: Among 44 cases, 29 (66%) showed a reduction or resolution of ITW and an increase in dorsiflexion
The natural history of idiopathic toe-walking: a long-term follow-up of fourteen conservatively treated children (2004)	Hirsch G, Wagner B <sup>(20)</sup>	Acta Paediatrica (3.8)	14	6.45 (3-9.9)	14.5	Combination of physiotherapy, stretching, casting, orthotics: - 100% combined with stretching. - 35.7% used with orthotics or casting. Among 14 cases, 8 (57.1%) children ceased ITW.
Idiopathic Toe-Walking: Does Treatment Alter the Natural History? (2000)	Eastwood D. M, Menelaus M.B, Dickens R.V.D, Broughton N. S, Cole W. S, Se M <sup>(32)</sup>	Journal of Pediatric (2.99)	136	Group: Observational: 4 (1.5-10) Group: Casting: 3.5 (1.5-10.3) Group: Surgical: 6.5 (2.5-14.5)	Group: Observational: 3.2 Group: Casting: 3.7 Group: Surgical: 7.9	Three groups (observational, casting, and surgical): - Observational: Improvement of ITW in 45% and resolution in 12%. - Casting: Improvement of ITW in 41% and resolution in 22%. - Surgical: Reduction of ITW by 50% and resolution by 37%.
Serial ankle casts for patients with idiopathic toewalking: effects on functional gait parameters (2019)	Thielemann F, Rockstroh G, Mehrholz J, Druschel C <sup>(33)</sup>	Journal of Children Orthopaedics (1.38)	10	(5-15)	< 1	Casting: - Improvement in gait analysis criteria.
Habitual toe-walkers. A clinical and electromyographic gait analysis (1977)	Griffin P. P, Wheelhouse W. W, Shiavi R, Bass W <sup>(35)</sup>	Journal of Bone and Joint Surgery (5.3)	6	6 (5-9)	<1	Casting: Improvement of ITW in 100%

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**Table 4.** Studies that investigated treatment options for ITW

Article (Publication Year)	Authors	Journal (Impact Factor)	Number of ITW sample	Mean age at the beginning of intervention (years)	Mean time of follow-up (years)	Conclusions of the study
Long-term gait outcomes following conservative management of idiopathic toe walking (2018) Group: Casting + BoNT-A : 7.2 (4.3-12.2)	Davies K, Black A, Hunt M, Holsti L <sup>(36)</sup>	Gait & Posture (2.4)	34	Group: Casting + BoNT-A : 7.2 (4.3-12.2) Group: Stretching: 8.7 (4.9-13) Group: Stretching: 8.7 (4.9-13)	13.4	Two groups: Casting + BoNT-A injection; Stretching - Casting + BoNT-A injection: Improvement of ITW in 74%. - Stretching: Improvement of ITW in 35%. - Improvement in gait analysis criteria observed in both groups.
Does botulinum toxin A improve the walking pattern in children with idiopathic toe-walking? (2010)	Engstrom P, Gutierrez-Farewik E. M, Bartonek A, Tedroff K, Orefelt C, Haglund-Akerlind Y <sup>(37)</sup>	Journal of Children's Orthopaedics (1.38)	15	(5-13), Median 9	1	BoNT-A injection + stretching: Improvement in 4 out of 11 cases (36.4%) and resolution in 3 out of 11 cases (27.3%)
IncobotulinumtoxinA Injection for Treating Children with Idiopathic Toe Walking: A Retrospective Efficacy and Safety Study (2022)	Fillipetti M, Picelli A, Di Censo R, Vantim S, Randazzo P. N, Sandrini G, Tassorelli C, De Icco R, Smania N, Tamburin S <sup>(38)</sup>	Toxins (4.2)	28	8.3 (Standard Deviation 3.1)	<1	Incobotulinumtoxin A: - Improvement in dorsiflexion
Botulinum Toxin A Does Not Improve the Results of Cast Treatment for Idiopathic Toe-Walking (2013)	Engstrom P, Bartonek A, Tedroff K, Orefelt C, Haglund-Akerlind Y, Gutierrez-Farewik E. M <sup>(39)</sup>	Journal of Bone and Joint Surgery-American (5.3)	47	9.4 (5-14.5)	1	Casting with and without BoNT-A injection: No improvement in gait analysis parameters was observed with the addition of BoNT-A injection.
Orthotic treatment of idiopathic toe walking with a lower leg orthosis with circular subtalar blocking (2021)	Berger N, Bauer M, Hapfelmeier A, Salzmann M, Prodinge P. M <sup>(42)</sup>	BMC Musculoskeletal Disorders (2.3)	22	7 (2.5-13.1)	2	Orthoses: - Improvement of ITW by 73% in 1 year and 64% in 2 years
Effects of wearing a foot orthosis on ankle function in children with idiopathic toe walking during gait (2022)	Brasiliano P, Alvini M, Di Stanislao E, Vannozzi G, Di Rosa G, Camomilla V <sup>(43)</sup>	Heliyon (3.7)	21	8.3 (5-12)	1.3	Orthosis: - Improvement of ITW by 82% - Enhancement of gait analysis criteria
A comparison of orthoses in the treatment of idiopathic toe walking: A randomized controlled trial (2015)	Herrin K, Geil M <sup>(44)</sup>	Prosthetics and Orthotics International (1.6)	18	Group: Ankle and Foot Orthosis: 5.4 Group: Foot Orthosis: 4.6	<1	2 groups (ankle-foot and foot orthoses): - Ankle-foot orthosis group: 63% improvement in ITW - Foot orthosis group: 38% improvement in ITW
Outcome after conservative and operative treatment of children with idiopathic toe walking: a systematic review of literature (2014)	Van Bommel A. F, Van de Graaf, V. A, Van den Beekom M. P. J, Vergroesen D. A <sup>(45)</sup>	Musculoskeletal Surgery (0.67)	298	Group: Casting 4.9 (3.3-6.8) Group: Surgical 9 (3.9-12.1)	Group: Casting 3.4 Group: Surgical 4.2	Surgery vs. Serial Casting: - Surgery resulted in higher dorsiflexion values. Gait improvement in 52.5% - Serial casting led to an improvement of 47.9%
Long term gait outcomes of surgically treated idiopathic toe walkers (2016)	McMulking M. L, Gordon A. B, Tompkins B. J, Caskey P. M, Baird G. O <sup>(46)</sup>	Gait & Posture (2.4)	8	9 (6.4-11.3)	5	Surgery: - 87.5% improvement in ITW (7/8 with first rocker) - Enhancement of gait analysis criteria
Outcome of Patients After Achilles Tendon Lengthening for Treatment of Idiopathic Toe Walking (2006)	Hemo Y, Macdessi S. J, Pierce R. A, Aiona M. D, Sussman M. D <sup>(47)</sup>	Journal of Children's Orthopaedics (1.38)	15	9 (4.2-13.1)	2.9	Surgery: - Enhancement of gait analysis criteria - May lead to knee hyperextension (recurvatum)

Other techniques have also been investigated, including using pyramidal insoles, serial casting, BoNT-A injections, and orthotics. To date, there is only one study published in English evaluating the outcomes of pyramidal insoles<sup>(11)</sup> and one review<sup>(29)</sup> where the main author cites four other studies published in German, reporting a success rate between 70%–95.8%<sup>(11,29)</sup>. Despite these studies demonstrating positive results, they often combined insoles with other therapies involving orthotics, BoNT-A injections, and physiotherapy. Furthermore, they did not have a control group, making it challenging to assess the efficacy of the treatment or whether the good outcomes were not related to the natural history of the disease.

Using serial casting has shown favorable outcomes in some studies<sup>(19,20,32,33)</sup>, including increased ankle dorsiflexion, resolution of ITW, and parental satisfaction with the final results. These positive effects have been observed even in older patients<sup>(34,35)</sup>. However, some authors still question whether these outcomes could result from the condition's natural progression<sup>(32,34,35)</sup>. Eastwood et al. (2000) suggest that 50% of children might experience spontaneous improvement<sup>(32)</sup>. Thielemann et al. (2019) used a functional gait analysis before and after serial casting and at a six-month follow-up visit to compare ten children with ITW submitted to serial casting with a control group. In contrast to the literature, they found complete normalization of the ITW gait compared to the healthy control group, which was observed shortly after treatment<sup>(33)</sup>.

A notable point in serial casting studies is the proposal by Fox et al. (2006)<sup>(19)</sup> to measure ankle dorsiflexion with the knee flexed, as this seems to be a more accurate indicator of improvement in ITW than measuring with the knee extended. Fox's study showed that while 100% of participants improved ankle dorsiflexion with the knee in extension, only 66% experienced improvement in flexion. Additionally, Davies' study (2018)<sup>(36)</sup> highlighted compensatory changes observed in older children, such as greater restriction of passive ankle dorsiflexion and compensatory knee hyperextension in both groups. These changes could be due to increased skeletal maturity or longer follow-up periods.

Regarding BoNT-A injections, some studies indicate positive outcomes with improvements in ankle dorsiflexion and other gait parameters<sup>(36,37,38)</sup>, while others do not show significant improvement in ITW<sup>(39,40)</sup>. Engström et al. conducted two studies<sup>(37,39)</sup>; the first showed a positive effect on ITW improvement when BoNT-A injection was used combined with isolated physiotherapy<sup>(37)</sup>, but the later study in 2013<sup>(39)</sup> found no superiority of BoNT-A injection combined with serial casting over serial casting without BoNT-A injection. Caserta et al. (2019)<sup>(27)</sup>, in a Cochrane review, concluded that there are not sufficient studies to confirm that combining BoNT-A injection with serial casting is superior to using serial casting alone. In addition, Sättilä et al. (2019)<sup>(41)</sup> conducted a randomized controlled trial in 30 children with ITW, dividing them into two groups: (1) a conservative treatment with firm shoes, night splints, a home stretching program, and physical

therapy and (2) the same approach with the addition of BoNT-A injections. They reported 38 adverse effects in the BoNT-A group (16 patients treated with 30 injections) and none in the conservative group. No significant differences between the groups in function or ankle range of motion were found in the 24 postoperative months.

Positive results in the short-term follow-up have been reported<sup>(42-44)</sup> regarding the use of orthotics for ITW. However, ongoing research aims to determine the most effective types of orthotics, suitable age groups, the optimal duration of use, and how long the effects of the intervention last, among other factors. There is no evidence to prove the standalone effectiveness of orthotics as a primary treatment option for ITW.

### Invasive therapies: Surgical procedures

There is a consensus that the surgical approach might have better outcomes in treating ITW, as shown in several studies<sup>(17,32,45,46)</sup>. However, studies still lack addressing the most suitable techniques for different age groups and severity levels. In the study by Westberry et al. (2020)<sup>(17)</sup>, which evaluated 26 patients classified as severe according to Alvarez's classification<sup>(16)</sup>, an 88% improvement was observed with the triceps surae surgical lengthening in zone II, and 100% improvement was seen in those operated in zone III. Among those operated in zone III, none required re-intervention. However, not all children had their symptoms completely resolved; out of the 21 children operated using this technique, 14 were subsequently classified as moderate and seven as mild. Among children undergoing lengthening in zone II, out of the 25 operated extremities, six extremities required revision.

Possible complications after surgery include tendonitis, wound dehiscence, and pain, among others. However, these complications have a low incidence and are usually easily resolved. Compensatory changes such as knee hyperextension (recurvatum) may also occur<sup>(29,47)</sup>, necessitating longer follow-up studies to assess the presence of further abnormalities and the impacts of each of these changes.

### Discussion

Despite the heterogeneity in the methodologies of the analyzed studies, certain observations can be noted. The determination of relevant etiological and prognostic factors is still under analysis. A trend towards higher prevalence in boys with a positive family history exists, but the inheritance of the condition remains inconclusive, and higher-level evidence studies are required to confirm these theories. In addition, the association of ITW with SPD and ASD is well established, nonetheless not as a causative factor since many children that present with ITW do not present these conditions.

Regarding the final age of spontaneous resolution, there is variability in the studies regarding the percentage of children exhibiting ITW. The number ranges from 30% to 88%; therefore, it is important to question the reference

age used by each author. Engström and Tedroff (2018)<sup>(8)</sup> use a milestone of 5.5 years, while Kelly et al. (1997)<sup>(41)</sup> use three years. Thus, some patients showing resolution in Engström's study might not have been considered by Kelly et al., making direct comparison difficult. Furthermore, the follow-up time of studies is questionable. In Engström's study (2018)<sup>(8)</sup>, children with neurodevelopmental disorders—which were not specified—were included in the ITW cohort. Despite this finding, most of the patients ceased toe walking spontaneously, thereby possibly overestimating the value of the ITW in the general population.

Regarding etiology and risk factors, authors such as Fox et al.<sup>(19)</sup> and Sobel et al.<sup>(14)</sup> did not find statistically significant evidence for correlations with other demographic data, such as sex, gestational age, and age at diagnosis, among others. However, it is important to consider that the lack of statistical relevance might be due to the small sample size of the cohorts, especially since sex seems to have clinical relevance in discussions from certain studies<sup>(14,19,21,22)</sup>. As for age, there appears to be a negative correlation between ankle dorsiflexion and age, a hypothesis supported by some studies<sup>(36,48)</sup>, which agrees with the normative data for normal individuals shown in two studies<sup>(49,50)</sup>.

Some authors have raised critical views about established standards of three-dimensional gait analysis, which warrant further investigation and studies to strengthen these opinions. For example, Brasiliano et al. (2023)<sup>(51)</sup> suggest increased accuracy using multi-segmented three-dimensional gait analysis incorporating foot models. Westberry et al.<sup>(17)</sup> proposed a modification to Alvarez's classification<sup>(16)</sup>, utilizing the sagittal ankle moment in kinetics to define the first rocker instead of kinematic assessment, as it provides a better representation of the foot's position relative to the tibia, yielding a more precise measure. However, it is important to recognize that three-dimensional gait analysis can only recognize the children's walking pattern in a "snapshot in time," which might not reflect the normal patient's walking pattern since these patients can voluntarily correct their gait at their will<sup>(6)</sup>. Further studies should compare habitual walking patterns using sensors and three-dimensional gait analysis.

Methods such as stretching, physiotherapy exercises, pyramidal insoles, and BoNT-A injections do not have strong evidence of effectiveness when used in isolation. Many studies have assessed these techniques in combination, making it difficult to distinguish the effect of each one in isolation<sup>(11,19,20,37)</sup>. It is important to question if the positive effects seen in conservative treatment methods are due to the small sample size, the short follow-up periods that might not account for future presentations, or if they are simply a result of the natural progression of the condition. Weighing the risks and benefits of each therapy may be the best approach for those options.

The most evidence-based conservative measure is serial casting, followed by orthotics. Given the heterogeneity across studies, randomized clinical trials are needed to define

optimal casting intervals and orthotic usage. Long-term effects also require investigation. Davies et al. (2018)<sup>(36)</sup> noted greater knee hyperextension in the stance phase and reduced flexion in the swing phase after serial casting. In contrast, Engström et al. (2013)<sup>(39)</sup> reported less knee hyperextension in the stance phase and increased knee flexion in the swing phase. Davies et al. attributed these differences to the varying follow-up periods: Davies' study spanned 12 years, while Engström's was only 12 months. The population in the former study also had a higher mean age than in the latter. Further research is necessary in this area, but serial casting seems a good option for gaining ankle dorsiflexion range of motion in mild contractures to improve ankle kinetics and kinematics in the short term.

There is growing evidence of the benefits of surgery in ITW, ranging from symptom reduction to complete resolution in the long term<sup>(17,32,45-47)</sup>. For severe cases aged near or above ten years, open or percutaneous lengthening in zone III is considered the best choice<sup>(17)</sup>. However, few robust studies adequately address these questions, demanding randomized clinical trials and better long-term follow-up of proposed interventions.

In our clinical experience, postoperative follow-up often reveals other pre-existing orthopedic conditions, such as increased tibial external rotation and flexible flatfeet. This underscores the importance of conducting a thorough physical exam before treatment and emphasizes the need to educate parents and children about these issues, which might be more evident after treatment. Further research is required to validate these findings, highlighting the significance of early assessment and the value of informing patients and their families about these potential concerns.

Finally, the lack of consensus in results is notable due to the heterogeneity in methodologies, as discussed by Caserta et al. (2019)<sup>(40)</sup> in their systematic review of outcomes and assessment tools, as well as in Bauer's (2022)<sup>(30)</sup> and Williams' (2020)<sup>(52)</sup> findings, which identified a lack of consensus among healthcare professionals regarding evidence-based therapies. Heterogeneity is seen in populations across studies<sup>(32,36,45)</sup>, the type of severity classification used<sup>(29,32,42)</sup>, previous treatments<sup>(46,47)</sup>, and different exclusion criteria<sup>(19,42)</sup>, among other differences, making results comparison unfeasible. For future studies, larger study populations, standardized techniques, follow-up durations, and defined outcome metrics are essential.

This study has some limitations. First, it was restricted to articles in English, excluding some studies in languages like Russian, Italian, Spanish, and others. Second, the methodological heterogeneity of the analyzed articles makes it difficult to explore and compare results. Third, the evaluation included studies other than randomized clinical trials, which decreases the overall evidence levels of the included studies.

## Conclusions


Despite the significant heterogeneity in methodologies across studies, it was possible to note a growing trend



toward investigating etiological factors, particularly genetic and neurosensory factors, and prognostic factors like age, Achilles tendon contractures, and sex. There appears to be a higher prevalence in boys with a positive family history, yet the genetic inheritance aspect remains inconclusive.

Novel classifications have been proposed based on anatomical features and three-dimensional gait analysis, aiming to differentiate severity, prognosis, and differential diagnoses. Treatment options encompass both conservative and surgical alternatives. Among conservative approaches, there is stronger evidence of gait improvement in equinus gait through serial

casting followed by orthotics, while the addition of BoNT-A injections has not shown better results. There is no evidence of sustained improvement with isolated physiotherapy exercises and stretching, but these may be recommended with other therapies. Surgical procedures are reserved for patients with Achilles tendon contractures, with better outcomes observed in severe cases, as per Alvarez's classification, who undergo lengthening in Zone III and are nearly ten years of age or older. However, more robust studies, preferably randomized clinical trials, are needed to establish clearer conclusions about treatment protocols for patients with ITW.

**Authors' contributions:** All authors read and approved the final manuscript.\*ORCID (Open Researcher and Contributor ID) LDLS \*(0009-0000-1020-0063), and TOG \*(0000-0001-9277-7746) Conceived and planned the activity that led to the study, wrote the article, participated in the review process; LRAA \*(0000-0003-3779-6914) Formatting of the article, bibliographic review; FCB \*(<https://orcid.org/0000-0001-5272-7998>), and FMB \*(0000-0002-4131-3017) participated in the review process. All authors read and approved the final manuscript.\*ORCID (Open Researcher and Contributor ID) 

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