CRITICALLY APPRAISED TOPIC

FOCUSED CLINICAL QUESTION

For children who are "idiopathic toe walkers" will corrective interventions (non–surgical) in the long-term be more effective for normalizing gait than not receiving treatment.

AUTHOR

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CLINICAL SCENARIO

A parent comes into the clinic with her 9 year old daughter, who toe walks, has been having back pain, physical activity is limited due to pain, and has gained weight due to her limited activity level. The parent's goals are for her daughter to have no pain with activities and stop the toe walking to prevent further problems down the road.

This clinical scenario is based off one of my nieces who has toe walked since she began walking. Gradually, as she has aged the idiopathic toe walking has become a problem that has decreased her quality of life. Soon she will be receiving physical therapy to address the problems toe walking has created.

Pediatric toe walking can lead to multiple musculoskeletal complications for patients later in life. Potential issues include: Achilles tendon shortening, decreased dorsiflexion, abnormal gait patterns, narrowed base of support, muscle weakness and compensatory strategies, pain, and activity limitations. Also, these physiological limitations for some children may cause psychological and social interaction barriers for "toe walkers."

SUMMARY OF SEARCH

- 10 studies met the inclusion/exclusion criteria with 3 studies reviewed
- The studies addressing idiopathic toe walking (ITW) are of poor quality and results need to be considered with caution as there is high variability and subjectivity.
- The conservative treatment best options are physical therapy, serial casting, and possibly orthosis.[1,2,5] Botulinum toxin A does not have strong evidence to support used for treament.[1,5]
- Conservative treatment is best done when the child is young (<7 years old) since this prevents loss of passive and active range of motion due to tightening of the triceps surae and produces greater beneficial gains in maximum dorsiflexion and normalizing gait.[5]
- Serial Casting produces the same results as serial casting with botulinum toxin A without the added fear and pain.[1]
- The ankle-foot orthosis (AFO) 100% prevents toe walking; however there is a high likelihood of reverting to previous gait pattern when removed.[2] The foot orthosis (FO) is less restrictive, but improvements were significantly maintained when removed and child/parent preferred the FO to the AFO.[2]

CLINICAL BOTTOM LINE

Conservative treatment methods are beneficial in reducing the effects of ITW. Currently, the treatment longevity is relatively short but they all improve children's functional ability and quality of life by improving passive dorsiflexion and normalizing gait. A clinician should exhaust conservative treatment methods and possibly repeat treatments while a child is developing in order to avoid surgery. There are a few treatment options that can be performed independently or in combination, including physical therapy, orthosis, and serial casting.

This critically appraised topic has been individually prepared as part of a course requirement and

has been peer-reviewed by one other independent course instructor

SEARCH STRATEGY

Terms used to guide the search strategy			
Patient/Client Group	**I**ntervention (or Assessment)	**C**omparison	**O**utcome(s)
Pediatric* Paediatric* Child* "Idiopathic toe walker" Toe walk* persistent toe-walk* Toe*	Hippotherapy Physical therapy Physiotherapy Exercise Rehabilitation Intervention Stretch* Non-invasive Not surgical treatment	No treatment	Gait Ambulat* Walk*

Final search strategy:

PubMed	***Number of Results***	***Limits Applied***
1. Pediatric* OR paediatric* OR child* 2. toe walk* OR toe-walk* OR (toe AND walk*) OR "idiopathic toe walker" OR (idiopathic AND (toe AND walk*)) OR (persistent AND (toe AND walking)) 3. toe-walker [MeSH term] 4. physical therapy OR physiotherapy OR exercise OR rehabilitation OR intervention OR Non-invasive OR treatment OR Hippotherapy OR stretch* 5. Surgical OR surgery 6. Ambulat* OR walk* OR gait 7. No treatment 8. #1 AND #2 9. #1 AND #2 AND #3 10. #1 AND #2 AND #3 AND #4 11. #1 AND #2 AND #4 12. #11 NOT #5 13. #11 AND #6 NOT #5 14. #11 AND #6 AND #7 NOT #5 15. #11 AND #6 AND #7 NOT #5 [limits applied] 16. #11 AND #6 AND #7 NOT #5 [limits applied] 17. #11 AND #6 AND #7 18. #11 AND #6 AND #7 [limits applied] 19. "Idiopathic toe walker" 20. "Idiopathic toe walker" [limits applied] 21. "Idiopathic toe walker" [limits applied]	1. 2397734 2. 1336 3. 3 4. 9171631 5. 3910656 6. 303464 7. 8779790 8. 285 9. 0 10. 0 11. 200 12. 114 13. 114 14. 67 15. 28 16. 17 17. 149 18. 35 19. 55 20. 25 21. 15	1-14. Any Language, Any Year, Clinical Trial, Review, Meta-Analysis, Systematic Review 15. Full text, published in the last 10 years, Meta- Analysis, Systematic Review, RCT, Review, Human, English 16. Full text, published in the last 5 years, Meta- Analysis, Systematic Review, RCT, Review, Human, English 17. Any Language, Any Year, Clinical Trial, Review, Meta-Analysis, Systematic Review 18. Full text, published in the last 5 years, Meta- Analysis, Systematic Review, RCT, Review, Human, English 19. No limits 20. Applied: English, Human, Full text, 10 years 21. Applied: English, Human, Full text, 5 years

Databases and Sites Searched **Search: "idiopathic toe walking"**	**Number of results**	**Limits applied, revised number of results (if applicable)**
PubMed	55	25 -Applied: English, Human, Full text, 10 years 15 - Applied: English, Human, Full text, 5 years

CINAHL	28	24 – Applied: English, Academic Journal 3 - Applied: English, Academic Journal, Full Text
WEB OF SCIENCE	68	30 - Applied: Review, Article, 2010-2013 7 – Applied: review, Article, 2010-2013, AND intervention
CHOCHRANE LIBRARY	1	No filters

INCLUSION and EXCLUSION CRITERIA

Inclusion Criteria
Systematic reviews, meta-analysis, randomized control trials, controlled trials, uncontrolled trials
Primary research studies
Case Studies (if research is limited)
Intervention is related to role a physical therapist would be involved with directly or indirectly
Studies paediatrics (18 year and under)
Adults idiopathic toe walking included only if persistent since childhood to determine long-term relationship between treatment vs. no treatment in paediatrics
Examine only kids who toe walk that is idiopathic and not cerebral palsy related
Published in English
Published up to November 2015
Exclusion Criteria
Surgical interventions
Abstracts, conferences, narrative review articles, dissertations,

RESULTS OF SEARCH

Summary of 10 articles retrieved that met inclusion and exclusion criteria

Author (Year)	Study quality score	Level of Evidence	Study design
Engstrom P, Bartonek A, Tedroff K, Orefelt C, Haglund- ├àkerlind Y, Gutierrez-Farewik E. (2013)[1]	Pedro 8/11	2a	RCT
Herrin & Geil (2015)[2]	Pedro 7/11	2a	Randomized Control Trial (RCT)
Fox A, Deakin S, Pettigrew G, Paton R. (2006)[3]	D&B 22/29 MINOR score 8	2b	Prospective Study Dec. 1999- Sept. 2003
Van Bemmel, (2014)[4]	AMSTAR 6/11	3a	Systematic review with observational (level 1-4)
van Kuijk, Annette A. A., Kosters R, Vugts M, Geurts ACH (2014)[5]	AMSTAR 7/11	3a	Systematic Review with 1 RCT (level 1) and 18 observational studies (level 4)
Engelbert R, Gorter JW, Uiterwaal C, van de Putte E, Helders P. (2011)[6]	D&B 23/29	3	Cross Sectional Study
Hirsch G, Wagner B. (2004)[7]	D&B 5/22	3	Retrospective Study, no

			RCT
Stricker SJ, Angulo JC. (1998)[8]	D&B 5/22 MINOR score 11	3	Retrospective Study, no RCT
Clark E, Sweeney J, K., Yocum A, McCoy S, W. (2010)[9]	D&B 14/29 MINORS score 11	4	Case Study of Non-concurrent Case-Series
Williams CM, Tinley P, Rawicki B. (2014)[10]	AMSTAR 5/11	5	Literature Review

BEST EVIDENCE

The following 3 studies were identified as the 'best' evidence and selected for critical appraisal. Reasons for selecting these studies were:

- **Recent Systematic Reviews and control trials that builds on previous ITW research**
- **Engstrom 2013[1]:**

  This RCT has high quality and level of evidence that meet the ITW inclusion and exclusion criteria, while being on the verge of being too aggressive for conservative treatment with the botulinum toxin A injections. The study examined subjects throughout a five year span and provided detailed information breakdown between the cast only/cast + botulinum toxin A groups and had a high retention rate throughout. There gait analysis technology allowed for a detailed examination of subjects gait and each subject went through a battery of assessments with a blinded investigator. This is the only study to compare casting with and without botulinum toxin A injections, which is important since parent(s) frequently are looking for the best treatment options for their child(ren). Furthermore, the blinding throughout the study keeps the internal validity high and the sample size is relatively large compared to other ITW study numbers.

- **Herrin 2015[2]:**
- This was the only study to examine just the effects of two different orthosis (ankle-foot orthosis and foot orthosis) that could be used to treat ITW in children. The researchers create a strong case for their results because they performed a parallel RCT, just underpowered at 76%, and with high technological resources to minimize bias and improve accuracy in assessing intervention outcomes. Also, there was a single orthotist for the casting, which minimized confounders, maintained internal validity, and consistency. The sample size was small, however efficient because of the broad age range used since ITW can change with age. Furthermore, in conjunction with this, they examined parent satisfaction, a strong key component to a child's compliance rate in wearing the orthosis. Also, this study is one of the few to examine conservative intervention options outside of applying passive, lengthening, stretches to the triceps surae and Achilles tendon prior to treating ITW with serial casting.

- **van Kuijk 2014[5]:**
- This is the only systematic review that used a RCT along with retrospective observations studies when examining ITW interventions. Although most of the articles reviewed are low-level evidence, van Kuijk et al. efficiently discussed pediatric ITW clinical treatment effects with. They were very deliberate and detailed in their inclusion criteria and methodological characteristics of studies reviewed, which allows for easy reproduction later and quick access to the various treatment options, both conservative and non-conservative. Figure 2 is and excellent overall visual picture for clinicians that provides a quick reference for patient age versus best treatment option to discuss with the parent(s). The benefit of ITW intervention is still being determined, but the most important information to obtain from the review is early intervention is key. Conservative treatment results are the most beneficial when 4 years old or younger at preventing ITW related complications, normalizing gait, and preventing surgical intervention.

SUMMARY OF BEST EVIDENCE

(1) Description and appraisal of Botulinum toxin A does not improve the results of cast treatment for idiopathic toe-walking: A randomized controlled trial by Engstrom P, Bartonek A, Tedroff K, Orefelt C, Haglund-Åkerlind Y, Gutierrez-Farewik E., 2013

Objective of the Study

The objective was to determine whether treatment of botulinum toxin A with below-the-knee walking casts or walking casts alone reduced toe-walking for 5-15 year old children and that the treatment outcome correlated with the degree of coexisting neuropsychiatric problems.

Study Design

- Randomized Control Trial
- Assessors and physical therapists were blinded to group allocation throughout the study, while parents and children knew the treatment they received following the baseline examination. The only assessor who knew the group allocation was the one performing the botulinum toxin A treatment.
- Outcomes for the children and parents were measured prior to treatment, 3 months post, and 12 months post treatment.
- All children received randomized treatment for four weeks in either the cast alone group (CA group) or cast following botulinum toxin-A group (CA+BX group). Treatment was based off randomized sealed envelopes with random permuted blocks of four. The medical statistician prepared the sealed envelopes and then a nurse completed the randomization prior to baseline examination.
- A statistical power analysis, based on the intention-to-treat principle, with a moderate effect size of 0.5 and alpha = 0.05 significance level was performed. They found power = 0.940 if n=50 and power = 0.881 if n=40.

Setting

Pediatric orthopaedics department at Karolinska University Hospital - Stockholm, Sweden

Participants

- Diagnosis: Idiopathic Toe-Walking (ITW)
- Number of subjects: 78 children met the inclusion criteria, 52 total children were randomized, although 5 withdrew (4 children afraid of BX, 1 child moved away) from the study, leaving N= 47 children
    - Cast Only group (CA) N= 26
    - Cast + Botulinum Toxin A group (CA+BX) N = 21
- Recruited: Patients admitted to the pediatric orthopaedics department due to ITW between November 2005 and April 2010
- Eligibility Criteria: All children included were evaluated by a pediatric orthopedic surgeon and considered to exhibit ITW, spent 25% or greater on his/her toes for a minimum of three months according to the parent's perception, there was no other explanation for toe-walking and no contractures, no history of previous treatment for ITW, and the parents consented to their child's participation.

Group Comparison	Total	Cast Only (CA)	Cast + Botulinum Toxin A (CA+BX)	P Value for differences between groups
No. of patients	47	26	21	0.07*
Gender Female Male	 18 29	 13 13	 5 16	
Age† (yr)	9.4 (5.0-14.5)	9.4 (5.4-13.6)	9.4 (5.0-14.5)	0.64
Family history of idiopathic toe walking (no.)	26/45 (58%)	17/25 (68%)	9/20 (45%)	0.30
Cesarean section delivery (no.)	4/45 (8.9%)	2/25 (8%)	2/20 (10%)	0.82
Age at independent walking† (mo)	12.4 (8-24)	12.4 (8-24)	12.3 (9-17)	0.90

**P value of the difference between the numbers of male and female subjects was determined with a chi-square test. †The values are given as the mean with the range in parentheses.*

Group Comparison:

- Prior to treatment there was no difference between groups in toe-walking frequency according to the parent's perception or the children's ankle dorsal extensor strength.
- There were no major differences between the gait analysis variables measured during pre treatment between groups.
- There were four children in the CA group whose passive ankle range of motion prevented reaching neutral by 20 degrees with extended knees and two in the CA+BX group unable to reach neutral by 5 degrees with extended knees. One child in the CA group was unable to reach neutral by 5 degrees in one leg with flexed knees.
- There were no hip or knee motion limitations in either group.

Intervention Investigated

CA Control Groups

- Treatment for all the CA children consisted of bilateral below-the-knee Softcast reinforced posteriorly with Scotchcast (3M) walking circular casts that extending from the toes to the proximal calf. They wore the cast for a total of four weeks.
- A single nurse with special cast technique education applied each cast with the focus on placing the ankle in neutral position.
- Stretching Program: After the casts were removed, a physical therapist gave the children/parents (CA and CA+BX group) oral and written home exercise program instructions for calf-muscle stretching to be completed five times per week along with heel walking a minimum of fifty steps per day.

CA+BX Experimental Group Only

- One to two weeks prior to being casted, the CA+BX group children received bilateral botulinum toxin-A injections with 12 units/kg body weight of Botox (Allergan). An hour prior to injection a 40 mg/kg of oral paracetamol was given and topical anesthetic cream (EMLA; AstraZeneca) was applied. An inhaled nitrous oxide sedative and pain reliever was given to the children just before the Botox injections. Each child received four injections per calf, "two in the proximal third of the lateral and medial gastrocnemius bellies and two distally in the gastrocnemius-soleus complex", for a total of eight injections administered via "electromyogram-amplifier guidance to ensure intramuscular position" (Engstrom, 2013, p 403).
- They wore the cast for a total of four weeks.

*Case application, treatment, and stretching same as mentioned above for the CA group

Outcome Measures

Gait Analysis (Primary)
A single physical therapist conducted and analyzed the children's 3-D gait analysis with fifteen markers and eight-camera system. Gait analysis was performed with children walking barefoot at a usual self-selected speed and manner.
Three trials which each consisting of one right and one left gait cycle were analyzed using 3-D kinematics and kinetics data and temporal-spatial parameters. A total of twenty-seven discrete kinematic and kinetic values were collected with the average values calculated for each child.
Each child's ITW severity was classified based on the system described by Alvarez et al. 2007, which examines the presence of the first ankle rocker, early third ankle rocker, and predominant first ankle plantar flexion moment during loading response.

Joint Range-of-Motion Measurements and Strength of Ankle Dorsal Extensors (Primary)
A single physical therapist prior to treatment, three months post, and twelve months post measured passive range of motion (PROM) at the hip, knee (knee extension and 90° flexion), and ankle joints with a goniometer. A handheld dynamometer measured ankle dorsal extensor strength after 5 seconds of resisted motion. Three measurements where recorded with the highest analyzed based on each child's weight.

The investigator completing the statistical analysis and interpretation of gait, strength, and range of motion data was blinded to all the children's group allocation.

Assessment of Neuropsychiatric Problems (Secondary)
A pediatric neurologist examined the children for neuropsychiatric disorders, performed a neurologic examination, and a neuropsychiatric assessment to rule out coexisting conditions, which none of the children displayed.

The parents completed a "Five to Fifteen" questionnaire (FTF) to identify any strengths or concerns associated with neuropsychiatric symptoms. The FTF questionnaire has eight domains and children with scores above the 90th percentile were classified as having difficulty with tasks.

Parents' Perception of Toe-Walking Frequency and Side Effects (Secondary)
The perception of time, spent toe walking while barefoot to the nearest quartile was conducted with each child's parents.

Main Findings

Gait
There were several significant improvements in gait for both groups at three and twelve months post treatment, but no significant differences between the groups (p = <0.001). On average, the foot position during the stance and swing phase improved to a neutral foot position due to improved maximal ankle dorsiflexion (CA -3.4° to 6.7°, CA+BX -3.3° to 7.9°). The maximal ankle dorsiflexion during stance improved by an average of 9 degrees. Also, dorsiflexion was observed later in the gait cycle (12 month post % of gait cycle - CA 33.9, CA+BX 38.1), which is closer to a typical gait cycle (30% of gait cycle) in children. Both groups demonstrated improved kinetic parameters: greater maximal ankle power generation (W/kg) at the ankle, CA 0.7 (p = <0.01) and CA + BX 1 (p = 0.034); greater ankle power absorption; and greater negative work (J/kg) at the ankle (CA 0.1 (p = 0.002) and CA+BX 0.1 (p = .018).
During the gait cycle, in midstance, there were positive changes in both groups. A small reduction in knee hyperextension, CA 2.3° (p = 0.008) and CA +BX 1.9° (p = 0.054). A small increase in knee flexion during the swing phase, CA 4.1° (p = 0.001) and CA+BX 2.8° (p = 0.038) and small improvement in knee extension moment (NM/kg) in stance phase improved, CA 0.2 (p = 0.001) and CA + BX 0.2 (p = <0.01). There were no significant changes in cadence, step length or walking speed.

ITW Severity Classification	Cast Only		Cast +Botulinum Toxin A	
	Pre-Treatment	12 month post-treatment	Pre-Treatment	12 months post-treatment
Mild	0	7	0	8
Moderate	9	17	4	10
Severe	17	2	17	12
Values are given as the number of patients				

The ITW classification severity in both groups was significantly improved between pretreatment and the post treatment evaluations, however there was no significant difference between the CA and CA+BX group results.

Parents' Perception of Toe-Walking Frequency and Side Effects
The CA+BX group parents noted a significantly lower ITW frequency than the CA group parents (p = 0.033) at three months post treatment, however the between-group factor was just outside significance (p=0.053). By the twelfth post treatment month there was no between-group difference, although six CA+BX group children (28.6%) and three CA group children (11.5%) had stopped toe walking according the parents.

Joint Passive Motion and Dorsiflexion Strength
There was no difference between the CA and CA+BX groups observed in either joint PROM or dorsal extensor strength, however both groups increased in ankle dorsiflexion (knees flexed and extended). Both groups demonstrated significant within group increases in dorsal extensor strength. The CA group had within- group improvements for pretreatment through the twelfth month testing (p = 0.011) and from the third month through the twelfth month evaluation (p = 0.031). The CA+BX group had within-group improvements for pretreatment through the third month (p = 0.01) and through the twelfth

month testing (p = 0.001).

Influence of Neuropsychiatric Problems on Treatment Effect
There were no difference between the two treatment groups and no correlation between FTF domain scores above the 90th percentile and treatment effect of the ankle angle at initial contact (r = 20.20, p = 0.18), ITW severity classification (r = 20.22, p = 0.14), or parents perception (r = 0.09, p = 0.56)

Original Authors' Conclusions

In regards to reducing idiopathic toe walking, botulinum toxin-A injections prior to below-the-knee casts for four weeks did not improve outcomes more than casting alone. There was no significant difference in improvement between children treated solely with casts and those treated with casts and botulinum toxin-A in regards to gait improvement, toe-walking classification, parent perception, passive ankle motion, or ankle dorsal extensor strength. Both groups improved at three and 12 months post casting but this did not stop toe walking in thirty-eight of the children.

Critical Appraisal

Validity

The study is a high quality RCT (level 2a evidence) that minimized potential sources of bias, maintained a high retention rate and high internal validity. Few of the weaknesses present in the study could have been predicted prior to its initiation. The greatest limitations were the total number of participants and the fact all the children came from one source instead of several pediatric orthopedic hospitals in the area. This potentially may have lead to a greater number of children meeting the inclusion criteria and parent willingness for their child's participation, which may have increased the effects size and the power. Drawbacks to the validity of the study were that the questionnaire regarding parents' perception of toe-walking frequency tool has not been validated. Possible confounders to the study include the limited number of female participants and parent's perceptions of ITW severity. Parent's perceptions are highly unreliable due to their emotional feelings towards their child, own preconceived belief of severity, knowledge, and attitude towards their child being diagnosed. In addition the exact manor the researchers performed the gait cycle analysis is unclear and may have created greater scores if a foot was counted in both the left and right gait cycles. Also, a minor limitation, creating possible contamination bias was that occasionally group allocation was revealed to the physical therapist by the child during treatment, which reduced the blindness of the therapist(s) who was conducting outcome measures or teaching the home stretching program. The largest question regarding this study is the true external validity of cast alone treatment, since no similar study has been conducted.

Interpretation of Results

The results indicate that casting with botulinum toxin-A injections is not necessary for treating idiopathic toe walking and that the parent's perception had no between group significance. However, parent opinion is important since they can provide information gathered outside the clinic. Neuropsychiatric problems were not an issue with the children in this study, but that may not always be the case for other ITW children. This study had relatively few subjects and no previous similar study to compare results, however the researchers were consistent with trying to prevent bias and maintain as close to a fully blinded study as possible when treating the children. The overall integrity of the study seems strong based off the study protocol and the acknowledgement of possible limitations. Although, providing calculations demonstrating the between-group correlation for joint PROM and dorsiflexion strength would have been beneficial.
Based on the high quality and internal validity of the study treating children with casting alone for conservative treatment is a good option prior to attempting to use botulinum toxin-A injections. This study indicates there is no need to provide a potentially fear provoking and painful injection, since the results indicated there is no statistically significant or clinically significant result by adding the botulinum toxin-A injections with casting. The within group changes are significant for both groups but the between group effect size is zero. Prior to treating a child with botulinum toxin-A injections further research on the correlation between improved dorsiflexion and relationship with casts would need to be assessed.

Implications of this study/evidence for clinical practice and future research

In regards to the clinical PICO question, this study demonstrates that children who are idiopathic toe walkers have a good chance of benefit from non-surgical cast treatment to improve their maximum dorsiflexion and do not need botulinum toxin-A treatment. The improved range provided from casting

would allow for greater dorsiflexion, improved ankle kinematics, and ankle rocker, which would create overall improvements in their gait cycle.

Further studies need to examine cast only to cast with botulinum toxin A treatment options to draw a more confident conclusion that there is no added benefit from adding botulinum toxin A injections with serial casting to decrease the idiopathic toe walking in the long term. Besides comparing casting and botulinum toxin-A treatment comparisons between the difference options for conservative ITW need to be further researched to assess treatment option clinical relevance and validity.

(2) Description and appraisal of A comparison of orthosis in the treatment of idiopathic toe walking: A randomized control trial by Herrin k and Geil M, 2015

Aim/Objective of the Study/Systematic Review:

The aim of the study was to determine the outcome differences for idiopathic toe walking between two orthoses.

Study Design

Parallel randomized control trial with no blinding

- Convenience sample
- Children randomly assigned Ankle Foot Orthosis (AFO) or Foot Orthosis (FO) after baseline assessment completion through a computer blocked randomization with 1:1 allocation ratio

Method

- Inclusion criteria: diagnosis of ITW, age 2-8 years, candidate for orthotic intervention
- Exclusion criteria: neurological condition diagnosis, prior treatment for Achilles tendon contractures, plantarflexion contracture preventing neutral ankle alignment (90 degrees)
- 9 children analyzed from each group.

Baseline and 6 weeks post-treatment

Post hoc power analysis 76% for group sample sizes of 9 and observing a 9.0 between the null hypothesis and AFO/FO groups heel rise time (alpha level 0.05)

Setting

Physical therapist and physicians clinics in the area around Georgia State University (GSU) Biomechanics Laboratory - Atlanta, Georgia

Participants

Diagnosis: Idiopathic Toe Walking

N = 18 Children

- Initial average passive dorsiflexion 3.75 degrees and full plantarflexion ROM, no between group difference (p = 0.50).
- Pre-treatment: 20% demonstrated initial contact with their forefoot, 80% had initial contact with the heel or flatfooted.
- No significant difference between group temporal and spatial parameters at baseline: cadence p = 0.854, step length p = 0.430, walking speed p = 0.551, and heel rise time p = 0.853)
- All children could walk normal if asked, according to the parents.
- Mean walking speed for all children was 1.27 m/s.
- AFO: 10 children, initial average toe walking age 12.1 ± 2.5 months
- FO: 9 children, initial average toe walking age 17.9 ± 13.4 months

Eligibility: treated by physical therapist or physician in clinic near GSU biomechanics lab, diagnosed with ITW, age range 2-8 years, never treated with orthotic intervention and a candidate, parent approval

Intervention Investigated

Control Group - Ankle Foot Orthosis)

- An orthotist provided all allocation enrollment, assignment, casting, modification, and fitting of orthosis.
- 10 children wore AFO for 6 weeks then were re-assessed.
- Instructed to wear brace constantly except for sports activities and sleep.
- AFO Brace: Orthosis fabrication standardized with AFO fabricated from $1/8^{th}$ polypropylene, 90 degree plantarflexion stop, free dorsiflexion, Tamarack joints, full heel posting, layover calf strap, ankle chafe and loop strap.
- Trim-lines placed midline at the calf and foot, full-length toe plate, medial distal footplate trim-line proximal to metatarsophalangeal joint at firsts/firth metatarsal head.

Experimental Group - Foot Orthosis

- 9 children wore FO for 6 weeks then were re-assessed.
- FO Brace: Orthosis fabrication standardized with FO customized with carbon fiber full-length firm footplates, cork base, 1/8 inch poron and 1/8 inch puff.
- *Treatment by orthotist and brace wearing instructions same as mentioned above.

Outcome Measures

3-D Gait Analysis

- *Completed with and without orthosis in shoes.*
- Analysis at 100Hz with a 7-camera Vicon Nexus System, 15 reflective markers to lower extremity with placement following the Vicon Plug-in-Gait sacrum model.
- Children ambulated at normal pace on 10-meter track for 5 trials.
- ITW defined as heel rise before 30% of gait cycle or toe initial contact.
- Cadence, step length, walking speed, heel rise time recorded from each trial were summarized.

L-test of Functional Mobility

- Completed with and without orthosis in shoes.

Parent Satisfaction Survey & Parent Orthotic and Prosthetic User Survey (only post-treatment)

- Parent perception regarding their child's ITW and orthoses.
- No additional information was provided about the survey.

Main Findings

3-D Gait Analysis

- Wearing Orthosis:
  - AFO group walked significantly faster 1.36 ± 0.25 m/s, $p = 0.006$, FO group 1.22 ± 0.19 m/s.
  - AFO was 100% effective at preventing toe initial contact.
  - FO resulted in 13% of initial contact being toe or flat-footed.
  - No significant difference between groups cadence ($p = 0.286$) or step length ($p = 0.194$).
  - AFO group mean heel rise at 35.5% ± 5.2% *(within normal limits).*
  - Heel rise significantly different ($p = 0.008$) between groups.
  - AFO significant within group improvement ($p < 0.001$)
  - FO significant within group improvement ($p<0.001$).
- Without Wearing Orthosis:
  - AFO significant within group improvement not sustained with shoes only ($p = 0.175$)
  - FO significant within group improvement maintained with orthosis removed ($p<0.001$).
  - Heel rise not significantly different ($p = 0.734$) between groups.
- No major difference between group temporal and spatial parameters at baseline: cadence $p = 0.286$, step length $p = 0.194$.

L-test

- No statistical between group difference in time completion (baseline $p = 0.62$, post-treatment with

orthosis and shoes p = 0.55, post-treatment with shoes only = 0.92).

Parent Satisfaction Survey Results

No difference in time spent wearing orthosis present between AFO and FO groups according to parent reported (p = 1.0) or parent satisfaction of gait (p = 0.38) or parent rating of orthosis effectiveness (p = 0.73).

Parent Orthotic and Prosthetic User Survey

- Parents significantly found donning FO easier (p = 0.049) and preferred FO due to ease of donning, appearance (p = 0.053), and reduced skin breakdown (p = 0.070).
- Compliance reported AFO 9.7 ± 1.48 h/ day and FO 11.33 ± 2.44 h/day, between group difference P = 0.23.

Original Authors' Conclusions

ITW gait was controlled by the AFO when worn but the less restrictive and controlling FO had greater out-of-brace effects on gait pattern when removed. Children and their parents preferred the rigid FO as opposed to the AFO for bracing.

Critical Appraisal

Validity

There has been no previous randomized controlled trial completed analyzing orthoses for ITW conservative treatment. Herrin and Geil's randomized control trial could be mistaken for a pilot study because of the relatively small sample size. However, the post hoc power analysis established the study at 76% power for group sizes of 9 subjects, so they were just slightly underpowered.

The study was limited by the recruitment schedule and grant funding term, which reduced the ability to determine if the wear time duration was sufficient and therefore the study's reliability. In addition, the researchers ITW indicator was early heel rise during the gait cycle, thus assuming that laboratory early heel rise after initial contact correlated with toe walking in other places. This is difficult to determine since ITW has an unknown etiology, but if true could be valuable in the clinic.

The age diversity could misrepresent ITW orthoses result validity since multiple gait patterns may be represented in the small group. Furthermore, selection bias occurred since the subjects were all selected based off being treated in a relatively small geographical area and the ability to travel to the biomechanics laboratory, which may not represent the true demographics or result for other children who toe walk.

Due to randomization method the AFO group was ten months older than the FO group, not a large enough difference for significance (p = 0.17). Although, this may have been a confounding factor for why the FO results were better. If completed with a larger sample size would the difference have favored one orthosis? The AFO treats mechanical equinus and the FO treats sensory-perceptual equinus. Other possible bias and confounders in the study comes from the parents, there was no indication of how the parents recorded their child's compliance wearing the orthosis except for the parent survey. It is difficult for anyone to remember exactly what occurred during a 6-week time frame, so the results reliability from the parent feedback may be skewed towards a greater indication of wearing the orthoses.

Interpretation of Results

Mechanically, the AFOs prevent initial toe contact without gross adaptation in proximal joints, such as excessive hip and knee flexion, whereas the FO's require less adaptation for the child to achieve initial toe contact. However, the AFO has poor carry-over results due to its restrictive nature and does not seem to teach the child a heel-toe gait pattern. The FO on the other hand, may allow for a greater learning response and decrease the sensory-perceptual effects that cause the child to resort to ITW. It also has the added benefit of being more visually appealing and easier to don/doff, meaning parents and children are more likely to wear it on a consistent basis.

This study had a relatively small subject size and was underpowered, which results in a higher variability and chance of a type II error occurring. It would be interesting to replicate the study but prolong the time the children wear the orthosis and add a brace that is similar to the AFO, slightly less

restrictive, and one that would be more accommodating to the child's and parents preferences.

Implications of this study/evidence for clinical practice and future research

Corrective orthotics, either AFO's or FO's may be beneficial in treating ITW to normalize gait in the long-term. Typically wearing orthotics is a relatively short phase of treatment, although the exact length of time for the best results is unknown. The FO's are easier for parents to don/doff their child, which may correlate to the children wearing the FO's more frequently, so this may be the best overall choice for the family. However, clinicians must remember, the FO does not control ITW from occurring. The AFO's prevents ITW when worn, but the reduced return leads to question its treatment value because of the poor return rate in this study.

Further research is necessary to determine the effects of orthosis intervention for ITW in children. Research needs to determine the appropriate length of time worn for orthosis and if a gradual decrease in the plantarflexion stop would demonstrate a higher return rate following the intervention. For now, the best treatment option may be a step-up or a step-down approach to orthosis. Start with the less restrictive FO then transition to the AFO if necessary or start with the rigid restrictive AFO then transfer to the FO after progress has occurred. Clinician's will need to base their approach on the child's ITW severity and family dynamics.

(3) Description and appraisal of Treatment for idiopathic toe walking: A systematic review of the literature by van Kuijk, Annette A. A., Kosters R, Vugts M, Geurts ACH, 2014

Aim/Objective of the Study/Systematic Review:

The aim of the study was to complete a systematic review of the literature that assesses the effects of current treatment options for ITW using the World Health Organization International classification of Functioning Disability, and Health for Children and youth (Who ICF-CY) 3 main levels: body structure and function, activities, and participation.

Study Design

Systematic Review

114 relevant citations, 19 studies met all criteria (1 randomized control trial, 8 case series, 1 qualitative study, 1 cross-sectional study, 1 historically controlled study, 7 cohort studies with 9 being prospective and 10 retrospective study designs, 1 OCEBM level 1 and 18 OCEBM level 4, MINORS scores range 2-18 for 18 of the studies (excluding the RCT))

Search Strategy:

- Systematic search in the data-bases of MEDLINE (January 1966–December 2013), Current Contents, CINAHL (January 1966–December 2013), and the Cochrane Library (January 1982–December 2013)
- MeSh headings and keywords used include: "equinus gait or walking", or "toe gait or walking", or "tiptoe gait or walking", or "forefoot gait or walking", or "short Achilles tendon", or "short tendo calcaneus" for the first keyword. A combination of the first word and either "idiopathic", or "idiosyncratic", or "habitual", or "persistent", or "dystonic" further identified articles. Lastly, relevant references were identified from the retrieved articles.

Selection Criteria:

- Inclusion - Studies with between-groups comparison or with-group pre-post treatment for ITW in children 2-18 years old, addressing clinical treatment efficacy, full-length articles in English, German, French, or Dutch language.
- Exclusion - Heterogeneous patient sample without ability to identify patients with ITW, studies not available in the Netherlands medical libraries.

Methods:

- Independently, two authors completed the study selection and methodological evaluation; a third reviewer was consulted when disagreement arose.
- The Oxford Centre for Evidence-Based Medicine (OCEBM) levels of evidence and the Methodological Index fro Non-Randomized Studies (MINORS) were used to evaluate and analyze studies following the ICF-CY.

Statistical Analysis:

- No meta-analysis or best-evidence synthesis due to inability to pool data, only 1 RCT.
- Outcomes analyzed based off WHO ICF-CY: body structure and function, activities, and participation.

Follow-up between and within studies ranged from 6 weeks to 22 years post-treatment

Setting

18 Hospital-based and 1 physical therapy (PT) practice-based populations with referral for ITW treatment. Systematic reviewers from Eindhoven, s-Hertogenbosch, and Nijmegen, Netherlands

Participants

475 subjects between all 19 studies, Age range 1.5-14.5 years old

Treatment Options

- PT: age 1.5 – 2 y/o, mean <7 years
- Casting: age .5 – 14.5, mean <7 years
- BTX-A: 3-14.5, mean age>7 years
- Surgical: 3-145, mean age >7

Participants were eligible if ITW was present with no neurological deficits, all used diagnosis of exclusion but this differed between the studies.

Surgical children had greater severity of structural equinus deformity compared to conservative treatment studies.

10 Retrospective studies: differences in patient characteristics were age and pre-treatment maximum ROM for ankle dorsiflexion

Intervention Investigated

Control

6 Compared Interventions (found on pg 947-950)

Eastwood et al., 2000

- **Observation** only; **Cast -** 6 weeks Bilateral (B) serial below- knee walking cast; **Surgery -** Baker's technique aponeurotic tendo-achilles lengthening (TAL) then 6 weeks of below-knee walking cast.

Engström et al., 2013

- **Cast**: 4 weeks (B) below- knee serial walking cast and then PT home exercise program (HEP) of stretching calf muscles 5 × per week and 50 steps walk on heels per day. **BTX-A+ Cast:** (B) triceps surae muscles 12 MU/kg BTX-A, 1–2 weeks afterwards cast for 4 weeks, and then PT HEP of stretching calf muscles 5 × per week and 50 steps walk on heels per day.

Hirsch & Wagner, 2004

- **PT**: passive stretching for triceps surae and tibialis anterior, exercises addressing dorsiflexion activation, HEP; **Cast**: 2–4 weeks of (B) below-knee casts and then intense PT and night splints.

Katz & Mubarak, 1984

- **PT**: 2 subjects completed tibialis anterior stretches; **Cast:** 5 subjects completed between 2-16 weeks of (B) below-knee dorsiflexion cut-out serial walking casts and then tibialis anterior stretches and wearing negative-heel shoes with brace.

Stott et al., 2004

- **Cas**t: 6 weeks (B) below-knee serial walking casts changes every 2 weeks and then HEP of passive stretching; **Cast +**

Surgery: casting with (B) percutaneous tendo-achilles lengthening or Baker's technique.

Stricker & Angulo, 1998

- **Observation** only; **Cast/Brace**: 6–12 weeks of (B) below-knee walking casts or 3–8 months of solid AFO and then HEP for stretching; **Surgery**: tendo-achilles lengthening (B) Baker's technique (n=4) or open technique and then 2–6 months cast and AFO.

Protocol for treatment within or between the studies was not standardized or provided in more detail.

Experimental

<u>13 studies had a single intervention</u> (found on pg 947-950)

1 PT motor control effectiveness

Clark et al., 2010

- **PT:** 9 total weeks of 2 days/week for a 1-hour session on motor control and a HEP of play adapted to the child's PT goals.

3 Serial casting effectiveness

Brouwer et al., 2000

- **Cast:** 5 weeks of (B) below-knee serial walking cast with changes every 1–2 weeks.

Fox et al., 2006

- **Cast:** 3-10 weeks (avg. 5.7) of (B) below-knee serial walking cast with changes every 2 weeks and then a PT created HEP for passive tibialis anterior stretching.

Griffin et al., 1977

- **Cast:** 6 weeks (n = 3) of (B) below-knee serial walking casts OR 8 weeks (n = 1) of (B) below-knee serial walking casts OR 6 weeks (n= 1) of (B) long leg casts OR 8 weeks (*n* = 1) of (B) long leg casts and then dorsiflexion exercises and gait training after cast removal for all subjects.

4 Botulinum toxin type-A (BTX-A) effectiveness

Brunt et al., 2004

- **BTX-A:** (B) 12 MU/ kg triceps surae muscles, max 400 MU. Then 20 days post-injection PT 2 time a week for gait training, active dorsiflexion exercises, passive stretching, and HEP.

Engström et al., 2010

- **BTX-A:** (B) 6 MU/ kg triceps surae muscles, max. 400 MU. Then stretching triceps surae muscles 5 time/week and 50 heel-toe steps walking daily.

Gormley et al., 1997

- **BTX-A:** (B) 7.5–10 Mu/kg triceps surae muscles and then serial casting until child reaches 10° of dorsiflexion.

Jacks et al., 2004

- **BTX-A:** (B) 10 MU/ kg triceps surae muscles and then serial walking cast, changed each week, until child reaches 10° of dorsiflexion. Afterwards a day and night articulated AFO and PT HEP for slowly stretching plantarflexors and strengthening dorsiflexors.

5 surgical Effectiveness

Hall et al., 1967

- **Surgery:** tendo-achilles lengthening (TAL), 3 weeks of weight-bearing, and then 6 weeks of below-knee cast.

Hemo et al., 2006

- **Surgery:** (B) TAL (3 percutaneous and12 open), 4-6 weeks below-knee casts, and then AFO initially wearing day and night with progression towards night only then stop wearing 1 year post-surgery.

Jahn et al., 2009

- **Surgery:** TAL (percutaneous or open) or vulpius.

Kogan & Smith, 2001

- **Surgery:** percutaneous TAL and then 1 month of below-knee casts.

McMulkin et al., 2006

- **Surgery:** TAL (6 percutaneous and 1 open) and 7 vulpius.

Protocol for treatment within or between the studies was not standardized.

Outcome Measures

- Multiple outcomes measures: passive ankle dorsiflexion ROM, gait (visual observation or comprehensive analysis), surface electromyography, and parent report, Parent Perception of gait

performance and activities.
- Early studies heavily relied on qualitative visual observation for gait while recent studies are using comprehensive gait analysis.
- Mostly post-treatment data gathered with either historical pre-treatment data or not collected.
- Post-treatment follow-up varied from 6 weeks to 22 years within and between studies.

**Data not provided on exact details of the outcome measures.*

Main Findings

Passive ankle range of motion

- Forest Plot Maximal Ankle dorsiflexion end ROM (fig 3. pg 952)
    - Children receiving physical therapy reached neutral dorsiflexion and on average reached 5° to 10° of maximum dorsiflexion.
    - Casting results in improved maximum dorsiflexion to around neutral (±5°) post-intervention.
    - Children requiring surgery often did not reach neutral maximum passive dorsiflexion (-5° to -10°) range post-surgery.
- Dorsiflexion increased immediately following and 6 weeks post serial casting and/or physical therapy termination.
- BTX-A treatment demonstrated not significant difference in maximum ankle dorsiflexion.
- Conservative treatment gains were short term, 2 years post-treatment children demonstrated a 6° decrease in maximal dorsiflexion on average following treatment conclusion.
- Surgical Achilles tendon lengthening increased long-term passive range of motion.

Gait Analysis

- Out-of-phase triceps surae activation demonstrated by children during ITW.
- Premature triceps surae activity during swing phase.
- In ITW there is early low amplitude tibialis anterior activity causing absent late swing and loading response activity. (van Kuijk et al. pg 954)
- Electromyographic pattern changes after treatment closer to a normal pattern.
- Significant kinematic increased ankle plantarflexion angle prior to the treatment intervention.
- Absent first-rocker prior to treatment for most children with ITW.
- BTX-A alone did not improve maximum passive dorsiflexion from pre-treatment ranges.
- Cast and BTX-A had no pelvis or hip deviations pre or post treatment.
- Surgical cases demonstrated increases external hip rotation and greater pelvic tilt for children needing tendo-achilles lengthening.
- After treatment there is significant increased ankle dorsiflexion and ankle power increase.

Parent perception

- Normal gait did not occur after ITW treatment.
- Parents reported good long-term effects with dorsiflexion maintained after surgical intervention.
- A quarter of parents are satisfied with results after after serial casting because it does not fully normalize gait.

Original Authors' Conclusions

Current evidence suggests serial casting and surgery is beneficial for improving passive ankle dorsiflexion. Walking kinetics and kinematics also improves, although normal gait appears not to be achieved. Botulinum toxin A does not improve outcome results. Lasting effects over one year is only maintained after surgery. Treatment option effectiveness for functional activities and social participation is unknown.

Critical Appraisal

Validity

The validity of multiple studies compromised the systematic review due to the use of historical pre-treatment data with the post-treatment studies data. This data was not collected from the individual subjects so comparing their results should only occur based off the post-treatment data. Also, ten of the studies were retrospective studies, so over half of the studies included where susceptible to high

subjectivity and information bias. Along the same lines, the immediate intermediate post-operative results were not necessarily documented, which reduced the validity and reliability of the independent studies results and the overall validity of the systematic review. The broad range for post-treatment time frame also calls into question the outcomes results, as there may have been several confounders through the years the researchers failed to exclude.

Fortunately, the systematic review researchers understood the limits of using observational studies and provided an age at intervention (fig. 2 pg 951) and forest plot of treatments (fig. 3 pg 952) that is very helpful at visualizing the vast differences in the 19 studies. The small sample sizes of each study may have created selection bias and not been a good indicator of the idiopathic toe walkers population. This is especially true for the surgical subject's as there tended to be even fewer subjects, the subjects were older, and it was an elective surgery. The surgeons may also have chosen subjects that they knew would have a favourable outcome, which further created selection bias and may indicate why long-term results favoured surgery.

Furthermore, the parent surveys create further subjective bias due their emotions, preconceived beliefs, knowledge and attitude. However, it is important to no discredit the parents reports as have the ability to see the child in their normal environment, on a regular basis, and understand their child's psyche better than the researchers.

Due to the limited methodological quality of ITW studies, a meta-analysis was unable to be performed and an accurate comparison of all the treatments was difficult. Also, in the systematic review of articles selection bias may have occurred because there was only access to studies available through the Netherlands medical libraries and the studies had to be written in Dutch, English, French, or German.

Interpretation of Results

- Children treated with physical therapy were the youngest, around 5 years old, with the least restricted dorsiflexion pre-treatment, and successfully gained on average 5° to 10° of maximum dorsiflexion that was maintained in the short term (~2years).
- Children receiving casting were around 7 years old or younger and demonstrated short-term sustainment of improved maximum dorsiflexion to around neutral (±5°) following intervention completion
- Children requiring surgery were older (>7 years) and had the most severe maximum passive dorsiflexion limitations, surgery improved maximum dorsiflexion but often theses children were slightly plantarflexed post-surgery, however some were able to reach neutral and results were sustained years later.

The authors thoroughly investigated the studies and used information available to complete the best comparison and results. Due to the quality and quantity of the studies available a more substantial analysis was not possible. However, this systematic review does shed light into the various treatments and potential range of results for healthcare providers and parents to consider before treating a child with idiopathic toe walking. The studies, independently, are not strong enough but together they create a stronger argument and more clinical significance for providing certain treatments in the clinic and avoiding others, such as botulinum toxin A. However, caution with results regarding ITW is strongly advised due to the poor quality studies and thus a poor quality systematic review.

Implications of this study/evidence for clinical practice and future research

For children who are "idiopathic toe walkers" will corrective interventions (non–surgical) in the long-term be more effective for normalizing gait than not receiving treatment.

Depending on the age of the child an initial conservative approach to treatment for ITW is appropriate. The best conservative treatment options are physical therapy and casting; BTX-A has not demonstrated to be an effective treatment option alone or in addition to serial casting. Conservative treatment alone is effective, although provides only short-term (~2 years) lasting results. A child's ITW may need to be treated conservatively again a few years later or a more aggressive treatment may be warranted. The preconceived effect of each treatment should be limited and clinician's cautions of creating too high and expectation for the parent(s) and child since the research backing up the various treatment methods for ITW is poor and highly variable.

EVIDENCE SYNTHESIS AND IMPLICATIONS

IMPLICALTIONS FOR PRACTISE

The overall quality of evidence for conservative treatment of ITW is poor. However, conservative intervention is more effective than not receiving treatment to correct ITW. Early intervention optimizes the potential that physical therapy, wearing orthoses, and/or casting will effectively reduce ITW, while BTX-A has not proven to be and effective treatment option.

In order for children to maintain their increased maximum dorsiflexion, clinicians must strongly iterate repetitively the necessity of parent(s)/child to continue their home exercise program. This is especially true for children who experience rapid growth in height and weight, as the muscle/tendon to bone length ratio may not be equal. Clinicians need to communicate to parents that conservative treatment may later require another round of treatment as the child ages to prevent an increase in ITW. Parent should be educated to monitor their child's ITW tendencies and seek assistance if their child demonstrates increased ITW or ability to walk heel to toe. Physical therapists should provide written HEP's for parent/child and observe the parent correctly assisting in exercises when required.

Conservative treatment options clinicians should be considering are physical therapy, serial casting, and short-term orthosis wearing.[1,2,5] These have demonstrated to be effecting in the short-term (~2 years) at increasing passive dorsiflexion, improving ankle rocker presence, maximum power generated at the ankle.[1,5]

IMPLICATIONS FOR FUTURE RESEARCH

The lack of a known ITW etiology creates challenges for researchers and hampers the validity and accuracy of treatment intervention effectiveness. The quality of evidence, including the systematic reviews and randomized control trials for ITW is poor. This is due to multiple studies being retrospective in nature, small sample sizes, a very limited number of RCT, and few studies with comparison or control groups. Multiple studies had minimal information about subject inclusion or exclusion criteria and did not provide a detailed analysis of the subjects, so it is impossible to determine if there are confounders outside of ITW that may be causing false-positive or false-negative results. The baseline subject groups are not fully comparable for heterogeneity comparison and due to the nature of ITW interventions completing blinded therapist or subject studies is difficult. Even the systematic reviews are weak due to the use of retrospective observational studies since there are few RCT. Today's research is still relying on older studies, fortunately, the research being reported since 2012-13 is improving and this should lead to higher quality systematic reviews in the upcoming years. Future research needs to have larger sample sizes, similar pre and post-treatment outcome measures, studies with comparisons between all treatment options with children in the same age range and ITW severity, and standardization of results so that a meta-analysis is possible to complete. Other possibilities to consider for ITW research include a more extensive analysis of various orthoses, use of BTX-A possible after serial casting to further increase maximum passive dorsiflexion range, and use of other treatment modalities in physical therapy. Options could include kinesio-taping to biofeedback, ultrasound, and electrical stimulation to assist with elongating the triceps surae, activation of the anterior tibialis, and gait normalization.

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