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REVIEW PAPER

Little Evidence for Conservative Toe Walking Interventions in Autism Spectrum Disorders: a Systematic Review

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Abstract

This systematic review summarizes the evidence about toe walking (TW) interventions in persons with autism. Following the PRISMA guidelines, a systematic search of MEDLINE, CINAHL, PsycINFO, The Cochrane Library, Google Scholar, and Opengrey was performed. Nine articles (all case reports or case series) were included. Methodological quality was assessed using the Mayo Evidence-Based Practice Centre tool. The included studies considered 17 subjects (16 males; age range: 4–15 years). All studies reported a reduction of TW frequency, but the follow-up was lacking in seven studies. There is a lack of high-quality studies with a sufficiently large and well-characterized sample to assess the effectiveness of TW interventions in autistic persons. These findings strongly support the need for further research in this area.

Keywords Toe walking · Autism spectrum disorder · Intervention · Treatment · Systematic review

Introduction

Autism spectrum disorder (ASD) is a complex neurodevelopmental disorder affecting 1 in 44 children (Maenner et al., 2021), resulting in an impairment of socio-communicative interaction, restricted interests, and repetitive behavior (American Psychiatric Association, 2013). Diagnostic criteria are described in the diagnostic and statistical manual of mental disorders–5th edition (American Psychiatric Association, 2013). Despite motor impairments are not currently included in the evaluation and diagnostic criteria of ASD, there is rising evidence that persons with ASD also have motor impairments. An epidemiological study reported a prevalence of 35.4% for motor impairments among 2,084 individuals with ASD (Licari et al., 2020). The authors observed a decline in motor scores with increasing age of diagnosis and found motor difficulties in children with (1/2 children) and without (1/3 children) intellectual impairments. The study of motor impairments in persons with autism is peculiar because motor-related impairments may become a significant barrier to a child's further social communication development as well as adaptive functioning (Bedford et al., 2016; Macdonald et al., 2013; Ohara et al., 2020). Moreover, motor delays can be one of the earliest markers of ASD (Biscaldi et al., 2014; Harris, 2017; Lloyd et al., 2013). Gross-motor function impairments have been reported in persons with ASD as visuomotor coordination deficits (Fleury et al., 2013; Kushki et al., 2011), bilateral coordination disorders (Green et al., 2009; Kaur et al., 2018; McPhillips et al., 2014), or difficulties with fine motor skills (Bhat et al., 2011; LeBarton & Iverson, 2013). Furthermore, postural capacity was found to be impaired in ASD individuals both with (Kohen-Raz et al., 1992; Perin et al., 2020) and without intellectual disability (Doumas et al., 2016; Fournier et al., 2010 and, 2014). Gait was found impaired in autistic individuals in terms of increased step width (Eggleston et al., 2020; Shetreat-Klein et al., 2014), more variable stride length (Nobile et al., 2011; Rinehart et al., 2006), differences in variability in each gait sub-phase (Eggleston et al., 2020),

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and toe walking (TW) (Accardo et al., 2014). TW is a diagnosis given to subjects who persist in walking on their toes after they should typically have achieved a heel-toe gait, and it should resolve by 3–7 years of age (Shetreat-Klein et al., 2014). To be diagnosed, TW should be manifested for more than 6 months (Accardo et al., 2014).

TW is the most common deformity in children with bilateral spastic cerebral palsy, with 83.3% (Horsch et al., 2019). TW can also be found in neuromuscular disorders (Hyde et al., 2000). The prevalence varies depending on the neuromuscular disorder; e.g., in Duchenne muscular dystrophy and Charcot-Marie-Tooth disease, TW has been identified as a relevant clinical concern (Rose et al., 2010). Furthermore, TW has also been described in the healthy population. Engelbert et al. (2011) reported a TW prevalence of 12% in a sample of 362 healthy subjects (8–20 years old), Engström and Tedroff (2012) described a TW prevalence of roughly 4.5% in 5 years old typically developed children, and Shetreat-Klein et al. (2014) reported a prevalence of 3% in healthy children ranging from 22 months to 10 years. Ming et al. (2007) and Barrow et al. (2011) reported a TW prevalence of 20% in autistic individuals. Recently, a broad retrospective study (Leyden et al., 2019) identified a higher prevalence of TW (8.4%) in ASD individuals than in typically developing children (0.47%). TW in ASD seems to be influenced by the severity of the diagnosis, with a 10% increased incidence of tight heel cords present in autistic children compared with a 3% increased incidence in children diagnosed with Asperger's (Barrow et al., 2011). Interestingly, Leyden et al. (2019) found no significant differences in TW prevalence in persons with autism with and without intellectual disability.

TW can be present with different levels of severity (Accardo & Barrow, 2015). Some authors described TW severity as intermittent or persistent (Ming et al., 2007); others graded TW using subjects' history and observation (absent, present in the past, intermittently present, or persistent). Tip toe behavior (TTB) can be present also during standing (Valagussa et al., 2017; Weber, 1978) and/or running (Robert, 2011; Valagussa et al., 2017). Valagussa et al. (2017) highlighted the existence of three mutually exclusive clinical patterns of TTB: (1) during standing, walking, and running; (2) during walking and running; and (3) only when running. The persistence of TW may lead to a higher risk of falling (Caselli et al., 1988), associated with a social or cosmetic impact (Pendharkar et al., 2008), and may affect the child's functional capabilities and quality of life (Calhoun et al., 2011). Moreover, it is suggested that TW persistence in autistic individuals may shorten the Achilles tendon (Accardo et al., 2014; Valagussa et al., 2020).

The etiology of TW in autism is still less understood (Accardo et al., 2014). Weber (1978) suggested that "TW arise from the fixation of a normal transient stage of development." Capute et al. (1979) proposed TW as a residual of a primitive walking pattern (e.g., positive support reflex or tonic labyrinthine reflex). Accardo et al. (2014) suggested TW could be related to a sensory disorder or vestibular issues.

A recent systematic review reported that posterior ankle–foot orthoses (AFO) increased ankle dorsiflexion at initial contact in children with cerebral palsy and TW (Lintanf et al., 2018). Additionally, surgery was considered adequate to improve TW in this condition (Rutz et al., 2020). Some RCTs have shown the effectiveness of interventions for TW in healthy individuals (idiopathic toe walking, ITW). Herrin and Geil (2016) compared the use of ankle–foot orthoses to full-length in-shoe orthoses. Sätilä et al. (2016) compared conservative care to conservative treatments combined with botulinum toxin injections. Engström et al. (2013) assessed the effectiveness of combining casts and botulinum toxin injections compared to casts alone in reducing TW.

Treatment of TW in autism draws from the anecdotal experience of treating children with other neurodevelopmental disabilities such as cerebral palsy and those with ITW (Accardo et al., 2014). Recently, Leyden et al. (2019) conducted a comprehensive retrospective analysis to determine how patients with and without ASD who have persistent TW are treated with various techniques. Among 2,221,009 pediatric patients included, 5,739 had a diagnosis of ASD, of which 484 also had a diagnosis of persistent TW (8.4% of subjects with ASD). On the other hand, 10,840 subjects presented a persistent TW without a neuro-orthopedic condition or ASD (i.e., typically developing children). The 57.2% ($n=5,995$) of these subjects did not report toe-walking interventions; 3,989 (38.1%) underwent physical therapy, 374 (3.6%) serial casting, and 122 (1.2%) underwent surgical correction. Concerning the individuals with ASD presenting TW, $n=287$ (59.3%) were referred to physical therapy and treated with stretching exercises at the lower limbs, specifically to the Achilles tendon; $n=36$ (7.4%) underwent serial casting; $n=16$ (3.3%) received surgery while the treatment was not reported in the remaining 30% ($n=145$) of subjects. When the authors compared the outcomes of individuals with ASD with TW versus typically developing children with TW, a significant difference ($p=0.041$) was shown in favor of ASD subjects. Despite these interventions, the 63.8% ($n=183$) of the ASD patients that were treated with physical therapy, the 47.2% ($n=17$) of patients that were treated with casting, and the 75% ($n=12$) of the patients that were treated surgically continued to toe-walk within 2 years of treatment. Therefore, from a clinical and research standpoint, it is relevant to know if all these interventions are supported by the current evidence about the TW management for persons with autism. To our knowledge, TW treatments were not systematically searched (Valagussa et al.,

2018), and the methodological quality of intervention studies on TW treatments was not assessed. Hence, the current study aimed to summarize the evidence on conservative, pharmacological, and surgical interventions for treating TW in autistic individuals by conducting a systematic review. Specifically, the research question for this study was: What is the evidence for the effect of interventions on TW for individuals with autism spectrum disorder? A systematic review of the literature was designed to answer this question, because it is recognized as the appropriate research method that objectively investigates and summarizes the existing evidence about a specific body of research. This systematic review was conducted according to the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA 2020) Statement (Page et al., 2021).

Methods

Protocol and Registration

The systematic review was *a-priori* registered on the International Prospective Register of Systematic Reviews (PROSPERO: number CRD42020176335).

Data Sources and Searches

The search was undertaken on MEDLINE (via PubMed), CINAHL, PsycINFO, The Cochrane Library, Google Scholar, and OpenGrey from databases inceptions until August 2021. Reference lists of included studies were controlled to identify additional studies. Medical subject headings (MeSH) and non-MeSH search terms were used, including synonyms for the keywords "toe walking" and "autism spectrum disorder." The search query was defined by two researchers (G.V. and D.P.). The search strategy was developed for the MEDLINE database and then was adapted for the others. No restrictions regarding language, year of publication, and age of population were applied. Specifics on the search queries for all the databases are available in Supplementary Material 1. On Google Scholar, two searches were run using two different keyword combinations, and then the first 100 hints of each of the two searches were considered (Chiarotto et al., 2016; Piscitelli et al., 2021).

Criteria for Considering Studies for This Review

Participants: as autism diagnostic criteria have changed over time, this systematic review considered any recognized diagnostic criteria for autistic individuals, i.e., from the "Kanner's autism" diagnosis to the different autism diagnoses labeled in editions of the diagnostic and statistical manual of mental disorders. Studies about the treatment of TW in persons with autism were considered without age limitation. Individuals should have had TW for more than 6 months, should walk with or without a heel-toe gait, and whether or not limited dorsiflexion of the ankle joint.

Interventions: conservative and/or surgical interventions were included.

Comparator(s)/control: comparisons of any intervention versus another or no intervention were considered.

Outcome measures: the primary outcome was defined as the decreasing of TW assessed by the time or the number of feet in TW (i.e., percentage of the time or number of feet engaged in TW during testing or monitoring), evaluated by objective and/or subjective judgment self-reported by the autistic individuals, caregivers, and healthcare professionals. Additional outcomes were (1) changes in Achilles tendon length expressed as improvement of ankle range of motion (passive and/or active), (2) recurrence of TW post-treatment, and (3) adverse events.

Types of studies: intervention study designs such as single-subject design, case reports, case series, single or double cohort clinical trials, and (non-)randomized (un-)controlled trials investigating the effect of the TW interventions were included. Furthermore, studies should be published in peer-reviewed journals. Abstracts, conference proceedings, and dissertations were excluded.

Study Selection

After deleting the duplicated records, the studies were screened independently by two authors (V.P. and S.B.) to identify records that potentially met the inclusion criteria outlined above. The full text was retrieved and independently assessed for eligibility by two authors (V.P. and S.B.). Any disagreement was resolved through discussion with a third reviewer (G.V.).

Assessment of Methodological Quality of Included Studies

Two authors (G.V. and D.P.) independently evaluated the methodological quality of the included studies. As all articles included in this systematic review were case reports or case series, the framework for appraisal, synthesis, and application of evidence developed by Murad et al. (2018) was used. This instrument considers four domains (selection, ascertainment, causality, and reporting) for a total of eight questions in order to assess the methodological quality (Table 1). Any disagreement between the two authors was resolved through discussion until consensus was reached or discussion with a third author (E.G.). Moreover, all included studies were independently assessed for accuracy, transparency, and usefulness of case reports using the CARE (CAse REport) guidelines checklist for case studies (Gagnier et al.,

Table 1 Results of the methodological quality assessment of case reports and case-series included in the systematic review

Questions	Studies								
	Marcus et al. (2010)	Persicke et al. (2014)	Barkocy et al. (2017)	Hodges et al. (2018)	Hodges et al. (2019)	Wilder et al. (2020)	Kratz (2020)	Shaw and Soto-Garcia (2021)	Barkocy et al. (2021)
I. Selection									
1. Does the participant(s) represent(s) the whole experience of the investigator (center) or is the selection method unclear to the extent that other participants with similar presentation may not have been reported?	No	No	No	No	No	No	No	No	Yes
II. Ascertainment									
2. Was the exposure adequately ascertained?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Was the outcome adequately ascertained?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes
III. Causality									
4. Were other alternative causes that may explain the observation ruled out?	Yes	No	No	No	No	No	No	No	No
5. Was there a challenge/rechallenge phenomenon?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
6. Was there a dose–response effect?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
7. Was follow-up long enough for outcomes to occur?	No	No	Yes	No	No	No	Yes	No	No
IV. Reporting									
8. Is the case(s) described with sufficient details to allow other investigators to replicate the research or to allow practitioners make inferences related to their own practice?	Yes†	Yes†	Yes	Yes	Yes	Yes†	No	No	Yes†

Legend: *N/A*, not applicable, as questions 5 and 6 are mostly relevant to cases of adverse drug events (Murad et al., 2018)

† treatment procedure description was reported with sufficient details, but clinical features of the case described were not sufficiently detailed to allow other investigators to make inference to their own practice

2013). The CARE guidelines is a 13-item checklist developed by an international expert panel to provide a framework to guide the completeness and transparency of case reports (See Table S1 for items description).

Data Extraction

After the initial assessment for eligibility, two reviewers (V.P. and S.B.) independently extracted the data from the included studies. We resolved discrepancies through discussion and, when necessary, by discussing with a third reviewer (G.V.). Since all the included studies were case reports or case series, the results were summarized qualitatively through a narrative synthesis of the findings. The following information was extracted: authors and year of publication, study design, and population characteristics (sample number, age, sex, diagnostic criteria used for ASD diagnosis, ASD severity, presence/absence of intellectual disability, language level, comprehension ability, comorbidities, and previous TW treatment). Moreover, TW treatment data were summarized as follows: study aim, type of intervention, device/technique employed, TW assessment methods, outcome measures, time estimate assessments (initial assessment, final assessment, presence/absence of follow-up), and results.

Results

Study Selection

The search strategy identified 619 studies. Nine eligible studies were identified (Fig. 1).

Assessment of the Methodological Quality of the Included Studies

Methodological quality assessment is presented in Table 1. Two studies (Barkocy et al., 2017; Marcus et al., 2010) satisfied 4 of 8 questions, six studies (Persicke et al., 2014; Hodges et al., 2018; Wilder et al., 2020; Hodges et al., 2019; Kratz, 2020; Barkocy et al., 2021) met 3 of 8 questions, and one study fulfilled only one question (Shaw & Soto-Garcia, 2021). The selection domain was met in one study (Barkocy et al., 2021); instead, the ascertainment domain (i.e., exposure and outcome) was satisfied all studies except in one study (Shaw & Soto-Garcia, 2021). The causality domain was not entirely satisfied in any study, and the reporting domain was judged as present in 7 studies (Marcus et al., 2010; Persicke et al., 2014; Barkocy et al., 2017; Hodges et al., 2018; Wilder et al., 2020; Hodges et al., 2019; Shaw & Soto-Garcia, 2021; Barkocy et al., 2021).

The CARE assessment is detailed in Table S1 (Supplementary Material 2).

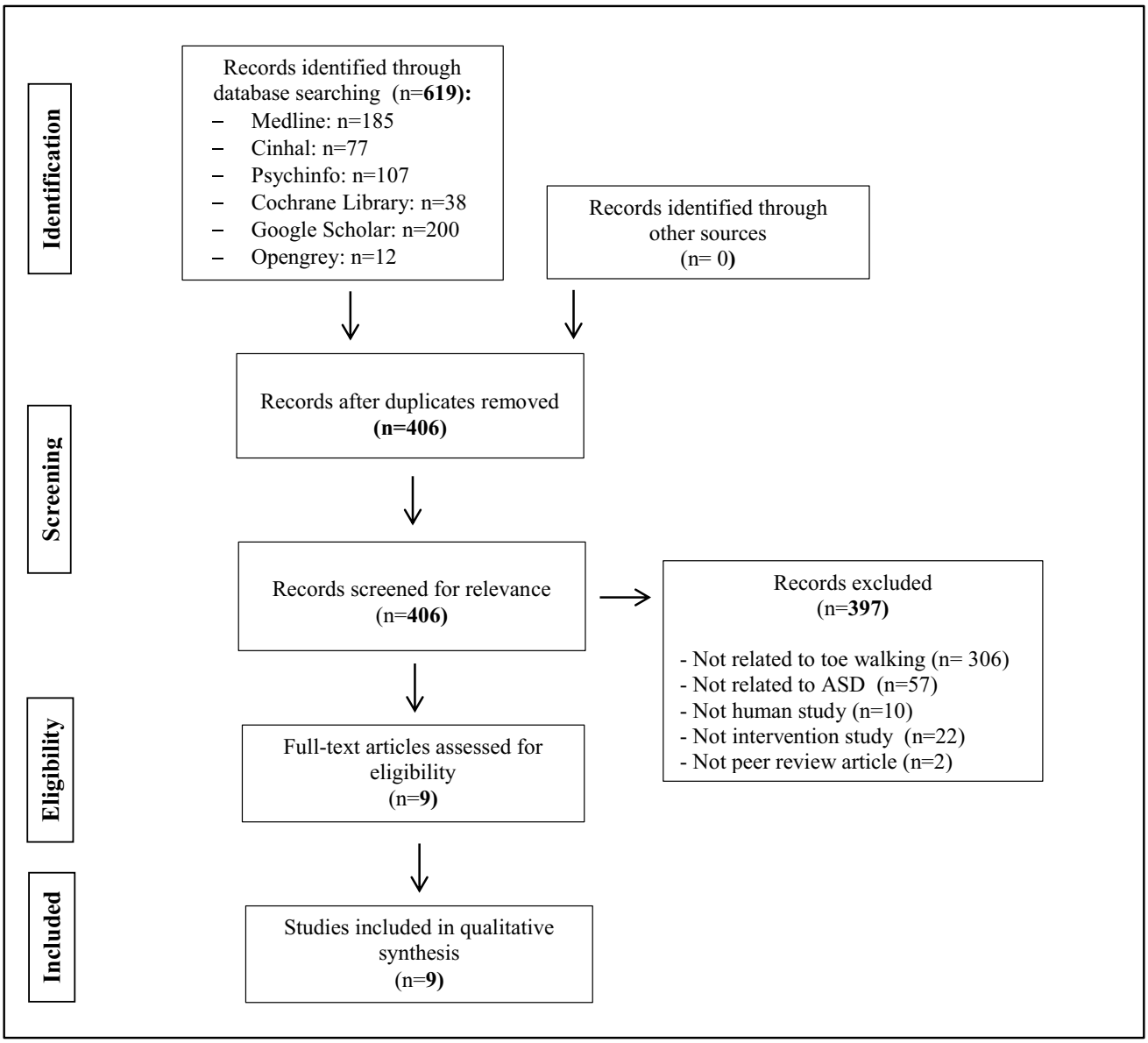

Fig. 1 Flow chart reporting the results of the search strategy and the selection of eligible and included articles

Study Characteristics

The details of the study characteristics are described in Table S2 (Supplementary Material 2). All the included studies were case reports or case series aimed to assess the effectiveness of a conservative intervention on TW frequency in persons with ASD. The included studies enrolled a total of 17 participants (16 males; age range: 4–15 years). One included study specified the diagnostic criteria used and the ASD severity of the individuals (Persicke et al., 2014). One study reported the presence of "moderate intellectual disability" in all the three involved participants. Kratz (2020) described the presence of "cognitive deficit" without the characterization of the impairment. The remaining studies ($n=7$) did not specify the participants' presence/absence of intellectual disability or intelligence quotient. No study reported the intellectual disability assessment method.

Seven included studies out of nine reported the language levels of the participants (Marcus et al., 2010; Persicke et al., 2014; Barkocy et al., 2017; Hodges et al., 2018, 2019; Wilder et al., 2020; Kratz, 2020). Comprehension ability was described in 5 articles; two studies described that the participants were not able to understand verbal instructions (Kratz, 2020; Persicke et al., 2014), while the subjects involved in 3 included studies had no comprehension problems (Barkocy et al., 2017; Hodges et al., 2018, 2019). Finally, four studies did not report the comprehension level (Marcus et al., 2010; Wilder et al., 2020; Shaw & Soto-Garcia, 2021; Barkocy et al., 2021).

Two studies reported the presence of other co-morbidities as mild cerebral palsy with epilepsy (Marcus et al., 2010) and dyspraxia and hypotonia (Kratz, 2020), while all the other studies did not specify the presence/absence of co-morbidities.

Seven out of seventeen participants had previous treatments because of TW persistence (Barkocy et al., 2017; Hodges et al., 2018; Kratz, 2020; Marcus et al., 2010; Persicke et al., 2014; Shaw & Soto-Garcia, 2021), but the proposed interventions did not influence it. Nine out of seventeen participants were not subjected to previous intervention for TW (Marcus et al., 2010; Wilder et al., 2020; Barkocy et al., 2021). Hodges' study (2019) did not report this information.

Toe Walking Interventions

The details about the interventions are described in Table 2.

Type of Interventions All the included studies assessed the effectiveness of conservative interventions. Barkocy et al., (2017 and 2021) used a serial casting approach followed by ankle–foot orthosis. Kratz (2020) utilized a single lymphatic drainage treatment, while Shaw and Soto-Garcia (2021) treated TW using cognitive-motor dual-tasking and primitive reflex integration exercises. The remaining five studies used behavioral treatments. In two out of five studies, acoustic feedback using "GaitSpot Auditory Shoe Squeakers" associated with positive reinforcement (verbal or tactile or small edible as *Chipsticks*) was proposed (Marcus et al., 2010; Wilder et al., 2020). Persicke et al. (2014) and Hodges et al. (2019) used a method for teaching behavior through positive reinforcement by a conditioned auditory stimulus (i.e., a "click" sound) to reinforce correct behaviors: the "TAGteach®" ™ method. The auditory stimulus was paired with an already established reinforcing item (small edible as *Chipsticks* or verbal or a favorite object) so that the "click" sound functions as a conditioned reinforcer. Finally, Hodges et al. (2018) extended previous research on stimulus control-based interventions to TW on a child with autism, using a wristband as a discriminative stimulus: in the presence of the wristband, appropriate walking was praised and TW was reprimanded.

Toe Walking Assessment Methods Most of the included studies used a video recording system to assess TW (Hodges et al., 2018, 2019; Marcus et al., 2010; Persicke et al., 2014; Wilder et al., 2020). Marcus et al. (2010) also employed pen and paper on a recording sheet to assess toe-walking steps in one of the three subjects involved in their study. Barkocy et al. (2017) used both gait analysis equipment and two scales (i.e., the Functional Mobility Scale and the Observational Gait Scale). Later, Barkocy et al. (2021) used both gait analysis equipment and two scales (i.e., the Functional Mobility Scale and the Patient Specific Function Scale). Kratz (2020) conducted the assessment through clinical observations and described the mobility skills using the Pediatric Evaluation of Disability Inventory; moreover, Kratz (2020) took photographs to record lower limbs alignment and feet placement before and after treatment. Finally, Shaw and Soto-Garcia (2021) used a clinical observation "based on quantifiable judgments by the child, parent, or healthcare professional."

Toe Walking Outcome Measures The outcome measures were different across the included studies. For the details of the outcome measure used in each study, see Table 2. Barkocy et al. (2017) used the Observational Gait Scale score, the Functional Mobility Scale score, the passive end-range ankle dorsiflexion angle (with the knee extended), and the kinematic data obtained using a gait analysis system. Later, Barkocy et al. (2021) used the Patient Specific Function Scale score, the Functional Mobility Scale score, the passive end-range ankle dorsiflexion angle (with the knee extended), and the kinematic data obtained using a gait analysis system. Kratz (2020) used as outcomes a clinical observation supported by photos and the mobility skills

Table 2 Detailed description of the proposed interventions in the included studies

Study	Study aim	Type of intervention	Device/technique employed	TW assessment methods	Outcome
1. Marcus et al. (2010)	To evaluate the effectiveness of a simplified habit reversal training with the use of GaitSpot Auditory Shoe Squeakers and differential reinforcement to reduce the frequency of TW in three subjects with ASD	Behavioral (simplified habit reversal) and differential reinforcement of incompatible behavior	GaitSpot Auditory Shoe Squeaker + positive reinforcement (social praise, edibles or tokens)	Video camera (2/3 subjects); pen and paper on a recording sheet (1/3 subject)	Percentage of 10 s intervals in 10 min sessions in which each participant engaged in TW
2. Persicke et al. (2014)	To evaluate the use of an acoustical conditioned reinforcer (TAG) in addition to minimal correction procedures to decrease TW for a child with ASD	Behavioral (conditioned stimulus)	Clicker device + positive reinforcement (potato chip) +correction procedure (downward pressure applied to shoulders)	Video camera	Percentage of flat-footed steps for each session in designated 20-foot-long areas
3. Barkocy et al. (2017)	To evaluate the effectiveness of serial casting to reduce TW in a child with ASD	Serial casting followed by AFOs	Cast made of semirigid fiberglass material, and ankle–foot orthosis	- Functional scales - Instrumental (Gait analysis)	- Observational Gait Scale score - Functional Mobility Scale score - Gait analysis data (kinematic, spatial and temporal parameters) - Passive Ankle dorsiflexion ROM
4. Hodges et al. (2018)	To examine the effect of a multiple schedule of reinforcement on TW exhibited by a child with autism using a wristband as a discriminative stimulus	Behavioral (stimulus control-based intervention)	Wristband + positive verbal reinforcement (praise) or negative verbal reinforcement (reprimands)	Video camera	Percentage of tip-toe steps in which the participant engaged in TW
5. Hodges et al. (2019)	Evaluating the effectiveness of an acoustical feedback to increase appropriate steps in a child with ASD	Behavioral (conditioned stimulus)	Clicker device + positive reinforcement (preferred items)	Video camera	Percentage of appropriate steps in sessions of 100 steps.
6. Wilder et al. (2020)	Evaluating the effectiveness of GaitSpot Squeakers to reduce TW in three subjects with ASD in the absence of other intervention components	Behavioral (conditioned stimulus)	GaitSpot Auditory Shoe Squeakers +positive reinforcement (edibles) (all three subjects) +correction procedure (downward pressure applied to shoulders) for one subject	Video camera	Percentage of TW steps during sessions of 100 and 1000 steps.
7. Kratz (2020)	To describe how lymphatic drainage can resolve TW gait and stance in a child with ASD	Lymphatic drainage massage	Lymphatic drainage massage using the *Chikly Method* 1 session of treatment that took 10–15 min per leg	- TW qualitative evaluation (reported by author) - Photos - Functional scale (PEDI)	- Clinical assessment of soleus-gastrocnemius muscle lengths - PEDI mobility subcategory score

Table 2 (continued)

Study	Study aim	Type of intervention	Device/technique employed	TW assessment methods	Outcome
8. Shaw and Soto-Garcia (2021)	To discuss the treatment of idiopathic toe-walking utilizing cognitive-motor dual-tasking and primitive reflex integration exercises	Active exercises (demonstrated during the visit and prescribed to be done at home) Vibration therapy	"lizard exercise" + vibration therapy + walking while performing cognitive or motor task	Gait observation ("based on quantifiable judgments by the child, parent, or healthcare professional")	- Time spent heel-toe walking - Occurrence of falling downstairs
9. Barkocy et al. (2021)	To determine the effect of a serial casting followed by Ankle Foot Orthoses treatment on walking kinematics and functional outcomes in autistic children who toe walk	Serial casting followed by AFOs	Cast made of semirigid fiberglass material, and ankle–foot orthosis	- Functional scales - Instrumental (Gait analysis)	Primary outcome: - Functional Mobility Scale score - Patient Specific Function Scale score Other outcome: - Gait analysis data (kinematic, spatial and temporal parameters) - Passive Ankle dorsiflexion ROM

ROM, range of movement. *PEDI*, Pediatric Evaluation of Disability Inventory. *AFO*, ankle foot orthoses. *TW*, toe walking. *ASD*, autism spectrum disorder

assessment using the mobility subsection of the Pediatric Evaluation of Disability Inventory. Marcus et al. (2010) calculated the percentage of 10-s intervals in 10-min sessions in which each participant engaged TW, dividing the number of occurrences of TW by the total number of 10 s windows TW exhibited. Hodges et al. (2018) and Wilder et al. (2020) used the percentage of steps engaged in TW during each assessment session as an assessment method, while Persicke et al. (2014) and Hodges et al. (2019) used the percentage of appropriate steps. Shaw and Soto-Garcia (2021) considered as outcome measure the time spent on heel-toe walking and the occurrence of falling downstairs.

Study Results Both pre- and post-treatment assessment details for all the included studies are reported in Table S3 (Supplementary Material 2). Serial casting followed by ankle–foot orthosis improved the administered functional scale scores, end-range passive ankle dorsiflexion range of movement, and the spatial and temporal parameters of gait measured by motion capture system (Barkocy et al., 2017 and 2021). A single treatment of manual lymphatic drainage on the lower limbs resulted in a full-feet placement on the floor with bilateral neutral dorsiflexion, a full-foot contact during gait, and an improvement in the mobility skills subsection of the Pediatric Evaluation of Disability Inventory (Kratz, 2020). Shaw and Soto-Garcia (2021) proposed a treatment based on cognitive-motor dual-tasking and primitive reflex integration exercises that reduced TW. Both the patient and mother stated that "he is now more comfortable within going downstairs and has not fallen since the second visit."

Behavioral treatments produced a TW improvement. The GaitSpot Auditory Shoe Squeakers (Marcus et al., 2010; Wilder et al., 2020), the use of a wristband (Hodges et al., 2018), and the TAGteach® (Hodges et al., 2019; Persicke et al., 2014) administered with or without positive reinforcement technique ± corrections reduced TW in the autistic individuals involved in the studies. Note that, in Marcus et al. (2010) study, one-third of the subjects completed the previously planned study phases.

Follow-up Barkocy et al. (2017) conducted a 2-year follow-up. Notably, the authors reported that the subjects continued to use the ankle–foot orthosis between the end of the study and the follow-up. Kratz (2020) performed a follow-up assessment after 1 year; the author described that the subject "continued with long term intensive occupational therapy" in the time between the end of the study and the follow-up. All the other studies (Hodges 2018, 2019; Marcus et al., 2010; Persicke et al., 2014; Wilder et al., 2020) did not report a follow-up.

Adverse Events and Drop-out Two individuals of Marcus' study (Marcus et al., 2010) and one subject of Wilder's study (Wilder et al., 2020) did not complete all treatment's study phases. One subject was unable to attend the study due to an injury unrelated to the study. The other could not reach the criteria established to complete all treatment's phases (Marcus et al., 2010). In Wilder et al. (2020) study, a subject had an unrelated medical issue, and the authors were unable to conduct the final study phase. Finally, the data collection protocol was modified for one participant of Barkocy et al. study (Barkocy et al., 2021) as the child experienced an adverse event at school that produced anxiety. For this reason, the data were collected for pre-intervention and post-intervention, the clinical evaluation was executed at home while the child was sleeping, and casting was done in the home environment.

Discussion

Study Design and Study Sample Characteristics

This systematic review aimed to present the current evidence regarding intervention effectiveness for TW in persons with autism. Despite a comprehensive search strategy, only nine studies were identified. All the included studies had a case report or case series study design and considered a total of 17 autistic individuals (16 males) ranging from 4 to 15 years. Only one out of nine studies (Persicke et al., 2014) sufficiently described the ASD diagnostic criteria and severity. Moreover, only two out of nine studies reported the presence/absence of comorbidities and gave information about intellectual disability (see CARE checklist assessment—Table S1 and Table S2) (Kratz, 2020; Marcus et al., 2010) without specifying the assessment methodology. Seven out of nine studies gave information about participants' language level, which was significantly diversified between study participants. Also, comprehension ability was found heterogeneous between studies. Even though TW is thought as a phenomenon related to sensory disturbance (Accardo et al., 2014; Valagussa et al., 2017), we noted that none of the included studies planned to assess it. Therefore, the individuals' deficit characteristics and severity were not clearly stated. All these findings together highlight the need to conduct higher quality design studies. Moreover, since an extreme variability of clinical manifestation characterizes ASD, greater relevance should be given for a better characterization of the sample when planning future studies regarding TW treatment, as previously suggested by Fournier et al. (2010).

Toe Walking Interventions

Four types of treatments were proposed to decrease TW in persons with autism: (1) behavioral interventions (Marcus et al., 2010, Persicke et al., 2014; Hodges et al., 2018; Wilder et al., 2020; Hodges et al., 2019), (2) use of serial casting followed by ankle–foot orthoses (Barkocy et al., 2017 and 2021), (3) use of a lymphatic drainage technique (Kratz, 2020), and (4) use of cognitive-motor dual-tasking and primitive reflex integration exercises (Shaw & Soto-Garcia, 2021).

Among the proposed behavioral interventions, two studies (six subjects) evaluated the efficacy of using GaitSpot Auditory Squeaker (Marcus et al., 2010; Wilder et al., 2020). This object was positioned under the participant's heel inside or under the sole of both shoes. When the subject walked adequately, the pressure given by the heel pushes the device, emitting auditory feedback. This procedure and edible and vocal positive reinforcement appeared to reduce the frequency of TW in the participants of Marcus et al. (2010) study. Although Wilder et al. (2020) aimed to study the efficacy of the GaitSpot device without adding other reinforcement components, only one out of three included subjects seemed to reduce TW only using this device. The other two participants also needed to introduce further treatment strategies (i.e., an edible reinforcement alone and an edible reinforcement plus manual correction). Thus, it would be clinically valuable to conduct future research to investigate the responders' characteristics to the GaitSpot treatment alone compared to responders to GaitSpot associated with another reinforcement. Moreover, Wilders et al. (2020) recognize as a limitation that they did not assess the extent to which the intervention effects generalized in the absence of the GaitSpot Auditory Squeaker. On the other hand, the squeaker sound could be valuable feedback because it could be used without healthcare professionals.

Two studies (Hodges et al., 2019; Persicke et al., 2014) adopted another behavioral correction technique called TAGteach® in addition to reinforcement procedures. It consists of using a "clicker" device manually squeezed by the examiner when the subject walks properly. For the two participants of the studies, there was a reduction in the percentage of TW. The authors added a manual correction with pressure on one of the two participant's shoulders to induce his heels to touch the ground (Persicke et al., 2014). Compared to the treatment with GaitSpot, TAGteach® requires another subject that makes the "click" sound every correct step. Moreover, being a procedure that requires careful and continuous observation by the examiner, it is easily prone to error (e.g., the examiner does not notice that a correct step has been taken and, as a consequence, does not squeeze the clicker or the examiner squeeze the device even when there is no complete support of the heel on the ground). A

limit of this type of intervention could be using this device in the context of everyday life. It may interfere with daily social activities (e.g., school) as the repeated "click" sound emission could be a nuisance in a quiet environment or, conversely, it may not be heard in a noisy environment. Moreover, it could be less socially acceptable because the TAGteach® technique is also used in other contexts, including animal training (TAGteach® International, 2012).

Hodges et al. (2018) adopted a type of behavioral intervention based on stimulus control using a wristband as a tactile stimulus worn on the participant's wrist. The tactile stimulus praised the subject when he was walking properly while giving a reprimand when showing TW. Compared to the previous ones (Hodges et al., 2019; Persicke et al., 2014), this type of intervention could be more acceptable in a social context.

Three other different treatment types were described. Serial casting followed by ankle–foot orthoses was proposed to increase the dorsiflexion of the tibiotarsal joint and reduce TW in two studies (Barkocy et al., 2017 and 2021). In particular, Barkocy et al. (2021) demonstrated the feasibility of such treatment in a population of autistic children even if a clear characterization of the study sample was missing. On the other hand, compared to the previous treatments, this type of intervention is more invasive since it limits joint mobility and the motor activity of the subjects. The second described treatment is specific lymphatic drainage called the "Chikly method" (Kratz, 2020). The author reported that the subject ultimately resolved its TW after one lymphatic drainage session. However, the author did not provide quantitative data. This technique showed different limits. Firstly, to be correctly applied, it requires specific healthcare professional training. Moreover, the underlying hypothesis that drove this intervention was not clearly stated. Notably, since about 60% of ASD subjects seem to show a sensorial disturbance (Tomchek & Dunn, 2007), including sensory deficits with characteristics of hypersensitivity, the application of manual technique or serial casting could create a sense of discomfort in a subject with these features, and an adverse reaction of avoidance or agitation could be provoked (Miller et al., 2007). Data about the sensory profile of the subjects involved in all the included studies were not provided.

The last treatment consisted of a series of cognitive-motor dual-tasking and primitive reflex integration exercises (Shaw & Soto-Garcia, 2021) executed during the treatment and prescribed as an exercise to perform at home. The authors recognized as a significant limitation of their study the lack of objective measurements to detect the effectiveness of treatments.

A limit presented by five out of nine studies is the absence of follow-up, not allowing for evaluating the efficacy of the treatments in the medium-long term. Moreover, in studies where a follow-up was planned, other interventions have been applied between the end of the treatment and the follow-up. Thus, it is not possible to infer that the reduction of TW frequency directly depends on the investigated treatment.

All the retrieved studies considered only conservative interventions. Despite pharmacological (i.e., botulinum toxin) and surgical interventions (i.e., muscle lengthening) are currently used in clinical practice to treat TW in persons with autism (Leyden et al., 2019), no studies about pharmacological and surgical effectiveness were found. All this suggests the need to plan studies about pharmacological and surgical interventions specifically addressed to persons with autism, as also previously suggested by Bartoky et al. (2017). Notably, as reported by a recent Cochrane review (Caserta et al., 2019), behavioral treatments and serial casting were also proposed to reduce ITW. In particular, the authors reported the results of a study (Engström et al., 2013) where the effectiveness of serial casting alone was compared to the use of serial casting associated with botulinum toxin: both procedures reduced ITW, but it was not possible to define the best treatment option.

The methodological quality of the included studies was assessed through the framework developed by Murad et al. (2018). This checklist does not provide a total score, and no cut-offs are yet available to classify the methodological quality. However, only two studies met half of the domains (i.e., four out of eight); one domain (i.e., selection domain) was only met in one study, while another domain (i.e., randomity) was not met in any included case report. Therefore, the methodological quality of the included studies may have some flaws.

Limitations and Strengths of the Study

Publication bias is a potential limitation of any review. However, a strong publication bias is unlikely in this review as several databases were queries, and no language or year of publication restrictions were applied. On the other hand, to our knowledge, this is the first systematic review addressed to summarize the evidence about the treatment of TW in ASD.

Research Implications

This systematic review evidenced several important and helpful points that should be incorporated and considered in future studies. Clinicians and researchers should better characterize the study sample in terms of conditions that may impact TW assessment and treatment. The following characteristics should be described: (I) ASD diagnostic criteria and ASD severity; (II) the presence/absence of comorbidities; (III) information about the intellectual disability level/IQ score and the methodology used to assess it; (IV)

information about the ability of language comprehension and expression; (V) information about previous treatments for TW and the obtained results; and (VI) information about sensory characteristics. Additionally, there is a need to perform higher-quality studies, such as prospective cohort studies and/or randomized controlled trials. Researchers should consider planning an effective follow-up strategy to evaluate the efficacy of the treatments in the medium-long term. Finally, no studies concerning pharmacological (e.g., botulinum toxin) and surgical interventions (e.g., muscle lengthening) for treating TW in autistic individuals were found; therefore, studies are needed to investigate their effect specifically in individuals with ASD.

A significant drawback in designing RCTs for interventions to improve TW in autism is that the etiology of TW is not well understood (Accardo et al., 2014). Therefore, the developed treatments tend to be symptom-based rather than etiology-based. Furthermore, the interventions presented in our systematic review should be investigated to verify their applicability to subjects with greater severity of ASD. Researchers should include individuals with a broad range of populations (e.g., from mild to severe intellectual disability) to enhance the generalizability of their studies. Concerning the outcomes, there is no gold standard measure for assessing interventions for TW. The most used outcome measure seems to be the time or the number of feet in TW (i.e., percentage of the time or number of feet engaged in TW during testing or monitoring), evaluated by objective and/or subjective measures, or kinematically changes of gait parameters measured by a motion capture system. Additionally, an indirect outcome measure is the quantity of foot range of movement in dorsiflexion performed with both the knee extended and flexed. To this end, the scientific community should set the standard for the outcomes used for assessing the effectiveness of treatment for TW, thus facilitating researchers and clinicians to use similar assessments.

Conclusion

There is a lack of studies with appropriate study designs and a sufficiently large and well-characterized sample to assess the effectiveness of conservative treatments for TW in persons with autism. The review highlights the absence of studies about the effectiveness of pharmacological and surgical interventions on TW individuals with ASD. The findings strongly support the need for further higher-quality studies addressed to evaluate the effectiveness of conservative, pharmacological, and surgical interventions for TW in ASD.

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Declarations

Ethics Committee Approval The manuscript is a systematic review that does not require ethics committee approval.

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