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RESEARCH ARTICLE

Quantitative assessment of tip-toe behavior in individuals with autism spectrum disorder and intellectual disability: A cross-sectional study

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Abstract

The term "toe walking" describes walking on the toes with a lack of heel strike upon initiation of the stance phase of gait. In individuals with autism spectrum disorder (ASD), this phenomenon, or "tip-toe behavior" (TTB), can be present in a substantial proportion of subjects even during standing. In this study, we investigated TTB in 50 persons with ASD (age range 4–26 years). We evaluated TTB through an observational/report-based assessment protocol. Subsequently, we employed a new structured video-based coding protocol based on standardized video recordings, focusing on static and dynamic conditions. Finally, the findings of the two protocols were compared. Twenty-four subjects with TTB were identified and classified according to three functional groups: TTB1, present only during running (6 subjects); TTB2, present during walking and running (11 subjects); and TTB3, present during standing, walking, and running (7 subjects). Moreover, we found that TTB3 subjects exhibited a significantly higher quantity of TTB compared with subjects in the TTB1 and TTB2 groups during both standing and walking tests. Additionally, a high quantity of TTB in the static test was found to be related to a high quantity of TTB in the dynamic test. Variables such as age, autism severity, intellectual disability, and gender were not significantly associated with the mean percent of TTB both in static and dynamic tests in multivariate analysis. This structured video-based coding approach appears feasible and useful for assessing TTB in individuals with ASD and it has the potential to provide insights into TTB trajectories and aid in designing possible interventions.

Lay Summary

Toe walking can be observed during the gait of individuals with autism spectrum disorder (ASD). As this motor behavior can also be present during standing, we used the term tip-toe behavior (TTB). TTB is an understudied sensorimotor phenomenon in ASD, and there is a lack of quantitative measures of TTB in ASD. This study proposed two standardized clinical tests to quantify TTB during walking and standing. These TTB measures could aid in understanding the natural history of TTB and in making decisions about TTB interventions.

KEYWORDS

assessment, autistic disorder, intellectual disability, toe walking, walking

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by persistent impairments in social interaction and the presence of restricted and repetitive patterns of behaviors, interests, or activities (American Psychiatric Association, 2013). ASD affects approximately 1 in 36 children aged 8 years in the United States, with 3.8 males diagnosed for every female (Maenner et al., 2023). An ASD diagnosis can be made after a comprehensive developmental history is taken and the subject's behavior is observed during interactions with other individuals (Risi et al., 2006).

Green et al. (2009) found up to 79%–83% of children with ASD have difficulty performing appropriate motor skills for their age. This seems to occur independently of the co-occurring presence of intellectual disability (ID) (Bhat et al., 2011). These motor impairments are clinically relevant because they can influence participation in activities crucial for the development of age-appropriate behavioral, cognitive, social, and communication skills (Bhat et al., 2011, 2022). Moreover, motor difficulties could interfere with participation in physical activity important for optimal health and wellness (Srinivasan et al., 2014).

Difficulties in motor skills performance could arise from specific motor impairments or differences in other domains that affect the way they learn motor skills, as suggested by de Moraes et al. (2017). This is corroborated by literature demonstrating impairments in postural control in people living with ASD with or without ID (Lim et al., 2017; Perin et al., 2020), difficulties in motor planning (Simermeyer & Ketcham, 2015), motor imitation (Williams et al., 2004; Zachor et al., 2010), and gait impairments, such as wider step width, slower walking speed, longer gait cycle, longer stance time, and longer step time, as summarized in a recent systematic review (Lum et al., 2021). Additionally, individuals with ASD may also manifest sensory processing differences, including tactile hypersensitivity and other sensory modulation impairments (Liu, 2013; Purpura et al., 2022; Tomchek et al., 2014; Valagussa et al., 2022). Interestingly, recent literature suggests that interventions aiming to improve motor skills have been effective in also improving social and communication dimensions (Bremer & Lloyd, 2016; Haghighi et al., 2022; Ketcheson et al., 2017).

Among the potential motor disorders in this population, toe walking (TW) is a clinically known phenomenon that can be found during gait observation. It is a term used to describe walking on the toes with a lack of heel strike upon initiation of the stance phase of gait (Ruzbarsky et al., 2016). Literature indicates that the term tip-toe behavior (TTB) might be more appropriate, as TTB can also manifest during standing (Valagussa et al., 2017; Weber, 1978) and/or running (Robert, 2011; Valagussa et al., 2017).

TW prevalence in individuals with ASD is about 20%–30% (Barrow et al., 2011; Ming et al., 2007; Shetreat-Klein et al., 2014). Moreover, TW was found more prevalent in individuals with ASD than in the general population (i.e., 33% vs. 3% and 8.5% vs. 0.47%) (Leyden et al., 2019; Shetreat-Klein et al., 2014).

TW occurs in varying degrees of severity (Accardo & Barrow, 2015). In a cohort study, using a qualitative assessment, Valagussa et al. (2017) described three mutually exclusive TTB clinical functional classes of increasing severity: (1) TTB present only during running, (2) TTB present during walking and running, and (3) TTB present during standing, walking, and running. Furthermore, it was suggested that the persistence and severity of TTB can impact the length of the Achilles tendon (Accardo et al., 2014; Valagussa et al., 2020). This, in turn, can influence motor behavior in terms of calf discomfort/pain during daily life activities, such as jogging or a higher risk of falling (Caselli et al., 1988), affecting the child's functional capabilities and quality of life (Calhoun et al., 2011). Moreover, TW can be associated with a social or cosmetic impact (Pendharkar et al., 2008).

As of now, various etiological hypotheses of TTB have been suggested. Weber (1978) proposed that "toe walking arises from the fixation of a normal transient stage of development"; later, Accardo et al. (2014) suggested TTB as a "residual of a primitive reflex (i.e., positive support reflex or tonic labyrinthine reflex)" or as a result of a vestibular issue. It was also hypothesized that TTB could be a sign of a sensory modulation or processing impairment. A reduction in TTB was observed when two individuals with ASD walked on different surfaces, as discovered by Wilder et al. (2022).

A systematic review describing the methods employed to assess TTB in individuals with ASD (Valagussa et al., 2018) highlighted several methodological issues in TTB assessment. The included studies presented heterogeneous information sources (i.e., information from parents, questionnaires administered to the parents, information collected by therapists' records or physicians' records, clinical observation by the medical doctor during office visits, video observation, and information provided by the main caregiver of the subject). Additionally, the qualitative descriptions of the TTB grading systems used varied among studies. Some used descriptions like "absent or present," "absent, present in the past, or present," "absent, present temporarily, present constantly," or "absent, present in the past, intermittently present, persistent." Furthermore, the procedures for TTB assessment were often not clearly outlined, which compromised the repeatability and comparability of the studies. Additionally, relying on a single observation to determine the presence or absence of TTB in individuals with ASD could be problematic due to the inherent behavioral variability observed in individuals with ASD. Finally, a quantitative assessment of TTB during walking was performed only by one study (a case study involving a single individual), while surprisingly, no study conducted a quantitative assessment of TTB during standing.

These concerns were further confirmed in the study by Gong et al. (2020). Moreover, Wilson et al. (2018) suggested that "in the assessment of motor function in ASD it is imperative the field continue to employ quantitative measures." Quantitative measures of motor function could be useful to assess a certain behavior and its trend across a lifespan, and thus they could become useful from a clinical and research standpoint.

Therefore, the primary aims of this cross-sectional study are: (1) to propose a quantitative approach to TTB assessment during both a static (i.e., in standing) and a dynamic (i.e., during walking) conditions in a sample of individuals with ASD and ID; (2) to assess the feasibility of this quantitative and ecological TTB assessment in individuals with ASD and ID; (3) to analyze and compare the quantitative characteristics of TTB subgroups, suggesting a possible clinical management. Furthermore, in a clinical setting, we have observed that individuals who exhibit a higher quantity of TTB while standing tend to also have a higher quantity of TTB during walking. Consequently, a secondary objective of this study is to evaluate the relationship between the quantity of TTB observed during the static and dynamic tests.

MATERIALS AND METHODS

Sample and study design

We conducted a cross-sectional study in a Neuropsychiatric Institute, Villa Santa Maria Institute, where there were 62 subjects (mean age: 13.25 years—SD 4.40; age range: 3.50–26.58 years; 55 males) with an ASD diagnosis.

The inclusion criteria were: (1) an ASD diagnosis according to the DSM-5 criteria; (2) a diagnosis confirmation established through ADOS-2.

The exclusion criteria were: (1) the presence of neurological comorbidities that could influence the performance during the quantitative assessment tests (i.e., cerebral palsy, spinal muscular atrophy, muscular dystrophy, Down syndrome, Rett syndrome, and blindness); (2) the subject was not able to conclude one out of three trials of one out of two planned quantitative tests.

Trained expert developmental psychologists, after performing clinical and diagnostic evaluations, because of the ID severity, decided to describe ID using the four classes ID classification to DSM-5 (i.e., mild, moderate, severe, and profound).

Parents or guardians of all participants signed an informed consent form to participate in the study. Ethical approval was obtained from the local IRB Insubria''s Ethics Committee (study n° 185/2019).

Measure and procedure

For this study, the evaluation of TTB was based only on the assessments of the current performance at the time of the recruitment. Historical information about the presence of TTB in the past was not obtained. Both an observational/report-based assessment and a structured video-based coding of TTB were administered to the individuals included in the study, following the procedure shown in Figure 1. The time interval between the two assessments (observational/report-based and structured video-based coding) had to be not more than 3 months.

Observational/report-based TTB assessment

A therapist assessed the presence/absence of TTB during standing and/or walking and/or running using both direct observation and a structured interview of the main caregiver living with the subject using a methodology previously described (Valagussa et al., 2017). This assessment classifies the subjects into three TTB clinical functional classes of increasing severity: TTB present only during running (TTB1), TTB present during walking and running (TTB2), and TTB present during standing, walking, and running (TTB3). A description of the testing and scoring procedure of the observational/report-based TTB assessment is available as Supplementary material 1.

Structured video-based coding of TTB

The quantity of TTB expression during the static and dynamic tests was calculated using two standardized tests. Both tests were conducted using a standardized video-recording approach. This approach was previously successfully used to assess TW in individuals with ASD by Marcus et al. (2010) and Persicke et al. (2014). Furthermore, a systematic review suggests that 2D video-based analysis is a reliable method for assessing foot strike patterns (de Oliveira et al., 2019).

Setting. The tests were conducted in a familiar setting with a known therapist to minimize potential biases associated with new environments or unfamiliar individuals. TTB was quantified during both standing and walking, with participants not wearing shoes but wearing socks. Linoleum flooring was chosen instead of standard flooring to eliminate the potential influence of a cold walking surface. Additionally, the height of the table was adjusted to match the subjects' umbilicus height prior to commencing the test.

During the dynamic test, the subject was video recorded on the sagittal plane. The camera was placed on the floor to have a good perspective to judge the foot contact on the floor, in a middle position between the starting point and a table situated 2 m away. The camera was positioned in a way that the video-recorded area encompassed the region between the pelvis and the feet of the individual. Additionally, a person was assigned to adjust the camera to track the walking movements during the test. During the static test, the subject was video recorded on the frontal plane (from the backside). The

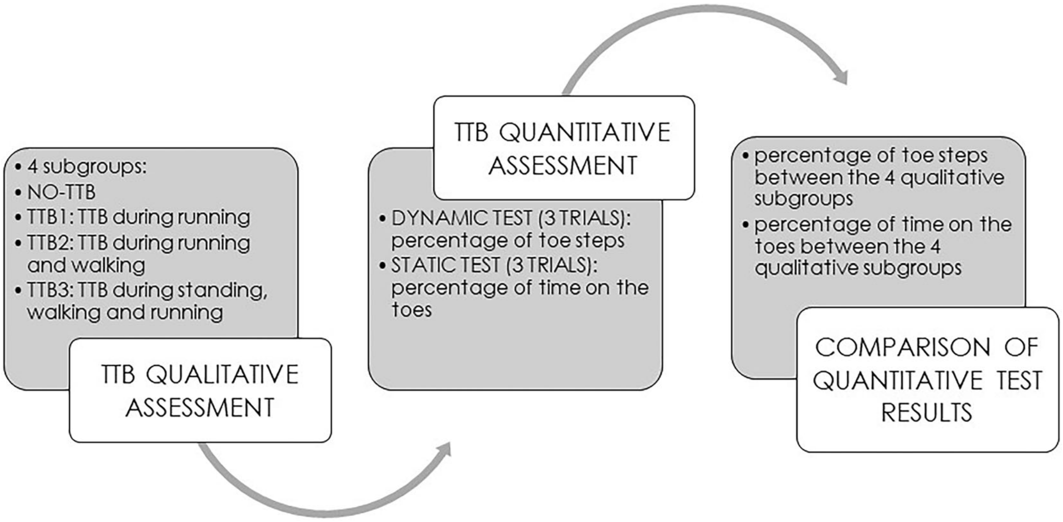

FIGURE 1 Schematic overview of the procedure used in the study. TTB, tip-toe behavior.

camera was placed on the floor and it was adjusted so that the video-recorded body area was between the pelvis and the feet of the individual. A person was involved to turn the camera to follow an eventual shift on the frontal plane during the test if necessary. Dynamic and static tests were repeated using the same setting on three separate days to decrease the bias related to the behavior variability of persons with ASD.

The two tests were masked in a play environment, using two tasks that corresponded to the dynamic and static tests, in the following sequence.

Assessment phase

Dynamic test: the therapist requested the subjects to transport an object (i.e., interlocking puzzle, shape sorter, etc.) previously chosen among his or her favorite toys from the starting point to the table situated 2 m away and back again for 15 consecutive times (total of 30 registered transfers). If at any point during this task, the subject was distracted and exited the area being filmed, or the subject jumped or run, an additional object to transport was added to assure 30 valid transfers were available for analysis. When the dynamic task was not executed with verbal cues, we used simple imitation strategies (e.g., walking beside the individual) and/or using hand contact.

Static test: the therapist asked the subjects to play with the previously transferred pieces while standing in front of the table for 3 min. During this task, the therapist stood beside the child to facilitate play if necessary. If at any point during the task, the subject was distracted and exited the area being filmed, additional time was added to insure 180 valid seconds were available for analysis.

Data extraction phase

All the video recordings were reviewed by a therapist not involved in the video-recording phase. The therapist was instructed on the criteria to evaluate the videos and trained using some videos of subjects not included in this study. During the evaluation of a video, if any doubts arose, the therapist could review the video or slow the speed of the video.

Dynamic test: the number of steps taken on the toes was calculated observing the video. We considered as toe step the absence of heel strike at initial contact and during the stance phase of walking (Sutherland et al., 1980). However, if during the phase of support of the foot, such support happened on the toes but was then followed by support of all the rest of the foot before the support of the opposite foot, this step was not to be counted as a step on the toes. Moreover, we did not consider the sequence of steps or the step that the subject used to reverse the direction: this sequence begins with the last foot support before changing direction and ends when the foot is oriented in the new direction. If during the task the subject was distracted and left the area being filmed, that part was not calculated. The counting of the steps was always done using the analysis of the first 30 valid tests. The percentage of toe steps was calculated comparing the number of the steps on the toes with the total number of steps. The average of the results of the three tests was then calculated.

Static test: the time (in seconds) spent weight-bearing on the toes during the 3 min was calculated, followed by calculating the time spent weight-bearing on the full sole bilaterally. The time spent weight-bearing on the full sole of one foot was calculated as the remaining time to reach 180 s. If during the assigned task the subject lost the position (e.g., walk away, sit down, or jump), the counting of the seconds was interrupted. Always the first valid 180 s were analyzed. Then, we calculated the percentage of time spent on each of the three conditions compared to the total time (180 s). The percentage of time spent on the toes compared to the total time (180 s) represented the TTB during standing. The average of the results of the three tests was then calculated.

A step-by-step procedure related to the dynamic and static tests data extraction phase is available in the Supplementary material 2.

Observational/report-based TTB assessment reliability
One expert therapist and one young therapist (graduate for a year) were recruited to determine the inter-rater reliability of the observational/report-based TTB assessment. Prior to initiating the assessments, the two raters received training from the senior investigator. During the first part of the training, written and verbal instructions for performing the observational/report-based TTB assessment were given to the two raters. Then, the two raters were asked to complete an assessment of two individuals (one with TTB and one without TTB), not included in the list of individuals used for the reliability assessment. The results of the analysis completed by each rater were then reviewed with the two raters during another training session; during that time any questions or concerns with the assessment procedures or scoring could be addressed.

Then, to establish inter-rater reliability of the observational/report-based TTB assessment, each of the two raters assessed 10 individuals with ASD and TTB and 10 NO-TTB individuals, presented in a randomized sequence. To establish intra-rater reliability, the expert therapist repeated the measurements after a week.

Structured video-based coding reliability
One expert therapist and one novice (last-year physiotherapy student) were recruited to determine the intra- and inter-rater reliability of the structured video-based coding. Prior to conducting the video-recording assessments, both raters underwent training provided by the senior investigator. During the first part of the training, written and verbal instructions for performing the structured video-based coding were given to the two raters. Then, the two raters were asked to complete an analysis of both a static and a dynamic test of two individuals (one with TTB and one without TTB), not included in the list of individuals used for the reliability assessment. The results of the analysis completed by each rater were then reviewed with the two raters during another training session; during that time any questions or concerns about the assessment could be addressed.

Then, to establish inter-rater reliability of structured video-based coding, each of the two raters evaluated 18 videos of the static test (9 videos of TTB individuals – 9 videos of NO-TTB individuals) and 18 videos of the dynamic test (9 videos of TTB individuals – 9 videos of NO-TTB individuals). The videos were presented in a randomized sequence. To assess intra-rater reliability, the expert therapist repeated the measurements after a week.

Statistical analysis

We presented the data as a percentage and mean ± standard deviation (SD) for nominal and continuous variables, respectively. Data were checked for normal distribution (i.e., Shapiro–Wilk test p-value >0.05 and by visual inspection of Q–Q plot). T-test was used if assumptions for data normality and homogeneity were met. Kruskal–Wallis test was used to compare the four TTB subgroups on the two outcomes. Single Mann–Whitney U test was adopted for the pairwise comparisons. p-values were adjusted for multiple comparisons using the false-discovery rate approach. Moreover, we used the Spearman correlation test to assess the correlation between the mean percentage values of time maintained on the toes during the static test and the mean percentage number of steps on the toes during the dynamic test. We performed a multivariable linear regression analysis to adjust the association of TTB subgroups with the mean percentage of time spent on the toes during the static quantitative test and the mean percentage of toe steps during the dynamic quantitative test for other demographic and clinical characteristics as age, ADOS severity (expressed as ADOS CSS values) and ID. Moreover, the Intraclass correlation coefficient (ICC) test (two-way mixed effects, single measurement, and absolute agreement) was performed to estimate the intra-rater agreement of the expert therapist in scoring both static and dynamic tests. The inter-rater agreement between the expert and the novice in scoring both static and dynamic tests was estimated using the ICC test (two-way random effects, single measurement, and absolute agreement). Finally, using Cohen's kappa coefficient, we estimated the intra-rater reliability of the expert therapist in scoring the observational/report-based TTB assessment and the inter-rater reliability of the observational/report-based TTB assessment between the expert and the young therapists. The significance level was set at a p-value <0.05. Data analysis was conducted using SPSS version 23.0 for Windows (IBM Corp., Armonk, NY, USA).

RESULTS

Motor impairments are underrecognized and underscreened from both clinical and research points of view, despite their prevalence and pervasiveness in individuals with ASD. Furthermore, very often, the proposed standardized motor assessment does not consider the contemporary presence of ID. Recent literature suggests that "the aetiology, natural history, and intervention efficacy for toe walking in ASD need to be further studied"

TABLE 1 Main demographic and clinical characteristics of the study sample.

		Total ($N = 50$)	NO-TTB ($n = 26$)	TTB ($n = 24$)	TTB class 1 ($n = 6$)	TTB class 2 ($n = 11$)	TTB class 3 ($n = 7$)
Age (years)	Range	3.53–26.59	7.28–26.59	3.53–17.81	9.17–17.81	3.88–16.93	3.53–15.26
	Mean (SD)	13.35 (4.49)	14.85 (4.31)	11.73 (4.18)	14.27 (2.83)	11.81 (4.35)	9.42 (3.99)
	Median	13.73	14.01	12.25	14.57	12.31	8.76
Sex	Male (*n*; %)	43; 86%	22	21	5	10	6
	Female (*n*; %)	7; 14%	4	3	1	1	1
ADOS total score	Mean (SD)	20.0 (5.46)	18 (5.71)	22.17 (4.31)	23.83 (3.6)	20.55 (4.61)	23.29 (3.99)
	Median	20.5	19	23	24.5	22	25
ADOS CSS	Mean (SD)	7.56 (1.68)	6.96 (1.54)	8.21 (1.62)	8.5 (1.76)	7.73 (1.68)	8.71 (1.38)
	Median	7	7	8	9	8	9
Intellectual disability	Mild (*n*)	3	2	1	0	1	0
	Moderate (*n*)	1	1	0	0	0	0
	Severe (*n*)	15	10	5	1	0	4
	Profound (*n*)	31	13	18	5	10	3

Abbreviations: CSS, calibrated severity score; SD, standard deviation; TTB, tip-toe behavior.

(Bhat & Kaznica, 2021), and "there is a need for more objective motor measures that can evaluate autistic individuals across a life span with a range of intellectual abilities" (Wilson, 2022). Considering this perspective, the proposal of a structured video-based coding approach for quantifying TTB in individuals with ASD who also have intellectual disabilities holds significance. This approach can be particularly relevant in clinical settings, offering a standardized method to assess TTB in a comprehensive and informative manner.

Participants

According to inclusion and exclusion criteria, we included 50 out of 62 individuals with ASD (43 males, 86%). Four subjects were not included because they had no diagnosis confirmation through ADOS-2, and eight subjects were excluded because they were not able to conclude one out of three trials of one of the planned quantitative tests for behavioral issues. All these subjects had a profound ID. Table 1 reports the demographic and clinical characteristics of the included subjects. The overall age of the sample was 13.35 years (SD 4.49). The mean age of the NO-TTB subjects was 14.85 years (4.31 SD) versus the mean age of 11.73 years (4.18 SD) of the TTB group ($p = 0.048$). We also assessed for the age difference between the three TTB subgroups using a Kruskal–Wallis test, but we did not find any significant difference ($p = 0.101$).

The overall ADOS calibrated severity score (CSS) of the study sample was 7.56 (1.68 SD). The ADOS CSS of the NO-TTB group was 6.96 (1.54 SD) versus an ADOS CSS of 8.21 (1.62 SD) in the TTB group ($p = 0.006$). No significant differences were found comparing the three TTB subgroups ($p = 0.417$). Finally, according to DSM-5 criteria, three subjects (6%) presented with a mild ID, 1 subject (2%) with a moderate ID, 15 subjects (30%) with a severe ID and 31 subjects (62%) with a profound ID.

Intra- and inter-rater reliability of the structured video-based coding of TTB

Intra-rater correlation of static and dynamic tests was 0.998 (95% confidence interval [CI], 0.994–0.999) and 0.999 (95% CI, 0.997–1). Inter-rater correlation of static and dynamic tests was, respectively, 0.978 (95% CI, 0.935–0.992) and 0.944 (95% CI, 0.860–0.979).

Intra- and inter-rater reliability of the observational/report-based TTB assessment

The Cohen's kappa coefficient of the observational/report-based TTB assessment between two observation of the expert therapist was 0.847 (95% CI, 0.647–1.000). The Cohen's kappa coefficient of the observational/report-based TTB assessment between the expert and the young therapists was 0.782 (95% CI, 0.566–0.998), showing a substantial agreement between the two raters.

Observational/report-based TTB assessment

According to the observational/report-based TTB assessment, 26 subjects with ASD (52%) resulted in the NO-

TABLE 2 Percentage values of static and dynamic tests in the total sample and the subgroups.

		Total ($N = 50$)	NO-TTB ($n = 26$)	TTB ($n = 24$)	TTB1 ($n = 6$)	TTB2 ($n = 11$)	TTB3 ($n = 7$)
Static test Bipodalic support	Mean (SD)	61.01 (27.15)	69.16 (25.75)	52.17 (26.33)	66.38 (18.67)	58.70 (23.85)	29.74 (23.49)
	Median	67.78	74.22	55.7	60.43	66.85	25.37
	Range (min–max)	0–100	4.07–99.44	0–100	50–100	10–85.37	0–68.7
Static test Monopodalic support	Mean (SD)	33.86 (23.65)	30.60 (25.68)	37.39 (21.22)	31.78 (18.77)	39.89 (22.74)	38.25 (22.95)
	Median	27.5	25.77	33.98	35.04	31.85	36.11
	Range (min–max)	0–95.37	0.56–95.37	0–89	0–50	14.44–89	4.33–78.06
Static test On the toes	Mean (SD)	5.16 (15.78)	0.24 (0.37)	10.49 (21.77)	1.85 (3.63)	1.3 (1.92)	32.34 (31.82)
	Median	0.19	0	1.58	0	0.93	19.44
	Range (min–max)	0–95.67	0–1.3	0–95.67	0–9.07	0–6.67	6.85–95.67
Dynamic test Heel/flat foot contact	Mean (SD)	88.22 (23)	99.32 (1.45)	76.19 (28.88)	96.27 (3.11)	88.85 (7.14)	39.07 (28.29)
	Median	98.98	100	88.18	95.9	89.72	38.02
	Range (min–max)	0–100	95.06–100	0–100	92.87–100	77–99.66	0–81.9
Dynamic test Toe steps	Mean (SD)	11.58 (23)	0.68 (1.45)	23.4 (29.06)	3.73 (3.11)	10.24/7.09)	60.93 (28.29)
	Median	1.03	0	9.95	4.1	9.62	61.98
	Range (min–max)	0–100	0–4.94	0–100	0–7.13	0.34–23	18.1–100

Note: All the reported values are percentage values.
Abbreviations: SD, standard deviation; TTB, tip-toe behavior; TTB1, TTB during running; TTB2, TTB during walking and running; TTB3, TTB during standing, walking, and running.

TTB subgroup, 6 (12%) subjects resulted in the TTB1 subgroup, 11 (22%) subjects resulted in the TTB2 subgroup, and 7 (14%) were in TTB3 subgroup.

Structured video-based coding of TTB: Static test results

At first glance, static test results in the study sample (Table 2) showed that the three possible modalities for support (i.e., bipodalic full foot support, monopodalic full foot support, and on the toes support) are expressed in the sample with high variability, suggesting that the subjects have very different responses to the tests. In particular, according to the static test results, the time percentage mean values on the toes were heterogeneous among the four groups ($p < 0.001$).

We found that TTB subjects expressed a significantly higher TTB when compared with NO-TTB subjects ($Z = -3.404$; $p = 0.001$). Moreover, the mean percentage time on toes was highest for TTB3 subgroup followed by TTB2 and then TTB1 subgroups. In fact, we found a significant difference between the following subgroups: NO-TTB versus TTB2, NO-TTB versus TTB3, TTB1 versus TTB3, and TTB2 versus TTB3 (see Table 3, Figure 2).

Structured video-based coding: Dynamic test results

Results of the dynamic test of the study sample (Table 2) showed that the two possible modalities for walking (i.e., toe steps or hell/flat foot contact) are expressed in the sample with high variability. This suggests that the subjects have very different responses to the tests.

According to the dynamic test, the mean percentage values of the steps on the toes were heterogeneous among the four groups ($p < 0.001$). We found that TTB subjects expressed a significantly higher mean percentage values of toe steps when compared with NO-TTB subjects ($Z = -5.453$; $p < 0.001$). Moreover, the mean percentage of steps on toes was highest for TTB3 subgroup followed by TTB2 and then TTB1 subgroups. In fact, a statistically significant difference between each of the four subgroups was observed (Table 3, Figure 3).

TABLE 3 Pair comparison between NO-TTB and the three TTB subgroups.

	Static test		Dynamic test	
Mann–Whitney test	***Z*** **score**	***p*-value**	***Z*** **score**	***p*-value**
NO-TTB vs. TTB1	−0.217	$p = 0.869$	−2.823	$p = 0.006$*
NO-TTB vs. TTB2	−2.577	$p = 0.015$*	−4.591	$p < 0.001$*
NO-TTB vs. TTB3	−4.211	$p < 0.001$*	−4.377	$p < 0.001$*
TTB1 vs. TTB2	−0.927	$p = 0.425$	−2.010	$p = 0.044$*
TTB1 vs. TTB3	−2.752	$p = 0.012$*	−3.000	$p = 0.005$*
TTB2 vs. TTB3	−3.496	$p < 0.001$*	−3.396	$p = 0.002$*

Abbreviation: TTB, tip-toe behavior.
*Statistically significant comparison; *p*-values were adjusted for multiple comparisons.

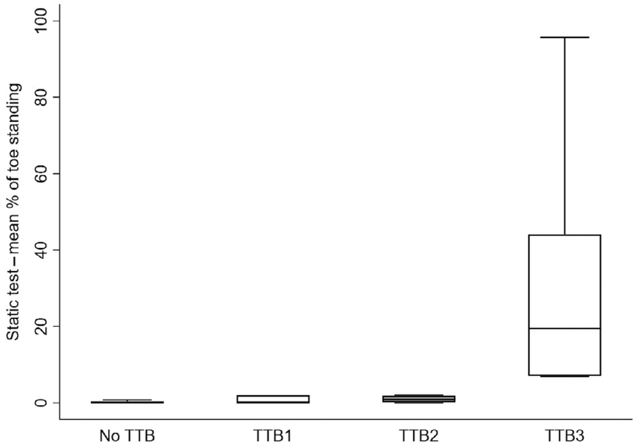

FIGURE 2 Static test: box plot of percentage of time maintained on the toes in the four subgroups. TTB, tip-toe behavior.

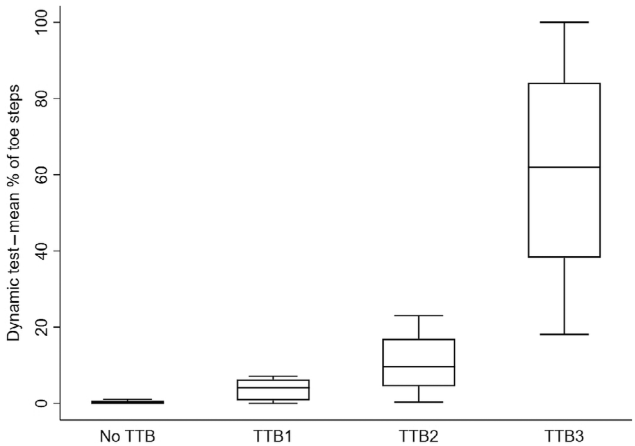

FIGURE 3 Dynamic test: box plot of percentage of toe steps in the four subgroups. TTB, tip-toe behavior.

Multivariate analysis

We found that the only factor in a statistically significant relationship with the mean percent of toe standing in the static test was the ownership to the TTB subgroups. Other variables such as age ($p = 0.19$), ADOS CCS score ($p = 0.31$), ID ($p = 0.68$), and gender ($p = 0.14$) were not significantly associated with the mean percent of toe standing in the static test. In the multivariable analysis for the dynamic test, the ownership to the TTB subgroups was confirmed as statistically significant ($p < 0.001$) after adjustment for the ID ($p = 0.030$). Other variables such as age ($p = 0.72$), ADOS CSS ($p = 0.32$) and gender ($p = 0.38$) were not significantly associated to mean percent of toe steps.

Structured video-based coding: Correlation between static and dynamic TTB test results

Because a secondary aim of this study was to assess the relation between mean time percentage values on the toes during the static test and the mean percentage number of the steps during the dynamic test, we performed a Spearman test, and we found a significant positive correlation ($r = 0.716$; $p < 0.001$) between the quantity of TTB in the static and the dynamic test.

DISCUSSION

Participants and methods

In this study, the proposed structured video-based coding tests effectively quantified TTB in a cohort of individuals with moderate–severe ASD and severe–profound ID. Only 13% of subjects were excluded due to their inability to complete one out of three trials for one of the designated quantitative tests. This suggests that the proposed assessment methodology remains feasible even for a sample with such specific clinical characteristics.

We used a video-recording approach. Several studies endorse the use of video recording for gait assessment in both orthopedic and neurological conditions (Brunnekreef et al., 2005; Damsted et al., 2015; Dickens & Smith, 2006; Wren et al., 2005). Furthermore, gait video recording has previously been utilized as an outcome measure to evaluate the efficacy of interventions in individuals with ASD and TTB, as evidenced by four case studies (Hodges et al., 2018; Marcus et al., 2010; Persicke et al., 2014; Wilder et al., 2020). The video-recording approach allows both reviewing the tasks and using a slow-motion technique. This increases the quality of the TTB assessment and permits a blind assessment. Another advantage of the video-recording system is that it does not require expensive technologies. Thus, it seems realistic to think that it can be used in a clinical setting and in low-resource context.

Qualitative assessment

The prevalence of TTB in our study sample is higher compared with previous studies (Barrow et al., 2011; Ming et al., 2007). A plausible explanation could be the absence of individuals with high-functioning ASD and the relatively high proportion of individuals with severe ID. This hypothesis is supported by the findings of Barrow et al. (2011) since they found a significantly lower prevalence of TW in Asperger syndrome subjects. Another contributing factor might be that our study's calculated prevalence rate includes individuals who exhibit TTB exclusively during running (six subjects); omitting these six subjects from the analysis would likely result in a lower prevalence percentage in our study.

Quantitative assessment

The paper aimed to describe the methods employed to assess TTB in individuals with ASD (Valagussa et al., 2018) and conducted a systematic review of literature up until May 2017. Remarkably, among the included studies, only one quantified TTB by calculating the mean percentage of flat-footed steps using a video-recording approach (Persicke et al., 2014). In the following years, using the same approach, other studies quantified TTB but they calculated the mean percentage of toe steps (Hodges et al., 2018; Wilder et al., 2020, 2022). Recently, Barkocy et al. (2021) quantified TTB using a gait analysis approach. All these last four studies, aiming to evaluate the effectiveness of a TTB treatment in individual with ASD, had a case-report/case-series study design, involving a total of 12 individuals with ASD with an age range between 4 and 12 years. Interestingly, none of the four studies reported information about the presence or not of ID and only one (Persicke et al., 2014) described the ASD severity (i.e., mild to moderate). Since the lack of these data and the variability of clinical manifestation that characterizes ASD, it is difficult to know if the proposed TTB assessment and treatment could be clinically performed also with individuals with severe ASD and ID.

Some studies suggest that shoes could decrease TTB, thus influencing the results of a quantitative test (Ming et al., 2007; Weber, 1978). To our knowledge, this is the first study that uses a structured video-based coding test to quantify TTB without shoes (albeit with socks) and using repeated trials. Moreover, this is the first study that proposes a standardized test to quantify TTB during the standing condition.

The proposed structured video-based coding test enabled us to assess the quantity at which TTB is manifested during standing and walking. As expected, in the individuals of the NO-TTB subgroup, TTB mean percentage values were found very close to zero in both the static and the dynamic tests. Conversely, in the static test, we found that individuals belonging to the TTB3 subgroup (i.e., TTB during standing, walking, and running) manifested TTB with a statistically significant higher mean time percentage than those belonging to the TTB2 subgroup (i.e., TTB during walking and running), TTB1 subgroup (i.e., TTB during running), and NO-TTB subgroup. Similarly, in the dynamic test, the TTB3 subgroup presented a higher mean percentage of steps on the toes than those belonging to the TTB2 subgroup, TTB1 subgroup, and NO-TTB subgroup. This supports the idea that individuals who manifest TTB during standing, walking, and running are likely to have a more severe TTB quantity compared with those in the other three subgroups.

The persistence of TTB in individuals with ASD is thought to be related to the Achilles' tendon shortening, potentially resulting in reduced ankle ROM in dorsiflexion. However, the reasons behind why some individuals develop TTB while others do not remain not entirely understood. In a previous study, Valagussa et al. (2020) evaluated the relationship between the three qualitative TTB clinical patterns of increasing severity and both the soleus and the gastrocnemius muscle lengths. They found that individuals who manifested TTB during standing, walking, and running (TTB3 subgroup) demonstrated a shortening of both soleus and gastrocnemius muscles compared with the individuals who manifested TTB only during running and NO-TTB subjects. Thus, it could be interesting to investigate in further studies the relationship between the quantity of TTB during static and dynamic tests and both soleus and gastrocnemius muscle lengths in individuals with ASD.

An additional potential factor contributing to the presence and/or persistence of TTB could be sensory issues. In a pilot study, Valagussa et al. (2022) examined sensory characteristics in a sample of ASD children and adolescents with or without TTB. Their findings indicated that individuals with ASD and TTB more frequently displayed the distinctive pattern of "under responsive/seeks sensation" compared with individuals with ASD and no TTB. This suggests that TTB might be a mechanism to enhance peripheral sensations in individuals with ASD.

During clinical practice, it is possible to observe that the variability of TTB individuals with ASD is very high, but to our knowledge, this clinical observation was never confirmed by studies where TTB was quantitatively measured. The results of the present study confirm the clinical observation.

The present study results highlight a significant positive correlation between the quantity of TTB in the static and the dynamic test. This suggests the clinical utility to check for the presence and the quantity of TTB during standing and not only during walking.

Study limitation and perspective

Our study presents some limitations. Despite the required time for administration of the test is short (i.e., roughly 15 min), two operators are requested for the procedure. Secondly, video recording may be considered as a subjective measure even if the use of video recording for gait assessment is supported by literature as previously described. A potential possible evolution of our assessment could be to quantify TTB using an instrumental approach (e.g., using wearable sensors that quantified foot–ground interaction); this could be also a way to confirm the validity of the results obtained with a video-recording approach. Lastly, our study considered a group of individuals affected by ASD and severe—profound ID. This decreases the generalizability of the results. On the other hand, this could be considered also a strength because of the specificity of the study sample. Moreover, even if we excluded from the study eight individuals with ASD because they could not conclude one out of three trials of one out of two planned quantitative tests, we also

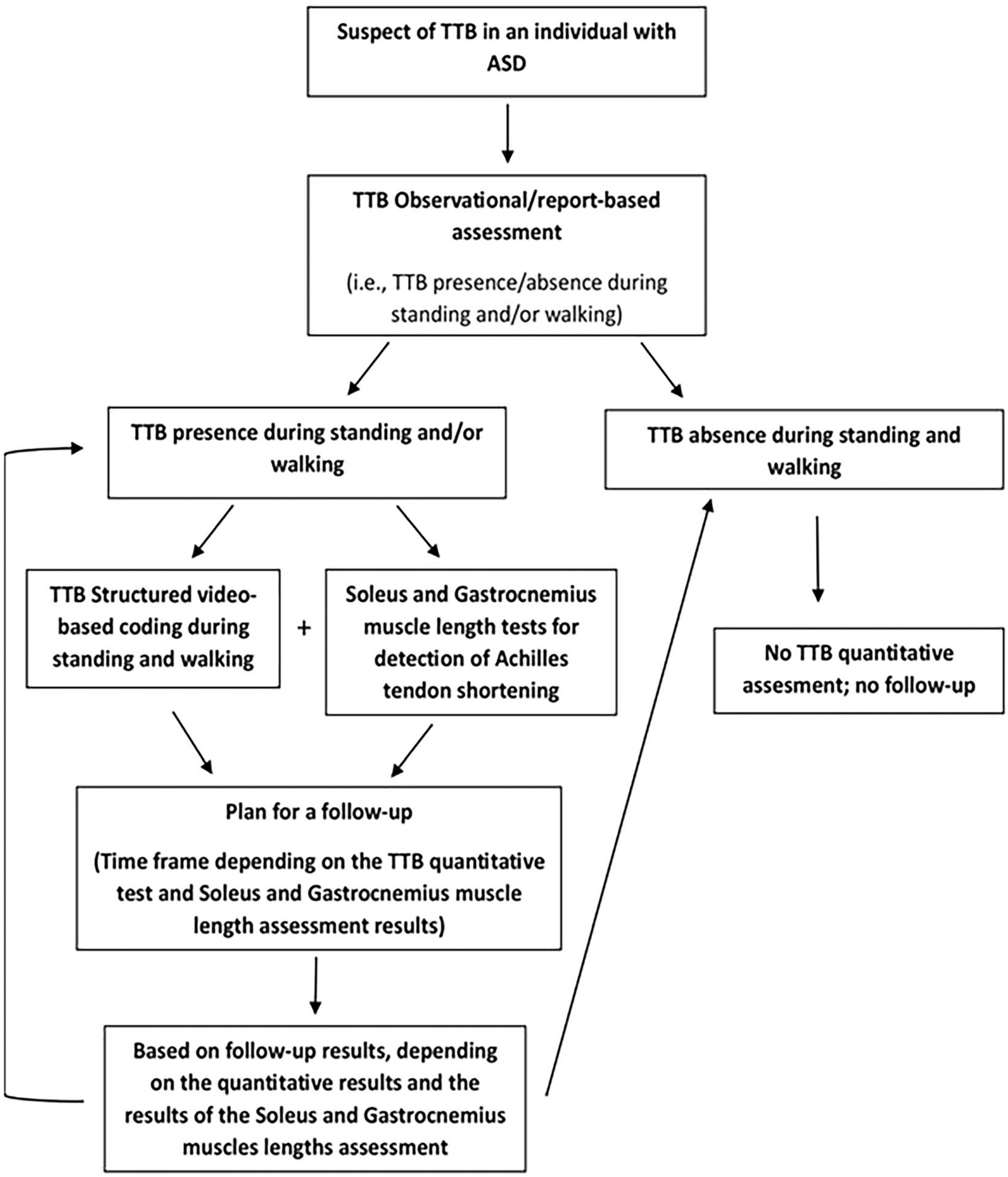

FIGURE 4 Flowchart of TTB assessment process. ASD, autism spectrum disorder; TTB, tip-toe behavior.

found the applicability of such an assessment also to individuals with ASD and severe to profound ID (i.e., 31 individuals with ASD and profound ID were able to conclude the quantitative assessment successfully). As such it is reasonable to hypothesize that the proposed assessment could be applied also to individuals with less severe ASD and without ID. Considering this perspective, it would be interesting to replicate this measurement in a similar sample.

In the future, conducting studies that employ both cross-sectional and prospective study designs to compare quantitative data related to TTB and Achilles' tendon length values could be highly interesting.

Clinical implications

Based on the empirical findings of our study, we propose a clinical assessment decision-making framework targeting individuals with ASD and TTB, as illustrated in Figure 4. This proposal is informed by a synthesis of both clinical observations and the quantitative assessments conducted within the scope of this research endeavor. To optimize the assessment process, we recommend commencing with qualitative evaluations. In instances where expeditious assessments are warranted, initiating the qualitative appraisal from a standing posture is advisable, as our analyses reveal that the presence of TTB during standing corresponds to its manifestation during walking and running as well. If TTB is not evident during the standing phase, our suggestion is to proceed with assessing its occurrence during walking. Conversely, if TTB is not detected during walking either, in accordance with prior research, it might be of limited clinical utility to evaluate TTB during running. This is due to the observation that TTB solely during running does not appear to correlate with Achilles tendon shortening, as demonstrated by Valagussa et al. (2020), thus potentially obviating the necessity for quantitative assessments. However, in instances where TTB is present during standing and/or walking, our recommendation is to incorporate quantitative assessment methods. Such an approach aids in quantifying the severity of TTB. Furthermore, conducting follow-up quantitative assessments could yield insights into the temporal trajectory of TTB. In addition to the foregoing, we propose the measurement of gastrocnemius and soleus muscle lengths, thereby facilitating an evaluation of the degree to which TTB presence influences Achilles tendon shortening. This multifaceted strategy could facilitate the decision-making process regarding the need for intervention in individuals presenting with ASD and TTB.

CONCLUSION

We proposed structured video-based coding tests to quantify TTB during both standing and walking in a sample of 50 individuals with ASD and severe ID. Our findings indicated that individuals with ASD who exhibit TTB during standing, walking, and running display higher levels of TTB than subjects in the other three subgroups during both standing and walking tests. Additionally, we observed that a higher TTB quantity in the static test was correlated with a higher TTB quantity in the dynamic test. Given these findings, the structured video-based coding approach we employed could be regarded as a valuable tool for clinical practice, facilitating the assessment, and monitoring of TTB in individuals with ASD and TTB. Moreover, this method could contribute to understanding the developmental trajectory of TTB in children. The resulting trajectory has the potential to guide clinicians in making informed decisions about whether or not to intervene. Furthermore, this approach could serve as an outcome measurement tool to evaluate the effectiveness of treatments aimed at reducing TTB in individuals with ASD. Further research is warranted to confirm the results.

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Our appreciation is for all persons with ASD and their families, who have taught us so much.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

Ethical approval was obtained from the local IRB Insubria's Ethics Committee (n° 185/2019). The study was conducted in line with principles of the Declaration of Helsinki.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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