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Early identification of toe walking gait in preschool children

- *Development and application of a quasi-automated video screening procedure*

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Abstract

Purpose **-** To develop and test the application of a quasi-automated screening procedure identifying probable toe walking in a large population of preschool children.

Methods **-** The proposed screening procedure was designed to identify children exhibiting signs of toe walking in a previously recruited cohort of preschool children (MiPS cohort). The procedure combines parent observation (step 1), objective parameters of foot contact during gait by an automated screening of 3-D video recordings (step 2), and clinical video screening of the children identified in step 1 and/or 2 (step 3).

Findings **–** From 879 children, gait trials were obtained from 87% (n=766). Step 1 (parent observation) identified 34 children with potential toe walking, step 2 (automated screening) 122. Fourteen were identified in both step 1 and 2. Thus, 142 children were selected for step 3 (clinical video screening), from which 41 children were classified as showing symmetric signs of toe walking, and five children were identified with asymmetrical signs of toe walking. Of the 41, five had been identified by step 1 only, 32 by step 2 only and four by both steps.

Interpretation **-** Application of a quasi-automated screening algorithm was feasible and may assist in early detection of toe walking. Disagreements found between parent reported toe walking and video screening, indicate added value in quasi-automated video screening. However, thresholds of heel lift and clinical criteria of toe walking in the algorithm and video screening need to be addressed and validated to confidently identify toe walking gait.

Keywords:

Toe walking

Gait

Video screening

Feasibility

Preschool children

1. Introduction

Toe walking is not uncommon in the immature gait patterns of children [1]. If a child continues to walk on their toes beyond the age of three, possible neurological causes should be examined[2]. Toe walking is typically associated with conditions such as cerebral palsy, muscular dystrophy and autism spectrum disorders[3]. But 2-5% of all children continue to walk on their toes with no apparent neuropsychiatric, neurodevelopmental or orthopaedic cause and are diagnosed with idiopathic toe walking (ITW) [4,5]. Prolonged toe walking may lead to structural changes such as shortening of the Achilles tendon or an underdeveloped calcaneus [6].

In clinical treatment, the prevailing assumption is that ITW often spontaneously dissolve or improve over time, although the natural history or progression of ITW is still unclear [7–9]. This assumption has fostered a clinical tradition of watchful waiting and reassurance [7,10]. Also, identifying ITW in early childhood may not be addressed in the health care system, and consequently, the prevalence of ITW among children is uncertain.

As toe walking gait, even apparent idiopathic, can be associated with several different impairments[11,12], focused early intervention addressing the underlying cause may yield positive results. In order to establish a practice of focused early intervention, it is important to identify and diagnose the children at an early age and describe potential associated underlying neurodevelopmental characteristics[3].

In children with ITW gait observations are often variable[13] making detection difficult. Some methods utilizing inertial sensors [14,15] or pressure-sensitive insoles[16] have been developed for toe walking detection, and 3-D gait analysis [17,18] plus pedographic measurements[19] are considered the gold standards for classification of toe walking. These methods can, however, be difficult to apply as a screening procedure of larger groups of children, due to feasibility issues and extensive equipment requirements. Alternatively, parent reported toe walking has commonly been applied, but this method assumes a parental insight in typical gait patterns and gait development which may not be present.

The present study represents the initial development of a novel quasi-automated screening procedure (QAS) combining parent reported toe walking, semi-automated video analysis, and visual clinical video confirmation. The long-term perspective is to develop an automated application to identify toe walking gait confidently in large cohorts of children. The specific aim of the present study is to explore the practical feasibility and describe added value and limitations of the QAS to identify probable toe walking gait in preschool children as an alternative to parent reported toe walking alone.

2. Methods

2.1 Setting and participants

This study was based on data extracted from the Motor skills in Preschools cohort (MiPS)[20] which is approved by the scientific ethical committees of Region Southern Denmark (Project-ID: 5-20150178).

All children born between 2011-2013 and attending one of 32 public preschools in the Municipality of Svendborg, Denmark had been invited in August 2016 to participate in the MiPS cohort. Prior to participation, parents received written information about the project, and written consent by a parent or legal guardian was obtained [20]. At the time of the current study, 879 children had been registered in the cohort and were included in the present analysis. A detailed protocol of the MiPS cohort has been published elsewhere[20]. The following data was extracted from the MiPS cohort: Parent questionnaires about children's'

toe walking and 3-D video recordings of gait. All data for this analysis was collected from March to May 2017.

The proposed QAS was a three step procedure designed to identify children exhibiting signs of toe walking by combining *Parent observation* (step 1), *Objective parameters of foot contact* during gait by 3-D video recordings (step 2) followed by *Visual clinical confirmation* of those children identified in step 1 and/or 2.

2.2 Clinical and biomechanical signs of Toe walking

The definition of toe walking used in the QAS procedure was based upon common clinical and biomechanical signs of toe walking, condensed into two primary indications of toe walking: i) absence of 1st rocker (heel strike) and ii) presence of early third ankle rocker/reversed 2nd rocker (Table 1) [13,17].

These two primary biomechanical signs formed the basis of the objective parameters extracted from gait during 3-D video recordings (step 2) and visual clinical confirmation of the video recordings (step 3).

For visual clinical confirmation (step 3), additional common toe walking characteristics from the literature were introduced as secondary indications to support an overall assessment of the child's gait pattern. The secondary indications were presence of i) peak knee extension at foot contact (FC) (Fig. 1), ii) asymmetries between sides during gait i.e. only toe walking on one leg, and finally iii) a bouncing gait i.e. larger vertical displacement of the center of mass [6,21–25]. These were descriptive and only relevant when at least one primary indication was observed.

Primary Indications	
Absence of 1st rocker	Absence of heel strike and/or ankle plantarflexion immediately after FC.
Early heel lift with early 3rd rocker (reversed 2nd rocker)	Interrupted ankle dorsiflexion and switch to plantarflexion within the first 25% of the stride cycle (before the thighs are parallel)
Secondary Indications	
Peak knee extension at Foot Contact	Full extension of the knee at FC
Side-to-Side symmetry	No limp or unilateral presence of the primary criteria
Bouncing Gait	Vertically oriented movement in mid-stance.

Table 1 – Descriptions of the formulated indications of toe walking on which the QAS step 2 and step 3 are based.

2.3 Data collection and variable definitions

2.3.1 Parent observation (step 1)

A parent toe walking questionnaire was administered to the parents by email as part of multiple questionnaires examining the child's well-being. Parents were asked if their child walked on their toes (*yes/no*) and if '*yes*' was asked how much time was spent walking on their toes (*Almost never, <25% of the time, 25-50% of the time, 50-75% of the time, 75-100% of the time*).

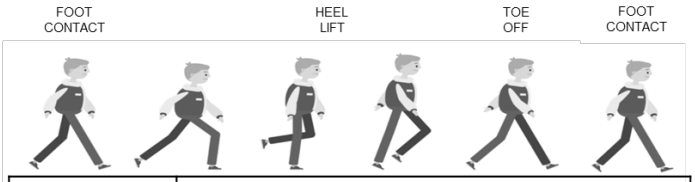
All children reported to toe walk either <25, 25-50, 50-75 or 75-100% of the time were selected for clinical video screening (step 3).

2.3.2 Automated Video Screening (step 2)

2.3.2.1 Data capture

The participating children were video-recorded as they walked back and forth on a 5-meter level walkway. The trials were recorded at 50Hz using eight synchronized Blackfly 808x608 digital cameras mounted on tripods in an oval area (5m. x 6m.). The Captury Live markerless system software (The Captury GmbH, Saarbrücken, Germany) was used to annotate gait events manually.

The children were instructed to walk to the end of the walkway and back by asking them to collect a teddy bear and return it. This instruction was to divert the child's focus from being observed (i.e. attention bias), as children walking on their toes are able to correct their gait deliberately [13,26,27]. One or two trials were used in the analysis. Each video recording was manually marked by the assessors in the Captury Live software (version 1.0.79) for the following gait events: Foot contact on either heel, flat foot or Toe as well as Heel Lift (HL) and Toe Off (Fig. 1).



FOOT CONTACT	The first frame with observed contact between foot and surface. Foot contact was marked as either; Heel, Flat foot of Toe.
HEEL LIFT	The first frame with observed lift of the heel.
TOE OFF	The last frame with observed contact between foot and surface.

Fig 1

Figure 1 – Gait cycle definitions: Illustration and definition of the gait events marked on the video trials in step 2 (automated video screening) and for clinical confirmation in step 3.

2.3.2.2 Automated identification of toe walking signs.

The main analysis was performed with a custom-made script (R, version 3.3.2) to identify all children marked with Foot Contact on flat foot or toe, representing a possible absence of 1st rocker in at least one stride. And to identify all children with early heel lift, indicating a reversed 2nd rocker/early 3rd rocker, in at least one stride. Timing of heel lift was calculated as the time from foot contact to heel lift relative to total stance phase for all stride cycles (Heel lift% = (heel lift frame – foot contact frame) / (toe off frame – foot contact frame)) (Fig. 1). Early heel lift was arbitrarily defined as 2 SD below the average heel Lift% of the entire MiPS cohort (mean heel lift% - 2SD).

All children identified with either foot contact on toe or flat foot and/or early heel Lift were selected for clinical video screening (step 3).

156 **2.3.3 Clinical video confirmation and classification (step 3)**

157 A clinical video screening served as a visual confirmation of the gait pattern of all children identified in step
158 1 and/or step 2. Three clinical researchers with 2-15 years of experience in clinical gait analysis were
159 selected to review the video recordings used in step 2. The clinicians were allowed to switch between camera
160 angles and use slow motion for the review. In a consensus session, the children were visually evaluated
161 based on the previously defined primary indications of toe walking (Table 1). All three clinicians were
162 blinded to why the children had been identified for the clinical video screening in step 1 and/or step 2.

163 Children exhibiting no signs of toe walking according to step 3 were classified as *typical gait* regardless of
164 results from step 1 and 2. Children exhibiting either early heel lift or foot strike with or without one or more
165 secondary indications according to step 3 were classified as *one primary indication* regardless of result from
166 step 1 and 2. Children exhibiting both early heel lift or foot strike with or without one or more secondary
167 indications according to step 3 were classified as *two primary indications* regardless of result from step 1 and
168 2. (Fig. 2).

169

170 **2.3.4 Control for false negatives**

171 Children with no indications of toe walking according to step 1 and 2 were classified as *typical gait* and not
172 reviewed in step 3**.** But to investigate the possibility of false negatives, a random sample of one hundred
173 children was selected from the group of children not identified in either of the initial two steps of the QAS
174 classifications. The gait trials of these children were reviewed by the same three clinical researchers in a
175 separate post hoc clinical video screening.

176

177 **2.4 Data summation and presentation**

178 Practical feasibility was examined by evaluating the time consumption for instruction, calibration, and
179 recording of the full gait trials and subsequently marking events on the trials. Furthermore, the time used for
180 clinical video confirmation (step 3) was evaluated.

181 Detailed results of the three steps in the QAS and the final classification are presented as clinical
182 observations. Results should therefore be read as descriptive tendencies, not statistically established [28].

183

184 **3. Results**

185 **3.1 Participants**

186 In total, 879 children were registered in the MiPS cohort with a mean age of 4.7±0.9 (mean±sd) years.
187 Response to the parent reported toe walking (step 1) was registered for 631 children (72%). Gait trials (step
188 2) were obtained of 766 (87%). Data from both step 1 and step 2 was available for 567 children (65%).
189 Children with data on toe walking from either parent reported toe walking and/or gait trials was included in
190 the study (830 children, 94%) (Fig. 2).

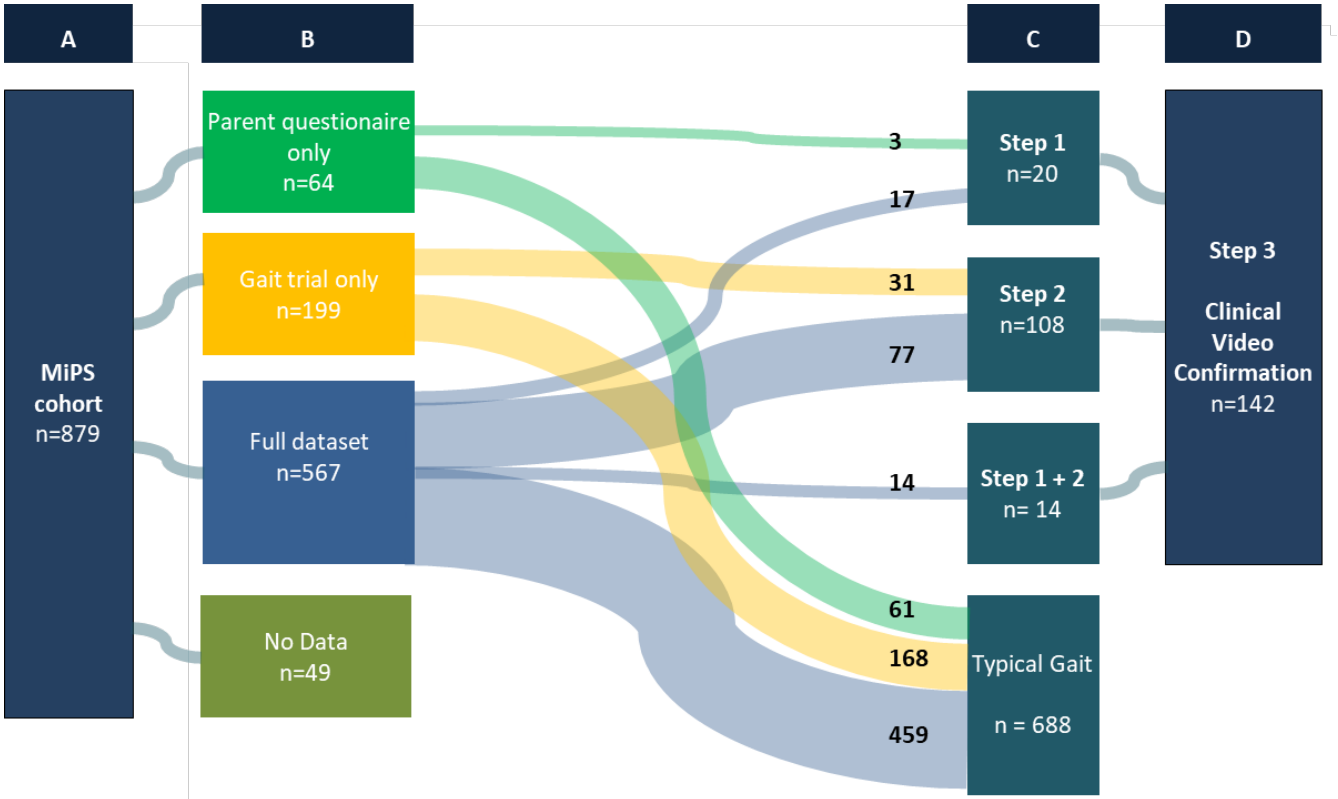


Fig. 2

Figure 2 – Flow diagram of study participants: Illustrating the available datasets for the children included in the QAS from the original cohort. Column A illustrates the full original MiPS cohort. Column B illustrates the available datasets for the QAS study. Column C illustrates the indications from step 1 (parent observation) and step 2 (automated video screening) from which the children reviewed in step 3 (column D) were selected. Children with no indication in step 1 or 2 were classified as typical gait, and not reviewed further. Detailed results from step 1 and 2 on the children reviewed in Step 3 and their final classification are illustrated in Fig. 3 in paragraph 3.5.

3.2 Practical feasibility

Time consumption for instruction, calibration, recording of the full movement trial, and marking events on the gait trials were 15 min. for each child.

722 children and 44 children completed two and one gait trials, respectively. The clinical video confirmation of 142 children (step 3) was estimated to one days' work for three clinicians.

3.3 QAS - Parent observation (step 1)

A total of 45 children were reported to toe walk by their parents. Eleven of these were reported as "*almost never*" and identified as typical gait (no sign of toe walking). One child was reported to toe walk, but with no report of time walking on the toes. This child was included in step 3. Finally, 34 children were identified with signs of toe walking according to step 1.

Twenty of the 34 children with signs of toe walking were identified and selected for clinical video confirmation (step 3) based on parent observation alone (fig 2). All but one of these had been reported to toe walk less than 25%. The exception was the one child with no report of time spent on toes.

Fourteen children were identified both by parent observation (step 1) and automated screening (step 2) (Fig. 3). Three of these were reported to toe walk 75-100% of the time, five were reported to toe walk 25-50% of the time and six were reported to toe walk less than 25% of the time.

3.4 Automated video screening (step 2)

One-hundred-and-eight children were identified with signs of toe walking based on the automated video screening only and 77 of these had full datasets available (both step 1 and step 2) (Fig. 2). Thirteen of the 108 children were identified with both early heel lift and divergent foot contract.

Three of the previously mentioned 14 children identified in both step 1 and 2 had been selected based upon both early heel lift and divergent foot contact (Fig. 3).

3.5 QAS - Clinical Video Screening (step 3)

Of the 142 children viewed in the clinical video screening (step 3) of the QAS, 46 children were classified with either one or two primary indications of toe walking (Fig. 3). Five of these with asymmetrical gait pattern.

A total of four were classified with two primary indications of toe walking. Three of these had also been selected with both early heel lift and divergent foot contact in the automated video screening (step 2).

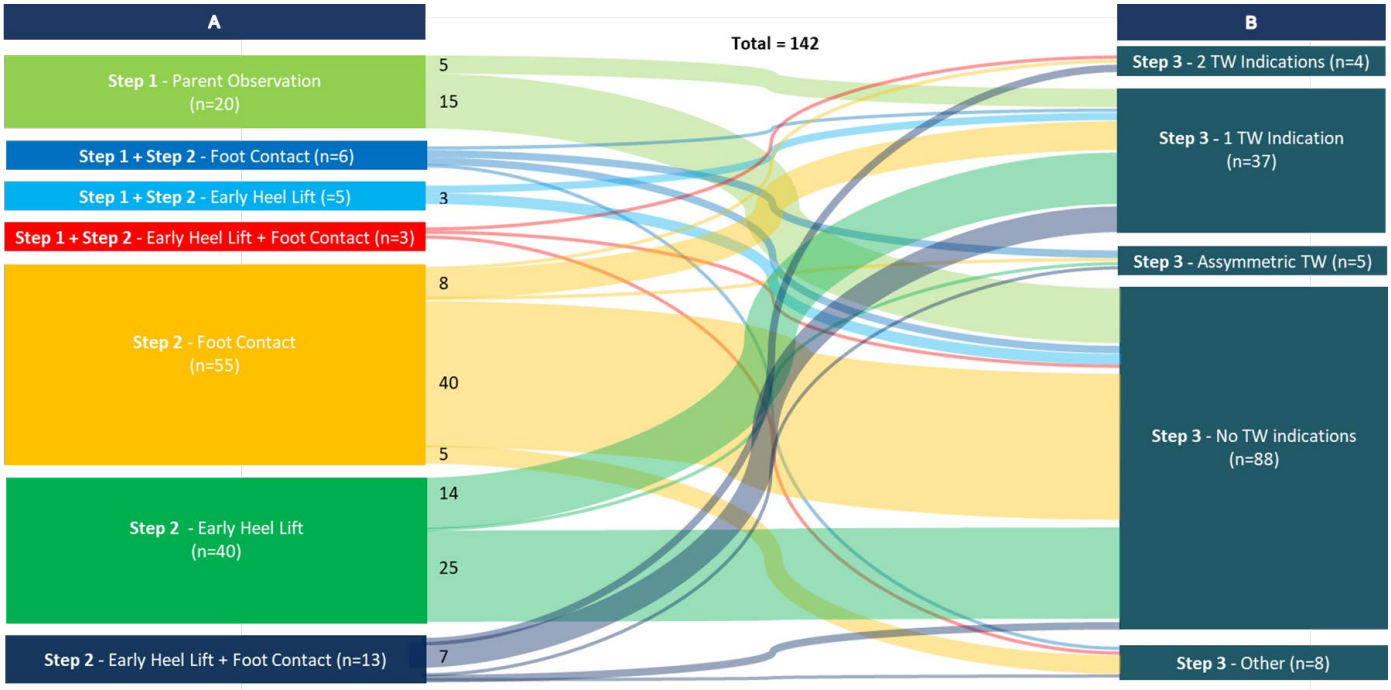


Fig. 3

Figure 3 – QAS final results: Column A illustrates the detailed results from step 1 (parent observation) and 2 (automated video screening) on the 142 children reviewed in Step 3 (clinical video confirmation). Column B illustrates the final classification of the children in column A. (TW=Toe walking)

Thirty-seven children were classified with one primary indication of toe walking in step 3. Seven with lack of 1^{rst} rocker as the primary indication, and thirty with Early heel lift with reversed 2nd rocker/early 3rd rocker. The largest proportion of these had been selected based on step 2 (Fig. 3). Eight out of 37 children classified with one primary indication of toe walking in step 3 were reported to toe walk by their parents (step 1).

There was a 74% risk of a child being classified in step 3 with indication of toe walking or other deviation in the QAS, if the child had been selected with at least two signs from steps 1 and 2 *(parent observation (step 1), early heel lift (step 2) and divergent foot contact (step 2).* Contrary, there was a 70% chance of being classified with no toe walking indications or other gait deviations, if the child had been selected with only one sign of toe walking according to step 1 and 2 (*parent observation or early heel lift or divergent foot contact*) (Fig.3).

From the random sample of 100 control children selected for the post hoc clinical video screening for false positive testing, one child was assessed as probable toe walker.

3.6 Added value of the QAS

According to our final QAS classification, the prevalence of toe walking was 6% of all children with recorded gait trials (Fig. 2). According to parent observation (step 1), the prevalence of children with signs of toe walking was 5% of responses to the toe walking questionnaire.

All but three children with parent reported toe walking had also been identified by the automated video screening. These three exceptions were reported to toe walk less than 25% of the time by the parents. Fourteen of the children classified with sign of toe walking according to the QAS final classification had no response registered for the parent observed toe walking. Ultimately, 54 children were identified with sign of toe walking or other gait deviation (Fig. 3). Forty-nine (91%) of these would have been identified solely by automated video screening (step 2). Thirteen would have been identified solely by parent observation (step 1).

Of the four children classified with two toe walking indications in step 3 (Table 2), all were identified by automated video screening (step 2), while only one of the four was identified by the parent reports. Similarly, five children were identified with one or two asymmetrical toe walking indications in step 3 (Fig. 3). All were identified in step 2, but only one in step 1.

QAS – 2 Primary Indications				
Case	**TW1**	**TW2**	**TW3**	**TW4**
Parent observation (step 1)				
Parent observed Toe Walking	<25%	No response	No response	No response
Automated Video Screening (step 2)				
FCF			25%	75%
FCT	100%	100%		
Early HL	100%	100%	50%	

Table 2 - This table unfolds the QAS results of the children classified with two indications of toe walking. The result of step 2 is reported as the percentage of the step deviation present amongst available steps on the video recordings. If nothing is noted, no markings of foot contact on flat foot or toe were made on the video recording and no early heel lift was detected.

A total of eight children were observed by their parents to toe walk more than 25% of the time in step 1. Five of these were also identified in step 2. Three were classified in step 3 with one or two primary indications of toe walking (Table 3), and one was classified as OTHER possible gait deviation (foot drop).

Parent Observed toe walking > 25%								
	TW5	**TW6**	**TW7**	**TW8**	***TW9***	***TW10***	***TW11***	***TW12***
Parent observed Toe walking	75-100%	75-100%	75-100%	25-50%	25-50%	25-50%	25-50%	25-50%
Automated video screening (Step 2)								
Foot Contact Flat		100%						12,5%
Foot Contact Toe			50%					
Early Heel lift	25%			25%				12,5%
Clinical video screening (Step 3)								
Lack of 1st rocker		+	+					
Early heel lift with reversed 2nd rocker/early 3rd rocker			+	+				

Table 3 - This table unfolds the QAS result of the children reported to toe walk more than 25% of the time.
The result of step 2 is reported as the percentage of the step deviation present amongst available steps on the video recordings. If nothing is noted, no markings of foot contact on flat foot or toe were made on the video recording and no early heel lift was detected.

4. Discussion

The present study aimed to develop a quasi-automated screening procedure (QAS) identifying probable toe walkers in a large population of preschool children. The novel screening procedure demonstrated that 5,5% in the cohort of preschool children had signs of toe walking. Furthermore, a divergence between toe walking reported by parents in step 1 and the remaining steps of the QAS was observed, suggesting that the additional steps contained in the full QAS adds value in the early identification of probable toe walking. In perspective, a quasi-automated screening procedure may assist in detecting toe walking gait in early childhood to guide early assessment and diagnosis and facilitate early intervention. However, validation of pathological toe walking in the children identified in the QAS and follow up of underlying causes is required before its clinical application and value can be confirmed.

4.1 Feasibility of the proposed QAS

Overall, the usability of the quasi-automated screening in large populations of children was good. Data in the form of video recorded gait trial and/or parent reported toe walking were obtained for 94% of children in the MiPS cohort (87% with gait trial). A high response rate of 72% on the parent reported toe walking was demonstrated. Thus, with the QAS we managed to obtain a very large dataset for further evaluation and condensing the population worth reviewing further for toe walking. In a research or clinical setting, the process of the QAS could reduce the further diagnostic process. Using the results conservatively, step 3 of the QAS could be expanded with the toe walking tool developed by Williams and colleagues to explore underlying causes [3].

The time consumption to record and mark events on each gait trial was completed along with several other movement tasks in 15 minutes. The clinical confirmation took 7 hours and therefore, the testing is the most time-consuming part of the process, but if included as part of a larger test-package in a cohort, the time consumption seems acceptable. Comparing to the golden standard methods of 3-D gait analysis or pedography[17,19] that can take up to 90 minutes without data processing and can be challenging to perform on young children, the QAS is less time consuming per child, and can be performed in a setting that is more familiar to the child. Also, considering the known variability of especially idiopathic toe walking [13], false negatives could also be an issue with 3-D gait analysis or pedography applied in a large cohort.

4.2 Added value of the QAS

The added value of combining multiple sources of data, and not only parent reported observations, is a strong argument for implementing the QAS as part of testing children for toe walking. Only 11 children with parent reported toe walking were ultimately classified with one or two primary indications of toe walking opposed to 41 based upon the automated screening procedure (step 2). Consequently, if only identifying by parent observation, 75% of the children we identified in the QAS could potentially be false negative. Furthermore, 23 children observed by parents to toe walk, could possibly be false positives. However, we cannot conclude that the automated screening procedure integrated in QAS *per see* improved the clinical accuracy of the toe walking diagnosis as five children with substantial parent observed toe walking, would be classified as typical gait if only relying on screening of video recordings. Furthermore, it is worth stating that the QAS is

based upon observational signs of toe walking and does not classify the severity of toe walking from kinematic and kinetic objective measures as is done by Alverez et al (2006)[17].

Our approach of combining multiple sources of data for the early identification of toe walking gait addresses a common challenge of missing data in larger cohorts. Especially with surveys, obtaining an acceptable response rate can be challenging. In the MiPS cohort long term follow-up on musculoskeletal pain in childhood and orthopedic treatments is conducted [20]. Information on whether a child has been walking on their toes or not would be important when analysing potential causes for musculoskeletal pain.

Based on these observations, combining the three steps in the QAS most likely adds value to the accuracy of identifying probable toe walkers in a large population of pre-school children.

4.3 Clinical perspectives of the QAS

The percentage of toe walking identified by parent observation was only 5% in our cohort, not far from the 5% classified with one or two symmetric indications of toe walking in step 3 (5,5% including asymmetric toe walking gait). Our results of prevalence of toe walking gait coincide well with a prevalence of 5% published in a Swedish cohort [29] detecting ITW by parent observation only. Caution should be made that no diagnostic examination has been made to the children identified in the QAS.

Despite similar level of prevalence as reported in the literature, the lack of full agreement between parent observed toe walking (step 1) and the final QAS classification, complicates valid prevalence rates of toe walking For that purpose, a study on criterion validity must be performed, which was not the purpose of the present study. However, the children in the MiPS cohort are followed up for musculoskeletal pain and orthopedic treatments in the future. Petitioning access to this follow-up data could be a valuable opportunity to clinically validate the QAS.

This study also suggests that some parents may be able to observe deviation from typical gait, but not differentiate between deviations. Out of three children that were reported to toe walk more than 75% of the time, one was classified as *other* gait deviation in the QAS, with probable foot drop, and one was classified as *asymmetrical* potentially indication a more severe gait deviation. Thus, it is possible that a QAS also can assist in identifying other early signs of neurologic and/or musculoskeletal issues, by adding clinical video confirmation.

4.4 Challenges and further development of the QAS

The initial investigations in the development of a quasi-automated screening algorithm revealed some issues to be addressed. Some disagreement was observed between the automated video screening (step 2) and the clinical confirmation (step 3) in terms of early heel lift. Early heel lift was arbitrarily defined as 2 SD below the average heel lift% of the entire MiPS cohort and determined on separate steps to ensure a bias towards more false positives than false negatives. It could be explored if the cut-off for early heel lift should be determined as the mean of all strides. This would exclude some children where one heel lift is clearly deviant from the rest and identify children with a consistent early heel lift. It should be noted that 74% of those children who exhibited two or three signs of toe walking according to step 1 and step 2 were classified with either toe walking indications or other deviations by the clinical researchers in step 3.

A challenge to be addressed in the further development of the QAS, is to avoid false negatives from step 3 amongst the children observed by their parents to toe walk (step 1). A solution could be longer and more gait trials to eliminate the potential bias of within-subject variation in toe walking pattern.

It should be noted that in most subjects who were classified with two primary indications or asymmetrical toe walking in QAS, almost full agreement was found between step 2 and the final classification both regarding early heel lift and divergent foot contact. This could indicate a better performance of the QAS on more severe toe walking.

4.5 Limitations

A limitation of the algorithm could be the quasi-automated character of the identification. The events in the gait trials were not marked by the same clinicians conducting the clinical video screening. However, this increases the usability and generalizability on larger populations, as dependency on few people would limit the implementation significantly due to the time consumption.

This evaluation of the QAS is as mentioned an early descriptive study to address challenges and possibilities of the method and therefore thorough validation is needed before possible implementation.

6. Conclusion

The application of a quasi-automated screening algorithm was feasible in a large population of preschool children and revealed concerns in relying on parent reported toe walking only. However, there is a need to address issues such as cut-offs in step 1 and 2 in the algorithm and to validate the prevalence of toe walking gait. In perspective, the QAS procedure as an addition to parental reporting may be able to assist in detecting children in a larger cohort who should be examined further for ITW and/or related musculoskeletal or neurologic disorders in early childhood and facilitate timely and effective interventions.

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