Critical Review:

Can pediatric idiopathic toe walking be associated with delays in language?

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This critical review examines the current research on the association between idiopathic toe walking and language delay. A completed literature search using computerized databases and examination of the retrieved articles yielded four studies. Overall, the literature review revealed an association between idiopathic toe walking and language delays, supporting idiopathic toe walking as a marker for developmental problems. Clinical implication and recommendations for future research are discussed.

Introduction

Idiopathic or persistent toe walking is a variation in gait that is characterized by a limited range of dorsiflexion, and bilateral ambulation from the initiation of independent walking (Sala, Shulman, Kennedy, Grant & Chu, 1999). Diagnosis of toe walking by an orthopedic surgeon or Physiotherapist is based on a physical examination of the characteristic walking pattern (Oetgen & Peden, 2012), and the exclusion of other neurodevelopmental motor-based disorders that could account for the effect on gait. This diagnosis of exclusion is given when neurodevelopmental disorders such as autism spectrum disorder (ASD), pervasive developmental disorder (PDD), spastic cerebral palsy, spinal dysraphism, myopathy and neuropathy have been ruled out (Shulman, Sala, Chu, McCaul, and Sandler, 1996). It is noteworthy that toe walking has long been reported with a high incidence in ASD and Asperger's (Barrow, Jaworski & Accardo, 2011). Interestingly, the incidence of toe walking in Asperger's is lower than that in other autism spectrum disorders (as defined in the DSM III-R; American Psychiatric Association, 1987), with the differentiating criterion for Asperger's being the absence of a language delay in the early years of development; however, toe walking can be found with both populations, indicating that persistent toe walking may be associated with more than ASD (Barrow, Jawirski & Accardo, 2011). In recent developmental pediatric literature, some research attention has focused on the association between persistent toe walking and the presence of language delays (Accardo and Whitman, 1989). Accardo et al. (1990) hypothesized that within a model of language delays and toe walking, autism represents only one extreme; therefore, children demonstrating toe walking, without the presence of developmental disorders, fit in this model and thus children have become an interest of current research.

This review will critically examine the literature on idiopathic toe walking in those children without a diagnosis of neurodevelopmental disorders as well as one study in which a range of comorbid diagnoses were documented in order to determine if idiopathic toe walking should be considered a 'red flag' for a developmental language assessment.

Objectives

The primary objective of this paper is to provide a critical review of the existing literature on the hypothesized association between idiopathic toe walking and delays in language. The secondary objective is to consider the extent to which idiopathic toe walking should be considered an indicator for assessment of language development.

Methods

Search Strategy

Computerized databases, including PubMed, Google Scholar and the Cochrane Library were searched using the following criteria, (idiopathic toe walking) AND ((developmental language) OR (language delay)). The search was limited to papers written in English and no restrictions on date of publication were used. Examination of the retrieved articles revealed additional studies for critical review.

Selection Criteria

Studies selected were required to examine the hypothesized association between idiopathic toe walking and language delays. No constraints were set on language assessment measures used or demographics.

Data Collection

Results of the literature search yielded four articles that are consistent with the aforementioned criteria. Of these

four articles, two are level 2b cohort studies and two are level 3 single group studies (Archibald, 2009).

Results

In a retrospective, cohort study, Accardo and Whitman (1989) investigated the relationship between idiopathic toe walking and developmental disorders, including language delay in 799 children (>18 months old) referred to a tertiary-level clinic for a multidisciplinary developmental assessment over a 6-year period, excluding children with a motor-based neuro-orthopedic diagnosis. Of the 799 children referred, 224 (28%) were toe walkers, as determined by parental report of ambulatory history or observation of toe walking during the child's visit to the clinic. Of relevance to the present review, the diagnosis of a language disorder was confirmed using standardized tests of receptive vocabulary and verbal intelligence. Based on clinical results from these and other tests, diagnostic groupings included autism, communication disorder, mental retardation and learning disability. Diagnoses were based on DSM-IIIR criteria for the first two groups, clinical judgment and IQ score for the mental retardation group, and an unspecified discrepancy model for the learning disability group. An appropriate ANOVA revealed significantly lower full IQ scores for children with than without toe walking regardless of disorder group. Appropriate odds ratio, chi-squared tests of association, and sensitivity and specificity analyses revealed significantly higher prevalence of toe walking in the diagnostic subgroups with more severe language delays (i.e., ASD=62.9%; communication disorders=40.2%; mental retardation=35.8%; learning disabilities= 20.0%).

The results of this study are limited to children referred for developmental diagnosis and thus cannot be generalized to the general pediatric population. Additional limitations to this study include the use of measures of intelligence and receptive vocabulary only to confirm clinical diagnosis of language disorders and the inclusion of categories of developmental delay that were not clearly defined. 'Presence' of toe walking was indicated by parental report and/or observation of toe walking by the researchers during the assessment and was not determined by a standardized assessment of gait. Additionally, duration of persistent toe walking was not operationally defined. Moreover, the researchers served as the primary investigators and the assessment measures were administered without blinding, possibly leading to diagnostic biases. Despite these limitations, this study was the first to investigate the possible association between toe walking and language delays and provides a suggestive level of evidence of the association.

Accardo, Morrow, Heaney, Whitman and Tomazic (1990) examined the association between toe walking and language delays in the general pediatric population. Included in this study were 163 children from the ambulatory pediatric clinic being seen for a routine well-child visit and who were found to have no neurological medical diagnosis. Language was assessed with the Clinical Linguistic and Auditory Milestones Scale (CLAMS; Capute, Shapiro, Wachtel, Gunther & Palmer, 1986) for children less than two years old, and with the Mecham Language Scale (Mecham, 1958) for those two years and older, and from these scores, a mean language quotient was calculated based on language and chronological age. Medical, academic and speech/language history was obtained through parental report. Toe walking history was confirmed by observation or parental report of toe walking for one month's duration, yielding 39 toe walkers in the study. Participants were divided into six groups given their toe-walking history and age. An appropriate ANOVA revealed a significantly lower mean language quotient for the toe walkers than for the non-toe walkers across all age groups. Odds ratios revealed significant associations between language delays and toe walking for the preschoolers and the school-aged participants, but not for infants. The specificity of toe walking and a low language score was 85%; however, the sensitivity was 32%, indicating that toe walking may identify those with a language impairment but absence of toe walking does not rule out a language impairment.

One strength of this study is the inclusion of participants from the general public as well as the operational definition of toe walking including its duration. However, despite these strengths, the validity of the gait and the language assessments is weakened, as they used outdated measures not commonly used in recent research in this area. Lastly, the researchers speculated that the data collected from the mean language scores may not be comparable across age groups as the sensitivity of the two assessments used to assess language abilities differ. Overall, this study provides a suggestive level of evidence that further supports the association between toe walking and lower language scores.

Engstrom, Van't Hooft and Tedroff (2012) investigated the association between idiopathic toe walking and neuropsychiatric symptoms, including language delay in a single group study with 51 children (31 boys; Mean age=9;1) referred for idiopathic toe walking to a pediatric orthopedic unit and compared the results to existing norms. In all cases, the toe walking had persisted for a minimum of one year prior to the study by parental report. Additional reports from parents

confirmed that none of the children were being followed for suspected intellectual disability or autism. Participants were evaluated by an orthopedic surgeon. Additional measures were completed by the pediatric neurologist and parents of which the language measures are the main focus here. All domains including three language domains (receptive and expressive language and communication) were measured using a clinical questionnaire with a 0 (does not apply), 1 (applies to some extent), 2 (definitely applies) point scale, with a high score indicating impairment. Results were analyzed by comparing the proportion of the group whose scores on the questionnaire were above the 90th percentile (the cut off indicating the presence of clinically apparent problems) or the 98th percentile (the cut off for the presence of major clinical problems). Results revealed that the group scored above the 98% percentile in language, indicating considerable difficulties. When questionnaire subdomains were considered, comparison of the confidence intervals to the normative group for receptive language, expressive language and communication did not show a difference relative to the normative group.

A strength of this study is the inclusion of a strict definition for the classification of toe walking, one that is consistent with the current orthopedic literature. Moreover, this study incorporated an historical age-matched normative group for comparison. However, this study is not without its limitations; there were more boys than girls included in the study, a detail that may have contributed to their overall results, as males are overrepresented with respect to neuropsychiatric problems. Also, no standardized language assessment measure was used, and results lacked clarity. Due to the limitations regarding the reliability of the language testing and the lack of clarity in the results, this study provides somewhat suggestive evidence for the association between idiopathic toe walking and language delays.

Shulman, Sala, Chu, McCaul and Sandler (1996) examined 13 children (Mean age: 3;9) referred to a pediatric orthopedic clinic in a prospective, descriptive single group study investigating the relationship between idiopathic toe walking and language delays. Children with suspected developmental problems such as autism were excluded. A complete multidisciplinary evaluation including neurology, developmental pediatrics, speech language pathology, occupational and physical therapy were done to rule out any underlying conditions as well as to fully assess the respective developmental areas. With regards to the language measures, a Speech Language Pathologist completed a motor speech exam, and 'gold' standard standardized tests of articulation, phonological analysis, and language. An informal assessment of pragmatics was also completed. A cut off of 6 months or below the expected score for age level was considered delayed. Moreover, a Physiotherapist evaluated ambulation through observation of gait. Data from the multidisciplinary assessments were analyzed for the proportion of children affected by the different variables.

Results specific to idiopathic toe walking and language abilities revealed that delays in receptive language were found in 7/13 (54%) children, with an average delay of 14 months; delays in expressive language were found in 8/13 (62%) children, with a mean delay of 12 months; 10/13 (77%) exhibited expressive or receptive delays; 5/13 (38%) had delays in both receptive and expressive language abilities. It should also be noted that delays in other developmental areas, such as fine motor, visuomotor and gross motor occurred. All children with delays in other areas other than language, also were found to have language delays.

Strengths of this study include a complete speech and language examination performed by a Speech Language Pathologist with specific and valid assessments and an examination of gait by a Physiotherapist and a Pediatric Orthopedists. Effort was made to rule out other possible contributing factors by excluding children with ASD or PDD from the sample. Nevertheless, other variables such as prenatal care and living were not controlled. The small sample size is representative of the area served by the clinic, but not of the general pediatric population, and no control group was used. Given the strengths and limitations, this study provides somewhat suggestive evidence of an association between idiopathic toe walking and language delay.

Discussion

Taken together, the results of the clinical appraisal on the four studies provide suggestive evidence in support of an association between idiopathic toe walking and language delays. However, no causal connections between toe walking and language development can be determined.

Common methodological limitations were found throughout the studies, which must be considered when considering the overall results of the research reviewed in this paper. Language assessments that are not considered 'gold' standard were completed in all studies except Shulman et al. (1996). A more sensitive and reliable measure of language may have provided more compelling and clinically significant evidence towards the association between idiopathic toe walking and

language delay. Moreover, another common limitation in all but one study, Shulman et al. (1996), lies in the unstandardized use of the term idiopathic toe walking, one which is inconsistent with the orthopedic and physiotherapy standard. A standardized protocol for diagnosing idiopathic toe walking combined with use of standardized language assessment measures could have provided a higher level of evidence in support of the hypothesized association.

With the exception of Accardo and Whitman (1989) and Shulman et al. (1996), the results from the reviewed literature can be generalized to the general pediatric population with reasonable confidence. This provides evidence for the consideration of reasonable inferences from the results to general clinical practice.

Additionally, only one of the studies reviewed, Accardo and Whitman (1989), examined toe walking among children with diagnosed disorders in which language delay is a feature, such as ASD. It could be argued that the toe walking in these populations may not be truly idiopathic, but rather associated with a developmental disorder. Nevertheless, it is noteworthy that this study focuses on individuals with language delay of a range of severity, some of which are not associated with a known neurological disorder.

Another common theme throughout the review literature is the diagnosis of language impairments and language-based learning disabilities strictly based on low language scores; however, an accurate description of language abilities may require more than low language scores and is best done through a complete, 'gold' standard language assessment.

The above considerations should be taken into account for future investigations into the association between idiopathic toe walking and language delays.

Clinical Implications

Despite the limited amount of research on this topic, the studies reviewed in this literature appraisal provide a juncture for future, specific and sensitive research. The main consideration that can be concluded is to encourage developmental pediatricians to refer children with idiopathic toe walking for complete language assessment by a Speech Language Pathologist. Additionally, if a Speech Language Pathologist receives a referral for a child who appears to have undiagnosed idiopathic toe walking, evidence suggests that an assessment of gait by an orthopedic surgeon or a Physiotherapist should be completed, followed by a 'gold' standard language assessment by the Speech Language Pathologist.

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