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ORIGINAL ARTICLE

Do children with attention deficit and hyperactivity disorder (ADHD) have a different gait pattern? Relationship between idiopathic toe-walking and ADHD☆

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Idiopathic toe-walking;
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Abstract
Introduction: Idiopathic toe-walking (ITW) is described as a gait pattern with no contact between the heels and the ground in children older than 3 years. The diagnosis is clinical, making it necessary to rule out other neurological and orthopaedic conditions. A relationship between ITW and vestibular dysfunction and/or proprioceptive sensibility has been proposed. Children with neurodevelopmental disorders (autism, language and cognitive disorders) often have ITW.
Objectives: To determine the frequency of ITW in children with attention deficit disorder and hyperactivity (ADHD).
Patients and method: A study was conducted on children diagnosed with ADHD, with normal neurological examination, with no alterations in MRI scan, cognitive disorder or autism. A complete clinical anamnesis was performed and Achilles shortening was measured with a goniometer.

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Results: The study included 312 children with a mean age of 11 years (73.7% boys). The ADHD combined subtype was the most frequent (53.8%), followed by the inattentive (44.9%), and hyperactive (1.3%). ITW was observed in 20.8% of patients, particularly in the combined subtype ($P = .054$). Only 32 of them (49.2%) had Achilles shortening. ITW was associated with sociability disorders ($P = .01$), absence of pain in legs ($P = .022$), and family history of ITW ($P = .004$). Only 11% had previously visited a doctor for this reason.
Conclusions: As in other neurodevelopmental disorders, children with ADHD have frequently more ITW and Achilles shortening than controls, especially if they presented with a social communication disorder or a family history of ITW. An early diagnosis is essential to establish effective treatments.

PALABRAS CLAVE
Marcha de puntillas idiopática;
Andadores de puntillas;
Trastorno por déficit de atención e hiperactividad;
Procesamiento sensorial

¿Caminan de manera diferente los niños con trastorno por déficit de atención hiperactividad (TDAH)? Relación entre marcha de puntillas idiopática y TDAH

Resumen
Introducción: La marcha de puntillas idiopática (MPI) se describe como el patrón de marcha sin apoyo del talón en niños mayores de 3 años. El diagnóstico es clínico y obliga a descartar otras enfermedades neurológicas y traumatológicas-ortopédicas. Se postula su relación con una disfunción vestibular o de sensibilidad propioceptiva. Los niños con trastornos del neurodesarrollo (trastorno del espectro autista, trastorno del lenguaje y cognitivo) presentan frecuentemente MPI.
Objetivos: Analizar la frecuencia de MPI en niños con trastorno por déficit de atención e hiperactividad (TDAH).
Pacientes y método: Estudio en niños diagnosticados de TDAH con exploración neurológica normal, sin alteraciones en neuroimagen ni trastorno cognitivo o trastorno del espectro autista. Se realizó anamnesis completa y se valoró la presencia de acortamiento aquíleo con goniómetro.
Resultados: Se analizó a 312 niños con edad media de 11 años, el 73,7% varones. El subtipo combinado fue el más frecuente (53,8%), seguido del inatento (44,9%) e hiperactivo (1,3%). Un 20,8% de los pacientes presentaban MPI, que era más frecuente en el subtipo combinado (p = 0,054). Solo 32 de estos (49,2%) presentaban acortamiento aquíleo. La presencia de MPI se relacionó con alteraciones en el área de la sociabilidad (p = 0,01), ausencia de dolor en miembros inferiores (p = 0,022) y antecedentes familiares de MPI (p = 0,004). Solo el 11% habían consultado por este motivo previamente.
Conclusiones: Al igual que en otros trastornos del neurodesarrollo, los niños con TDAH presentan con mayor frecuencia MPI y acortamiento aquíleo, especialmente entre aquellos con trastornos de comunicación social o antecedentes familiares de MPI. Es fundamental una identificación precoz para instaurar tratamientos eficaces.

Introduction

Idiopathic toe walking (ITW) is defined as a gait pattern in children aged more than 3 years characterised by the bilateral absence of heel contact with the ground (''walking on tiptoes''). It is a clinical diagnosis that requires ruling out other neurologic or traumatologic–orthopaedic conditions that may cause a similar gait.[1,2] Before age 2 years, this pattern is considered normal, and it resolves spontaneously in most children, as demonstrated by a longitudinal study by Engström et al. with follow up of a cohort of Swedish children through age 5 years. Only 30 of the 70 children who toe-walked at the beginning of the study continued to do so at age 5 years.[3]

Based on several studies, the prevalence of ITW in healthy children is estimated at 7–24%.

In addition to pain and increased instability, children with persistent ITW lose the ability to dorsally flex the ankle, which results in a shortened Achilles tendon.

When it comes to the aetiology of ITW, a hereditary pattern of ITW has been described since the earliest studies. Other studies have proposed impairments in sensory processing and proprioception as the cause of this gait pattern.[4–6]

The increased prevalence of ITW in children with autism spectrum disorder is well known, but several studies have also reported an association of ITW with other neurodevelopmental disorders. In a sample of 163 children, Accardo

et al. found that those with ITW were more likely to have language disorders.[7,8] A study in 26 children with ITW by Futagi et al. found that half had neurodevelopmental disorders (mainly hyperactivity and motor impairment) and 4 were in follow up for intellectual disability.[9] Another study conducted by Schulman et al. also found an association between ITW in children with language disorders and fine and gross motor delays and visuomotor delays.[10]

Just as the incidence of ITW is higher in children with neurodevelopmental disorders, there is also a higher probability of its occurrence in association with different learning disorders. Thus, Engström et al. found that out of 51 children with ITW, 39% had motor disorders, 17% impairment in executive function, 25.5% perceptual disorders, 23.5% memory and language disorders, 25% poor social skills and 21.6% emotional/behavioural disorders.[11]

In Spain, ITW has also been described as a marker of impaired psychomotor development. For instance, a study comparing 56 Spanish school children aged 3–6 years who walked on their toes to children who did not exhibit this gait pattern by means of a psychometric test (CUMANIN) found that the children who toe-walked had a lower score in the developmental questionnaire overall and in specific subscales such as psychomotor development, memory, and verbal and nonverbal development.[12]

The scarcity of studies addressing the presence of ITW in association with one of the most prevalent neurodevelopmental disorders in Spain, attention-deficit hyperactivity disorder (ADHD), motivated us to conduct this study. Thus, our aim was to assess the potential association of ADHD with the presence of ITW.

Materials and methods

We analysed data for children aged 6–18 years that received an ADHD diagnosis based on the criteria of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) and with a normal neurological evaluation assessed in the Paediatric Neurology clinic of the Hospital Fundación Jiménez Díaz (Madrid) between January and December 2015.

Every patient included in the study had undergone a psychometric assessment that included a cognitive evaluation and screening for learning disorders. We excluded patients with any of the following: intellectual disability (IQ < 70), pyramidal signs on neurologic assessment, presence of significant abnormalities in neuroimaging, autism spectrum disorder, chromosomal disorders or other defined genetic syndromes, or neurologic diseases accounting for the presence of ADHD symptoms (poorly controlled seizures, epileptic encephalopathies such as continuous spike and wave during sleep, and others).

We took a detailed history of ADHD symptoms, including an assessment of their severity by means of the Clinical Global Impression scale (CGI) and the Children's global assessment scale (C-GAS). We also analysed developmental, learning and social characteristics, and the presence of tics, developmental coordination disorders and other factors associated with ITW (family history, pain in lower extremities and frequent falling).

Furthermore, we took biometric measurements with a 360° goniometer with 2 moveable arms measuring 17.5 cm each, with an angle scale marked at 2° intervals and a linear scale accurate to 1 mm. In each patient, we measured the popliteal angle and the passive ankle dorsiflexion both with an extended and a flexed knee. We defined shortening of the Achilles as a passive ankle dorsiflexion with an extended knee of less than −10° (Fig. 1).

Figure 1 Measurement of ankle dorsiflexion with the goniometer.

We performed the statistical analysis with the software SPSS version 22, using the χ^2 test.

Results

Sample characteristics

We analysed 312 patients with an ADHD diagnosis. The mean age was 11 years ± 2.8 standard deviations (SD), and 73.7% of the patients were male. Preterm birth was reported by 11.2%, of who most (51.4%) were born between 32 and 37 weeks' gestation, all with birth weights of more than 2000 g. None of the patients had intellectual disability, and the mean intellectual quotient was 102 ± 15 SD. The ADHD subtype was combined in 53.8%, predominantly inattentive in 44.9% and predominantly hyperactive in 1.3%. Of all patients, 15.4% had tics (10.3% simple or complex motor tics; 1.3% vocal tics; 2.6% a combination of tics and 1.3% Tourette syndrome).

Most patients (87%) had mild-to-moderate symptoms. We observed that those with combined ADHD had the highest severity based on the CGI scale, an association that was statistically significant ($P = .023$); nearly half of these patients (48%) had moderate-to-severe ADHD.

The mean score in the other scale used to assess the intensity of symptoms (C-GAS) was 60 ± 7 SD, which confirms the mild-to-moderate severity in most patients in our sample.

As for the presence of comorbidities, we found impairments in social interaction, identified through the *Autism Diagnostic Observation Schedule* (ADOS-1) and without meeting the criteria for autism spectrum disorder, in 10.9%, and language disorders in 33% (of who 17.6% had simple delays in language and 10.9% specific language disorders). When we assessed reading and writing, we found that 44% of the patients had some type of disorder and 27.6% problems in both areas. Also, 15.1% of patients had impairments in

motor coordination, most of them mild. Thirty-four percent of the 312 children in the study had some type of psychiatric comorbidity, with oppositional-defiant disorder being the most frequent diagnosis.

Probability of toe walking

Of all the patients with ADHD in our sample, 20.8% (65/312) exhibited toe walking (continuous in 53.8% and intermittent in 27.2%) (Fig. 2). Toe walking was most frequent in patients with the combined subtype (37/65), followed by patients with predominantly inattentive ADHD (26/65) and predominantly hyperactive ADHD (2/65). Although the frequency of ITW in patients with combined ADHD was higher, the difference was not statistically significant ($P = .054$).

Not all children with ITW had shortening of the Achilles tendon, in fact, we only found an ankle dorsiflexion of less than -10° in 32 (49%).

On the other hand, we observed that 45% of patients with functional equinus had a family history of ITW in a first-degree relative.

Patients with ITW did not have significant symptoms secondary to the abnormal gait pattern. Thus, 64.6% of patients with ITW did not report pain in the lower extremities, and 86.1% did not experience frequent falls.

Most patients with ITW (89%) had not sought medical care for this condition and only 3 (4%) were receiving specific treatment for it (physical therapy or botulinum toxin).

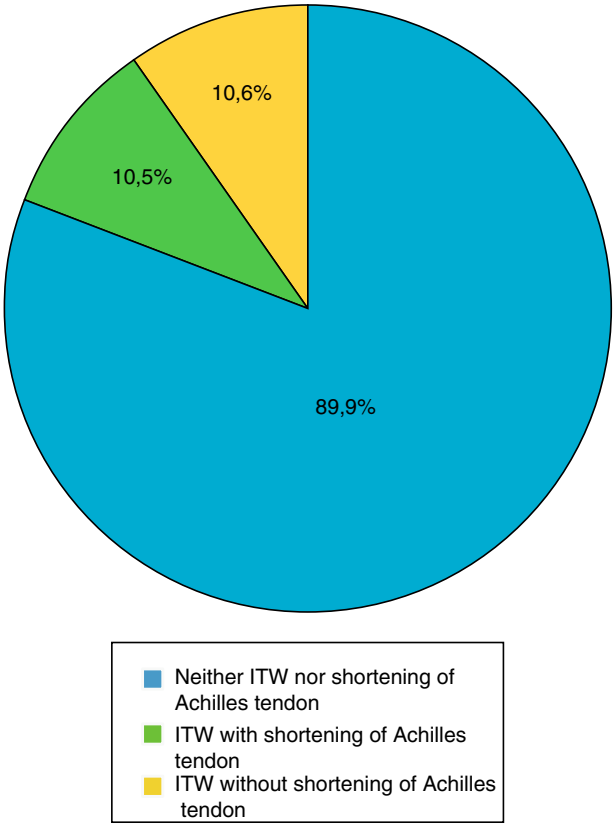

Figure 2 Percentage of patients with ADHD and ITW. ITW, idiopathic toe walking.

Aspects associated with the presence of toe walking

We analysed the association between the presence of ITW and the characteristics of our patients, and present the results in Table 1.

The presence of ITW was not significantly associated with sex ($P = .838$), preterm birth ($P = .128$), severity of ADHD symptoms ($P = .329$), presence of tics ($P = .434$), family history of ADHD ($P = .542$) or age.

It is known that patients with ADHD frequently have comorbidities, especially impairments in social interaction, language, reading and writing, psychomotor skills and other psychiatric disorders, so we analysed the presence of these disorders in our sample of children with ADHD and their association with ITW. The only neurodevelopmental disorder for which we found a significant association with ITW was impairment in social interaction ($P = .01$) (after ruling out autism spectrum disorder). However, we did not find an association between ITW and language disorders ($P = .593$), reading and writing disorders ($P = .768$), impairment in motor skills ($P = .069$) or psychiatric disorders ($P = .165$).

When we analysed the characteristics of ITW, we found that its presence was associated with a positive family history of ITW in a first-degree relative ($P = .004$), which supports the hypothesis that ITW could be hereditary. We also found a statistically significant association with the absence of pain in the lower extremities ($P = .022$). However, we did not find an association with a personal history of frequent falling.

Discussion

The incidence of ITW is greater in patients with neurodevelopmental disorders compared to the general population. This association has been well defined, as we have noted above, with ITW found in patients with autism spectrum disorder (19–63%), language disorders (16–22%), cognitive disorders (30%) and learning disorders (20%). However, there is no previous study analysing the prevalence of ITW in patients with ADHD, the disorder that is the leading reason for seeking psychiatric care and found in 3.5% of school-aged children in Spain.[13]

In our sample, 20.8% of children with ADHD had ITW. Although this gait pattern was more frequent in patients with the combined type of ADHD, the differences between ADHD subtypes were not statistically significant. On the other hand, we found that not all patients with ITW had shortening of the Achilles tendon at the time of the assessment, although it is possible that they would have developed it eventually, since there is a progressive loss of ankle dorsiflexion in patients with ITW, as demonstrated in the work of Engelbert, frequently leading to shortening of the Achilles tendon or other tendons in the dorsal musculature of the lower extremities.[14]

We found a statistically significant association between ITW and the presence of impairments in social interaction (in the absence of autism spectrum disorder), which was consistent with the higher prevalence of ITW in patients with autism spectrum disorder. We also found a strong association

Table 1 Association between idiopathic toe walking and other clinical variables.

Sex	.838	Developmental coordination disorder	.069
ADHD subtype	.054	Language disorder	.593
Preterm birth	.128	Disorder of reading and writing	.768
Severity of ADHD (CGI)	.329	Family history of ITW	.004
Impairment in social interaction (without ASD)	.01	Family history of ADHD	.542
Psychiatric comorbidity	.165	Frequent falls	.106
Tics	.434	Pain	.022

P-values.

between the presence of ITW and a positive family history of functional equinus in first-degree relatives, which supports the hypothesis that ITW is hereditary. But we did not find an association with other variables under study, such as sex, preterm birth, severity of ADHD, tics, language disorders, motor coordination, reading and writing or other psychiatric comorbidities.

It seems that the presence of ITW in children with neurodevelopmental disorders may stem from impairments in proprioception and sensory processing. Thus, in 1978 Montgomery and Gauger presented a series of cases that suggested that impairments in sensory processing and proprioception may be the main cause of ITW, although their results were not statistically significant.[15]

Later on, in 2011, Williams et al. performed a study where they assessed patients with and without ITW on a vibration platform and observed that patients with ITW had lower vibration perception thresholds compared to patients with normal gait, as well as poorer coordination. These data support the hypothesis that ITW results from impairments in sensory processing due to which patients require increased proprioceptive input to be able to maintain postural stability.[5]

Although many of our patients with ITW experienced pain, most of them had never sought medical care for it. We think that the early diagnosis of ITW is essential in order to implement effective treatments (physical therapy, botulinum toxin, orthotics, etc.) capable of preventing the associated symptoms (pain, instability) or surgeries required to treat shortened tendons.

This is the first study that demonstrates the high probability of ITW in children with ADHD. This association is usually underdiagnosed and may be a significant cause of morbidity in these patients.

Limitations

The main limitations of our study are the lack of a control group and that the sample was obtained from a hospital setting, which may have resulted in a bias towards greater disease severity (although we did not find a significant association between the severity of ADHD symptoms and the presence of ITW).

Conflicts of interest

The authors have no conflicts of interest to declare.

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