



Serial casting in the treatment of idiopathic toe-walkers and review of the literature

Anna FOX, Sue DEAKIN, Gill PETTIGREW, Robin PATON

From Blackburn Royal Infirmary, Blackburn, United Kingdom

Idiopathic toe-walking is defined as persistent toe-walking in a normal child in the absence of developmental, neurological or neuromuscular conditions. True idiopathic toe-walking is a rare referral, representing approximately 1:100 new patients seen in the Paediatric Orthopaedic Clinic.

A prospective study of idiopathic toe-walking (ITW) was organised between 1999 and 2003. Patients underwent full history, neurological examination and assessment of ankle dorsiflexion, followed by below-knee weight-bearing casting.

Forty four developmentally normal children with no delay in walking age were in this study. There was an age range on presentation from 2 years to 14 years 4 months, with median 60.5 months. Sixty eight percent were male. Thirty four percent had a family history of the condition. Following casting, 66% of patients had improved gait on patient and clinician determined outcomes, with the majority of children ceasing to toe-walk. Ankle dorsiflexion significantly improved in those children who were successfully treated ($p = 0.001$).

Keywords : idiopathic toe-walking ; serial casting.

INTRODUCTION

Idiopathic tip-toe-walking is regarded as persistent toe-walking after the age of 2 in a developmentally normal child who can usually stand flat-footed when not walking. Other conditions such as spastic diplegia, neuropathy, myopathy, autism and

developmental disorders may present as tip-toe-walking and idiopathic toe-walking is a diagnosis of exclusion (25). This study was set up as a prospective research project to determine the demographics and clinical presentation of toe-walkers to the Orthopaedic department of a District General Hospital.

PATIENTS AND METHODS

All consecutive children over the age of 2 years with the preliminary diagnosis of idiopathic toe-walking, seen in the Paediatric Orthopaedic Clinic of the senior author (RWP) between December 1999 and September 2003, were invited to participate in a prospective study. Research & Development committee approval and parental and ethical committee consent were obtained.

■ Anna Fox, MRCS, Senior House Officer in Orthopaedics.
■ Gill Pettigrew, MCSP Dip RGRT, Paediatric Physiotherapist.

■ Robin Paton, FRCS (Orth), Consultant Orthopaedic Surgeon.

Blackburn Royal Infirmary.

■ Sue Deakin, FRCS (Tr & Orth), Consultant Orthopaedic Surgeon.

West Suffolk Hospitals.

Correspondence : Anna E Fox, 5 Manthorpe Avenue, Roe Green, Worsley, Manchester M28 2AZ, United Kingdom.

E-mail : annafox2k@btopenworld.com.

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Cases with known neuromuscular disorders and autism were excluded from the study. Inclusion criteria were : age over 2 years, main complaint of toe-walking (based on parental observation), independent walking achieved, no known diagnosis of neuromuscular disorder, neurological condition, developmental delay, and finally no other known orthopaedic problems.

At the initial visit, the child's birth history, family history and motor development were reviewed. Specifically parents were questioned about the age of independent walking, the use of a baby walker, and the age at which toe-walking was first noticed. They were asked to complete the Gillette score, a 10 point mobility/disability assessment system which is used in the assessment of cerebral palsy to identify potential gait and motor skill anomalies. Although the Gillette functional assessment questionnaire has been shown to be reliable and valid for children with neuromuscular disorders (21), we acknowledge it is not validated for use as a discriminatory test between idiopathic and other causes of toe-walking. Despite this we found it useful to identify potential gait and motor skill dysfunction. The percentage of time spent toe-walking was estimated to the nearest quartile. The presence of pain, balance, shoe wear problems and difficulties were noted. Each child underwent a full neurological examination by the senior author (RWP) specifically looking at : 1) muscle bulk, 2) tone, 3) power, 4) sensation, 5) deep tendon reflexes and 6) Gower's test. In addition full orthopaedic examination of the spine, hips, legs and feet was performed assessing range of movement, alignment and laxity. Goniometer measurements of the range of dorsiflexion of the ankle with the knee flexed and extended were performed. Each child was assessed by a Consultant Orthopaedic Surgeon (RWP) and a Paediatric Physiotherapist (GP).

Treatment protocol

All cases were treated with below knee walking casts set in plantigrade with serial cast changes at 2 weekly intervals. On removal of casts, all children were assessed by the Paediatric Physiotherapist (GP) who instructed parents how to perform passive Achilles' tendon stretching exercises. The children were clinically reviewed by the senior author (RWP) and the Paediatric Physiotherapist (GP) at 3 months post-removal of the cast, then 6 monthly intervals thereafter. Outcome was assessed with regards to toe-walking, and ankle dorsiflexion. If the parents were satisfied and plantar grade walking was present, the child was discharged ; although if problems returned, access to the clinic was encouraged.

Statistical Analysis

Demographic data of the successfully treated toe-walkers were compared to the continued toe-walkers using the Mann-Whitney U-test for continuous non-parametric data, and Fisher's exact test for categorical data where sample sizes were small. Changes in ankle dorsiflexion from initial to post-treatment values were analysed using the Wilcoxon matched pairs signed rank sum test. Finally, pre- and post-treatment ankle dorsiflexion were correlated to identify any relationship between fixed ankle equinus and age. A p value of < 0.05 was considered to be significant.

RESULTS

Forty four children were eligible for this study, all referred with bilateral toe-walking as their predominant complaint. Age at presentation ranged from 24 months to 172 months (14 years 4 months) with a median age of 60.5 months and mean age of 73 +/- 42 months. Thirty (68.2%) patients were male. The majority of children (35/44) had been referred by their general practitioner, however the remaining 9 came from other sources such as Community Paediatricians, Paediatricians and Rheumatologists (table I).

Family history

A family history of idiopathic toe-walking was reported in 34.1% of cases (95% CI 21.9 to 48.9) (fig 1). The incidence of a positive family history was 43.3% in males compared to 14.3% in females (p = 0.07). Congenital talipes equinovarus was reported in one father, but this was not present in the child.

Birth history and development

Gestation at birth ranged from 26 to 42 weeks with median 40 weeks and mean 39.1 weeks +/- 2.69. Only 5 children were born prematurely, defined as 36 weeks gestation or less, the youngest being at 26 weeks. Thirty-five (79.5%) were born vaginally and with 2 requiring forceps assistance. Nine were delivered by Caesarean section, 3 for breech presentation, 1 for small pelvis, 4 for foetal

Table I. — Patient demographics and clinical presentation

Male : Female	30 : 14
% Males	68.2%
Mean Gestation at birth (weeks)	39.1 +/- 2.69 (95% CI 38.3 to 39.9)
Median Gestation (weeks)	40 (IQR 38 to 40)
% Premature birth (< 37 weeks)	11.4%
% Family history of ITW	34.1%
Mean age independent walking (months)	12.8 +/- 2.71 (95% CI 11.8 to 13.7)
Median age independent walking (months)	12 (IQR 11 to 13.5)
Mean age parents noticed TW (months)	29 +/- 30.34 (95% CI 19.1 to 38.8)
Median age parents noticed TW (months)	14 (IQR 12 to 27)
Mean age presented to clinic (months)	73 +/- 41.8 (95% CI 60.2 to 85.7)
Median age presented to clinic (months)	60.5 (IQR 38 to 101.5)
Clinical complaints :	
Foot pain	27.3%
Foot-wear problems	31.8%
Poor balance	45.5%

Family history of toe-walking (n=15)

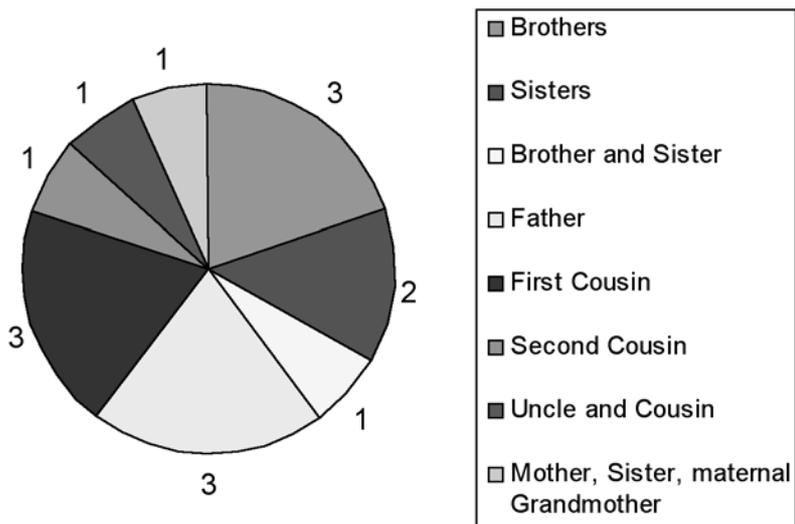


Fig. 1. — Family members affected with idiopathic toe-walking

distress and 1 as the mother was an insulin-dependent diabetic. Post-partum stay ranged from 24 hours to 2 weeks in all but one case. Nine children were kept in for 1 week or more, but in the majority (seven) of cases this was due to maternal complications. The child born prematurely at 26 weeks spent 6 weeks in neonatal intensive care. There were no recorded cases of tight tendo

Achilles contractures at birth. 8 children were referred post-natally for Orthopaedic opinions. One child was diagnosed with a unilateral mild flexible Metatarsus Adductus deformity of the foot which did not require intervention. Two children were referred with suspected congenital talipes equinovarus which was excluded. Five of the children were referred following breech presentation for

exclusion of hip dysplasia. Three children had hyperbilirubinaemia of the newborn presenting with jaundice, which settled.

All of the 44 children achieved independent walking within the normal time period with median 12 months and mean of 12.8 +/- 2.71 months. A baby-walker was used in 63.6% at some stage during walking development. Three parents noticed mild learning delays and 2 reported symptoms of dyspraxia.

The age at which toe-walking was noticed by parents, ranged from 11 months to 11 years (table I). Parents noticed in 65.9% that their child had toe-walked from the age of 2 years or below.

Presentation and examination

The age of the child at presentation ranged from 24 months to 172 months with median 60.5, and mean 73 +/- 42 months. The difference between the age at which parents noticed the toe-walking, and the age at presentation confirmed a significant ($p < 0.0001$) delay in identifying the abnormality and seeking medical advice in the majority of cases.

Time spent on tip toes as estimated by parents was 100% for over half of all cases (25), with 75% for 11 patients, 50% for 7 and 25% for 1 child.

Symptom wise, few patients reported problems, and of those who did, the vast majority were non-specific and vague. Chief complaint was poor balance (46%), unsurprisingly as toe-walking disrupts the normal steady tripod configuration of the foot. Foot-wear problems (32%) and foot pain (27%) were also reported. On the functional Gillette score, 41 children scored the maximum of 10 ("walks, runs and climbs on level and uneven terrain without difficulty or assistance"). The remaining 3 all scored 9 ("Walks outside the home for community distances, easily gets around on level ground, curbs, and uneven terrain, but has difficulty or requires minimal assistance with running, climbing, and/or stairs").

Orthopaedic and neurological examination revealed no abnormal clinical findings in any of the 44 cases. Specifically there was no evidence of increased muscle tone or altered reflexes, and nothing

to indicate a neuro-muscular explanation for the toe-walking. No evidence of foot abnormalities were noted for the 3 children who had previously received an Orthopaedic opinion.

No significant differences were noted between patient's left and right ankle dorsiflexion. Ankle dorsiflexion at presentation with the knee in extension ranged from -10 to 20 degrees with median 0 and mean 1.1 +/- 5.1. With the knee held in flexion to allow relaxation of the gastrocnemius muscle, ankle dorsiflexion ranged from 0 to 20 degrees with median 5 and mean 4.8 +/- 5.2. With the knee in extension, only 5/44 children did not have passive dorsiflexion to neutral plantigrade, however all could reach neutral when the knee was flexed. Correlating age with range of passive dorsiflexion, no significant association was noted between age at presentation and ankle dorsiflexion with the knee extended ($r = -0.159$), however a weak association was observed when the knee was flexed ($r = -0.43$).

Outcome of treatment

Bilateral walking casts set at neutral were placed on all 44 children for between 3 and 10 weeks, median 6 and mean 5.7 +/- 1.1. Children underwent serial cast changes at 2 weekly intervals until there was reduced resistance to passive ankle dorsiflexion. Only one complication was reported with the casting, plaster sores caused by the proximal end of the cast. Mean follow-up was 14 months.

Following casting 29 (66%) children either completely stopped toe-walking or improved sufficiently to satisfy their parents (table II). Although numbers were small, younger children appeared more likely to improve.

Comparing the pre-casting range of ankle dorsiflexion of those toe-walkers who improved following casting, and those who continued to toe-walk, initially there was no significant difference in dorsiflexion in either knee extension ($p = 0.97$) or knee flexion ($p = 0.79$). Following casting, ankle dorsiflexion in those patients whose toe-walking improved compared with those whose didn't, was better. Although this was highly significant when measured with the knee flexed ($p = 0.001$) and not with the knee extended ($p = 0.23$). This is due to

Table II. — Outcome following casting, results stratified by age

Age at treatment	N =	Successful	Unsuccessful	%
All ages	44	29	15	66
< 3 years	11	9	2	82
3-5 years	9	6	3	67
5-8 years	12	6	6	50
8-12 years	10	6	4	60
> 12 years	2	2	0	100

ankle dorsiflexion with the knee extended significantly improving in both groups of patients ($p < 0.0001$ and $p = 0.01$), regardless of whether or not their toe-walking improved, but dorsiflexion with knee in flexion only significantly improved in those whose toe-walking improved ($p = 0.0001$ versus $p = 0.36$). This implied that casting not only stretches the gastrocnemius muscle, but also stretches other posterior ankle soft tissues such as the soleus muscle and tendo Achilles' (table III).

This would lead us to conclude that casting does significantly improve ankle dorsiflexion, but it is the improvement in ankle dorsiflexion with the knee flexed that is important in clinical outcome.

Finally we compared the demographics of those successfully treated and those continued toe-walkers to identify any risk factors that may associate with a poorer outcome. No significant differences were noted between the two groups of patients, although longer periods in casts showed a trend towards a better result (table IV).

DISCUSSION

Idiopathic toe-walking is not a common referral to a General Paediatric Orthopaedic clinic in a District general Hospital. There were 44 pure idiopathic toe-walkers in a 4-year period, equating to approximately 1 :100 new referrals. Idiopathic toe-walking is principally a diagnosis of exclusion. The literature published on this subject has shown in some cases toe-walking to be a self-limiting condition with improvement in adulthood, however associations with other developmental pathology have been reported and underlying neuro-muscular aetiology must be sought (2, 8, 12, 14, 26, 29).

Table III. — Ankle dorsiflexion before and after casting

Mean Pretreatment ankle dorsiflexion (degrees)	
Knee flexed	4.8 +/- 5.23
Knee extended	1.1 +/- 5.08
Mean Post-treatment ankle dorsiflexion (degrees)	
Knee flexed	9.9 +/- 5.58
Knee extended	5.8 +/- 5.57
Mean Post-treatment ankle dorsiflexion (degrees)	
<i>Continued Toe-walkers :</i>	
Knee flexed	6.8 +/- 5.94
Knee extended	5.0 +/- 5.98
<i>Stopped or rare Toe-walkers :</i>	
Knee flexed	11.3 +/- 4.73
Knee extended	6.2 +/- 5.40

During normal childhood walking development, independent standing and independent walking are achieved by, on average, 9.5 and 13 months respectively. Although toe-walking is not usual or a predominant feature in early stages of development, for some children it can be regarded as a normal, temporary, variant, often familial, prior to the age of 2 years. Initially the child ambulates with the feet wide apart to provide a stable base for a relatively high centre of gravity. Initial footstrike occurs with the ankle in plantarflexion and but gradually develops into ankle dorsiflexion and heel-strike (28, 31). This is usually completed by the age of five. Three studies have investigated the kinematics of idiopathic toe-walking (12, 18, 29). The primary gait abnormalities occur at the ankle with loss of heel strike at initial contact. During swing phase the ankle is initially dorsiflexed, with sudden plantarflexion midway secondary to gastrocnemius activity, which leads to the child landing forefoot first. This differs from mild spastic diplegia, where ankle dorsiflexion at floor contact is adequate, but the child lands on the forefoot due to excessive knee flexion (13). Gait patterns are significantly different in ITW and cerebral palsy (CP), however when normal children are asked to toe walk, they produce similar gait kinematics to idiopathic toe-walkers (6, 18). Electromyographic studies have found premature onset of gastrocnemius activity prior to foot contact and continuing into stance, with inhibition of tibialis anterior in swing phase. Normally at heel-strike and during stance

Table IV. — Differences between successfully and unsuccessfully treated patients

Variable	Successful N = 29	Unsuccessful N = 15	Significance
Male sex	65.5%	66.7%	NS, p = 0.63
Length of stay in hospital (median)	5 days	5 days	NS, p = 0.75
Median gestation	40 weeks	40 weeks	NS, p = 0.83
Median age toe-walking noticed	12 months	17 months	NS, p = 0.33
Median age presented to clinic	58 months	67 months	NS, p = 0.53
Median length of time on toes	100%	100%	NS, p = 0.48
Median age independent walking	12 months	13 months	NS, p = 0.33
Median length of time in cast	6 weeks	6 weeks	NS, p = 0.09

tibialis anterior is active. Gastrocnemius onset occurs to cause plantarflexion and take-off. Tibialis anterior controls dorsiflexion at swing phase by concentric contraction allowing heel-strike, then eccentrically contracts to aid plantarflexion along with gastrocnemius (4, 15, 22, 24).

Isolated contracture of the tendo Achilles causing toe-walking was first reported in 1967 under the name "Congenital short tendo calcaneus" in 20 children with significant fixed ankle equinus (12). Peri-operatively shortening of the tendinous portion of the triceps surae was observed. A subsequent genealogical study reported on a 5-year-old male toe-walker with fixed equinus, who although neurologically intact, stuttered and had awkward hand movements. This led to postulation that "short tendo calcaneus" affected males more severely than females and an autosomal dominant inheritance with variable gene expression was suggested as the underlying anomaly (20). In subsequent studies the incidence of males affected has consistently been higher than females, reportedly lying between 50 and 80% (8, 13, 16, 18, 27, 30). The majority of studies have observed a positive family history with the incidence varying from between 10 and 88% (9, 11, 12, 14-16, 27, 30). The patient demographics in this study did not vary, the incidence of family history being 34.1%, and males affected 68.2%. However most of the previous studies are retrospective whereas this study has the advantage of being prospective. Incidentally, we observed a higher proportion of male patients with a positive family history when compared to female patients ($p = 0.07$). This would add further weight to the theory of genetic disposition. This study reported a

lower incidence of prematurity (11.4%) than has been previously observed.

Toe-walking may occur in the absence of tendo Achilles contracture although many studies hypothesise that contracture occurs as a result of toe-walking, rather than the cause (9). Indeed it is unclear whether we are talking about two separate clinical entities, or a spectrum of the same disease pathology. Shulman *et al* (26) reported a weak negative correlation between ankle dorsiflexion and age ($R = -0.46$) agreeing with our findings. It would seem sensible to conclude that, whether or not contracture or toe-walking occurs first, continued toe-walking must contribute to continuing shortening of the tendo Achilles, and vice versa. But is toe-walking simply an idiopathic harmless occurrence, an unidentified neurological or neuromuscular condition, or a developmental problem?

Toe-walking has been found to be predominant in those with autism, and/or language delay (1, 2, 32). Shulman *et al* (26) studied toe-walkers with normal neurological examination, finding speech and language deficits and developmental delays. At first glance toe-walking may appear to be a benign condition, however developmental delays may be associated, and the more subtle signs missed. Orthopaedic surgeons need to be aware of these associated factors reported in the paediatric literature although it is arguable whether or not early paediatric opinion should be sought.

Although it seems most likely there is some form of underlying subclinical neuropathic or neuromuscular process occurring in toe-walking, at present this is unproven, although many studies have reported findings which would seem to

support this theory (7, 9, 20). In the original study by Hall *et al* (12), muscle biopsies were reported as normal. A more recent histological study into muscle biopsies taken from toe-walkers referred for a neurological opinion, found similar changes to those seen in congenital talipes equinovarus, the commonest abnormality being a predominance of type 1 muscle fibres in gastrocnemius biopsies. Angulated atrophic fibres and basement membrane thickening were also observed (7). This appears to support the theory of a neuropathic process; however this was a group of patients, who had all been referred for a neurological opinion although no neurological problem was identified. Furrer and Deonna (9) classified 4 categories of toe-walkers: 1) those with clear cut pyramidal tract signs (spastic diplegia), 2) those with no clinical findings (habitual), 3) those with limited dorsiflexion only (congenital short tendo Achilles), and 4) those difficult to classify into any of the other 3 categories. Importantly, toe-walking may be an early indication of serious progressive neuromuscular disease, differential diagnoses including myopathic, neurological and developmental disorders. A systemic approach must be used which includes detailed history, thorough neurological examination and where necessary appropriate investigations such as creatine kinase and in a minority of cases MRI scanning of the brain and spine. Some authors suggest creatine kinase levels are required in all cases of abnormal gait to exclude Duchenne muscular dystrophy (23). At what stage toe-walking with no apparent neurological problem should be investigated is currently down to clinical judgement. None of the children in this study showed delay in development and all reached independent walking satisfactorily. Gillette scoring confirmed that there were no limitations to activity.

At present no adequate long-term prospective studies exist that compare treatment options available for the idiopathic toe-walking child. Indeed the natural history of the condition is not fully understood, and only a few studies address persisting problems in the skeletally mature adult patient (29). Proposed treatments range from casting/orthotic devices to tendo Achilles and gastrocnemius lengthening, and more recently botulinum

toxin injection. Auditory feedback devices have been used, but with variable improvement (5). In addition studies have reported on groups of patients in whom spontaneous correction of toe-walking had occurred without any treatment (8).

The value of casting remains controversial in the literature, and is thought to produce its effect by stimulating an increase in the number of sarcomeres present in the calf musculature (10). Katz and Mubarak (16) found improvements in gait in 5 out of 6 developmentally normal children treated with casts. Brower *et al* (3) compared children with CP and ITW treated by casting, finding that despite more restricted dorsiflexion in the latter, casting reduced resistance to passive stretching, and results were better maintained in the ITW group. In addition no deleterious side effects on muscle function were seen. Despite this a retrospective study by Eastwood *et al* (8), found casting/orthotic treatment had no advantage over conservative treatment, with similar outcomes, although pretreatment status and severity was unknown. This finding was supported by a study by Stricker and Angulo (30). More recently, Brunt *et al* (4) reported on the use of botulinum toxin injection into the gastrocnemius and soleus muscles, followed by casting. This treatment is well recognised in cerebral palsy (17), and appears to produce improvement in the gait pattern of the small number of idiopathic toe-walkers studied (4).

Despite varying degrees of success with casting therapy, better sustained results have been widely reported using tendo Achilles lengthening surgery. Aponeurotic (Baker's), open Z and percutaneous tendo Achilles lengthening operations restore musculotendinous length. However persisting problems may occur with under or over-lengthening of the Achilles tendon. In Hall's original paper all 20 patients underwent Achilles tendon lengthening surgery, and at 3 years all walked with a normal heel-toe gait with normal ankle dorsiflexion (12). Kogan and Smith (19) treated 15 idiopathic toe-walkers with percutaneous lengthening, following which all could initially heel-toe walk, although 2 developed Achilles tendonitis. Eastwood *et al* (8) found significant improvements in gait for 72% of toe-walkers undergoing Baker's procedure but only

37% achieved normal gait. Stricker and Angulo (30) also noted significantly better outcomes with surgery.

Although gait can improve and modify into adulthood, normal gait is rarely achieved either secondary to casting or surgery. It would appear that with no treatment, idiopathic toe-walking persists with some improvement although normal gait is rarely achieved. Stott *et al* (29) showed that there are persistent changes in ankle kinematics secondary to serial casting and percutaneous lengthening of the Achilles tendon in skeletally mature individuals who were treated as children with idiopathic toe-walking. It was noted that parental satisfaction was high despite normal gait not being achieved. This suggests that despite treatment, the gait will never be normal (29). Hicks *et al* (13) are of the opinion that in persistent toe-walkers, compensation occurs through the development of hind-foot valgus and out-toeing, resulting in external tibial torsion. This would suggest that idiopathic toe-walking is even more common in the community with only a small number being referred (iceberg effect), although more epidemiological work is required to assess its true incidence in the community. At present we do not know the natural history of ITW, and if compensation does occur, are there any other long-term effects in withholding treatment? Lengthening of the triceps surae can theoretically weaken push-off and would be debilitating if a calcaneal gait develops maybe causing a greater problem than persisting toe-walking. It seems reasonable to suppose that gait can be modified in the young child by casting or orthotic devices, but the older child who may have developed tendo Achilles contracture, requires surgical correction. In this study casting resulted in definite improvement in ankle dorsiflexion, but only when the knee was held in flexion and in those who stopped toe-walking. Results were better in the younger age group, suggesting an element of gait retraining as well as muscle stretching. Initial ankle dorsiflexion weakly correlated with age and would support theories that continuing toe-walking leads to worsening tendo Achilles contracture.

Further epidemiological studies to assess the incidence in the community and to review the

natural history of those not treated should be advocated. It appears illogical to be advocating treatment in a benign, non-debilitating condition where the incidence and natural history is not well understood. There are no adequate prospective long-term studies randomising idiopathic toe-walking into different treatment groups: no treatment, casting or Achilles tendon lengthening, and further studies should seek to rectify this.

CONCLUSION

Casting does appear to benefit idiopathic toe-walkers in the short-term by improving ankle dorsiflexion and stopping toe-walking in the majority of cases. This study shows that non-invasive treatment should be considered in the first instance, reserving surgical options for resistant cases, however we acknowledge further randomised trials are required.

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