Severe crouch gait in spastic diplegia can be prevented

A POPULATION-BASED STUDY

We studied the prevalence of severe crouch gait over a 15-year period in a defined population of children with spastic diplegia and Gross Motor Function Classification System levels II and III, to determine if there had been a decrease following changes to the management of equinus gait. These changes were replacing observational with three-dimensional gait analysis, replacing single level with multilevel surgery, and replacing gastrocsoleus lengthening with gastrocnemius recession. Of 464 children and adolescents with spastic diplegia who underwent three-dimensional gait analysis, 27 had severe crouch gait. Seventeen of these had been managed by isolated lengthening of the gastrocsoleus. Following changes in the management of equinus gait, the prevalence of severe crouch gait decreased from 25% and stabilised at a significantly lower rate, fluctuating between 0% and 4% annually (p < 0.001).

We conclude that severe crouch gait in this population was precipitated by isolated lengthening of the gastrocsoleus. These findings may be relevant to other surgical populations, as severe crouch gait may be a useful way to monitor the quality of the surgical management of abnormal gait in children with cerebral palsy and spastic diplegia.

In spastic diplegic cerebral palsy, toe-walking may delay the ability to walk independently or cause tripping, falling and functional impairment.3,4 Spastic equinus can be managed variously by injections of Botulinum neurotoxin A (BoNT-A), ankle-foot orthoses (AFO), serial casting with physiotherapy or a combination of these treatments.3-6 When spastic equinus progresses to fixed shortening of the gastrocsoleus, operative intervention is often required.

The traditional treatment is isolated lengthening of the gastrocsoleus by various forms of gastrocsoleus recession. Of 464 children and adolescents with spastic diplegia who underwent three-dimensional gait analysis, 27 had severe crouch gait. Seventeen of these had been managed by isolated lengthening of the gastrocsoleus. Following changes in the management of equinus gait, the prevalence of severe crouch gait decreased from 25% and stabilised at a significantly lower rate, fluctuating between 0% and 4% annually (p < 0.001).

We conclude that severe crouch gait in this population was precipitated by isolated lengthening of the gastrocsoleus. These findings may be relevant to other surgical populations, as severe crouch gait may be a useful way to monitor the quality of the surgical management of abnormal gait in children with cerebral palsy and spastic diplegia.
Severe crouch gait was defined as knee flexion > 30° throughout the stance phase of gait, combined with excessive dorsiflexion at the ankle and incomplete extension at the hip. Kinematic data may be used to develop a categorical classification for severe crouch gait which is useful for studying prevalence trends, and other studies have suggested a possible causal relationship between isolated lengthening of the gastrocnemius and subsequent development of severe crouch gait. Based on previous investigations, the ‘at risk’ population was defined as children aged six years and over with spastic diplegia and levels II or III according to the Gross Motor Function Classification System (GMFCS). Children at GMFCS level I may develop crouch gait but this is usually not severe and can often be managed by simple methods. Children at level IV have borderline walking abilities between the ages of six and twelve years and are not expected to be long-term ambulators. Children with spastic hemiplegia do not develop severe crouch gait because the uninvolved side maintains an extended posture. Finally, crouch gait rarely becomes severe or symptomatic before the age of six years.

From the State-wide Cerebral Palsy Register we included children with spastic diplegia and GMFCS levels II or III who fulfilled the criteria for severe crouch gait on sagittal kinematics, had at least one 3D gait analysis between 1995 and 2009 and were aged six to 18 years at the time of 3D gait analysis.

We excluded those who failed to meet inclusion criteria, had inadequate or missing documentation or were born in or moved to other states without follow-up.

The combination of a State-wide Cerebral Palsy Register, a gait analysis laboratory and a tertiary children’s hospital gave us the opportunity to make this a population-based study. Children with spastic diplegia, GMFCS II and III were identified from the register and their records cross checked with the gait laboratory and orthopaedic department databases. Positive confirmation of severe crouch gait was always from sagittal kinematics. However, exclusion of severe crouch gait was based on sagittal kinematics, video gait analysis or clinical assessment.

Following identification of children and adolescents fulfilling the above criteria, gait laboratory records and hospital records were reviewed by the first author (CV). Sagittal kinematic data, with patient-identifying information removed, were presented to an orthopaedic surgeon and a senior physiotherapist (HKG, JR) for classification as severe crouch or other gait pattern. Possible contributory causes of severe crouch gait were identified including previous operations on the gastrocnemius and history of BoNT-A injections. During the study period and in the preceding five years, equinus deformities were managed by three different surgical techniques (Fig. 1). The Hoke technique of lengthening the tendo Achillis and Baker technique of gastrocnemius recession were according to the authors’ description.
described by Strayer⁷ was modified. The gastrocnemius aponeurosis was separated from the underlying soleus fascia and divided transversely, according to the original description. The range of passive dorsiflexion was checked and, if satisfactory, nothing more was done. However, if equinus was not fully corrected, the soleus was lengthened by a single transverse cut in the fascia, from medial to lateral, at the same level as the division of the gastrocnemius aponeurosis.

Given the known delay between surgical lengthening of the gastrocsoleus and development of calcaneus at the ankle and crouch gait, it was considered necessary to investigate the surgical management of equinus gait for the five years before the study period (Fig. 2).

**Statistical analysis.** A generalised linear model with a logarithmic link and binomial distribution was used to model the risk of severe crouch gait, with indicator variables for three five-year periods, 1995 to 1999, 2000 to 2004 and 2005 to 2009 inclusive as the only covariates. This type of regression produces coefficients which can be exponentiated to express results as risk ratios. The results were reported as risk ratios with associated 95% confidence intervals (CI). Analyses were performed using Stata Statistical Software Release 11 (StataCorp, College Station, Texas) by a statistician (KS) with biomedical experience.

**Results**
From the Cerebral Palsy Register between 1995 and 2009, 585 children aged six to 18 years who had spastic diplegia and GMFCS level II or III were identified. When cross-checked with gait laboratory records, 464 of these (79%) had one or more 3D gait analyses. Of the 121 children who had not had a 3D gait analysis, detailed clinical records were available for 117 (20%). A total of 68 had attended for a video-based gait analysis and 49 had been seen in outpatient clinics and were classified as having either a mild spastic gait not requiring surgical intervention or dystonia. None had a severe crouch gait and the majority were offered BoNT-A or other non-operative treatment. The status of the remaining four children was unknown.

Of the 464 children who had 3D gait analysis, 27 fulfilled the criteria for severe crouch gait (Fig. 3). There were nine girls and 18 boys with a mean age of 12 years (7 to 17) at the time of gait analysis. A total of 12 were GMFCS II and 15 were GMFCS III. The age at diagnosis of severe crouch gait and presentation for gait analysis was no different in the first three years compared with the subsequent 12 (p = 0.66). In all, 17 of the children had lengthening of the gastrocsoleus, with a mean interval between the operation and development of symptomatic crouch gait of six years (1.5 to 13), of whom nine had bilateral lengthenings of tendo Achilles and eight had bilateral Baker type lengthenings.⁹,¹⁰,¹⁷,³² There was strong evidence for a decrease in the risk of severe crouch gait when the first and second five-year periods were compared (p < 0.001). The risk was estimated as 91% lower in the second five-year period compared with the first (95% CI 71% to 97%) (Figs 3 and 4).

Between 1990 and 1994 all operations for equinus were performed at a single level as isolated gastrocsoleus lengthening by the Baker technique or lengthening of the tendo Achillis. Between 1990 and 1994, 51 children had lengthening of the tendo Achillis (41 percutaneous Hoke slide lengthenings and ten open Z-lengthenings) and 47 had Baker’s
lengthening of the gastrocsoleus fascia. There was a transition in 1995, when four children had multilevel surgery with Strayer lengthenings (Fig. 2). Since 1996, the most common type of gastrocsoleus lengthening was by the Strayer or its modified technique as part of single event multilevel surgery and guided by 3D gait analysis (Fig. 2).
After 1995, a minority of children, with a severe equinus contracture and a negative Silfverskiold test had a Baker recession or lengthening of the tendo Achillis, according to surgeon or family preference (Fig. 2).

Other changes in the overall management of equinus gait after 1994 are summarised in Table I. Serial casting for spastic equinus was replaced by BoNT-A. Intensive rehabilitation was provided by community-based physiotherapists, funded by a Post Intervention Physiotherapy Fund. Hinged AFOs were replaced by solid AFOs and post-operative progress was monitored by regular assessments in the gait laboratory rather than the outpatient clinic.

Before 1994, the standard AFO used in our centre was hinged, because ankle motion was perceived as being of functional benefit. Following the introduction of single event multilevel surgery, solid AFOs were prescribed routinely for a minimum of 12 months post-operatively. The transition to a hinged, or no AFO was allowed only when gait data indicated satisfactory plantar flexion, knee-extension coupling and minimal risk of crouch gait. We do not have objective data on compliance with the AFO prescription. However, given the close and frequent contact between gait laboratory physiotherapists, parents and community-based physiotherapists, we think compliance was satisfactory.

Discussion

In a report from our centre in 2001, single level gastrosoleus lengthening for equinus gait in children with cerebral palsy resulted in calcaneus at the ankle in 36% at five to ten year follow-up. In many children, this was associated with disabling crouch gait because of loss of effective plantar flexion knee-extension coupling. Crouch gait often progresses rapidly during the adolescent growth spurt. In severe cases, there may be increased reliance on walking aids, wheelchairs and eventually loss of independent ambulation. Although reconstructive surgery for severe crouch gait is effective, it is invasive, with a significant complication rate and prolonged rehabilitation. Many short-term studies of single and multilevel surgery for equinus gait in spastic diplegia report satisfactory outcomes. In the longer term, the rate of recurrent equinus deformity can be tracked by the need for revision surgery. However, we believe that only a long-term study using objective outcome measures could identify the insidious problem of over-lengthening and crouch gait. Not all adolescents with crouch gait have reconstructive surgery. In those who do, the period from surgery for equinus can be so long that neither the patient nor surgeon may consider the obvious connection.

Given the arduous rehabilitation after reconstruction, it would be preferable to prevent crouch than treat it. This study found convincing evidence of a major and sustained decrease in the prevalence of severe crouch gait in a defined population over a 15-year period (Fig. 4). Many aspects of equinus gait management were changed in our centre in 1995, following the opening of the gait laboratory. These are summarised in Table I and some have been described in detail elsewhere. Although randomised clinical trials have a role in the short-term evaluation of multilevel surgery, long-term changes in management cannot be evaluated in this way. For these purposes, a population-based cohort study, with long-term follow-up and objective outcome measures is the best design.

Of the factors in Table I, we think the three most important are instrumented replacing observational gait analysis, multilevel replacing single level surgery and gastrocnemius recession replacing lengthening of the tendo Achillis and Baker gastrosoleus recession. However, changes in the funding and organisation of post-operative rehabilitation, the prescription of AFOs and post-operative monitoring may also have played a role. A weakness of our study was the lack of detailed data about these ancillary factors. These may interact in that the gait laboratory allows a more accurate surgical prescription to be devised and a precise analysis of both short-term and longer term outcomes. Signs of over-lengthening of the gastrocnemius can be detected when they are mild and asymptomatic. This allows corrective steps to be taken, including the selection of more conservative methods of gastrocnemius recession or differential lengthening of the gastrocnemius by the modified Strayer technique. In 1995, 25% of children with spastic diplegia GMFCS II and III had
severe crouch gait. From 1999 onwards the annual rate fluctuated between 0% and 4%. It took about five years for the rate of severe crouch gait to fall and become level at a much lower rate. Our interpretation of these results is that the high prevalence of severe crouch gait during the first five years was mostly iatrogenic, with single level surgery by lengthening of the tendo Achillis responsible for, or contributing to most cases. However, in each five-year period, there were patients who had no previous lengthening of gastrocsoleus. In all of these, severe lever-arm disease was found, consisting of varying degrees of increased femoral neck anteversion, patella alta, external tibial torsion and pes valgus. Severe crouch gait is therefore part of the natural history of spastic diplegia but, in this community, the majority of cases were associated with lengthening of the tendo Achillis. Significantly, there were no cases of severe crouch gait following Strayer regression, which was always performed as part of single event multilevel surgery. Improvements in surgical outcomes for equinus gait were not restricted to those who did not develop severe crouch gait. Other flexed-knee gait patterns were also reduced, including milder forms of crouch. However, as there is no widely accepted definition for ‘mild’ or ‘moderate’ crouch gait, we restricted our study to ‘severe crouch gait’. The strengths of this study are that it was population-based, used a valid and reliable description of severe crouch gait and was of sufficient duration to detect changes in surgical practice and gait patterns. The weaknesses were other changes in the management of equinus, the lack of gait studies for the entire population and the exclusion of milder forms of crouch gait and other flexed knee gait patterns.

Severe crouch gait is part iatrogenic and may be preventable. Its prevalence may be a useful way to monitor the quality of surgical management and rehabilitation of gait dysfunction in children with cerebral palsy and spastic diplegia.

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References


