The role for hip surveillance in children with cerebral palsy

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Abstract Spastic hip displacement is the second most common deformity seen in children with cerebral palsy (CP), and the long-term effects can be debilitating. Progressive hip displacement leading to dislocation can result in severe pain as well as impaired function and quality of life. Recent population-based studies have demonstrated that a child's Gross Motor Functional Classification System (GMFCS) level is most predictive for identifying hips "at-risk" for progressive lateral displacement. As a result, in many developed countries, hip surveillance has now been adopted as an integral piece of the comprehensive care puzzle for the management of children with spastic hip displacement. This paper reviews the spectrum of treatments available for progressive hip displacement, examines the current literature on the success of hip surveillance, and illustrates an example of a current hip surveillance program stratified by the GMFCS level.

Keywords Pediatric · Cerebral palsy · Hip surveillance · Gross motor functional classification system · Proximal femoral varus osteotomy · Adductor surgery · Botulinum toxin · Hip salvage surgery

Introduction

Cerebral palsy (CP) describes a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain [1]. The incidence of CP is approximately 2 per 1000 live births and is the most common cause of physical disability affecting children in developed countries [2]. The incidence of spastic hip displacement in CP is related to the severity of involvement, varying from 1 % in children with spastic hemiplegia, up to 75 % in those with spastic quadriplegia [3, 4]. Two large population-based studies have reported the overall incidence of hip displacement to be approximately 35 % across all children with CP [2, 5].

The most useful development in the classification of CP in recent years has been the creation of the Gross Motor Function Classification System (GMFCS) [6]. The GMFCS is a five-level ordinal grading system based on the assessment of self-initiated movement with emphasis on function with regard to sitting and walking. The risk of progression from hip displacement to dislocation is related to severity of neurologic involvement [7, 8] and walking ability [3, 9], and is directly related to gross motor function as graded by the GMFCS [2].

The primary goal of hip surveillance programs is to identify children "at risk" of hip displacement, to monitor their hip development over time, and to offer early appropriate intervention [10]. The natural history of spastic hip disease is progressive lateral displacement of the hip secondary to impaired mobility and spastic hypertonia of the hip adductor and flexor musculature [10]. Hip dislocation may lead to pain, functional impairment affecting the ability to sit, stand, and walk, as well as an impaired quality of life [4, 5, 7, 11–17]. Early identification of these "at-risk"

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children is critical; however, timely referral and triage by an orthopedic surgeon is just as important. Identification of progressive hip displacement has limited value unless effective intervention is available [18••]. Early identification and orthopedic intervention has been shown to alter treatment outcomes, reduce the number of reconstructive surgeries required, and avoid the need for salvage surgery [2, 10, 17, 19]. As a result, hip surveillance has become an integral part of evidence-based care for children with CP in many developed countries based on growing evidence supporting surveillance programs and their outcomes [10, 12, 14, 15, 19, 20].

Pathophysiology of hip displacement

The majority of children with CP are born with anatomically “normal” hips without evidence of hip displacement or dislocation [21]. However, the natural history of spastic hip disease places children at risk for progressive lateral hip displacement [22]. In children with CP, this displacement is commonly referred to as “silent subluxation,” as children are often not symptomatic until the hip is dislocated and painful.

The neurologic lesion associated with CP manifests as a non-progressive or static encephalopathy; however, the associated musculoskeletal pathology is progressive, resulting in contractures of muscle tendon units, bony torsional deformity, and ultimately, joint instability [1, 23]. Asymmetric muscle spasticity has long been felt to be a major contributor to hip instability in children with CP. Sharrard et al. [24] demonstrated that progressive limitation of abduction, often associated with flexion deformity, was an indicator of early instability of the hip. In their study, they found that no hip with radiologic evidence of subluxation had abduction greater than 45°, suggesting limited abduction, and possibly spasticity, plays some role in the development of hip instability.

Two critical components to proximal femoral anatomy are anteversion of the femoral neck in the transverse plane and the femoral neck-shaft angle in the coronal plane [25, 26]. Robin et al. [27] demonstrated that these two deformities were closely related to a child’s GMFCS level. In their study, they propose that increased femoral anteversion and neck-shaft angle may be caused by persistent fetal alignment due to delayed walking and limitations in gross motor function. The combination of elevated femoral neck anteversion and neck-shaft angle contributes to the increased risk of hip displacement.

Soo et al. [2] demonstrated a linear relationship between the rate of hip displacement and a child’s GMFCS level. Their population-based study showed that GMFCS V children demonstrated an incidence of hip displacement of 90 % while there were no cases of hip displacement in 114

children with cerebral palsy who were GMFCS level I. Hagglund came to very similar conclusions with respect to his population-based study of children with CP in Sweden [14]. Much can be learned from these population-based studies; knowledge and identification of groups of children with “hips at risk” for displacement are critical for planning surveillance programs and initiating early intervention [2, 10, 12].

Clinical assessment

Clinical examination is a vital component of hip surveillance in children with spastic hip disease. Important elements of the assessment include: assignment of GMFCS level (because this defines the level of risk), spasticity in major muscle groups (Modified Ashworth scale) [33], pain, difficulties with sitting, standing, or walking, and examination of the child’s spine [5], pelvis [34], and lower limb musculoskeletal system [35].

Musculoskeletal hip joint measurements should include both passive and dynamic range of motion recordings. Measurements by a number of observers are more reliable than by a single observer, and experienced observers employing a standardized method improve reliability [29, 36, 37]. Standard clinical examination includes: hip flexion and extension, abduction in full extension and 90° of flexion, and internal/external rotation in the prone position. Presence of a hip flexion contracture (using the Thomas test), knee flexion contracture, and hamstring contracture (knee popliteal angle—unilateral and bilateral) should be noted. In children with spastic diplegia, excessive anterior pelvic tilt will produce a “hamstring shift” between unilateral and bilateral knee popliteal angle in conjunction with an apparent hip flexion contracture [38–40]. Dynamic muscle spasticity using the Modified Tardieu Scale is applied to the hip adductors and hamstring muscles [41]. A comprehensive examination also includes examination of the spine for scoliosis and assessment of leg length discrepancy. In the setting of a leg length discrepancy, the distinction should be made between a real shortening as in the case of a unilateral hip dislocation, or an apparent shortening from pelvic obliquity or a windswept hip deformity.

Radiographic examination

Clinical examination alone is insufficient to evaluate hip displacement in children with CP [28]. Decisions for treatment and surveillance must be made in conjunction with a well-taken anteroposterior (AP) radiograph of the pelvis and hip joint with the child in the supine position. For hip

surveillance to be successful, a standardized radiographic technique must be followed, ensuring reliability between interval radiographs and between patients. In children with CP, excessive femoral anteversion, hip flexion, and adduction contractures are often present. Recognition and correct position in these circumstances is necessary to generate consistent radiographs. Unrecognized hip flexion contracture will create a lordotic pelvis (excessive anterior pelvic tilt), which can be corrected by raising the legs on pillows to flatten the lumbar spine [12]. Positioning the legs parallel to each other will address adduction or abduction contractures, and pointing the patellae upwards will correct for excessive femoral anteversion [10]. A film-focus distance of 115 cm has been suggested to standardize film magnification between patients [8].

The most accepted and reproducible measurement for hip displacement is Reimer's migration percentage (MP) [42, 43], which is a measure of the femoral head's containment with the acetabulum in the coronal plane [13]. This measurement is obtained by identifying both Hilgenreiner's line and Perkin's line and then measuring, as a percentage, the amount of ossified femoral head that has migrated beyond Perkin's line laterally (Fig. 1). In cases of moderate to severe hip displacement, there is often a "gothic-arch" deformity of the lateral acetabular margin, and in these cases, the midpoint of the lateral acetabular margin is used instead of Perkin's line [43]. The MP is a linear measure of hip displacement and is the most valid, reliable, and useful measure of hip displacement in children with CP [28, 42–45]. In the setting of hip surveillance, reliability of this measure can be improved with special training of the recording health care providers [45].

Fig. 1 The Migration Percentage (MP) is obtained by identifying Hilgenreiner's line (H) and Perkin's line (P) and then measuring as a percentage the amount of ossified femoral head that has migrated beyond Perkin's line laterally ($A/B \times 100$). The acetabular index (AI) is the angle between the slope of the acetabulum and Hilgenreiner's line. (From Dobson et al. [10]; with permission)

The acetabular index (AI) [46] is one of several measures of acetabular shape and development and is probably the most commonly used acetabular measure for spastic hip disease. This index measures, in degrees, the angle between the slope of the acetabulum and Hilgenreiner's line. However, when measuring the AI in children with CP, observers must be cognizant of increased acetabular anteversion, which is common. In this scenario, the apex of the acetabulum may not be the most lateral point of the acetabulum [28]. While some studies have demonstrated utility of the AI in predicting hip instability [28], others have concluded that it has a low interobserver reliability with increased variability based on patient positioning [5, 12, 14, 20].

In 2008, a new CP hip classification scheme was introduced that included both quantitative (MP) and qualitative features (femoral head and acetabular deformity, pelvic obliquity, and break in Shenton's line) of femoroacetabular deformity [47, 48]. This six level ordinal grading scheme is applied to children with CP at skeletal maturity (closure of tri-radiate cartilage) (Fig. 2). Similar to Severin's developmental dysplasia of the hip classification [49], this new CP hip classification system (CPHCS) is applicable to young and old children through common MP measurements. Initially, the CPHCS was designed for children who had reached skeletal maturity (closure of the triradiate cartilage), but recently Gose et al. [50] has validated the CPHCS for hip disease in children with CP aged 2–7.

When considering treatment for children with spastic hip disease and associated femoroacetabular deformity, additional modalities may be useful. Computed Tomography (CT) with three-dimensional reconstruction provides a comprehensive evaluation of proximal femoral and acetabular morphology and may be useful when areas of acetabular deficiency exist. Magnetic Resonance Imaging (MRI) may also be employed to aid the surgeon in decision-making between reconstruction and salvage procedures. The delayed gadolinium-enhanced magnetic resonance imaging of cartilage (dGEMRIC) technique has been shown to identify early signs of hip osteoarthritis and has been found to correlate well with clinical symptoms of hip pain [51]. We now use dGEMRIC imaging in cases where significant femoroacetabular incongruity and cartilage loss exist. The information from this technique enables surgeons to decide preoperatively whether reconstructive or salvage surgery is most appropriate.

Management options

The primary goals of treating hip displacement in children with CP are to maintain flexible, well-located, and painless

Cerebral Palsy Hip Classification

Grade I: Normal Hip– Migration Percentage <10% <ol style="list-style-type: none"> 1. Shenton's arch intact 2. Femoral head round (within 2mm using Mose circles) 3. Acetabulum – normal acetabular development with a normal sourcil, an everted lateral margin and normal tear drop development 4. Pelvic obliquity less than 10 degrees
Grade II: Near Normal Hip– Migration Percentage ≥10% ≤15% <ol style="list-style-type: none"> 1. Shenton's arch intact 2. Femoral head round or almost round 3. Acetabulum – normal or near normal development 4. Pelvic obliquity less than 10 degrees
Grade III–Migration Percentage >15% ≤30% <ol style="list-style-type: none"> 1. Shenton's arch intact or broken by less than or equal to 5mm 2. Femoral head round or mildly flattened 3. Acetabulum normal or mildly dysplastic including blunting of the acetabular margin and a widened tear drop 4. Pelvic obliquity less than 10 degrees
Grade IV–Migration Percentage >30% <100% <ol style="list-style-type: none"> 1. Shenton's arch broken by more than 5mm 2. Femoral head variable deformity – Appendix 1 3. Acetabulum variable deformity – Appendix 2 4. Pelvic obliquity variable – Appendix 3
Grade V–Migration Percentage ≥100% <ol style="list-style-type: none"> 1. Shenton's arch completely disrupted 2. Femoral head variable deformity – Appendix 1 3. Acetabulum variable deformity – Appendix 2 4. Pelvic obliquity variable – Appendix 3
Grade VI: Salvage Surgery <ol style="list-style-type: none"> 1. Valgus osteotomy 2. Arthrodesis 3. Excision arthroplasty (Castle) ± valgus osteotomy (McHale) 4. Replacement arthroplasty

Fig. 2 A new hip classification system for children with CP at skeletal maturity. (From Robin et al. [47•]; with permission)

hips with a symmetrical range of motion. Previous studies have illustrated the importance of establishing a concentric

femoral head within the acetabulum before age five for normal hip joint development to occur [52–54]. Unlike

developmental dysplasia of the hip (DDH), children with CP typically are born with normal anatomic hips, but due to a combination of delayed motor milestones and asymmetric muscle tone, they develop hip displacement at a later age [55]. Moreover, displacement of the hip is silent in the initial stages and difficult to detect by physical examination alone [10, 28].

The surgical treatment of spastic hip disease is guided by the degree of displacement of the femoral head and acetabular dysplasia. Treatment is stratified according to MP, acetabular dysplasia, and severity of muscle spasticity into three broad categories: preventive, reconstructive, and salvage surgeries. Accepted indications for preventive surgery include MP >40 %, or an increase in MP >10 % over the last year, and hip abduction <30° [9, 10, 44]. Reconstructive surgery is indicated when the MP is >50 % and there is evidence of hip subluxation or early dislocation without evidence of degenerative changes to the femoral head. Finally, salvage surgery is indicated for those who present with painful and degenerative dislocated hips or where reconstruction has previously failed or now is not an option due to the degree of degeneration [55].

Preventative treatments are directed at neutralizing the deforming forces across the hip joint that lead to progressive displacement. Non-surgical management using physical therapy, bracing, and Botulinum toxin A injections initially were believed to delay the progression of hip displacement; however, a recent randomized clinical trial has demonstrated that these therapies are not effective in preventing hip displacement [30, 56]. Preventative surgery involves soft tissue lengthening of tight structures around the hip, starting with the tendon of adductor longus and including the gracilis tendon, iliopsoas tendon, and adductor brevis muscle as necessary [57]. Historically, neurectomy of the anterior branch of the obturator nerve has been described; however, this procedure is associated with over-correction and development of abduction contractures. Phenolization of the anterior branch of the obturator nerve is now accepted as a safer alternative and still considered a preventative measure [32, 58].

The reported success of isolated soft tissue lengthening for the treatment of hip displacement is quite promising within the literature. Presedo et al. [59] reported a success rate of approximately 88 % for a population of children with spastic cerebral palsy if the MP was <40°, with an overall success rate at 7 years of 70 %. Shore et al. [32] recently demonstrated that the success of preventative surgery for hip displacement is directly related to a patient's GMFCS level. In their study, the overall success rate at a mean follow up of 7 years was 32 %. However, the success rate was predicted by GMFCS level, with a 94 % at GMFCS II decreasing to 14 % at GMFCS V (Fig. 3).

Fig. 3 The success of preventative surgery for hip displacement is directly related to a patient's GMFCS level. (From Shore et al. [32]; with permission)

When a child initially presents with significant hip subluxation, or after a failed previous soft tissue procedure, bony reconstruction is recommended. Reconstructive options include femoral varus derotation osteotomy, pelvic osteotomy (Dega, San Diego or Pemberton), or a combination of both with open hip reduction. Soft tissue procedures alone will not prevent dislocation of the spastic hip when the MP is greater than 50 % [42, 53, 60, 61] and will not address the deformities of increased femoral anteversion and neck shaft angle [27]. Proximal femoral osteotomy allows for simultaneous correction of increased neck shaft angle and excessive femoral anteversion commonly seen in nonambulatory children with CP [27, 62]. In most patients, femoral osteotomy alone is enough to address hip displacement in children with CP; however, when the displacement is long-standing and associated with significant acetabular dysplasia, consideration of simultaneous pelvic osteotomy is warranted [63, 64]. Once dysplasia has developed, the acetabulum has a limited ability to remodel [21, 65]. Several different acetabuloplasty techniques have been described for the treatment of acetabular dysplasia in children with CP; however, variations of the Dega are widely accepted to generate the most superior results, providing posterior and lateral coverage [9, 64, 66, 67]. The osteotomy should be centered above and directed away from the deficient area to allow for maximal correction at that site. In children in whom the triradiate cartilage is closed, the acetabulum may be redirected with a Ganz periacetabular osteotomy; however, in order for this type of osteotomy to be successful the joint must be congruent. If the joint is incongruent, consider using a Chiari or shelf osteotomy.

Salvage surgery is the only option for treatment of a painful long-standing hip dislocation and requires removal of the destroyed cartilage surface of the misshapen femoral head [68]. Pain and difficulty with perineal hygiene are the two most common indications for surgery in this

group of patients. Salvage procedures include proximal femoral resection with soft tissue interposition, valgus osteotomy with femoral head resection, hip arthrodesis, and replacement arthroplasty with mixed results, each one carrying their own set of risks and potential complications [69–72]. At best, salvage procedures produce modest results. Efforts should be directed toward identifying patients at risk of dislocation early and preventing the need for salvage surgery rather than perfecting these surgical techniques.

Evidence of hip surveillance

Hip surveillance is a screening program aiming to identify and monitor the critical early indicators of progressive hip displacement [10]. The first suggestion that hip dislocation in children with CP could be prevented was published nearly 60 years ago [73]. Since that time, several authors have reported mixed experience with hip surveillance [2–5, 9, 10, 12, 14, 19, 20, 22, 28, 43, 45, 65, 74–80]. In establishing a hip surveillance protocol, reliance on clinical examination alone is not sufficient, and regular radiographic examination is also important [28]. Early indicators for hip displacement include a patient's GMFCS level, age, gait classification, and migration percentage (MP) [29]. Not surprisingly, isolated techniques to reduce spasticity or address adductor contracture alone have been ultimately disappointing in preventing hip displacement in higher level GMFCS children [30–32]. Hip surveillance identifies children with progressive hip displacement but does not dictate the timing or type of intervention [18•]. Treatment must be tailored for the individual child taking into consideration all of the above-mentioned factors.

A recent detailed review of the literature on hip surveillance demonstrated 25 English articles in press from 1980 to 2011 [18•]. There were no randomized controlled clinical trials within this review. However, there have been four population-based studies [2, 5, 15, 19] that demonstrate high ascertainment, diagnostic reliability, and generate a highly representative population sample of children with CP. Within these four studies, the incidence of hip displacement was similar and appeared to be intimately related to CP sub-type and gross motor function.

Gordon and Simkiss preformed the only systematic review on the evidence for hip surveillance in children with CP [20]. In their review, only six articles met the inclusion criteria, and all were observational studies not worthy of meta-analysis. Within these articles, the epidemiology and study design was variable with a mixture of prospective and retrospective studies from tertiary care centers [10, 28,

44, 75] and regional institutions [13, 14]. All studies used radiological measurements to monitor hip displacement, with Reimer's migration index being the most reliable technique. All six identified children with spastic quadriplegia as an "at-risk" cohort who would benefit most from a hip surveillance program. The conclusion was that progression of the MP greater than 7 % per annum was ominous, requiring careful monitoring and orthopaedic referral. The authors recommend that all children with bilateral cerebral palsy should have a hip radiograph at or prior to 30 months of age [20], similar to Scrutton and Baird [13].

Australian surveillance guidelines

The *Australian Standards of Care for Hip Surveillance in Children with Cerebral Palsy* is the first consensus statement published on hip surveillance [29]. Between 2007 and 2008, clinicians working in the area of hip management in children with CP came together to formalize a consensus statement. The process of creating the consensus statement occurred in three stages [18•]. First a draft document was generated by a working party of physicians and therapists with national representation. The second stage involved presentation of the document at the national CP conference, where professionals attending were surveyed and feedback regarding the guidelines was given. Finally, specific orthopaedic surgeons across Australia and New Zealand who did not attend the national meeting were targeted and asked to complete a detailed survey. Results from 124 medical and allied health professionals were grouped according to experience, discipline, and volume of practice to generate the consensus statement, which is outlined in Fig. 4. These standards are recommended guidelines for screening, monitoring, and triaging children to orthopedic services as part of an overall prevention program of hip displacement. The conclusions from this consensus statement were for referral to an orthopedic surgeon when (1) the MP is greater than 30 %, or (2) if the MP is unstable (changing 10° per year), or (3) if there is associated pain in the hip and (4) if other orthopedic conditions have been identified [29].

Conclusions

Progressive lateral hip displacement in children with spastic hip disease is common and can progress silently. Clinical examination alone is not a sensitive enough measure to detect early hip displacement. Hip surveillance programs identify critical early indicators of

Fig. 4 Recommended Frequency of Hip Surveillance According to GMFCS (Adapted from Wynter et al. [29])

GMFCS 1:

- Initial AP Pelvis at 12-24 months (or at age of identification)
- Repeat clinical and radiographic review at 3 years[†]
 - If GMFCS level has changed - alter surveillance accordingly
- Repeat clinical & radiographic review at 5 years[†]
 - If GMFCS level has changed - alter surveillance accordingly
 - If remains GMFCS level 1 – discharge from hip surveillance

GMFCS 2:

- Initial AP Pelvis at 12-24 months (or at age of identification)
- Repeat clinical & radiographic review via yearly surveillance until MP stability
 - If MP unstable/abnormal – continue yearly surveillance
 - If MP stability established – review at 4-5 years
- Repeat clinical & radiographic review at 4- 5 years[†]
 - If GMFCS level has changed - alter surveillance accordingly
 - If MP stable – review at 8-10 years
 - If MP unstable – continue yearly surveillance until stability
- Repeat clinical & radiographic review at 8-10 years[†]
 - If MP stable – discharge

GMFCS 3:

- Initial AP Pelvis at 12-24 months (or at age of identification)
- Repeat clinical & radiographic review 6 months later[†]
 - If MP unstable/abnormal – continue yearly surveillance
 - If MP stability established – review at 4-5 years
- Repeat clinical & radiographic review at 4- 5 years[†]
 - If GMFCS level has changed - alter surveillance accordingly
 - If MP stable – review at 8-10 years
 - If MP unstable – continue yearly surveillance until stability
- Repeat clinical & radiographic review at 8-10 years[†]
 - If MP stable – discharge
 - If MP unstable – continue yearly surveillance until stability established

GMFCS 4:

- Initial AP Pelvis at 12-24 months (or at age of identification)
- Repeat clinical & radiographic review every 6 months[†]
 - If GMFCS level has changed - alter surveillance accordingly
 - If MP unstable/abnormal – continue 6 month surveillance until stability
 - If MP stability established – continue yearly surveillance
- Repeat clinical & radiographic review at 7 years[†]
 - If MP stable (<30%) & GMFCS stable – review at pre-puberty
 - Continue yearly surveillance from pre-puberty to skeletal maturity
- Independent of MP, if evidence of pelvic obliquity, scoliosis continue 6 month surveillance until skeletal maturity

GMFCS 5:

- Initial AP Pelvis at 12-24 months (or at age of identification)
- Continue 6 monthly surveillance until 7 years[†]
 - If GMFCS level has changed - alter surveillance accordingly
 - If MP stable (<30%) & GMFCS stable – review yearly until maturity
- Independent of MP, if evidence of pelvic obliquity, scoliosis continue 6 month surveillance until skeletal maturity

Legend:

[†]Verify GMFCS Level

AP = Anteroposterior

GMFCS = Gross Motor Functional Classification Level

MP = Migration Percentage

progressive hip displacement, and early detection and appropriate surgical intervention of spastic hip displacement can

prevent hip dislocation and the need for more invasive surgery.

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