Review Article

Hip Surveillance in Children With Cerebral Palsy

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Abstract

Hip dysplasia is common in children with cerebral palsy (CP), especially in those children with notable functional impairment. Severity of hip dysplasia has been shown to correlate with higher Gross Motor Function Classification System levels. Migration percentage measured on AP pelvis radiographs is the key radiographic measure quantifying hip displacement in CP. Hip surveillance programs for children with CP exist in Europe, Australia, and parts of Canada and have been adopted as standard of care. These programs have demonstrated improved detection of hip subluxation and appropriate early intervention with a resultant decrease in the number of painful dislocations. Hip surveillance programs provide healthcare providers with guidance for a schedule of obtaining hip radiographs based on patients' age, Gross Motor Function Classification System level, and migration percentage. Although systematic surveillance programs have yet to be adopted in the United States, several centers and organizations are currently investigating the potential and efficacy of hip screening in CP.

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Cerebral palsy (CP) is a descriptive term for a group of disorders mostly affecting motor function, which are usually attributed to nonprogressive disturbances that occurred in the developing child's brain. Hip dysplasia is the second most common orthopaedic issue for children with CP (after equinus) that can potentially cause notable pain and disability.[1,2] Hip dislocation may lead to pain, functional impairment affecting the ability to sit, stand, or walk, and impaired quality of life.[1-4] The overall incidence of hip displacement in children with CP has been estimated to be approximately 35%.[1,5] The severity of hip pathology is directly related to the severity of neurologic involvement[6] and is highly correlated with gross motor impairment as graded by the Gross Motor Function Classification System (GMFCS).[7]

A true hip surveillance program is a population-based, targeted evaluation of children with CP with a regular schedule of physical examination and hip radiographs. The targets of hip surveillance programs are the entire population at risk.[2] The primary goal of such programs is to ensure that progressive hip displacement is detected early enough to enable timely referral for orthopaedic assessment and treatment.[8] Early identification and orthopaedic intervention has been shown to alter treatment outcomes, reduce the number of required reconstructive surgeries, and avoid the need for future salvage surgery.[5,9,10]

Based on the growing evidence supporting hip surveillance for children with CP, many developed countries have adopted national hip surveillance programs.[2,8-12] However, no such established national surveillance

program exists in the United States. This lack of a uniform approach for the screening of this relatively common pathology likely results in tremendous national, state, and hospital practice variation.[13]

The purpose of this article is to review the epidemiology and pathophysiology of neuromuscular hip dysplasia in CP, define hip surveillance in the setting of this patient population, to describe some of the best executed international hip surveillance programs, and to discuss efforts here in America that are being undertaken to begin the process of implementing such screening programs.

Pathophysiology and Clinical Relevance of Hip Dysplasia in Cerebral Palsy

Studies of the geometry of the hip in children with CP have demonstrated notable differences when compared with typically developing children. In spastic children, muscle imbalance causes abnormal forces across the hip joint.[1,5] Over time, increased activity of the adductors, hip flexors, and hamstrings abnormally alter the joint reactive force, gradually causing the proximal femur to subluxate from the acetabulum.[8,9] These forces also create anatomic differences and prevent the physiologic remodeling of immature hip geometry including increased anteversion, increased neck-shaft valgus (increased head-shaft angle), epiphyseal valgus, and acetabular dysplasia. The resulting pathology can lead to hip subluxation and eventual dislocation without appropriate monitoring and treatment. The risk and severity of hip dysplasia is directly correlated with the degree of neurologic impairment.

Proactive treatment of subluxated hips in ambulatory children is universally accepted because well-reduced hips provide a stable platform for walking in stance phase.[8,9] Intuitively, a reduced hip has a potentially greater range of motion in abduction when compared with a dysplastic hip. Furthermore, there is a theoretical reduction in the rate of osteoarthritis in well-maintained hips because deformation of the femoral head and damage to the articular cartilage occurs in long-standing dislocation as a result of the contact pressure created by the abductor musculature.[14]

The rationale for hip management in nonambulatory children with CP differ. The maintenance of adequate range of motion is important for proper perineal care.[2,4] For nonambulatory patients who retain the ability to transfer or stand, a reduced hip provides a more stable platform and improves the likelihood of maintaining this activity with aging, which may maintain and improve quality of life.[15,16] Hip subluxation and dislocation frequently lead to pelvic obliquity in this population, negatively affecting seating balance and tolerance. This pelvic obliquity may contribute to the creation or progression of neuromuscular scoliosis.

The treatment or prevention of pain is the main goal of proactive hip surveillance and treatment in all children with CP. Bagg's study is commonly cited because it supports the concept that dislocation and subluxation can cause hip pain in older patients with CP.[4] More recent works by Ramstead and Wawrzuta support an increased incidence of hip pain in patients with greater radiographic hip subluxation and greater GMFCS levels.[17,18] Because of this potential for pain with neuromuscular hip subluxation or dislocation, preventing notable hip subluxation in all affected patients is a reasonable goal.

However, early displacement is rarely painful, and advocates of hip surveillance do not use pain as a deciding factor in the management of neuromuscular hip dysplasia. Treatment of neuromuscular hip subluxation should begin when displacement is noted on radiographs. The treatment may include continued radiographic surveillance, soft-tissue surgery, or hip reconstruction depending upon age and percent displacement.

Clinical examination is an important aspect in the assessment of a child with CP. Caregivers may notice a reduction in abduction or increased difficulty in perineal hygiene that may be due to increased tone and/or hip subluxation. Although loss of abduction is associated with hip dislocation, clinical examination alone is insufficient to assess the amount of subluxation; the degree of hip abduction does not correlate with radiographic measures of subluxation.[8,9] Nonetheless, when passive hip abduction is less than 30°, hip subluxation is likely and radiographs are clearly warranted.

Radiographic Assessment

All current hip surveillance protocols rely upon radiographic measures and not physical examination. An AP radiograph of the pelvis is needed to assess the hips. In routine surveillance, an orthogonal view is not required, such as in frog-leg lateral. The pelvic radiograph is obtained supine with the legs near parallel, the patellae facing upward, and the spine supported to reduce lumbar lordosis from hip flexion contractures.[20] The rotation of the pelvis should be minimized. Although improper positioning of the pelvis may result in incorrect measurements, in radiographs that are reasonably obtained, more error can be noted between examiners than because of small alterations in technique[19] (Figure 1).

The migration percentage (MP) is the most commonly used measurement to quantify hip subluxation. This is a measurement of the relative amount of femoral head lateral to the acetabulum and has been shown to

Figure 1

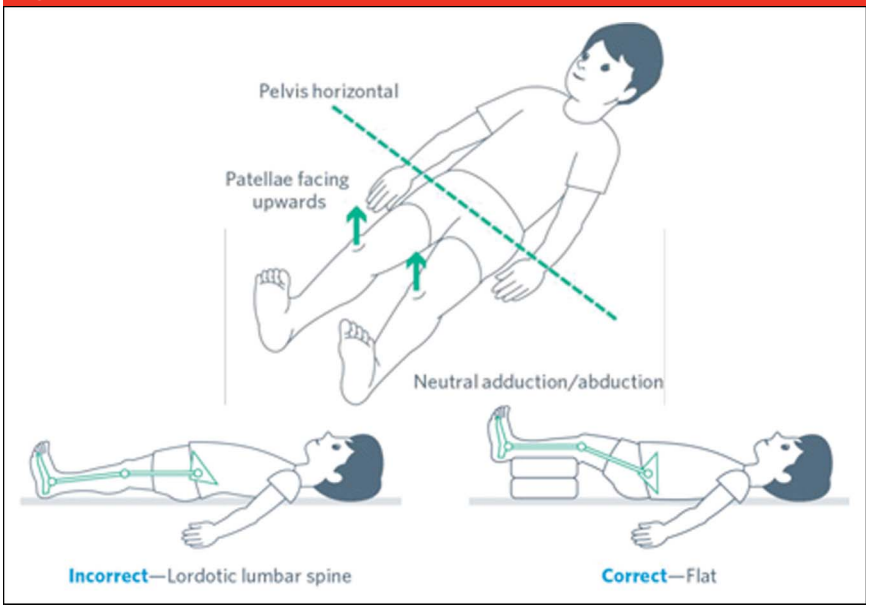

Illustration showing AP Pelvis radiograph Positioning. (Copyright Bill Reid, The Royal Children's Hospital, Melbourne, Australia.)

Figure 2

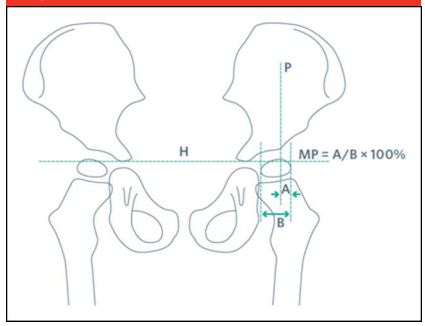

Illustration showing Reimer's Migration Percentage (MP).[8] (Copyright Bill Reid, The Royal Children's Hospital, Melbourne, Australia.)

be a reproducible measure.[21,22] Using Perkin's Line as a marker of the lateral margin of the weight-bearing portion of the acetabulum, the width of the femoral epiphysis is measured (Figure 2). Dividing the portion of the epiphysis lateral to Perkin's Line by the entire width results in the MP. An accurate measure of the subluxation should be recorded at each visit. The practice of measuring MP has been shown to be reliable and valid.[12,22-24]

The use of the acetabular index, or Sharp's angle in skeletally mature patients, to measure acetabular dysplasia is also helpful. This is measured as the angle subtended from Hilgenreiner Line and the line connecting the lateral edge of the acetabular sourcil and the triradiate cartilage.

Risk and Gross Motor Function Classification System Level

The risk of hip subluxation and eventual dislocation is directly correlated with the GMFCS[7,19] (Figure 3). GMFCS is a five- level scale used to describe the ambulatory ability of the patient: 1 is ambulatory without assistive devices and 5 is non-ambulatory without head control, requiring a caregiver for wheelchair control. Forty-seven percent of GMFCS V patients had greater than 40% subluxation at age 7 years.[2,6] Throughout childhood, the total risk for displacement >30% (the threshold for a hip at risk for continued displacement) is approximately 90% in GMFCS V, 70% in GMFCS IV, 40% in GMFCS III, and 15% in GMFCS II patients.[5,19] The risk in GMFCS I is considered to be negligible, except in hemiplegic patients with hip involvement, the so-designated Winter Gage Hicks (WGH) type IV hemiplegia.[25] These patients with hemiplegia are those with gait impairments including hip and knee flexion, internal hip rotation, and equinus. These patients require continued surveillance until skeletal maturity or stability.

International Hip Surveillance Models

Organized hip surveillance efforts in the United States have been limited despite its recognized importance by North American pediatric orthopaedic surgeons.[5,13] In contrast, national and regional surveillance programs have been incorporated successfully in Australia, parts of Europe, and British Columbia[8,24,26,27] as part of evidence-based care for children with CP.[12] Although the details of each surveillance schedule vary, every program largely considers age and GMFCS in schedule determination—frequency of radiographic and clinical follow-up—because both are correlated with dislocation risk.[1,3,7] As stated, each use the MP as the primary radiologic measure of subluxation.

Scandinavian Model

The first national hip surveillance model was established in southern Sweden in 1994 and is now used across Scandinavia (CPUP Program).[26] Clinical evaluations are standardized and completed by local physical and occupational therapists every 6 months in all children with a diagnosis of CP until the age of 6 years, and then yearly thereafter.

Examination includes an evaluation of gross and fine motor function as well as upper and lower extremity range of motion. Radiographic surveillance schedule is determined by the GMFCS level and evaluated by an orthopaedic consultant.

GMFCS I: no radiographic evaluation.

GMFCS II: evaluation at age 2 and 6 years.

GMFCS III-V: annual radiographic evaluation that commences immediately after diagnosis and continues through age 8 years. After 8 years of age, the interval between radiographs is individualized based on previous clinical and radiographic findings. After closure of the triradiate cartilage, if MP < 33%, radiographic examinations are discontinued.

Preventive surgery including soft-tissue release, varus osteotomy of the proximal femur, and/or pelvic osteotomy are recommended in the presence of subluxation, defined as a MP > 33%.[26] However, specific surgical decision making is left to each patient's local orthopaedic surgeon. More recently, the CPUP score was established in conjunction with this surveillance model to better categorize patients by risk of subsequent subluxation/dislocation to further guide surgical decision making.[28] This score incorporates MP, age, GMFCS and head-shaft angle, a measure of coxa valga, but the development of this score has not altered surveillance guidelines as of yet.

Australian Model

The most recognized international model is the Australian model, which was developed by a national working group and external consensus process between 2006 and 2008, implemented in 2008 and revised in 2014 based on intervening data collected in aggregate.[8,29] Surveillance commences between 12 and 24 months for *all* children identified with CP, regardless of the projected GMFCS level. Surveillance is conducted by physical therapists who complete a clinical assessment including an evaluation for changes in functionality, pain, and/or changes in gross motor function. The physical examination includes an evaluation of the spine, leg lengths, hip range of motion, and gait. As in the Swedish model, the clinical examination is augmented with a single standard AP pelvis radiograph.

After the initial assessment, the surveillance schedule differs by the GMFCS level,[8,11] and all patients are referred for orthopaedic consultation in the presence of hip pain and/or an unstable (increasing) MP and/or MP above 30%.

GMFCS I: repeat clinical evaluations without radiographic examination at age 3 and 5 years. If the GMFCS level is confirmed, the patient may be discharged from surveillance unless the patient has hemiplegic CP classified as WGH IV.

GMFCS II: repeat clinical and radiographic evaluation 12 months after commencement. If MP is stable, repeat evaluation at 4 to 5 years of age; if MP is unstable/abnormal, repeat yearly until stabilized. At 4 to 5 years, if MP is stable, repeat evaluation at 8 to 10 years; if MP is unstable/abnormal, repeat yearly until stabilized. At 8 to 10 years, if MP is stable, discharge from surveillance; if unstable/abnormal, repeat yearly until stabilized or the patient has reached skeletal maturity.

GMFCS III-V: repeat evaluation 6 months after commencement. If MP is stable, repeat evaluations every 12 months; if unstable/abnormal, repeat every 6 months until stabilized. For GMFCS III/IV, at 7 years, if MP is stable, radiographic examination is discontinued until pre-puberty, wherein radiographs will recommence every 12 months

Figure 3

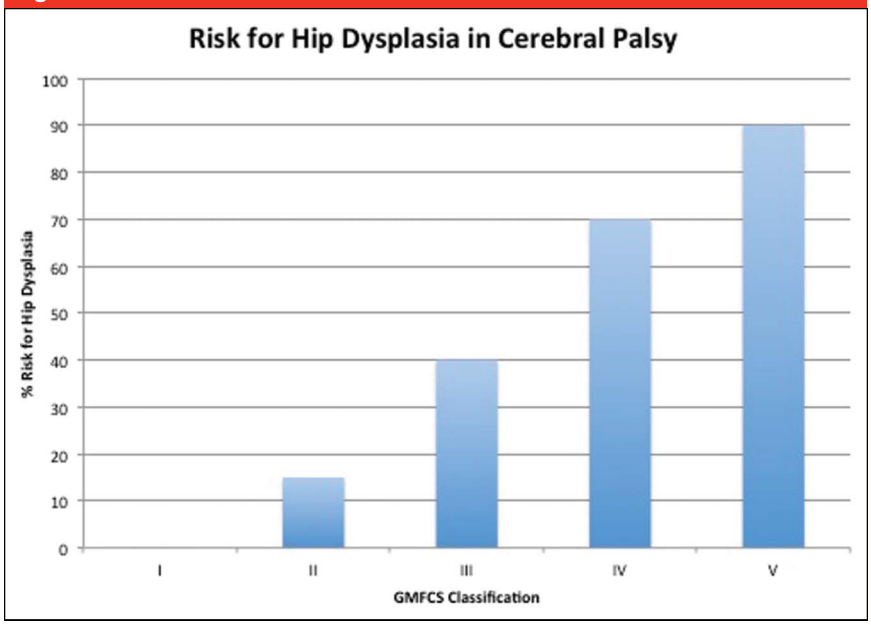

Chart showing the risk of Hip Dysplasia as a Function of Gross Motor Function Classification System (GMFCS).[19]

until skeletal maturity; if MP is unstable/abnormal, repeat every 6 months until stabilized. For GMFCS V, yearly radiographs are obtained until skeletal maturity, regardless of MP.

WGH IV: Regardless of GMFCS classification, hemiplegic patients classified as WGH IV have an increased risk of late dislocation. Review WGH classification at age 5 and 10 years. If confirmed type IV, repeat clinical and radiographic examination at age 10 and yearly thereafter until skeletal maturity.

For GMFCS III-V and WGH IV, surveillance should continue yearly *after* skeletal maturity in the presence of progressive scoliosis, pelvic obliquity, deteriorating gait, or an abnormal MP. If at any time an individual patient's GMFCS level classification changes, his/her surveillance schedule should reflect these changes.

British Columbian Model

The most recently established program was initiated in 2012 and is primarily based on the surveillance practices and outcomes in Australia and Sweden.[27] Clinical evaluations are standardized and completed by each patient's physical therapist. This evaluation includes verifying any difficulty with pain, function, or care, determination of GMFCS, and hip range of motion. Clinical examinations commence at diagnosis for all children identified with CP, and follow-up evaluations are completed according to the GMFCS.

GMFCS I-II: clinical examination repeated yearly until age 5 years. Standard AP pelvic radiograph at 5 years. If radiographs appear normal, discharge from surveillance.

GMFCS III: after the initial clinical examination, repeat examination and commence radiographs at 24 months of age and yearly thereafter until age 6. After age 6 to skeletal maturity, repeat clinical examination yearly and radiographic examination every 24 months.

GMFCS IV-V: after the initial clinical examination, repeat examination and commence radiographs at 24 months of age and biannually thereafter until age 6. After age 6 to skeletal maturity, repeat evaluation (clinical and radiographic) yearly.

WGH IV: schedule per the GMFCS level I–II until age 5. Thereafter, yearly clinical and radiographic examinations until skeletal maturity are recommended.

Referral to an orthopaedic consultant for preventive surgical consideration occurs when MP is greater than 30%, hip abduction ≤30°, the patient experiences clinical deterioration in hip abduction or extension, the patient/caregiver reports worsening in function, and/or the patient has an increase in hip pain.

International Hip Surveillance Outcomes

Both the Australian and Swedish protocols have been successful in identifying children at risk and preventing dislocations in their respective populations. However, as was noted by Hägglund et al,[30] the success of these models is in large part dependent on the setting in which they are implemented. Each require the identification of all children with CP, universal access to surveillance, prompt access to orthopaedic surgeons, and buy in from caregivers, providers, and policy makers. Nevertheless, their outcomes are encouraging.

After implementation of surveillance in 1994, the cohort of children diagnosed with CP born in Sweden between 1992 and 1997 were compared with a historical cohort of children who were not enrolled in a surveillance program (born between 1990 and 1991). The incidence of dislocation dropped from 9% in the control population to 0.5% in the first surveilled cohort.[2] Children born between 1998 and 2007 were included in a 20-year review, and the dislocation rate in this cohort was 0%.[30] Preventive surgery (soft-tissue release or varus femoral osteotomy) was undertaken in 15% of the first study group and 12% of the second study group. Repeat surgeries were common in both study groups and not considered a failure of treatment. No salvage surgery was required in either study group. Based on these results, the authors of both follow-up studies conclude that preventing dislocation is eminently possible.[30]

The complementary comparison between the surveilled cohort in Sweden and a cohort in Norway receiving typical presurveillance care revealed a stunning difference in dislocation rates—0.7% versus 15.1%, respectively, despite a markedly lower surgical rate in the surveilled population (31.6% versus 44.5%). On average, the surveilled population underwent preventive surgery 2 years younger than the nonsurveilled population.[30]

Five years after the initial implementation of the Australian consensus statement, a review of pooled data from several participating states highlighted the efficacy of the surveillance protocol, wherein all children with progression of MP > 30% were identified and referred for orthopaedic consultation in a timely manner.[8] This is consistent with pilot data from Dobson,[9] who demonstrated that with early on-going surveillance, the need for salvage surgery was eliminated. Surveillance led to an overall increase in soft-tissue/preventive surgery, a decrease in reconstructive/osseous surgery, and the eradication of dislocations, with an overall decrease in age at preventive surgery

(4.2 versus 8.3 years), which parallels what was seen in Scandinavia.

Data from Queensland further support the Australian model. Of 1,115 children included for study, 314 (28%) presented with or developed hip subluxation (MP ≥ 30%) during surveillance, and *all* 314 were referred for orthopaedic review with a 0% resultant dislocation rate.[10]

Cerebral Palsy Hip Surveillance Efforts in the United States

Currently, no formal hip surveillance programs exist in the United States. The biggest hurdle in establishing a formal national hip surveillance program in the United States is the identification of all children at risk for CP and neuromuscular hip dysplasia. Currently, no nationwide surveillance program exists for CP and other similar disorders. The Centers for Disease Control have CP surveillance programs in four states for epidemiological research purposes only (Georgia, Wisconsin, Alabama, and Missouri). The United States does not have programs similar to those of Australia or Scandinavia, wherein all children with CP are identified and followed up in a registry-type format. This identification and continued follow-up is the key primary step in developing a care program that can effectively systematically evaluate these patients for hip pathology.

Although true hip surveillance remains elusive in the United States in the present day, many pediatric orthopaedic centers use their own internal hip radiographic schedule parameters, which, in large part, are based on established formal international hip surveillance programs. Many pediatric hospitals have made the screening of these children with appropriate radiographic examinations a priority. Using a cross-disciplinary approach, children with CP are identified by therapists, physiatrists, and pediatric orthopaedic surgeons, and have appropriate hip radiographs ordered and reviewed, according to familiar schedules dictated by the patient's age and GMFCS level.

Short of more formalized surveillance programs, the quality of hip screening and treatment of hip pathology in children with CP is subject to great center-to-center variability. The attitudes of pediatric orthopaedic surgeons toward the importance of hip health in CP drive some of that variation of care. Shore et al[13] recently investigated the attitudes and opinions of members of the Pediatric Orthopedic Society of North American (POSNA) by performing a society-wide survey.

With the help of the POSNA Quality/Safety/Value committee, a survey was developed and circulated to all POSNA members. Member attitudes and practices regarding CP hip surveillance were evaluated. More than 350 POSNA members completed the survey, for an approximate 35% response rate. Highlights from the survey include that most respondents treated pediatric patients exclusively (97%), worked in academic practice (70%), and were affiliated with a university (76%). Only 18% of respondents currently followed a regular neuromuscular hip surveillance program, half of which (44%, 30/69) had adopted the Australian CP hip surveillance guidelines. Despite this lack of surveillance implementation, respondents agreed that a dislocated hip in a child with CP was painful (90% agreement) and should be prevented by an active hip surveillance program (93% agreement). Furthermore, 93% of respondents indicated that they would follow a national surveillance program if one was in place. Age (79%), GMFCS (81%), and MP (78%) were all identified as critical elements of a hip surveillance program. Most respondents felt that a hip at risk for hip displacement had an MP between 20% and 30% (57% of respondents); furthermore, respondents felt that surgery should be used once the MP exceeded 40% (50% of respondents).[13]

The American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) is a multi-disciplinary professional educational society aimed at the scientific education of health professionals and researchers across a broad range of specialties dedicated to the well-being of people with and at risk for CP and other childhood-onset disabilities. In 2014, AACPDM began a thorough process of developing clinical pathways to help guide their members in a variety of aspects of the clinical care of children with CP, which included a Hip Surveillance Care Pathway.[31]

According to AACPDM, "a care pathway is a practical summary, including an algorithm, of evidence informed guidelines or the best evidence, for an aspect of care/services for individuals with childhood-onset disabilities to inform clinical practice." The algorithm is developed by an expert-panel consensus focusing on the current level of evidence in the literature. They use a "traffic signal" concept that is color coded to the level of current evidence, with "green light" indicating effective, "yellow light" indicating probably/possibly effective, "purple light" indicating expert opinion, and "red light" indicating not effective based on the current available literature.[31]

The care pathway includes the definition of hip surveillance, why it is important in children with CP, what should be involved in a routine hip surveillance program, the frequency of evaluation, and the desired referrals to pediatric orthopaedic surgeons that should be the goal of a comprehensive program. A reference section highlights the current evidence in the literature, and a practical tools section discusses proper

Figure 4

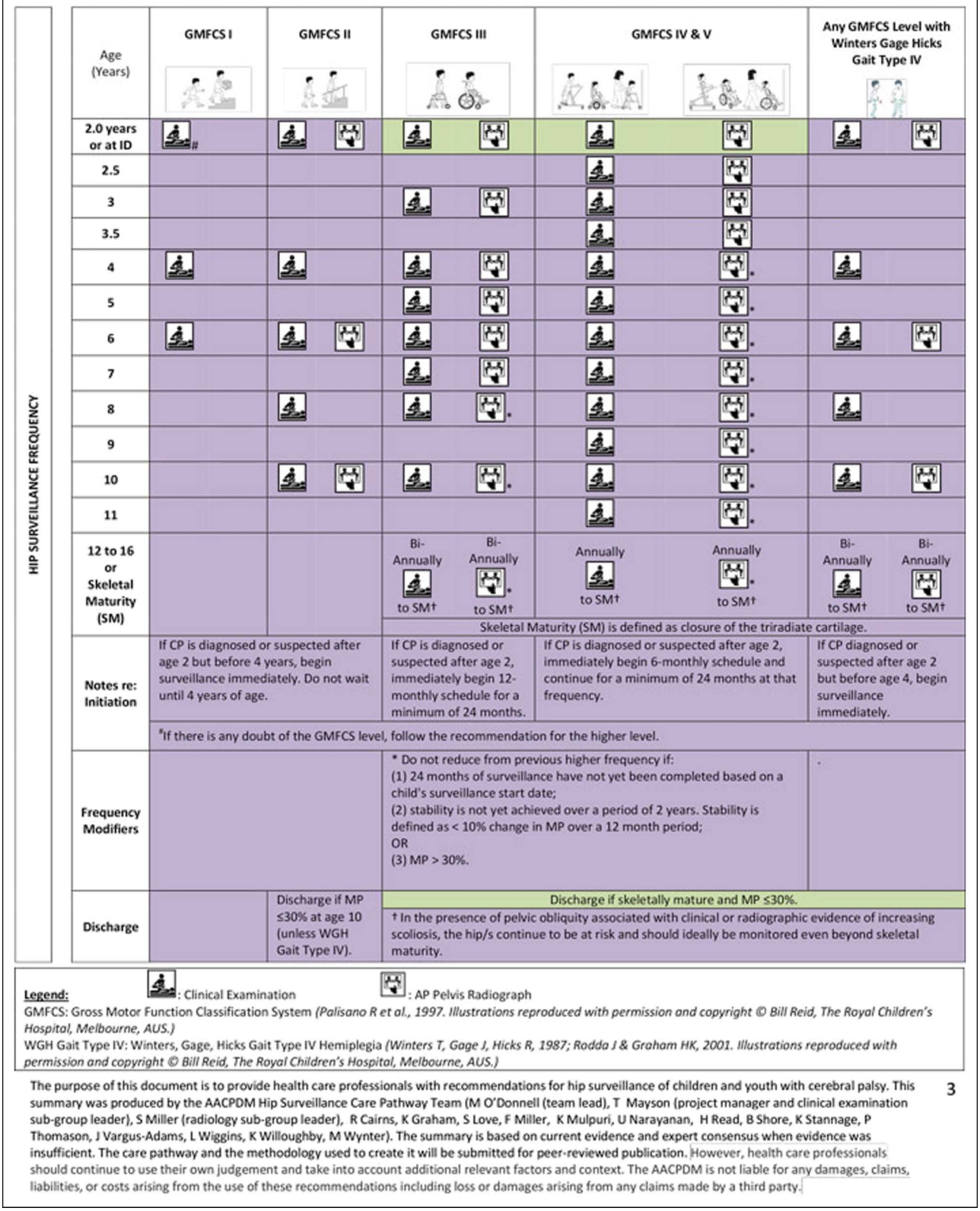

Table showing American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) hip surveillance care Pathway. GMFCS = Gross Motor Function Classification System. (Copyright AACPDM 2017.)

radiographic techniques and appropriate radiographic measurements.

The policy of AACPDM is to revisit and maintain the care pathways every four years, to ensure that new evidence affects the endorsed recommendations for the care of children with CP. As such, there is a comments section on the AACPDM website directed at feedback for the care pathways. As it currently stands, these tools are meant to be a living, evolving document over time. The current recommended examination and radiograph screening schedule, based on age, GMFCS level, and previous MP measurement are included in Figure 4.[31]

In the spirit of the multidisciplinary nature of AACPDM, the consensus panel of 17 experts represented many different specialties, including seven pediatric orthopaedic surgeons, which included four POSNA members, two from the United States. Although the work by the AACPDM is an excellent first step toward improving the awareness of hip surveillance in North America, many patients with CP in the United States are likely cared for by pediatric orthopaedic surgeons who may not be members of AACPDM. As indicated by the survey by Shore et al, POSNA members indicate that they would likely respond to and use a vetted hip surveillance program in the United States. The 2018 POSNA Clinical Trials grant was recently awarded to the authors of that survey to develop a POSNA recommendation statement on hip surveillance in CP that includes broad representation across its membership.

Summary

Systematic hip surveillance programs are popular internationally and are well-accepted as standards of care for children with CP in those countries. Evidence suggests that the use of these monitoring systems can prevent salvage surgery and improve the outcome of the treatment of hip dysplasia in these patients. Efforts to adopt these proactive strategies of hip monitoring in CP are evolving in the United States through the work of organizations like AACPDM and POSNA.

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