Screening for Developmental Dysplasia of the Hip:

A Systematic Literature Review for the

U.S. Preventive Services Task Force

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Abstract

Background: Developmental dysplasia of the hip (DDH) represents a spectrum of anatomic abnormalities that can result in permanent disability.

Objective: We sought to gather and synthesize the published evidence regarding screening for DDH by primary care providers.

Methods: We performed a systematic review of the literature using a best evidence approach as used by the U.S. Preventive Services Task Force. The review focused on screening relevant to primary care in infants from birth to 6 months of age, and on interventions employed before 1 year of age.

Results: The literature on screening and interventions for DDH suffers from significant methodological shortcomings. No published trials directly link screening to improved functional outcomes. Clinical examination and ultrasound identify somewhat different groups of newborns at risk for DDH. A significant proportion of hip abnormalities identified through clinical examination or ultrasound in the newborn period will spontaneously resolve. Very few studies examine the functional outcomes of patients who have undergone therapy for DDH. Due to the high rate and unpredictable nature of spontaneous resolution of DDH and the absence of rigorous comparative studies, the effectiveness of interventions is not known. All surgical and nonsurgical interventions have been associated with avascular necrosis of the femoral head, the most common and most severe harm associated with treatment of DDH.

Conclusion: Screening with clinical examination or ultrasound can identify newborns at increased risk for DDH, but due to the high rate of spontaneous resolution of neonatal hip instability and dysplasia and the lack of evidence of the effectiveness of intervention on functional outcomes, the net benefits of screening are not clear.

Key words: developmental dysplasia of the hip, DDH, hip dysplasia, mass screening, infants, systematic review

INTRODUCTION

Developmental dysplasia of the hip (DDH) represents a spectrum of anatomical abnormalities in which the femoral head and the acetabulum are in improper alignment and/or grow abnormally. The precise definition of DDH is controversial.[1, 2] The spectrum includes hips that are dysplastic, subluxated, dislocatable and dislocated. Clinical instability of the hip is the traditional hallmark of the disorder. In an unstable hip, the femoral head and acetabulum may not have a normal tight, concentric anatomic relationship, which can lead to abnormal growth of the hip joint and may result in permanent disability. DDH can lead to premature degenerative joint disease, impaired walking, and chronic pain.

Estimates of the incidence of DDH in infants vary between 1.5 and 20 per 1000 births.[3] The incidence of DDH in infants is influenced by a number of factors, including diagnostic criteria, gender, genetic and racial factors, and age of the population in question.[4] The reported incidence has increased significantly since the advent of clinical and sonographic screening, suggesting possible overdiagnosis.[2] In addition to a higher prevalence of DDH in females, reported risk factors for the development of DDH include a family history of DDH, breech intrauterine positioning, and additional *in utero* postural deformities.[5-7] However, the majority of cases of DDH have no identifiable risk factors.[8]

Self-limited hip instability is a common finding in newborns.[9] More than 80% of clinically unstable hips noted at birth have been shown to resolve spontaneously.[10] Because of the potential for subsequent impairment and the widespread belief that earlier treatment leads to improved outcomes, screening newborns for DDH has become commonplace. However, the high rate of spontaneous resolution raises uncertainty about

the most appropriate plan of action when a newborn has a positive screening examination for an unstable hip.

Intervention for DDH includes both nonsurgical and surgical options. A variety of abduction devices are used to treat DDH nonsurgically, with the Pavlik method among the most common. These devices place the legs and hips in an abducted and flexed position in an effort to promote proper alignment and stabilization of the hip joint. The duration of treatment varies from center to center. Complications of nonsurgical therapy are not trivial, with avascular necrosis of the femoral head among the most serious.[3]

Surgical intervention may be necessary when DDH is severe, when it is diagnosed late, or after an unsuccessful trial of nonsurgical methods.[11] Many surgical procedures are used to treat DDH, most of which involve manual reduction of the femoral head into the acetabulum, with or without additional procedures on the adductor and/or iliopsoas tendons, the femur, or the acetabulum. Preoperative management may include a period of traction, and postoperative management typically includes a period of fixed positioning in a spica cast. The duration and specific approach to pre- and post-operative management are highly variable. Surgical intervention places the hip at risk of avascular necrosis, in addition to standard operative risks including general anesthesia, intraoperative complications, and post-operative wound infections.

This evidence synthesis assesses the literature on screening and intervention for developmental dysplasia of the hip. It was conducted for the U.S. Preventive Services Task Force (USPSTF), which had no previous recommendations for this condition. Two systematic reviews of DDH have been published previously, one by the Canadian Task Force on Preventive Health Care (CTFPHC)[3] and another by the American Academy of

Pediatrics (AAP).[1, 4] This evidence synthesis summarizes this previous work as applicable, and incorporates studies published since these reviews were completed.

METHODS

The analytic framework and key questions (Figure 1) guiding the literature review were developed in consultation with liaisons from the USPSTF. We focused on screening in infants from birth through 6 months of age. The overarching question (KQ1) considers direct evidence linking screening to improved patient outcomes. The remaining key questions examine critical links in the logic underlying screening. To be effective, screening must identify cases of DDH earlier than they would be identified in the usual course of care (KQ2, 3). In addition, early identification must lead to earlier treatment, and earlier treatment must lead to better functional outcomes than late treatment (KQ5). Finally, the benefits of early identification and treatment must outweigh the harms of screening and of the treatments themselves (KQ4, 6). Finally, pending sufficient evidence of effectiveness, evidence regarding the cost-effectiveness of screening is considered.

Literature Search Strategy

Two recent systematic reviews of screening for DDH, by the AAP and the CTFPHC, targeted several questions also relevant to this review. We utilized the previous reviews to focus the search strategy and eligibility criteria for our review.[12] When questions had substantial overlap, we reviewed all studies identified in these

reviews and searched the literature for studies published subsequently (after 1996 for the AAP review and 2000 for the CTFPHC review).

Additionally, relevant studies were identified from multiple searches of MEDLINE (1966 to January 2005) and the Cochrane Library databases through June of 2004. Specific search strategies are available from the authors. Additional articles were obtained by reviewing reference lists of other pertinent studies, reviews, editorials, and websites, and by consulting experts. This strategy was modified for assessments of screening modalities in Key Question 3, in which we focused our review on the relevant literature beginning in 1996, the year in which the AAP review concluded.

Inclusion/Exclusion Criteria

Investigators reviewed all abstracts identified in the searches and the previous systematic reviews and determined eligibility by applying inclusion and exclusion criteria specific to key questions.[12] Full-text papers of included abstracts were then reviewed for relevance. Eligible studies had English-language abstracts, were applicable to U.S. clinical practice, and provided primary data relevant to key questions. Non-English literature with English abstracts was reviewed to identify any controlled trials. We excluded so-called teratological DDH, that occurring in children with neuromuscular disorders or other congenital malformations. For all included studies, initial screening had to be conducted in children less than 6 months of age, and screening studies needed to be prospective, primary care based or population based in design. Studies of risk factors also had to be primary care based or population based. Intervention and outcomes studies had to report results of children diagnosed before 6 months of age, and

interventions had to be employed earlier than 1 year of age on average. For intervention studies, we were particularly interested in functional outcomes, including: gait, pain, physical functioning, activity level, peer relations, family relations, school and occupational performance. For noninvasive interventions, another potential benefit is a reduced need for surgery later in childhood. Therefore, intervention studies were eligible if they reported one of these functional outcomes and/or a subsequent need for surgery. Studies that reported only radiological reports of anatomic structural relationships and development, which have not been shown to be valid predictors of functional outcomes, were excluded (indicated by a dotted line in the analytic framework). For avascular necrosis (AVN), the predominant harm from interventions, studies needed to report the rate of this complication in the treated patient population, meet age-based inclusion criteria, have at least 1 year of follow-up, and not experience excessive (>50%) loss to follow-up.

We used a "best evidence" approach[13]; that is, for each key question, we included studies with weaker designs only if better-designed studies were not available. Case reports, series with 5 or fewer subjects, editorials, letters, nonsystematic review articles, and commentaries were excluded from the evidence review.

Most studies of DDH are observational, uncontrolled or poorly controlled, and have significant flaws in design. To assess the quality of these studies, we considered the following: study design, clarity of diagnostic standards, comparability of subjects, variation in screening approach and/or intervention protocol, duration of follow-up, loss to follow-up, efforts to control for confounding and minimize bias, masking of outcome assessment, and validity and standardization of outcomes measured.[14]

Size of Literature Reviewed

Investigators reviewed 1,145 abstracts of English-language articles identified by the searches, excluding 679 citations on first review. Review of an additional 544 abstracts of non-English language articles identified no controlled trials. A total of 466 full-text articles were retrieved and reviewed; 416 were from the electronic searches and 50 were from reference lists or experts' suggestions (list of expert reviewers available upon request from the authors). The following met inclusion criteria: thirteen papers about risk factors; 59 about screening, including 3 controlled trials; 5 about harms of screening; 47 about interventions and harms of interventions, including no controlled trials; and 8 about cost.

RESULTS

Key Question 1. Does screening for DDH lead to improved outcomes (including reduced need for surgery and improved functional outcomes

such as: gait, physical functioning, activity level, peer relations, family relations, school and occupational performance)?

There are no prospective studies—either randomized or observational—comparing a screened to a non-screened population with measurement of functional outcomes after an adequate period of follow-up. There are also no controlled trials that compare surgical or nonsurgical treatment for early DDH to observation only.

In theory, early application of noninvasive treatments (e.g., a harness) to obtain a concentric and stable reduction of the femoral head in the acetabulum may obviate the need for surgery later on. However, the evidence that screening leads to a reduced rate of surgery is weak and indirect. The 2000 CTFPHC report, citing several descriptive studies, concluded "With serial clinical examination, the operative rate for DDH has decreased by more than 50% to 0.2-0.7% per 1000."[3] It should be noted that this reduction was observed at an ecological level: descriptive studies in screened populations were compared, indirectly, to unscreened populations or to historical rates. The studies were not comparative and did not report functional outcomes. In addition, while some studies suggest that surgical rates have declined since the adoption of universal screening programs, they do not indicate why. The decline might be attributable to increased rates of screening, but other factors, such as wider use of a period of observation before recommending surgery, could also account for the declining use of these surgical procedures.

The outcome measure used in many studies was the proportion of infants and children with DDH who had surgical intervention. If screening identifies more cases than usual care, it could reduce this proportion even if the same number of cases required

surgery as before. For this reason it is difficult to determine whether a decrease in the surgical rate over time reflects the efficacy of noninvasive intervention or the inclusion of additional cases in the denominator who are at little or no risk of requiring surgery.

The findings are also inconsistent: some studies observed a decrease in operative rates,[15-18] while others saw no change[19, 20] or an increase.[21-23] Ascertainment of cases was often flawed, and the studies span several decades, making it difficult to assess whether the varied results represent artifacts of data quality, secular trends, or differences in local practice styles.[24] These studies are also limited because they typically do not follow the screen-negative population with the same vigilance as the screen positive population, and experience significant loss to follow-up in the screen positive population that can bias the outcomes.

More recent studies also have conflicting results. In 1998, the MRC Working Party on Congenital Dislocation of the Hip reported operative rates in a randomly selected, population-based survey of 20% of all births in the U.K.[24] After adjustment for differences in ascertainment that had been overlooked in previous reports, the incidence of a first operative procedure for congenital dislocation of the hip was similar before and after screening was introduced (pre-screening rate range 0.66 – 0.85 per 1000, post-screening rate 0.78 per 1000 live births, 95% CI 0.72-0.84 per 1000). Even in the screening era, 70% of the cases reported by surgeons to the registry had not been detected by screening. In 1999, Australian investigators reported the operative rate in the post-screening era using an existing perinatal database and an inpatient discharge database to identify infants with congenital dislocation of the hip.[25] In contrast to the U.K. study above, they reported an operative rate of 0.46 per 1000 live births and found that 97.6%

of congenital dislocation cases were diagnosed before 3 months of age. The causes behind conflicting findings such as in these two studies are unknown.

Key Question 2. Can infants at high risk for DDH be identified, and does this group warrant a different approach to screening than children at average risk?

Risk factors are considered an adjunct to, rather than a substitute for, universal screening by physical examination. For example, the AAP recommends using risk factors to identify newborns whose risk for DDH may exceed the comfort level of physicians, prompting additional screening using ultrasound. The rationale for this approach is that, in high-risk newborns, clinical examination alone can miss many cases of DDH that ultrasound may be able to identify. The assumptions underlying this approach are (1) risk factors can identify a group of newborns at a high risk of DDH and (2) ultrasound is more sensitive than clinical examination for identifying infants at risk of complications from DDH.

In case control and observational studies, breech positioning at delivery, family history of DDH, and female gender have been most consistently shown to have an association with the diagnosis of DDH. Additional risk factors may include maternal primiparity, high birthweight, oligohydramnios, and congenital anomalies.

Primary care and population-based cohort studies[26-36] that include one or more of the major risk factors are summarized in **Table 1**. Consistently, only a minority (10-27%) of all infants diagnosed with DDH in population-based studies have identified risk factors (with the exception of female gender)[30, 32, 33, 35] and among those with risk factors,

between 1% and 10% have DDH.[30, 33, 35] This wide range illustrates the impact of the reference standard on the relative importance of risk factors. Those studies with a more strict standard for diagnosing "true" DDH, for instance limited to those patients that receive treatment, demonstrate substantially lower rates of DDH among those with risk factors. For example, a recent cohort study of 29,323 births at one hospital, the prevalence of treated DDH was 20/1000 in breech females, versus 110/1000 in this group if the diagnosis of DDH had been based upon an abnormal clinical exam. Additional rates of DDH using the more strict reference standard: 12/1000 in family history positive females, 4/1000 in breech males, 5/1000 and 0.3/1000 in females and males with no risk factors, respectively.[28]

Lehmann and colleagues conducted a meta-analysis of studies published through 1996 to estimate the probability of having a positive screening test for the three leading risk factors.[1] Breech females (84/1000) had a dramatically higher than average risk (calculated at 8.6/1000 for all newborns) of being screen-positive, followed by family history positive females (24/1000), breech males (18/1000), females with no risk factors (14/1000), and males with no risk factors having the lowest risk (3/1000). When considering these prevalence estimates, it should be noted that the reference standard used in Lehmann's synthesis was a positive Barlow or Ortolani test at the newborn screening examination. While this is a commonly used measure of the disorder, it may overestimate the number of infants with "true" DDH, i.e. those that do not spontaneously resolve and thus require therapy. The substantial differences in prevalence between the AAP review and the previous population-based study is likely to reflect different diagnostic standards, and impacts the predictive value of risk factors for DDH. Further

implications of the lack of a practically applied "gold standard" for diagnosing DDH is discussed in greater detail under KQ3.

Several potential biases should be considered in evaluating risk factor data. In studies where the examiner is aware of patients' risk factor status, the diagnosis of DDH may be overestimated due to more careful or thorough examinations or more aggressive follow-up and reexamination in infants with known risk factors. Moreover, in retrospective studies researchers apply criteria to improve the reliability of their record review; this approach, while necessary to conduct such a study, reduces the influence of an equivocal or inaccurate history. A predictor such as family history may be less reliable in a prospective, practice-based study than in case control studies which exclude patients (charts) that have equivocal or incomplete information about it. Finally, investigators' awareness of the subjects' final diagnoses could influence the way risk factor information is handled in retrospective studies.

Key Question 3. What is the accuracy of screening tests for DDH, and does screening for DDH lead to early identification of children with DDH?

The most common methods of screening for DDH involve the physical examination of the hips and lower extremities. Provocative testing includes the Barlow and Ortolani maneuvers, which involve adduction of the flexed hip with gentle posterior force, and abduction of the flexed hip with gentle anterior force, respectively. The Barlow test attempts to identify a dislocatable hip,[10, 37] while the Ortolani exam attempts to relocate a dislocated hip.[38] Due to variations in technique, the Barlow and Ortolani tests have been shown to have a high degree of operator dependence.[39] In addition,

confusion about the identification of a “click” versus a “clunk” on these tests, and the significance of each of these findings, can lead to disparate conclusions between examiners. Additional findings sometimes reported on clinical examinations for DDH in infants include asymmetry of gluteal and thigh skin folds, discrepant leg lengths, and diminished range of motion (particularly abduction) in an affected hip.[4]

To measure sensitivity of a test directly in a prospective study, infants who had negative initial screening tests must be followed and examined at older ages to identify false negative initial test results. Measuring sensitivity is also difficult because results of the Barlow test can be classified into several levels, rather than just two (“positive” or “negative”). Conversely, measuring specificity and false positives is difficult because, in most studies, all infants who have a positive screening test are treated with a nonsurgical intervention; the great majority improve, and it is impossible to say how many of them “responded” and how many of them did not have DDH in the first place.

Assessing the impact of a screening program on the rate of late diagnosis of DDH provides an indirect measure of sensitivity. It is apparent that screening tests performed soon after birth identify some individuals at risk of developing DDH sooner than they would otherwise be identified: most children would otherwise not come to medical attention until they present with crawling or gait delays or disturbances. However, it is difficult to quantify the impact of screening tests on the incidence of late diagnosis with the available literature. Studies of the impact of screening programs on the frequency of late diagnosis have had mixed results.[16-18, 21, 25, 40-52] Most of these studies report the experience of a screening program in a defined geographic or hospital service area over many years. The comparisons are ecological, and these studies have the same

methodological problems as those that examined the effect of screening on rates of surgical treatment (discussed above under KQ1). Some studies in this group reported that, after a screening program was adopted, late diagnosis was very rare, while others report that screening had no effect on the rate of late diagnosis, and that unexplained fluctuations in late diagnosis rates were observed from year to year within the post-screening era (Figure 2).[16-18, 20-22, 29, 33, 40, 45, 50, 53]

The lack of a practical confirmatory "gold standard" diagnostic test for DDH makes it difficult to assess—or define—false positives. Various reference standards appear in the literature, including positive clinical examination, ultrasound confirmation, radiographic confirmation, arthrography, persistence of abnormal findings on serial exam or ultrasound over weeks to months, diagnosis by an orthopedist, and use of treatment. The most meaningful reference standard defines "true" DDH as "those neonatal hips, which, if left untreated, would develop any kind of dysplasia and, therefore, are to be included in the determination of DDH incidence."[2]

To apply this standard, a cohort study must follow infants for a long enough period without applying any treatment, in order to determine whether or not the abnormal findings persist and lead to clinical problems. In one good-quality prospective cohort study that followed untreated infants for 2 to 6 weeks, approximately 9 of 10 infants with initially abnormal ultrasound examinations revert to normal.[2] Similarly, by 2 - 4 weeks of age, over 60% of infants identified at birth by abnormal clinical examination (Barlow or Ortolani tests) have reverted to normal when judged by repeat clinical examination or by ultrasound examination.[10, 37, 54] Longer prospective studies[28, 53-59] and a systematic review of observational studies of ultrasound screening[60] demonstrate that in untreated

hips, mild dysplasia without frank instability usually (consistently over 90%) resolves spontaneously between 6 weeks and 6 months.

The clinical exam approach to diagnosis for DDH shifts over time. Barlow and Ortolani tests become less sensitive as infants age, due to factors including increased strength, bulk, and size.[3, 4] In their place, assessment of hip abduction becomes the preferred examination, because infants with dislocated hips have increased contractures of the hip adductors.[4] In general, the specificity of examination improves as infants age, because the hips of the newborn infant are more likely to exhibit transient and clinically insignificant laxity than they will subsequently.[37] Two recent studies provide indirect insight into the changing signs of DDH as the infant ages. In a study of 1071 referred infants at one center, only 2 of 34 (6%) hips in patients with positive Barlow or Ortolani tests, confirmed as dislocatable by ultrasound, had any limitation in abduction at 1-2 weeks of age, suggesting that limited abduction has poor sensitivity in newborns.[61] Specificity of limited hip abduction in newborns was also poor: among 203 1-2 week old infants with limited abduction, <20% had abnormalities on ultrasound. These findings contrasted with older children: of the eight patients who presented after six months of age with dislocatable hips, hip abduction was limited in 7 (87.5%). In the second study, a prospective observational study limited to infants greater than 3 months of age (N=683), unilateral limited hip abduction had a sensitivity of 69% (156/226), and a specificity of 54% (247/457).[62] The reference standard in this study was any ultrasound abnormality; among the subset of subluxable and dislocatable hips, sensitivity of limited hip abduction was > 82%. Of the 136 patients with limited abduction and normal ultrasound findings at the initial exam, none showed exam or gait abnormalities at 5 years of age. Though not

conclusive, these studies suggest that hip abduction is a relatively insensitive and nonspecific marker of DDH in early infancy, but becomes more accurate after 3-6 months of age and with more severely affected hips.

Additional physical examination findings sometimes linked to DDH include asymmetrical gluteal and thigh skinfolds, and leg length discrepancy. No studies from the past 40 years were identified which assessed the value of these findings in diagnosing DDH. In 1962, Barlow pointed out the lack of utility of asymmetric skin folds due to their poor sensitivity and specificity,[10] and in 1961 Palmén studied 500 random newborns, finding that 27% had no thigh skinfolds, 40% were symmetrical, and 33% asymmetrical; 4 of these 500 babies had an abnormal provocative test of stability, of which 2 had symmetrical skinfolds.[63] Based on this scarce and unsupportive literature, it is difficult to conclude that these additional findings on exam are useful.

The degree of training and experience with the clinical examination of the hip in infants has been shown to be a strong predictor of the test characteristics. Pediatricians have been shown to have a case identification rate of 8/1000, whereas orthopedists identify approximately 11/1000.[1] Two studies show that having duplicate blinded examinations by a pediatrician and an orthopedist improves the sensitivity, specificity, and predictive value of clinical exam screening.[64, 65] Additional studies show that well-trained non-physicians, including physiotherapists and neonatal nurse practitioners, perform at least as well as physician examiners, and better than physician trainees.[66-68] In one single site longitudinal study, as the number of pediatricians involved in screening infants increased (holding steady the overall number of newborns screened), a greater number of cases of DDH were missed despite an increased rate of suspected cases

identified.[69] In other words, both sensitivity and specificity suffered when there was less centralized oversight of the newborn screening program and when fewer infants were screened, on average, by each pediatrician.

Studies comparing pediatricians with orthopedic surgeons often employ a study design in which the orthopedist reviews a subset of hips found to be positive or questionable by a previous examiner. This second exam may happen days after the initial examination. Also, the surgeons often have at their disposal the results of ultrasonography, and their clinical examination is not blinded from the ultrasound exam. Not surprisingly, such studies show a higher sensitivity and specificity of clinical examination in the hands of the specialist.

Use of Imaging to Screen for DDH

In addition to the clinical examination, ultrasonography and radiography are also used to screen for DDH. The use of ultrasonography and/or radiography in screening has been controversial, particularly due to reports of high false positive rates leading to unnecessary and potentially harmful follow-up and intervention.[70] Despite the controversy, ultrasound has been widely incorporated into DDH screening programs in many developed countries.[71, 72] Ultrasound methods include both static and dynamic assessments of the hip. As is the case with clinical examination, all imaging methods used to screen for DDH are variably subjective and operator-dependent.

In the first 4-6 months of life, ultrasound has been deemed to be a more appropriate test than radiographs for anatomic hip abnormalities as well as instability of the hip, due to incomplete ossification of the femoral head in early infancy. No study

addressed the comparative value of ultrasound to radiograph. However, there is strong endorsement of the superiority of ultrasound in the early months of life in the literature, ranging from historical studies reporting on timing of ossification and analyzing the technical challenges of hip radiography in the young infant,[63, 73] to contemporary systematic reviews.[1, 4]

However, ultrasound screening is not without its shortcomings. In addition to the high rate of identification of nonpathological hip findings summarized above, the most widely used ultrasound-grading system, the Graf classification,[74] has come under scrutiny. The Graf score is used in the vast majority of the screening literature to differentiate normal hips from immature hips, minor dysplasia, or major dysplasia; and stable from unstable, subluxable, and dislocatable/dislocated. Many studies base treatment decisions on these classifications. A study examining the reliability of Graf classification found that among normal hips, intra- and inter-observer reliability is quite high, with a 98% chance of having the same assessment on future readings. However, among ultrasounds read as abnormal by at least one person, intra-observer reliability was moderate (kappa = 0.41) and inter-observer reliability was fair (kappa = 0.28). In addition, knowledge of the patients' history and physical exam vs. blinded review of the ultrasound lowered the intra-observer kappa from 0.41 to 0.37.[75]

Another study found moderate agreement between observers with subjective ultrasound reading (kappa = 0.5), but this decreased to 0.3 when objective measurements of anatomic relationships were conducted. Grading of dynamic hip stability showed only moderate agreement between examiners (kappa 0.42) even when dislocated and dislocatable hips were grouped together. This study estimated that the decision to treat

would have been affected in 2.4% of cases due to discordance between reviewers.[76] Considerable effort had been given to standardizing ultrasound assessment in this study, including a training session and 100 repetitions of conducting measurements before the start of the study. Still another study found ultrasound reliability to be similarly suspect, with kappas ranging from 0.52 -0.68 and 0.09 to 0.30 for intraobserver and interobserver agreement, respectively, across seven anatomic measures used in grading DDH.[77] These findings raise concerns about the operator dependence of this evaluation for DDH, and may shed light on the variability of ultrasound screen positive rates found in the literature.

While there are no trials or comparative studies of a screened to an unscreened population, 2 randomized controlled trials[78, 79] and 1 nonrandomized controlled trial[53] provide some insight into the accuracy of clinical and ultrasound examinations. These trials reported data about test performance of one screening strategy versus another (Table 2). The first randomized controlled trial (RCT) compared universal ultrasound screening to selective screening at a population level.[78] In the trial, patients at the University of Trondheim, Norway were randomized over a 5 year period to one of two groups. In the first group, each of the 7840 patients received clinical exam and ultrasound. In the other group, 7689 received clinical exam alone or, if they had risk factors (abnormal exam, breech, family history, foot deformities), ultrasound and clinical exam. In the selective ultrasound group, 5 infants presented between 5-6 months with previously undiagnosed DDH, whereas in the universal screening group there was only 1 case of late diagnosis. In all these late-presenting cases, treatment with an abduction brace was implemented and the hips were reported to be normal upon follow-up, with

none requiring subsequent surgery. Overall treatment rates were equivalent in the two groups.

The second RCT[79] included 629 patients who had been diagnosed with unstable hips on screening examination and were referred to 33 specialty centers in the United Kingdom (UK). The subjects were randomized within the specialty centers to receive ultrasonographic hip examination (n=314) or clinical assessment alone (n=315). A total of 90% of patients in the ultrasound group received an ultrasound in the first 8 weeks of life; 8% in the no-ultrasound group received an ultrasound. Compared to those in the ultrasound group, infants in the no-ultrasound group were treated more often (50% vs. 40%) and earlier (98/150 vs. 42/117 treated in the first 2 weeks of life). The need for surgical treatment (8% vs. 7%), age at surgical treatment (31 vs. 29 weeks), mean number of visits at outpatient clinics (4 in each), total hip-related hospitalizations (30 vs. 23) and the occurrence of definite or suspected avascular necrosis (5 vs. 8) were not significantly different between the two groups. Thus, despite a higher rate and earlier initiation of treatment in the clinical examination only group, the non-functional "outcomes" of the two groups were quite similar. This suggests that, in the specialty setting, clinical examination alone may lead to a greater degree of unnecessary treatment than that which occurs when an abnormal clinical examination is followed up with evaluation by ultrasound.

A nonrandomized controlled trial conducted in 1994 compared 3613 infants in a universal ultrasound screening program to 4388 in a selective screening program, and 3924 who received only clinical examination.[53] In the selective ultrasound cohort, a positive clinical examination was considered to be a risk factor prompting ultrasound.

The authors concluded that the universal ultrasound cohort had a significantly higher treatment rate overall, but no higher rate among high risk infants. There was a nonsignificant trend toward a lower rate of cases diagnosed after 1 month of age in the universal screening patients. Among those not treated, many more children with mildly dysplastic hips were identified by ultrasound, resulting in more follow-up visits and ultrasounds for a greater number of patients without persistent DDH in the universal screening approach.

Table 3 includes studies of population-based or primary care clinic-based cohorts screened by clinical examination as well as ultrasound screening, published since the 1996 endpoint of the AAP review.[28, 70, 72, 80-83] Despite variation in the reference standards used in these studies, several important findings emerge. First, a high proportion of hips diagnosed with minor findings of dysplasia undergo spontaneous resolution**.** It is important to note that minor dysplasia is not identified by clinical exam, but only by ultrasound. Due to the identification of anatomic variations that are marginal and self-limited, the potential exists for over-treatment on the basis of ultrasound. On the other hand, in 4 of the 7 studies in Table 3, 38% - 87% of abnormal findings on clinical exam were not DDH, leading to a similar risk of unnecessary therapy on the basis of clinical examination.[72, 80, 81, 83] Very few of these studies followed patients longitudinally, particularly those patients who did not screen positive by exam or ultrasound.

Key Question 4. What are the adverse effects of screening?

Dislocation. While it has been suggested that the examination of already-lax newborn hips might cause injury or dislocation,[84] we identified little research that sought to test

this hypothesis. Three studies provide some insight.[85-87] An autopsy study examined 10 hips in stillborn infants, 4 of them full term and one at 28 weeks gestation, and found that after repeated (up to 30) "forceful" (amount of force not quantified) Barlow maneuvers six of the hips became lax.[85] Upon further study, it was determined that if the vacuum present in the joint capsule is disrupted, the hip becomes readily dislocatable.[85] A second study used an anatomic hip model and examiners ranging from clinicians with "many years" of experience to pediatric home visiting nurses who had just completed a training course in hip examination. This study reported that the average maximum force applied during the Barlow maneuver was 3 times that necessary to dislocate a dislocatable joint, and was consistently excessive across all levels of experience.[86] A study with living patients used dynamic ultrasound to monitor laxity during 4 successive examinations with Barlow and Ortolani and found no increased laxity over the course of these exams.[87] However, different examiners conducted each exam, so within-subject trends in stability may reflect differences between examiners as much as changes in the joints themselves.[87]

Radiation Exposure. A single center study of radiation exposure and increased theoretical risk of fatal cancers or reproductive defects reported the radiographic history of 173 patients who completed a course of treatment for DDH between 1980 and 1993. Based upon cumulative radiation exposure, males and females with DDH who had surgery (a marker for significantly elevated levels of exposure) were calculated to have a 0.09% and 0.12% increased risk of fatal leukemia and a 0.23% and 0.5% increased risk of reproductive defects, respectively.[88] There was no increased risk of fatal breast cancer in either gender. Attributable risks in nonsurgical DDH patients were approximately 1/2 to 1/3 of those reported for surgical patients. Given changes in technology and

management in the time interval since this data was gathered, it is not clear whether the level of radiation exposure documented in this study is still applicable.

Psychosocial. We found no published studies that sought to identify or quantify the psychosocial stresses of the diagnosis of DDH. No evidence was identified regarding adverse effects suffered by the child or family from false positive identification. Presumably, there is a cost borne by the family and/or society for the follow-up evaluation that ensues, but this has not been quantified. Other adverse effects may be experienced, but are not represented in the literature.

Key Question 5. Does early diagnosis of DDH lead to early intervention, and does early intervention reduce the need for surgery or improve functional outcomes?

10 different nonsurgical abduction devices are represented in the published literature and 23 different surgical procedures are used to treat DDH (see Evidence Report[12] for a complete listing). The indications and timing of treatment, and the protocol for the selected treatment modality vary from study to study, further obfuscating attempts at clarifying effectiveness. These circumstances are characteristic of interventions that have not been evaluated, or proven effective, in controlled trials.[89] Because no experimental or prospective cohort studies compare intervention with no intervention, the net benefits and harms of interventions for DDH are unclear, not only for infants diagnosed early but for all children.[90]

Poor functional outcomes from hip pathology may not manifest for decades. Thus, functional outcomes have not commonly been measured. Even when measured,

the effect of interventions on functional outcomes is unknown because of *1)* the absence of an appropriate comparison cohort and *2)* the substantial risk of bias stemming from short duration of follow-up, significant loss to follow-up, and/or nonstandardized, unblinded assessment methods without adequate rigor to ensure their validity (e.g. the surgeon's subjective report of the patient's function and pain). Due to these methodological problems, the evidence assessing whether interventions improve functional outcomes is poor, and study details have been excluded. Details about intervention studies[91-103] that included any assessment of functional outcomes are included in the Evidence Report.[12]

Given the absence of direct evidence from controlled trials, the case for the effectiveness of early intervention rests on less secure grounds, as follows:

1. Biological plausibility. It is biologically plausible that placing the femoral head into the acetabulum would facilitate normal development. While they are retrospective, careful analyses of late-presentation cases provide convincing fair quality evidence that late-presentation dislocations are often accompanied by premature arthritis, indicating that, at least in some cases, untreated DDH can have serious consequences.[104-106]

Based on this information, it is reasonable to hypothesize that relocating hips long before clinical symptoms occur may prevent morbidity and improve function. Unfortunately, an understanding of the effectiveness of interventions for DDH is confounded by the fact that many unstable and dysplastic hips undergo spontaneous resolution.[10] Thus, without a study design that includes an untreated cohort, the benefit attributable to an intervention remains in doubt.

Although the number of studies is small, it is clear that untreated DDH has an unpredictable course, with outcomes that are not universally poor. Among 628 Navajo infants born in a single region from 1955 to 1961, 548 were examined and radiographed during the first four years of life (20% in the first 6 months of life, but none as neonates).[107, 108] Eighteen (3.3% of those examined) were found to have hip dysplasia (including subluxation, but not including frank dislocation) by accepted radiographic criteria. None were treated. Seventeen of these 18 children were followed for seven to 19 years, and all had stable hips with normal x-rays.[108] When 10 of these patients were followed up at 33-37 years of age, none were aware that they had ever had a problem with their hips. While 6 did report a history of mild hip pain, this did not correlate with the degree of abnormality on x-ray. Additionally, all patients had normal function, engaged in light to heavy labor and were able to contribute to society without limitations.[107] Another study followed 51 consecutive patients with a normal clinical examination but evidence of dysplasia on x-ray. Altogether, 6 patients were lost over 5 years of follow-up. Forty-four affected hips (number of patients not reported) were normal after 5 years, 4 had undergone successful abduction therapy, and 20 were borderline on repeat imaging. No progression to subluxation or dislocation was noted in any of the hips.[109]

2. Reduced need for surgery. Early noninvasive intervention may reduce the need for surgery. This is a key observation that underlies previous recommendations favoring screening for DDH. As discussed earlier, however (KQ1), the evidence supporting this assertion is conflicting. More over, the need for surgery is a moving target: when they are observed, reductions in surgical rates might have occurred because of changing

indications or because of wider use of a period of observation prior to surgery, rather than because of screening itself.

Earlier intervention may reduce the risk of complications. Several observational studies examined the impact of age at the time of intervention.[25, 81, 95, 110-113] In one small study that included children initiating therapy for DDH from birth through 4 months of age, duration of treatment increased in a dose response fashion as the age at initiation of treatment increased, holding the severity of DDH steady.[81] In a separate series of patients undergoing surgery for DDH (70% of whom had failed therapy with a Pavlik harness), those 6-9 months of age (18 patients) required no additional corrective surgeries, whereas 29% of patients 10-11 months of age, 13% of patients 12-14 months of age, 26% of patients 15-18 months of age, and 30% of patients 19-24 months of age required additional surgical interventions.[110] Another study, based upon unadjusted analysis, reported that the average age of DDH cases complicated by avascular necrosis was > 15 months, whereas uncomplicated cases averaged 11 months of age.[111] Two additional studies found that intervention initiated after 6 months of age was associated with significantly higher rates of avascular necrosis.[95, 112] In a study that focused on late diagnosis of DDH, closed reduction failed in a similar proportion of cases in children 0-3 months as those 3-6 months, but failed significantly more frequently after 6 months of age (no upper age limit was identified, potentially biasing these conclusions).[113] Finally, a study of 55 children who underwent operative procedures for DDH between 1988 and 1998 found that procedures were less invasive in children less than 6 months. All children greater than 12 months undergoing a procedure for DDH required an osteotomy,

the most invasive procedure.[25] While inconclusive, these studies provide fair evidence that initiation of interventions after 6 months of age may carry added risks of harms.

In contrast, three retrospective observational studies did not support an effect of age on success of treatment.[94, 114, 115] The first reviewed the rate of success of closed reduction, and showed no difference among patients treated with this intervention at less than 6 months, 7-12 months, or 13-18 months.[114] A study limited to 168 children with hip subluxation or dislocation and a minimum follow up of 5 years, compared children in whom a Pavlik harness was successful with those requiring closed reduction and those who eventually required open reduction, and found that age was not a predictive factor of the success of nonsurgical therapy.[115] Finally, a study of 75 children with DDH treated within the first 14 weeks of life with the Pavlik method showed that age at initiation (ranging from 5 to 13 weeks) had no influence on duration of treatment, success rate, or AVN outcome at 1 year of age.[94]

It is possible that some relevant literature was excluded because we limited the review to studies in children whose intervention began within their first year of life. Within this period, conclusive evidence of a clear benefit of earlier intervention is elusive. The design of the studies cannot exclude other plausible explanations for the association between age at intervention and rates of surgery. One of these explanations is that passive abduction therapy may be less effective as children become stronger and more mobile beyond 6 months of age. Another is that the early-treated group includes a high proportion of children with mild disease that would have recovered without intervention, while the older children have persistent disease that would not have responded even if they had been treated earlier.

3. Improved radiographic appearance. Use of noninvasive treatments is often associated with improvements in radiographic or sonographic appearance. While radiographic reduction may be an essential step in the causal pathway from congenital dislocation to prevention of serious complications, radiographic outcomes have not been shown to be valid or reliable surrogates for functional outcomes. The most commonly used and widely accepted radiographic assessment is a 6-level scale initially described by Severin in 1941, based upon radiological appearance of hips in 16-24 year olds.[116] One study examined the validity of the Severin classification with functional outcomes in patients who had received surgery for dislocation of the hip, at an average of 31 years post-intervention.[117] The study found that x-ray findings (normal position of femoral neck and head, degree of arthritis and shape of the femoral head) were poorly correlated with the outcomes of range of motion and pain.

Two studies assessed the reliability of the Severin classification.[118, 119] Ali et al found intraobserver reliability among pediatric orthopedists in the UK with 7 or more years experience to be moderate to substantial (kappa ranging from .58 to .77), and interobserver reliability to be poor to slight in the intermediate Severin classes of II and III (kappa 0.19 to 0.20) and moderate (kappa 0.44 to 0.54) in the disparate Severin classifications of I (normal) and V (marginal dislocation). Ward found even less reassuring results.[119] Blinded assessments by pediatric orthopedists in this study were assessed by dichotomous observer groups as well as multi-rater groups, and found kappa scores in the range of 0.0 to 0.29 across the range of Severin classes, and no higher than 0.56 for overall agreement across any two surgeons. Even more concerning, the operating surgeon's unblinded scores showed uniform poor reliability (kappa 0.02 to

0.21) when compared to each of the blinded observer's scores. Despite the absence of studies supporting the reliability of radiographic measures, intervention studies rarely included blinded or repeated assessments of radiographic outcomes. Due to highly suspect validity and reliability, studies that reported only radiographic outcomes were excluded from further review.

4. Closer follow-up. Diagnosis leads to attentive follow-up of infants with DDH, facilitating quick detection and intervention. Thus, children undergoing early noninvasive therapy may benefit from closer follow-up and the physician's ability to react to a deteriorating condition more rapidly. Though limited, available evidence supports the notion that a high proportion of families follow through with initial referral.[29] However, we could not determine how many families adhere to ongoing follow-up.

Underlying the effectiveness of early diagnosis and early intervention is the degree to which families adhere to medical recommendations. One study assessed failure to follow-up with a specialty appointment after identification of newborns with an abnormality on exam or the presence of a risk factor for DDH.[29] This specialty clinic, a part of Britain's National Health System, followed a systematic approach to contacting non-attenders, including up to 2 letters to the family explaining the reason for referral, safety of ultrasound, and offering an appointment the following week, followed by contact with the general practitioner to persuade the family. With this approach, nearly 95% of patients followed up. The groups with the highest follow-up rate (>98%) included those with an unstable hip at the newborn exam and those with a positive family history. It may be unlikely that the average orthopedic clinic in the United States will

achieve an equivalent rate of follow-up, given access barriers and less robust efforts at contacting those who initially miss scheduled appointments.

A second study, based in the U.S., examined the rates of parental adherence to recommended abduction therapy with the Pavlik harness.[120] Of 32 patients treated by the same physician, only 2 families reported strict adherence to the physician's orders in a post-treatment questionnaire. Nonadherence was defined as failure to do one or more of the following: a) full-time use during the initial period of reduction when the hip was not stable, b) altering or deliberating misplacing the harness, c) discontinuing use of the harness for prolonged periods of time without permission. Nearly two-thirds of the mothers participating in the study had a college education or advanced degree; their age range was 17-40 years (average age 29 years). Harness therapy failed in 3 out of the 32 patients, and by the authors' report these cases were not more egregious in their degree of noncompliance than successfully treated children. The single exception was a mother who routinely removed or adjusted the harness because the child could not fit into a car seat due to limited adduction.[120]

Key Question 6. What are the adverse effects of early diagnosis and/or intervention?

Good quality literature examining harms of intervention for DDH would include a comparison of 2 or more (ideally randomized) cohorts, each exposed to a standardized intervention and followed over sufficient time (with limited loss to follow-up) to ensure complete ascertainment of the potential harms with an assessment of the effect of the measured harms on patient outcomes. Unfortunately, these studies have not yet been

conducted. In their absence, we reviewed the fair quality literature on adverse effects of both nonsurgical and surgical interventions.

The most well described adverse effect from interventions aimed at treating DDH is AVN of the femoral head. This is the most common adverse effect for both abduction therapy and surgical interventions. AVN severity ranges from a persistent but asymptomatic radiographic finding to a severe condition that causes growth arrest and can lead to eventual destruction of the joint. The rates described in the literature for AVN vary greatly for abduction therapy as well as surgical interventions. (Figure 3).[91-95, 97, 99, 101-103, 112, 121-129] The reasons for these disparate findings are not straightforward, and most likely relate to a complex and confounded set of variables including but not limited to the wide spectrum of the disorder, heterogeneous populations studied (age at intervention, specific type of DDH, previous interventions received), the variety of interventions and the poorly standardized approach to interventions (particularly the pre- and post-intervention phase of management), variable training and talent among the treating physicians, different lengths of follow-up across studies, and disparate approaches to follow-up in different health care systems. As calculated in the AAP review, meta-analytic rates of AVN range from 13.5 - 109/ 1000 infants who undergo treatment (non-surgical vs. surgical rates not specified).[1]

Additional harms from abduction therapy that have been addressed in the literature are typically mild and self-limited, and include rash, pressure sores, and femoral nerve palsy**.** All surgical interventions carry the risks inherent in general anesthesia, and those that involve open surgery also include the generic surgical risks of infection,

excessive bleeding, and wrong site surgery, though these receive scant review in the published literature and thus cannot be quantified.

A fair quality study assessing the long-term psychological impact on children of successfully treated DDH showed that parents and teachers found that children with DDH were more "disordered" than peers with no hospitalizations, 1 hospitalization, and multiple hospitalizations on the domains of "health," "habits," and "behavior."[130] This 1983 study implies (but does not quantify) extended hospitalizations for children with DDH as a rule, and thus may not be generalizable to the impact of treatment today.

Key Question 7. What cost-effectiveness issues apply to screening for DDH?

Several economic analyses of screening for DDH have been published.[79, 90, 131-136] Most concern the marginal benefit of ultrasound screening in relation to screening with clinical examination.[79, 90, 132, 133, 136] None of the available studies used quality adjusted life years, and none used models based upon U.S. data or the U.S. health care system. These analyses demonstrate that the economic impact of ultrasound screening is complex, reflecting that ultrasound may have mixed effects on diagnosis of DDH: it may identify false positive clinical examinations, reducing or shortening the duration of unnecessary treatments, but it also identifies many abnormalities in infants who have normal physical examinations, potentially leading to more early treatment and greater follow-up costs. The mixed results of the economic studies largely reflect mixed results of the clinical studies on which they are based. The best quality economic study, derived from a RCT (in the UK) of clinical exam screening versus clinical exam plus ultrasound, maintained

detailed records of utilization of medical services and related costs.[79] While the costs of ultrasound were predictably higher in the cohort receiving ultrasound, hospitalization costs in this group were lower. In sum, the overall direct medical costs for the two approaches were not statistically significantly different.[79] This study did not report indirect costs, such as missed work by the family, nor did it include the costs of long-term follow-up or complications.

DISCUSSION

As a condition that can result in impaired functional outcomes for children and adults, DDH merits the attention of primary care clinicians. However, there is no direct evidence that screening improves functional outcomes, and the evidence for several links in the analytic framework is weak. Table 4 summarizes the quality of the evidence.

The definition of DDH is variable, including dislocated, dislocatable, subluxable, and dysplastic hips. The benefits of early intervention are based on expert opinion along with mixed evidence that later diagnosis results in a greater likelihood for surgical intervention, and more complications. Using indirect comparisons, some studies suggest that earlier diagnosis is associated with better results, but these findings could be the result of lead-time bias, that is, the identification of DDH in a group of younger patients, in whom a higher rate of spontaneous resolution may lead to better outcomes, rather than the effect of earlier intervention. The outcomes of screened infants have not been compared to those of unscreened infants in an experimental or observational study.

Despite a paucity of evidence supporting its value in improving outcomes, universal screening for DDH is a well-established approach to the disorder. However, the approach to screening varies significantly. In addition to physical examination with

the provocative tests of Barlow and Ortolani and evaluation of range of motion emphasizing abduction of the hip, static and dynamic ultrasound are employed to identify anatomic abnormalities and stability of the hip, respectively. Some have recommended risk stratification to inform selective use of ultrasound, with females in breech positioning at delivery found to have the highest rate of clinical hip instability (84/1000). Yet, when a more conservative reference standard for DDH is employed, the value of ultrasound as an aid to diagnosis in those with risk factors is less conclusive. Some health systems have elected to employ universal ultrasound screening in an effort to reduce the incidence of late diagnosis of DDH. The use of ultrasound to further evaluate hips found to be unstable on clinical exam may reduce the rate of unnecessary treatment, but may also lead to higher rates of follow-up for hips that will ultimately spontaneously normalize. The reliability of DDH classification by ultrasound is questionable. Theoretical harms from screening include examiner induced hip pathology with vigorous provocative testing, elevated risk of certain cancers from increased radiation exposure from follow-up radiographic tests, and parental psychosocial stress from the diagnosis and therapy. None of these has been quantified in patients/families in clinical studies published to date beyond anecdotes.

It is known that a significant number of hips with positive screening tests, both by physical examination and by ultrasound, will normalize over time without intervention. This is particularly true of ultrasound in hips that are stable on clinical exam of the neonate: more than 90% of abnormal ultrasound findings in this situation have been shown to normalize spontaneously. While limited fair quality evidence exists to support the value of initiating treatment within the first 6 months of life, there is little to suggest

that immediate treatment in the neonatal period is associated with improved outcomes or a reduced need for subsequent surgery. However, no study has examined the effect of timing of treatment initiation, controlling for the degree of hip instability.

First-line intervention includes abduction bracing of the hips, which attempts to induce passive alignment of the hip. Several devices are used for abduction, with a wide range of institutional protocols. Failure of abduction therapy, or the occasional case of dislocated and clinically irreducible hips at presentation, leads to surgical intervention. The indications and protocols for surgery vary widely, as do the pre- and post-operative approaches to management.

Estimates of the effectiveness of therapy are confounded by spontaneous resolution of hip dysplasia, which has only rarely been assessed and never in a prospective or comparative fashion. The impact of interventions on functional outcomes is rarely addressed in the literature, and when addressed is of poor quality due to a lack of standardization within studies, and the absence of validated functional outcome measures across studies.

The most significant and common adverse effect of both nonsurgical and surgical intervention for hip dysplasia is avascular necrosis of the femoral head, which can lead to growth arrest and eventual destruction of the hip joint. The balance of benefits and harms of intervention is obscured by significant gaps in the available evidence. Assessment of the cost effectiveness of screening for DDH similarly requires more conclusive information about effectiveness.

Future Research

While the body of literature on screening and intervention for DDH has significant flaws, several recent studies provide valuable information on the screening evaluation of DDH. However, conclusive evidence is still absent. A more complete understanding of the natural history of spontaneous resolution of hip instability and dysplasia is needed to develop an evidence-based strategy for conducting screening and implementing therapy at the optimal time. Given the infrequent nature of DDH, multicenter studies of interventions that measure functional outcomes in a standardized fashion are needed. Studies designed to assess whether any clearly defined, reliable radiological markers predict functional outcomes would be a valuable step. Even more valuable would be patient-centered research that seeks to understand patient and family preferences as they relate to the process of care and short and long-term outcomes of DDH.

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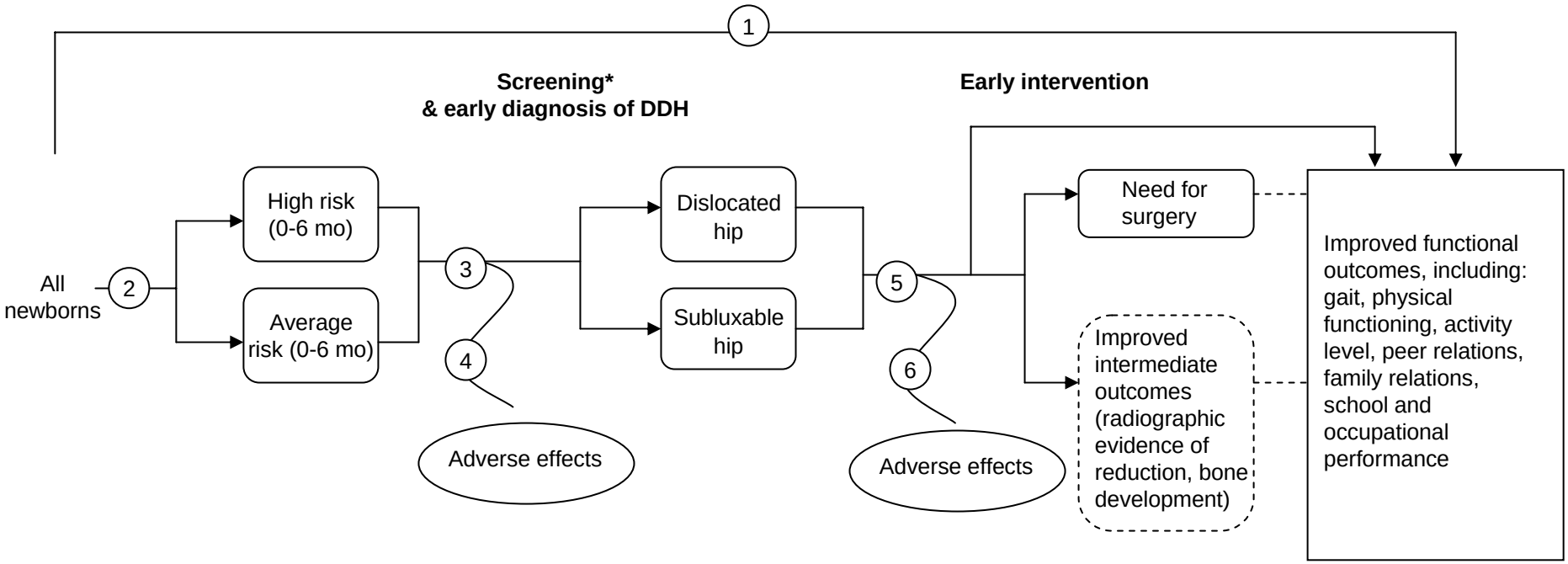
FIGURE 1. ANALYTIC FRAMEWORK
Screening for Developmental Dysplasia of the Hip (DDH)



*Screening examination (Barlow/Ortolani, Assymetry, ROM) and Radiographic evaluation (ultrasound, x-ray)

KEY QUESTIONS
Screening for Developmental Dysplasia of the Hip (DDH)

Arrow 1: Does screening for DDH lead to improved outcomes (including reduced need for surgery and improved functional outcomes such as: gait, physical functioning, activity level, peer relations, family relations, school and occupational performance)?

- **Arrow 2**: Can infants at high risk for DDH be identified, and does this group warrant a different approach to screening than children at average risk?
- **Arrow 3**: Does screening for DDH lead to early identification of children with DDH?
  - a) What is the accuracy of clinical examination and ultrasound?
  - b) How does the age of the child affect screening parameters?
  - c) How does the educational level and training of the screener impact screening?
- **Arrow 4**: What are the adverse effects of screening?
- **Arrow 5**: Does early diagnosis of DDH lead to early intervention, and does early intervention reduce the need for surgery or improve functional outcomes?
  - a) Is the likelihood of surgical intervention reduced in children diagnosed at an earlier age?
- **Arrow 6**: What are the adverse effects of early diagnosis and/or intervention?
- **Key question 7 (no arrow)**: What cost-effectiveness issues apply to screening for DDH?

FIGURE 2. VARIATION IN LATE DETECTION RATE BY YEAR OF STUDY PUBLICATION AND SCREENING METHOD

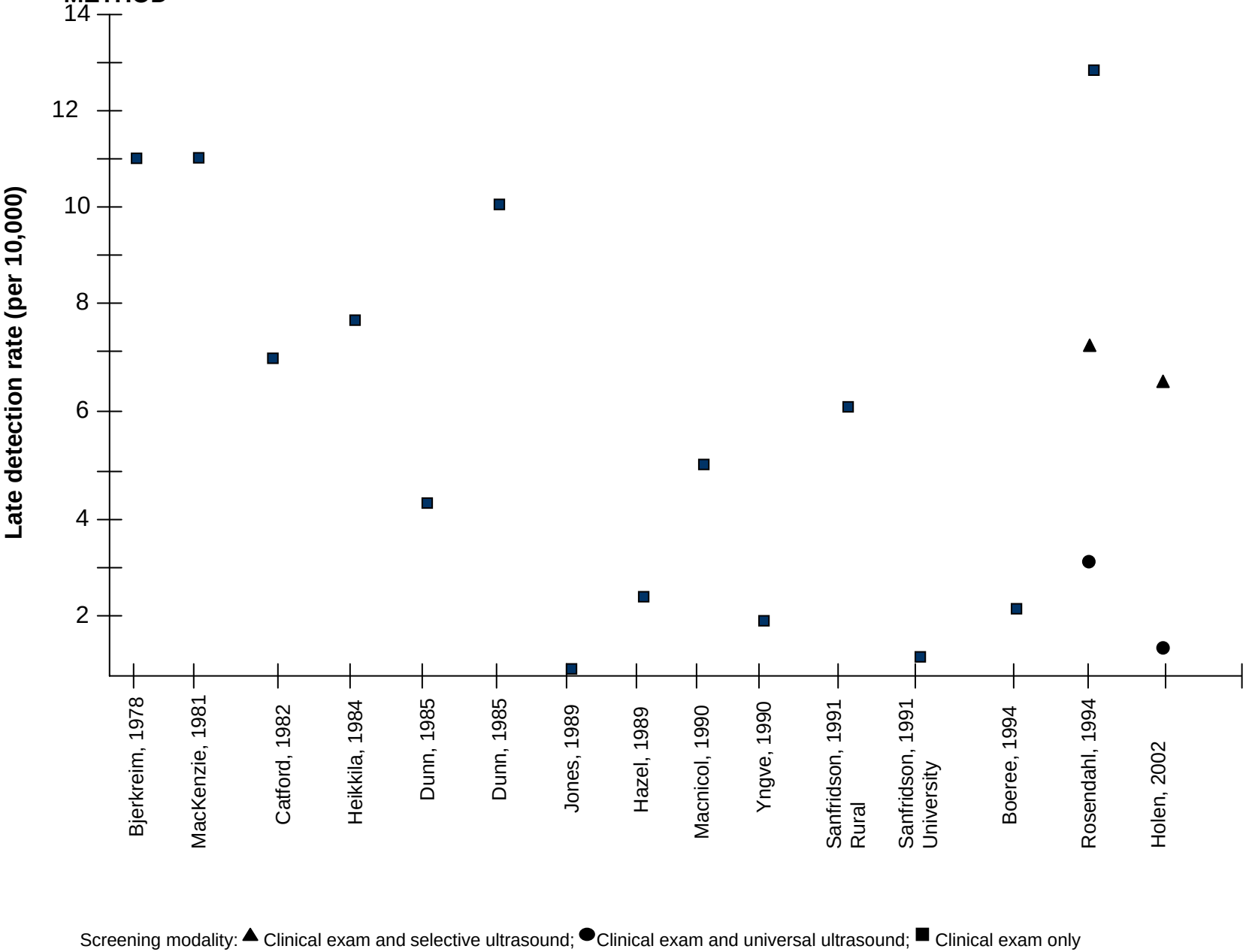
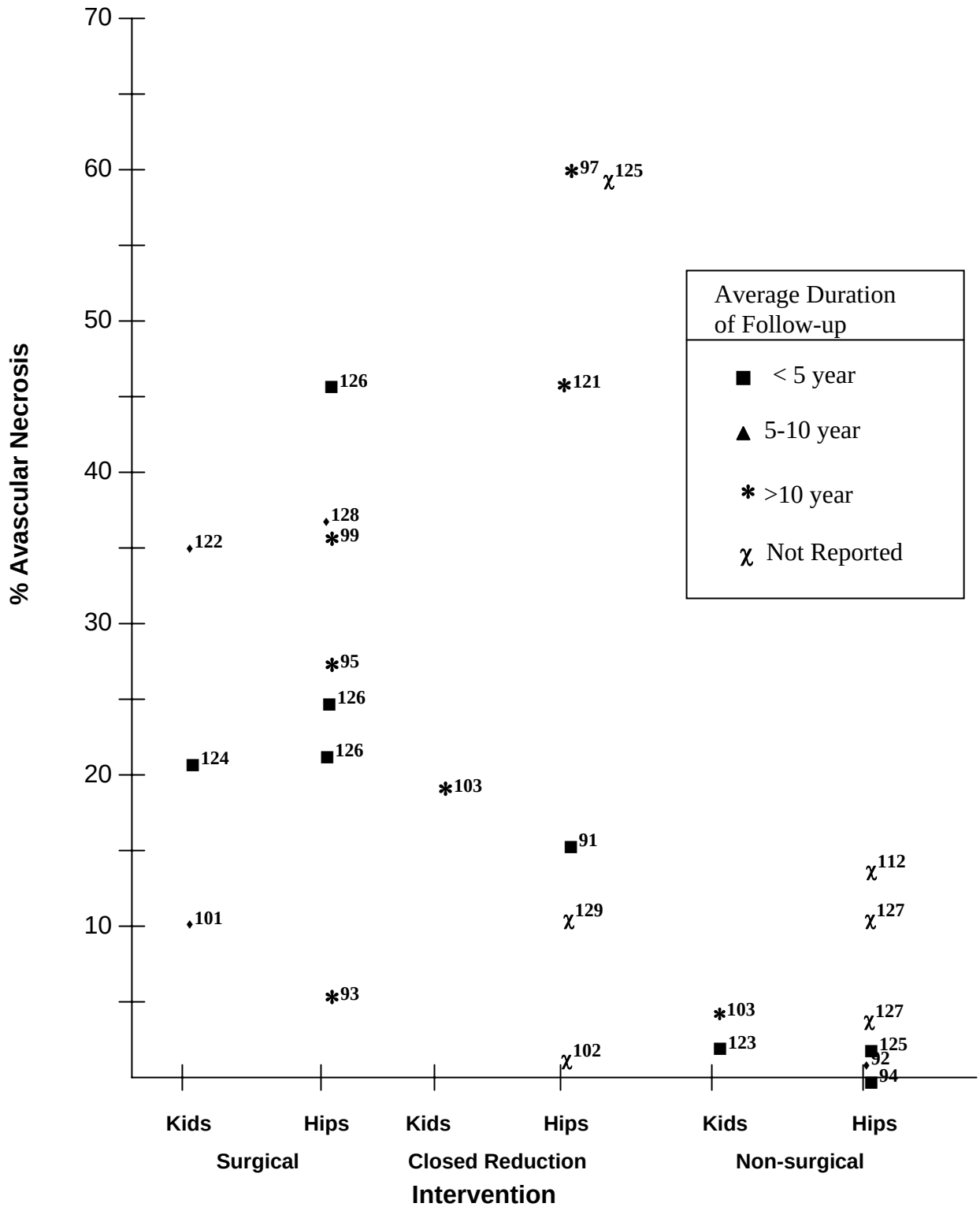


FIGURE 3. RANGE OF PUBLISHED RATES OF AVASCULAR NECROSIS



Note: due to the fact that studies were inconsistent in reporting % of children or % of hips with AVN, both are included in this figure.

TABLE 1. Risk Factors

Author, Year	Overall *N*	No. With DDH	Risk Factor	Relative Risk	Patient With Risk Factor Who Have DDH, %	DDH Positive Cases With Risk Factor, %	Quality Rating
Andersson, 2001	6,571	78 D or I*	Breech	3.72	D or I: 3.89%	D or I : 12.8%	Fair
		13 Treated		11.08	Treated: 1.56%	Treated: 30%	
Artz, 1975	23408	312	Breech	6.35	6.64%	22.10%	Fair
			First Born	1.31	1.71%	68%	
			Female	4.15	2.15%	79.50%	
Bache, 2002	29,323	2340 based on screening; exam 92 treated	Breech	1.95, 4.14[†]	7.8%, 1.3%[†]	27%	Good
			Family History	3.4, 3.8[†]	13.4%, 1.2%[†]	7.60%	
			Female	1.7, 1.9[†]	6.6%, 0.59%[†]	91%	
			Breech Female	2.8, 6.6[†]	11.0%, 2.0%[†]	14%	
			Family History and Female	5.1, 3.7[†]	20.2%, 1.2%[†]	2.2%	
			Birth Weight > 4 kg	1.6, 1.8[†]	6.1%, .54%[†]	NR	
Boere-Boonekamp, 1998	1,968	72	Breech	1.35	5.00%	4.2%	Fair
			Family History	2.59	9.6%	11.1%	
Boeree, 1994	26,952	118	Breech	6.98	3.0%	10.2%	Fair
			Family History	24.9	10.7%	20.8%	

			Foot Deformity	4.42	1.90%	2.5%	
Goss, 2002	5,166	100	Breech	5.2	10.1%	24%	Fair
			Family History	NR	NR	25%	
			Female	3.3	6.4%	77%	
Holen, 1996	408	25	Breech	5.55	6.1%	NR	Fair
Jones, 1989	3289	51	Breech	4.97	7.7%	11.8%	Fair
			Family History	10.8	16.7%	5.9%	
Miranda, 1988	49,937	317	Breech	4.72	NR	17.4%	Fair
			First born	1.29	NR	53.0%	
			Female	1.67	NR	81.1%	
Sahin, 2004	5,798	10	Breech	NA	<1% (1/111) overall	10% overall	Fair
			Family history	NA			
			Muscle/skeletal deformity	NA			
			Swaddling	NA			
Walter, 1992	1,772	8	Breech	8.24 overall	4.12% overall	5% overall	Fair
			Family history				
			Postural abnormalities				
			Oligohydramnios				

*D or I = dislocated or dislocatable

† = Ultrasound positive, Treated

TABLE 2 Comparison of Screening Approaches Represented in RCTs

Study,Year Setting	Clinical Exam Only				Selective Ultrasound				Universal Ultrasound			
	N	Treatment Rate*	Late Diagnosis Rate*	Subsequent Treatment Required*†	N	Treatment Rate*	Late Diagnosis Rate*	Subsequent Treatment Required*†	N	Treatment Rate*	Late Diagnosis Rate*	Subsequent Treatment Required*†
Elbourne, 2002 Infants < 43 days of age, with hip instability on initial exam, Referred to 33 clinics, UK 1994-98	315	492	NR	63					314	385	NR	54
Holen, 2002 Newborns 1-3 days old in hospital, Norway 1988-92					7689	8.6	0.65	0.13	7640	9.6	0.13	0.26
Rosendahl, 1994 Newborns in maternity hospital, Norway 1988-90	3924	18	2.6	1.2	4388	20	2.1	0.7	3618	34	1.4	0.3

NR, not reported.

*Number per 1,000 children

†Additional treatment required, indicating primary treatment was unsuccessful.

TABLE 3 Population-based Comparative Studies: Clinical Exam Versus Ultrasound

Author (Year)	N	Clinical examiners (Number of Examiners)	Reference standard for DDH	Clinical exam instability rate/1000 children	Ultrasound positive rate/1,000 children	Treatment rate/1000 children	% with DDH identified only by exam	% with DDH identified only by ultrasound	% with pos. exam and neg. ultrasound*	Late diag-nosis rate/1000 children	Rate/timing of spontaneous resolution	Follow-up of initially negative tests
Bache, 2002	29,323	Not specified	Requires intervention	NR	65.9 (hips) (39 subluxable/ dislocated)	3.1 (hips)	0%	65%	18%	0	96% of hips with ultrasound abnormalities at birth by 6 weeks	Unclear
Blalik, 1998	4321	Neonatologist (NR)	Requires intervention	15.2	55.3	6.2 (hips)	0%	52%	2%	NR	90.3% of hips with dysplasia or instability by 6 weeks	NR
Gianna-kopoulou, 2002	6140	Pediatrician (2)	Ultrasound: abnormality	17.9	12.2	10.6	NA	32%	41%	NR	10/75 hips (10/10 with physiological dysplasia) within 4 weeks	NR
Paton, 1999	20,452	Pediatrician (NR)	Ultrasound: dislocation	14	1.8	NR	NA	31%	87%	0.4	NR	Unclear
Riboni, 2003	8896	Neonatologist (NR)	Ultrasound: abnormality	2.1	28	3.8	NA	56%	58%	2.1 DDH / 0.6 more severe than dysplasia	206/215 with borderline dyplasia by 1 month	Yes (83%), at 3 months

TABLE 3 Population-based Comparative Studies: Clinical Exam Versus Ultrasound, Continued

Author (Year)	N	Clinical examiners (Number of Examiners)	Reference standard for DDH	Clinical exam instab-ility rate/1000 children	Ultrasound positive rate/1,000 children	Treat-ment rate/1000 children	% with DDH identified only by exam	% with DDH identified only by ultra-sound	% with pos. exam and neg. ultrasound*	Late diag-nosis rate/1000 children	Rate/timing of spontan-eous resolution	Follow-up of initially nega-tive tests
Rosen-berg, 1998	9199	Neonatologist (NR)	Clinical exam or ultrasound: instability	14.5	68.2	NR	5%	50%	NA	NR	NR	NR
Rosen-Dahl, 1996	3613	Physicians with ≥ 2 yrs pediatric experience (8)	Clinical exam: dislocatable Ultrasound: "major" dyplasia	19.1	30.4(23.8 dislocatable/ dislocated)	34	11%	28%	38%	0.2	13/16 with minor dysplasia by 1-2 months	Unclear

NR, not reported; NA, not applicable.
*Independent of standard used for diagnosis of DDH.

TABLE 4. Evidence Summary

Arrow	Key Question	Level and Type of Evidence
1	Does screening for DDH lead to reduced need for surgery or improved functional outcomes?	Poor: No controlled studies have compared screening with no screening to determine whether there is an impact on functional outcomes. There is conflicting evidence from ecologic studies that screening reduces rates of surgery.
2	Can infants at high risk for DDH be identified, and does this group warrant a different approach to screening than children at average risk?	Fair: In case-control and cohort studies, family history, breech presentation, and clinical instability are consistently associated with an increased risk of DDH. Most infants with DDH do not have risk factors. No practice-based, prospective studies on on the performance of risk assessment instruments are available.
3	Does screening for DDH lead to early identification of children with DDH?	See 3a, 3b, 3c below.
3a	What is the sensitivity, specificity, and predictive value of screening exams? (e.g. Barlow/Ortolani, other exam findings, ultrasonography, and radiographs).	Poor: Ascertainment of test characteristics is unreliable, because definitions of a positive test vary, and most studies did not use an independent standard to determine disease status. Low risk/ screen negative patients are followed with less intensity than high risk/screen positive patients. High rates of spontaneous resolution have been reported. Fair: Most hip dysplasia identified by early ultrasound will resolve spontaneously in first weeks of life.
3b	How does the age of the child affect screening parameters?	Fair: Limited hip abduction becomes a more sensitive sign of DDH after the first several months of life.
3c	How does the educational level and training of the screener impact screening?	Fair: Experience with the clinical examination of the hip in infants predicts screen positive rates and accuracy of exam, but few head-to-head comparisons without biases have been conducted. Consistent but limited amount of evidence that well-trained non-physicians can interpret clinical examination findings as well as pediatricians and better than physicians-in-training.
4	What are the adverse effects of screening?	Poor: In theory, forceful exam of already-lax newborn hips might cause injury or dislocation, but there is limited and conflicting evidence regarding this hypothesis.
5	Does early diagnosis of DDH lead to early intervention, and does early intervention lead to improved functional outcomes? Is the likelihood of surgical intervention reduced in children diagnosed at an earlier age?	Fair: Early diagnosis leads to early intervention. Evidence of the effectiveness of intervention is inconclusive, due to 1) high rate of spontaneous resolution, 2) absence of comparative studies of intervention vs. no intervention, 3) variation in surgical indications and protocols. Few studies examine functional outcomes in a valid and reliable fashion. Fair-poor: Evidence is limited and mixed on the effect of earlier diagnosis on likelihood of surgery.

6	What are the adverse effects of early diagnosis and/or surgical and non-surgical interventions?	Fair: All nonsurgical and surgical interventions are associated with a risk of avascular necrosis. Many nonsurgical interventions are in use, but data are insufficient to determine whether there are differences among them. This is also true of surgical interventions.