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Incidence of Neonatal Developmental Dysplasia of the Hip and Late Detection Rates Based on Screening Strategy
A Systematic Review and Meta-analysis

Ilari Kuitunen, PhD; Mikko M. Uimonen, PhD; Marjut Haapanen, MD; Reijo Sund, PhD; Ilkka Helenius, PhD; Ville T. Ponkilainen, PhD

Abstract

IMPORTANCE Universal ultrasonographic screening for developmental dysplasia of the hip (DDH) has gained increasing popularity despite the lack of benefit in terms of reducing the rates of late-detected cases (age ≥12 weeks) in randomized clinical trials.

OBJECTIVE To report the reported incidence of DDH in the English scientific literature and compare rates of late-detected cases in settings with different DDH screening strategies.

DATA SOURCES PubMed, Scopus, and Web of Science databases were searched on November 25 and 27, 2021. No time filters were used in the search.

STUDY SELECTION All observational studies reporting the incidence of early-detected or late-detected (age ≥12 weeks) DDH were included. Non-English reports were excluded if the abstract did not include enough information to be included for analysis.

DATA EXTRACTION AND SYNTHESIS The number of newborns screened and the detection rates were extracted. Meta-analysis calculated the pooled incidence of DDH per 1000 newborns with 95% CIs using a random- or fixed-effects model. This study is reported according to the PRISMA and MOOSE guidelines.

MAIN OUTCOMES AND MEASURES The main outcome measures were early detection, early treatment, late detection, and operative treatment incidences.

RESULTS A total of 1899 studies were identified, 203 full texts were assessed, and 76 studies with 16 901 079 infants were included in final analyses. The early detection rate was 8.4 (95% CI, 4.8-14.8) infants with DDH per 1000 newborns with clinical screening, 4.4 (95% CI, 2.4-8.0) infants with DDH per 1000 newborns with selective ultrasonographic screening, and 23.0 (95% CI, 15.7-33.4) infants with DDH per 1000 newborns with universal ultrasonographic screening. Rates for nonoperative treatment were 5.5 (95% CI, 2.1-14) treatments per 1000 newborns with clinical screening, 3.1 (95% CI, 2.0-4.8) treatments per 1000 newborns with selective ultrasonographic screening, and 9.8 (95% CI, 6.7-14.4) treatments per 1000 newborns with universal ultrasonographic screening. The incidence of late-detected DDH was 0.5 (95% CI, 0.2-1.5) infants with DDH per 1000 newborns with clinical screening, 0.6 (95% CI, 0.3-1.3) infants with DDH per 1000 newborns with selective ultrasonographic screening, and 0.2 (95% CI, 0.0-0.8) infants with DDH per 1000 newborns with universal ultrasonographic screening. The corresponding incidences of operative treatment were 0.2 (95% CI, 0.0-0.9) operations per 1000 newborns with clinical screening, 0.5 (95% CI, 0.4-0.7) operations per 1000 newborns with selective ultrasonographic screening, and 0.4 (95% CI, 0.2-0.7) operations per 1000 newborns with universal ultrasonographic screening.

(continued)

Key Points

Question Is universal ultrasonographic screening of developmental dysplasia of the hip associated with detecting more patients and reducing the late operative treatment rates compared with selective ultrasonographic and clinical screening?

Findings In this systematic review and meta-analysis including 76 studies with 16 901 079 patients, the early detection rate in universal ultrasonographic screening was significantly higher than in selective ultrasonographic or clinical screening. However, there were no significant differences in the incidences of late-detected patients and operative treatment among the screening strategies.

Meaning These findings challenge the rationale for universal ultrasonographic screening, as it was associated with higher initial detection rate without reducing the late detection rate.

+ Supplemental content

Author affiliations and article information are listed at the end of this article.

Abstract (continued)

CONCLUSIONS AND RELEVANCE This meta-analysis found that early detection rates and nonoperative treatments were higher with universal screening. The late detection and operative treatment rates with universal screening were similar to those among selectively and clinically screened newborns. Based on these results, universal screening may cause initial overtreatment without reducing the rates of late detection and operative treatment.

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Introduction

Developmental dysplasia of the hip (DDH) includes the following newborn hip findings: "clicky" hip, clinical instability, clinical reversible and irreversible dislocation, immature ultrasonographic findings, and potential dislocation on dynamic ultrasonographic examination.[1,2] Since Graf[3] introduced ultrasonographic classification of the hip joint in 1980, the role of ultrasonography in diagnosis and screening has increased.[4,5] The rationale for this has been that clinical examination does not detect all infants with DDH.[6] At the moment, universal ultrasonographic screening is especially popular in Central Europe.[7]

The initiation of ultrasonographic screening has led to increased rates of harness and splinting treatments owing to increasing DDH detection.[8] However, 97% of immature hips detected via ultrasonography normalize during the first months without any treatment.[9] Observational studies have reported that after the initiation of universal ultrasonographic screening, the rates of late-detected DDH and the need for operative treatment have been lower compared with the prescreening era.[10] Nonetheless, randomized clinical trials have not found universal screening beneficial or effective in reducing the rate of late-detected DDH.[11,12] A Cochrane review[13] in 2013 concluded that the universal screening increases the rate of early treatment but does not reduce the rate of late dislocation. In addition, cost-effectiveness analyses have not recommended the use of universal ultrasonographic screening.[14,15]

Universal screening has been claimed to reduce the need for total hip replacements (THRs) in the future because early treatment may reduce the risk of acetabular dysplasia. However, this has not been widely studied. To our knowledge, there is only 1 large-scale nationwide register study,[16] conducted in Norway, which did not find any relevant differences in the rates of THR in young adulthood based on the DDH screening status in infancy. The study by Engesaeter et al[16] was based on clinical screening. Because the optimal DDH screening strategy remains controversial, we aim to report on the incidences of DDH and late-detected DDH and compare the detection and treatment rates among the different screening strategies.

Methods

We conducted a systematic review and meta-analysis of observational studies reporting the incidence of DDH. We initially searched the literature for new clinical trials systematically but did not find any new randomized trials to add to the most recent Cochrane review[13]; therefore, we decided to focus on observational studies. The search process and search strategy for the randomized clinical trials are presented in eFigure 1 in Supplement 1. The protocol for this study has been sent for registration to PROSPERO (registration ID 300705), but owing to COVID-19–related delays, it has not yet been published. The protocol can also be found in Supplement 2.We report this study according to the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) reporting guideline and Meta-analysis of Observational Studies in Epidemiology (MOOSE) reporting guideline.

Search Strategy

We searched PubMed, Web of Science, and Scopus on November 25 and 27, 2021. We used the following search phrase: (hip) AND (dysplasia) AND (incidence or epidemiolog*) AND (country [eg, Australia or Australian]). The complete screening process is described in eTable 1 in Supplement 1.

We used Covidence systematic review software (Veritas Health Innovation) for the screening process. Each title and abstract was screened independently by 2 of 4 authors participating in screening (I.K., M.M.U., M.H., and V.T.P.). Disagreements were resolved by mutual agreement or third-party opinion by another of the 4 participating authors who did not participate in the study's initial assessment. Full texts were similarly assessed independently by 2 authors and disagreements were resolved similarly (by mutual agreement or third-party opinion).

Inclusion Criteria and Main Outcomes and Measures

We included all observational studies (cohort studies, case series, cross-sectional studies, and register-based studies with prospective or retrospective design) that reported the number of patients screened and the event rates for any of the following main outcomes: early-detected DDH (age <12 weeks), nonoperatively treated DDH (ie, splinting or harness treatment), late-detected DDH (age ≥12 weeks), or operatively treated DDH (including closed or open reductions and pelvic or proximal femoral osteotomies).

We excluded studies that did not report the number of patients screened or did not specify the screening method used. We excluded studies focusing only on preterm or breech-delivered newborns or twins. We also excluded non-English reports if all the necessary information was not reported in the abstract in English. The main outcome measures were the country- and screening-stratified incidences of early-detected DDH (age <12 weeks), initial nonoperative treatment rate, incidence of late-detected DDH (age ≥12 weeks), and operative treatment rate.

Data Extraction

We extracted the data to an Excel 2021 spreadsheet (Microsoft). One author extracted the data (I.K. or V.T.P.), and disagreements were resolved via a second opinion of the other participating author. We chose this extraction method, although it may cause more errors than double data extraction.[17] We gathered the following information: author, study period, study region, study design, screening method, number of patients screened, screening coverage, number of infants with clinically diagnosed DDH, number of infants with DDH diagnosed via ultrasonography, number of nonoperative treatments, number of operative treatments, and number of infants with late-detected DDH. We stratified the screening process into 3 categories: clinical screening, selective ultrasonographic screening (includes strategies of risk-based ultrasonography and ultrasonography based on clinical findings), and universal ultrasonographic screening. If a study reported incidences prior to and after the implementation of a screening protocol, we included only the after values because this was the current protocol used. The number of dysplastic hips was extracted either per patient or per hip, as reported by the authors. If the study reported only the findings according to Graf classification,[3] we labeled classes IIb to IV as dysplastic and classes I and IIa as nondysplastic.

Statistical Analysis

Some of the included studies reported DDH results per hip and not per patient. Thus, we calculated the ratio of dysplastic hips per patient from 7 studies[1,18-23] that included both hips and patients, resulting in a ratio of 1.305 dysplastic hips per patient. The number of hips were multiplied with the ratio to calculate comparable incidences per patient if the number of patients was not presented.

In the meta-analysis, we calculated the pooled incidence of DDH per patient by using a generalized linear mixed model. A random-effects model was used owing to the high heterogeneity (measured with I^2). Incidences are reported per 1000 newborns with 95% CIs. All analyses were performed using R version 4.0.3 (R Project for Statistical Computing), and the pooled incidences were calculated using the function metaprop from the meta package version 5.1-1.

As we included observational studies that reported prevalence data and did not necessarily compared interventions between groups or preintervention vs postintervention periods, risk of bias in nonrandomized studies of interventions (ROBINS-I) was not suitable.[24] We uses Critical Appraisal Tool for Prevalence Studies by Joanna Briggs Institute.[25] Statistical differences were determined with overlapping 95% CIs.

Results

Included Studies

Our initial search retrieved 1899 results, and we assessed 203 full texts. Ultimately, 76 studies[1,4-6,8,18-23,26-90] with 16 901 079 patients were included for analysis (eTable 1 in Supplement 1). Of these studies, 15 concerned clinical screening, 29 concerned selective ultrasound, and 32 concerned universal ultrasound (eTable 2 in Supplement 1). Most studies were conducted in Europe (25 studies [59%]) or Asia (21 studies [28%]). Most of the studies were retrospective (43 studies [57%]) and used single-institute data (60 studies [79%]). Risk of bias was analyzed in 8 domains and overall (eTable 3 in Supplement 1). Most issues were detected in the domains of proper identification of the condition and if the condition was measured similarly to all (eFigure 2 in Supplement 1). None of the studies were excluded based on the risk of bias estimate.

Incidence of Early-Detected DDH

A total of 60 studies[1,4,5,8,18-23,26-75] including 8 670 492 newborns assessed the incidence of early-detected DDH. Of these, 10 studies[26-35] with 551 894 newborns used clinical screening, 21 studies[4,5,36-54] with 7 884 989 newborns used selective screening, and 29 studies[1,8,18-23,55-75] with 233 609 newborns used universal ultrasonographic screening. The highest reported incidence was 120.3 (95% CI, 110.9-130.5) infants with DDH per 1000 newborns, and the lowest was 0.2 (95% 0.1-0.4) infants with DDH per 1000 newborns (**Figure 1**). The total pooled incidence estimates were 23.0 (95% CI, 15.7-33.4) infants with DDH per 1000 newborns among those with universal ultrasonographic screening, 4.4 (95% CI, 2.4-8.0) infants with DDH per 1000 newborns among those with selective ultrasonographic screening, and 8.4 (95% CI, 4.8-14.8) infants with DDH per 1000 newborns with clinical screening. The greatest variation was observed among the universally screened populations (Figure 1). Country-specific incidences were highest in Europe (Slovakia and France) and the Middle East (Israel and Iran) (**Figure 2**). Most countries had not published results in English in indexed peer-reviewed literature regarding the incidence of DDH.

Incidence of Nonoperative Treatment

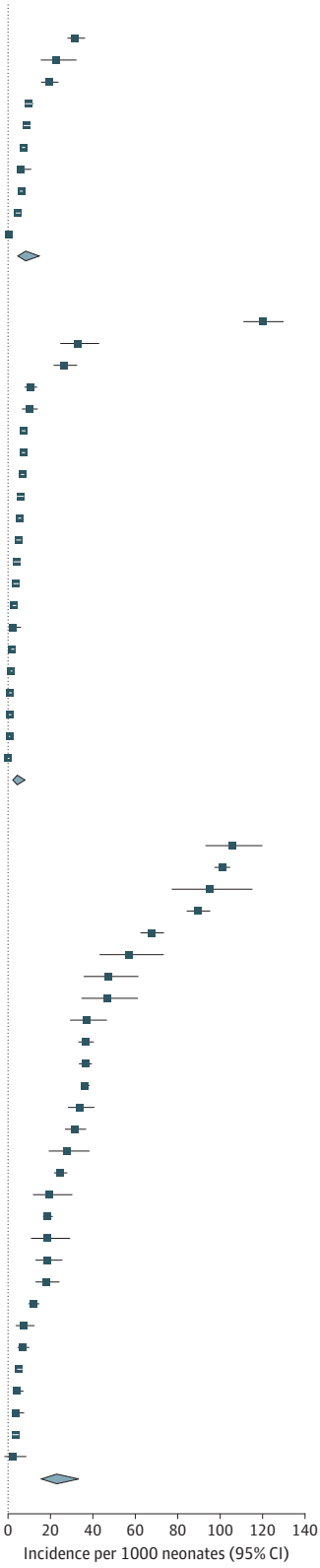
A total of 43 studies[5,8,18,20-23,28,29,31,35-39,42-53,55,56,59-61,64,65,67,68,72,74,76-80] including 4 488 951 newborns assessed the incidence of nonoperatively treated DDH. Of these, 5 studies[28,29,31,35,76] with 457 752 newborns used clinical screening, 20 studies[5,36-39,42-53,77-79] with 3 852 022 newborns used selective screening, and 18 studies[8,18,20-23,55,56,59-61,64,65,67,68,72,74,80] with 173 476 newborns used universal ultrasonographic screening. The highest reported incidence was 34.0 (95% CI, 28.5-40.6) treatments per 1000 newborns and the lowest was 0.1 (95% CI, 0.0-0.3) treatments per 1000 newborns (**Figure 3**). The total pooled incidence estimates were 9.8 (95% CI, 6.7-14.4) treatments per 1000 newborns with universal ultrasonographic screening, 3.1 (95% CI, 2.0-4.8) treatments per 1000 newborns with selective ultrasonographic screening, and 5.4 (95% CI, 2.1-14.0) treatments per 1000 newborns with clinical screening. The greatest variation was observed among the universally screened populations (Figure 1).

Incidence of Late-Detected DDH

A total of 43 studies[6,8,20-23,28-31,33,36-38,40-44,46,48,50-53,56,57,61,68,72,74,76-87] including 6 913 795 newborns assessed the incidence of late-detected DDH. Of these, 10 studies[28-31,33,76,81-84] with 2 126 583 newborns used clinical screening, 21 studies[6,36-38,40-44,46,48,50-53,77-79,85-87] with

Figure 1. Forest Plot of the Incidence of Early-Detected Developmental Dysplasia of the Hip in Individual Studies Stratified by Screening Strategy

Source	No. events	Total	Incidence per 1000 neonates (95% CI)
Clinical screening			
Mamouri et al,[26] 2003	207	6575	31.5 (27.4-36.0)
Hesaraki,[27] 2017	32	1400	22.9 (15.7-32.1)
Goss,[28] 2002	100	5166	19.4 (15.8-23.5)
Burger et al,[29] 1990	140	14264	9.8 (8.3-11.6)
Yiv et al,[30] 1997	173	19622	8.8 (7.6-10.2)
Sharpe et al,[31] 2005	1281	176427	7.3 (6.9-7.7)
Ishikawa,[32] 2008	13	2083	6.2 (3.3-10.6)
Azzopardi et al,[33] 2017	1878	302666	6.2 (5.9-6.5)
Ang et al,[34] 1997	96	20295	4.7 (3.8-5.8)
Chotigavanichaya et al,[35] 2012	2	3396	0.6 (0.1-2.1)
Random-effects model	NA	551894	8.4 (4.8-14.8)
I^2 = 99%			
Selective ultrasonographic screening			
Güler et al,[36] 2016	575	4782	120.2 (111.2-129.8)
Gyurkovits et al,[37] 2019	54	1636	33.0 (24.9-42.8)
Moosa et al,[38] 2009	101	3786	26.7 (21.8-32.3)
Giannakopoulou et al,[39] 2002	65	6140	10.6 (8.2-13.5)
Geertsema et al,[40] 2018	35	3536	9.9 (6.9-13.7)
Donnelly et al,[41] 2015	561	75 856	7.4 (6.8-8.0)
Clarke et al,[42] 2012	774	107440	7.2 (6.7-7.7)
Studer et al,[43] 2016	777	114155	6.8 (6.3-7.3)
Kural et al,[44] 2019	56	9758	5.7 (4.3-7.4)
Wilf-Miron et al,[5] 2017	625	115918	5.4 (5.0-5.8)
Wenger et al,[45] 2013	111	22517	4.9 (4.1-5.9)
Phelan et al,[46] 2014	33	8317	4.0 (2.7-5.6)
Zenios et al,[47] 2000	57	14625	3.9 (3.0-5.0)
Woodacre et al,[48] 2016	113	37233	3.0 (2.5-3.6)
Walter et al,[49] 1992	4	1772	2.3 (0.6-5.8)
Paton et al,[50] 2005	77	34723	2.2 (1.8-2.8)
Degnan et al,[4] 2021	6808	4126060	1.7 (1.6-1.7)
Tong et al,[51] 2011	33	31055	1.1 (0.7-1.5)
Kumar et al,[52] 2016	20	23295	0.9 (0.5-1.3)
Den et al,[53] 2021	1879	3117610	0.6 (0.6-0.6)
Sirisabya et al,[54] 2019	4	24825	0.2 (0-0.4)
Random-effects model	NA	7884989	4.4 (2.4-8.0)
I^2 = 100%			
Universal ultrasonographic screening			
Kokavec and Bialik,[55] 2007	231	2178	106.1 (93.4-119.8)
Bache et al,[56] 2003	2961	29323	101.0 (97.6-104.5)
De Marino et al,[57] 1994	95	1000	95.0 (77.5-114.9)
Bialik and Berant,[58] 1997	1009	11249	89.7 (84.5-95.1)
Bialik et al,[18] 1999	613	9030	67.9 (62.8-73.3)
Bhalvani and Madhuri,[59] 2011	57	1000	57.0 (43.5-73.2)
Cekic et al,[60] 2015	55	1162	47.3 (35.9-61.2)
Vafaee et al,[1] 2017	50	1073	46.6 (34.8-61.0)
Boere-Boonekamp et al,[61] 1998	77	2066	37.3 (29.5-46.4)
Wirth et al,[62] 2004	453	12331	36.7 (33.5-40.2)
Peled et al,[63] 2008	658	18067	36.4 (33.7-39.3)
Peled et al,[64] 2008	1652	45497	36.3 (34.6-38.1)
Rosendahl et al,[65] 1996	123	3613	34.0 (28.4-40.5)
Arti et al,[66] 2013	180	5701	31.6 (27.2-36.4)
Gharehdaghi et al,[67] 2011	36	1300	27.7 (19.5-38.1)
Schams et al,[68] 2017	291	11820	24.6 (21.9-27.6)
Colta et al,[19] 2016	20	1021	19.6 (12.0-30.1)
Treiber et al,[22] 2008	324	17393	18.6 (16.7-20.7)
Köse et al,[20] 2006	18	975	18.5 (11.0-29.0)
Buonsenso et al,[69] 2021	37	2000	18.5 (13.1-25.4)
Lange et al,[70] 2017	46	2534	18.2 (13.3-24.1)
Munkhuu et al,[21] 2013	100	8356	12.0 (9.7-14.5)
Krolo et al,[71] 2003	15	2010	7.5 (4.2-12.3)
Puol et al,[72] 1998	32	4568	7.0 (4.8-9.9)
Treiber et al,[23] 2021	118	21676	5.4 (4.5-6.5)
Olsen et al,[8] 2018	19	4245	4.5 (2.7-7.0)
Kolb et al,[73] 2015	11	2678	4.1 (2.1-7.3)
Riboni et al,[74] 2003	30	8896	3.4 (2.3-4.8)
Mureşan et al,[75] 2019	2	847	2.4 (0.3-8.5)
Random-effects model	NA	233609	23.0 (15.7-33.4)
I^2 = 99%			



A generalized linear random-effects model was used to calculate pooled incidences per 1000 newborns in each screening group. NA indicates not applicable.

4 649 086 newborns used selective screening studies, and 12 studies[8,20-23,56,57,61,68,72,74,80] with 138 126 newborns used universal ultrasonographic screening. The highest reported incidence was 13.0 (95% CI, 9.7-17.4) infants per 1000 newborns and the lowest was 0.0 (95% CI, 0.0-0.0) infants per 1000 newborns (Figure 3). The total incidence estimates were 0.2 (95% CI, 0.0-0.8) infants per 1000 newborns in universal ultrasonographic screening, 0.6 (95% CI, 0.3-1.3) infants per 1000 newborns in selective ultrasonographic screening, and 0.5 (95% CI, 0.2-1.5) infants per 1000 newborns per 1000 newborns in clinical screening. The greatest variation in incidences was observed among the selectively screened populations (**Figure 4**).

Incidence of Operatively Treated DDH

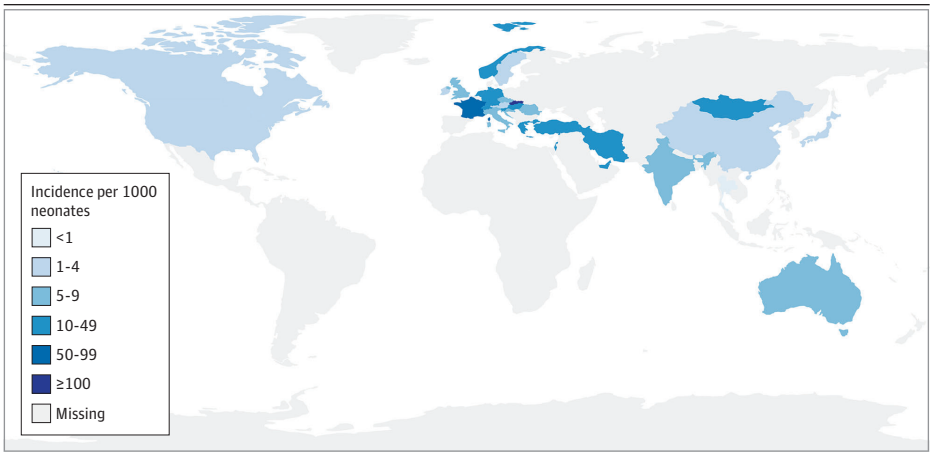
A total of 30 studies[6,8,20,22,23,29,34,37-39,42-44,46-48,50,52,56,62,63,74,77,79,80,82,85,88,89] with 6 497 382 newborns assessed the incidence of operatively treated DDH. Of these, 3 studies[29,34,82] with 64 559 newborns used clinical screening, 16 studies[6,37-39,42-44,46-48,50,52,77,79,85,88] with 855 109 newborns used selective screening, and 11 studies[8,20,22,23,56,62,63,74,80,89] with 5 577 714 newborns used universal ultrasonographic screening. The highest reported incidence was 2.6 (95% CI, 1.8-2.7) operations per 1000 newborns (Figure 3). The total incidence estimates in the random-effects models were 0.4 (95% CI, 0.2-0.7) operations per 1000 newborns with universal ultrasonographic screening, 0.5 (95% CI, 0.4-1.7) operations per 1000 newborns with selective ultrasonographic screening, and 0.2 (95% CI, 0.0-0.9) operations per 1000 newborns with clinical screening (**Figure 5**).

Discussion

This systematic review and meta-analysis found that the incidence of DDH detection and nonoperative treatment among newborns given universal ultrasonographic screening was higher than among those given selective or clinical screening. The incidences of late-detected DDH and operatively treated DDH were similar among all screening strategies. The highest DDH burden was found in Europe and the Middle Eastern area of Asia.

Our results are in line with those of previous randomized clinical trials and a meta-analysis of randomized studies.[11-13] However, the use of universal ultrasonographic screening has been increasing, and a recent narrative review stated that, based on the excellent results achieved in observational studies, universal ultrasonography is an effective way to reduce the rate of late-detected DDH[7]; however, this was not seen in our current systematic review and meta-analysis of observational studies.

Figure 2. Geographic Variation in the Incidence of Early-Detected Developmental Dysplasia of the Hip

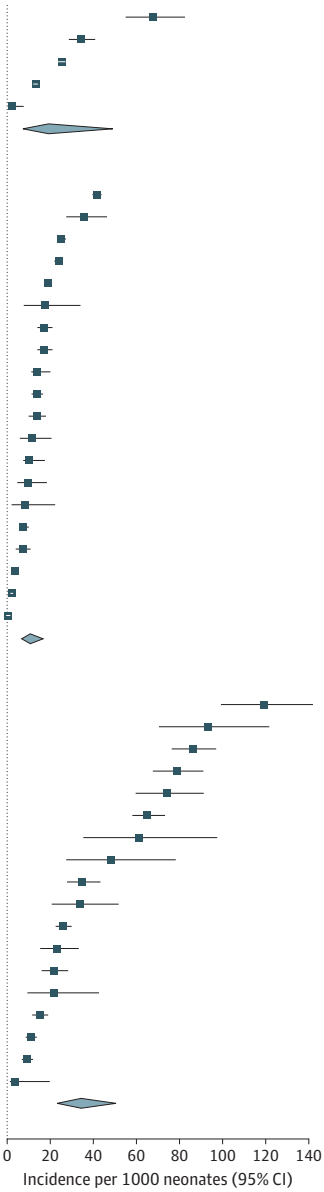


Incidences reported per 1000 newborns. Incidence calculated based on either the clinical or ultrasonographic findings per patient, depending on the screening strategy in use.

The early detection rates and nonoperative treatment rates were the lowest in studies with selective ultrasonographic screening. This has previously been shown also in a randomized clinical trial by Elbourne et al,[91] which found that ultrasonography confirmation of clinically suspected DDH reduced nonoperative treatment rates. However, in the study by Elbourne et al,[91] the rate of later operative treatment was lower as well, but their follow-up only included patients who were initially referred owing to clinical DDH and did not include patients with late-detected clinical DDH. Thus, based on our findings and the results of the randomized clinical trial performed by Elbourne et al,[91] it may be that the ultrasonographic confirmation of clinically suspected DDH could reduce the unnecessary nonoperative treatment rate.[91] The strength of a selective screening policy is that it confirms the clinically suspicious cases of DDH, which could be treated based on the clinical examination findings. Another advantage is that if the selective screening is performed in delayed

Figure 3. Forest Plot of the Incidence of Initial Nonoperative Treatment for Developmental Dysplasia of the Hip in Individual Studies Stratified by the Screening Strategy

Source	No. events	Total	Incidence per 1000 neonates (95% CI)
Clinical screening			
Goss,[28] 2002	100	5166	9.4 (15.8-23.5)
Burger et al,[29] 1990	140	14264	9.8 (8.3-11.6)
Sharpe et al,[31] 2005	1281	176427	7.3 (6.9-7.7)
Pollet et al,[76] 2016	970	258499	3.8 (3.5-4.0)
Chotigavanichaya et al,[35] 2012	2	3396	0.6 (0.1-2.1)
Random-effects model	NA	457752	5.5 (2.1-14.0)
I^2 = 99%			
Selective ultrasonographic screening			
Milligan and Cosgrove,[77] 2020	2027	170580	11.9 (11.4-12.4)
Giannakopoulou et al,[39] 2002	63	6140	10.3 (7.9-13.1)
Clarke et al,[42] 2012	774	107440	7.2 (6.7-7.7)
Studer et al,[43] 2016	777	114155	6.8 (6.3-7.3)
Wilf-Miron et al,[5] 2017	625	115918	5.4 (5.0-5.8)
Walter et al,[49] 1992	9	1772	5.1 (2.3-9.6)
Reidy et al,[78] 2019	114	23112	4.9 (4.1-5.9)
Wenger et al,[45] 2013	111	22517	4.9 (4.1-5.9)
Phelan al,[46] 2014	33	8317	4.0 (2.7-5.6)
Woodacre et al,[48] 2016	147	37233	3.9 (3.3-4.6)
Zenios et al,[47] 2000	57	14625	3.9 (3.0-5.0)
Tyagi et al,[79] 2016	12	3618	3.3 (1.7-5.8)
Güler et al,[36] 2016	14	4782	2.9 (1.6-4.9)
Moosa et al,[38] 2009	11	3786	2.9 (1.5-5.2)
Gyurkovits et al,[37] 2019	4	1636	2.4 (0.7-6.2)
Paton et al,[50] 2005	77	34723	2.2 (1.8-2.8)
Kural et al,[44] 2019	19	9758	1.9 (1.2-3.0)
Tong et al,[51] 2011	30	31005	1.0 (0.7-1.4)
Den et al,[53] 2021	1879	3117610	0.6 (0.6-0.6)
Kishore Kumar et al,[52] 2016	2	23295	0.1 (0-0.3)
Random-effects model	NA	3852022	3.1 (2.0-4.8)
I^2 = 100%			
Universal ultrasonographic screening			
Rosendahl et al,[65] 1996	123	3613	34.0 (28.4-40.5)
Boere-Boonekamp et al,[61] 1998	55	2066	26.6 (20.1-34.5)
Schams et al,[68] 2017	291	11820	24.6 (21.9-27.6)
Munkhuu et al,[21] 2013	188	8356	22.5 (19.4-25.9)
Olsen et al,[8] 2018	90	4245	21.2 (17.1-26.0)
Treiber et al,[22] 2008	324	17393	18.6 (16.7-20.7)
Köse et al,[20] 2006	17	975	17.4 (10.2-27.8)
Çekiç et al,[60] 2015	16	1162	13.8 (7.9-22.3)
Bialik et al,[18] 1999	90	9030	10.0 (8.0-12.2)
Kokavec and Bialik,[55] 2007	21	2178	9.6 (6.0-14.7)
Biedermann et al,[80] 2018	209	27808	7.5 (6.5-8.6)
Puol et al,[72] 1998	30	4568	6.6 (4.4-9.4)
Riboni et al,[74] 2003	55	8896	6.2 (4.7-8.0)
Gharehdaghi et al,[67] 2011	8	1300	6.2 (2.7-12.1)
Peled et al,[63] 2008	78	18067	4.3 (3.4-5.4)
Bache et al,[56] 2003	93	29323	3.2 (2.6-3.9)
Treiber et al,[23] 2021	56	21676	2.6 (2.0-3.4)
Bhalvani and Madhuri,[59] 2011	1	1000	1.0 (0-5.6)
Random-effects model	NA	173476	9.8 (6.7-14.4)
I^2 = 98%			



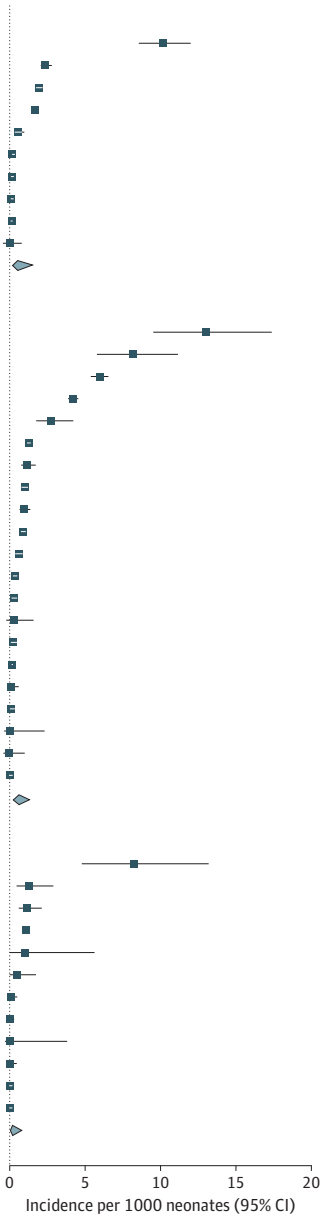
Nonoperative treatment included harness or splinting. A random-effects model was used to calculate the pooled incidences per 1000 newborns in each screening group. NA indicates not applicable.

schedule for patients with hips with clinical suspicion, many of these hips have had time to mature and stabilize.[42] Studies have shown that most immature hips diagnosed via ultrasonography mature without any interventions during the first weeks of life.[92] However, it must be noted that in 1 of the largest studies reporting late detection rates before and after selective screening implementation in England,[87] the rate remained unchanged compared with prior clinical screening policy. The presented late detection rate (1.3 per 1000 neonates) was also extremely high, as studies with clinical screening only have had much lower rates (eg, a study in Sweden[84] that found a rate of 0.12 per 1000 neonates).

It is obvious that clinical screening will miss a small percentage of patients with DDH (eg, bilateral or teratologic DDH) who could have been treated nonoperatively and will therefore undergo

Figure 4. Forest Plot of the Incidence of Late-Detected Developmental Dysplasia of the Hip in Individual Studies Stratified by Screening Strategy

Source	No. events	Total	Incidence per 1000 neonates (95% CI)
Clinical screening			
Burger et al,[29] 1990	145	14 264	10.2 (8.6-12.0)
Bjerkreim and Johansen,[81] 1987	197	82 574	2.4 (2.1-2.7)
Pollet et al,[76] 2016	499	258 499	1.9 (1.8-2.1)
Yiv et al,[30] 1997	33	19 622	1.7 (1.2-2.4)
Lisle et al,[82] 2012	17	30 000	0.6 (0.3-0.9)
Azzopardi et al,[33] 2017	67	302 666	0.2 (0.2-0.3)
Sharpe et al,[31] 2005	27	176 427	0.2 (0.1-0.2)
Barik et al,[83] 2021	32	223 776	0.1 (0.1-0.2)
Wenger et al,[84] 2019	126	1 013 589	0.1 (0.1-0.1)
Goss,[28] 2002	0	5166	0 (0-0.7)
Random-effects model	NA	2 126 583	0.5 (0.2-1.5)
I^2 = 100%			
Selective ultrasonographic screening			
Geertsema et al,[40] 2018	46	3536	13.0 (9.5-17.3)
Güler et al,[36] 2016	39	4782	8.2 (5.8-11.1)
Donnelly et al,[41] 2015	451	75 856	5.9 (5.4-6.5)
Milligan and Cosgrove,[77] 2020	713	170 580	4.2 (3.9-4.5)
Phelan et al,[46] 2014	23	8317	2.8 (1.8-4.1)
Broadhurst et al,[87] 2019	754	589 062	1.3 (1.2-1.4)
Reidy et al,[78] 2019	27	23 112	1.2 (0.8-1.7)
Studer et al,[43] 2016	111	114 155	1.0 (0.8-1.2)
Woodacre et al,[48] 2016	35	37 233	0.9 (0.7-1.3)
Maxwell et al,[85] 2002	154	173 746	0.9 (0.8-1.0)
Kamath et al,[86] 2007	29	51 166	0.6 (0.4-0.8)
Clarke et al,[42] 2012	37	107 440	0.3 (0.2-0.5)
Talbot et al,[6] 2017	18	64 670	0.3 (0.2-0.4)
Tyagi et al,[79] 2016	1	3618	0.3 (0-1.5)
Paton et al,[50] 2005	8	34 723	0.2 (0.1-0.5)
Den et al,[53] 2021	480	3 117 610	0.2 (0.1-0.2)
Kural et al,[44] 2019	1	9758	0.1 (0-0.6)
Tong et al,[51] 2011	3	31 005	0.1 (0-0.3)
Gyurkovits et al,[37] 2019	0	1636	0 (0-2.3)
Moosa et al,[38] 2009	0	3786	0 (0-1.0)
Kishore Kumar et al,[52] 2016	0	23 295	0 (0-0.2)
Random-effects model	NA	4 649 086	0.6 (0.3-1.3)
I^2 = 100%			
Universal ultrasonographic screening			
Boere-Boonekamp et al,[61] 1998	17	2066	8.2 (4.8-13.1)
Puol et al,[72] 1998	6	4568	1.3 (0.5-2.9)
Riboni et al,[74] 2003	10	8896	1.1 (0.5-2.1)
Treiber et al,[22] 2008	19	17 393	1.1 (0.7-1.7)
Cervone de Martino et al,[57] 1994	1	1000	1.0 (0-5.6)
Olsen et al,[8] 2018	2	4245	0.5 (0.1-1.7)
Schams et al,[68] 2017	1	11 820	0.1 (0-0.5)
Treiber et al,[23] 2021	1	21 676	0 (0-0.3)
Köse et al,[20] 2006	0	975	0 (0-3.8)
Munkhuu et al,[21] 2013	0	8356	0 (0-0.4)
Biedermann et al,[80] 2018	0	27 808	0 (0-0.1)
Bache et al,[56] 2003	0	29 323	0 (0-0.1)
Random-effects model	NA	138 126	0.2 (0-0.8)
I^2 = 85%			

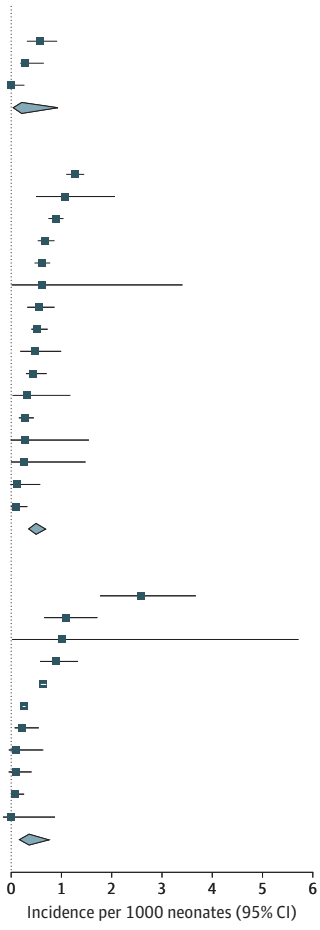


Late detection was defined as age 12 weeks or older. A random-effects model was used to calculate pooled incidences per 1000 newborns in each screening group. NA indicates not applicable.

operative treatment at a later stage. The earlier the DDH is detected, the better the outcome, with less complications in early phase and less likely progress to total hip replacement in adolescence or young adulthood.[93,94] However, the late detection rate and the need for operative treatment are not zero with universal ultrasonographic screening either, because ultrasonography does not detect all patients with clinical instability, and the reproducibility of ultrasonographic findings varies among examiners.[95,96] For example, nationwide studies from Germany and Austria presented late detected and operated rates for DDH of 0.16 to 0.26 per 1000 neonates, whereas a Swedish study reported a rate of 0.12 per 1000 neonates in clinical screening.[84,90,97] Therefore, the cost-effectiveness of universal ultrasonographic screening should be critically assessed. Indeed, previous cost-effectiveness studies have stated that universal ultrasonographic screening is not justified based on cost-effectiveness alone but that it could be used in countries with high incidences of DDH to reduce the suffering of a small proportion of patients.[2,14,15,49] Compared with many globally established cost-effective screening and prevention programs for other diseases (eg, vaccination programs and preterm birth prevention programs) intended to reduce child mortality rates and severe morbidity, given the results of our analysis, universal ultrasonographic screening should be considered carefully and locally, based on the current screening effectiveness and resources. Improvements to clinical

Figure 5. Forest Plot of the Incidence of Operative Treatment of Developmental Dysplasia of the Hip in Individual Studies Stratified by Screening Strategy

Source	No. events	Total	Incidence per 1000 neonates (95% CI)
Clinical screening			
Lisle et al,[82] 2012	17	30000	0.6 (0.3-0.9)
Ang et al,[34] 1997	6	20295	0.3 (0.1-0.6)
Burger et al,[29] 1990	0	14264	0.0 (0-0.3)
Random-effects model	NA	64559	0.2 (0-0.9)
I^2 = 0%			
Selective ultrasonographic screening			
Milligan and Cosgrove,[77] 2020	216	170580	1.3 (1.1-1.4)
Phelan et al,[46] 2014	9	8317	1.1 (0.5-2.1)
Maxwell et al,[85] 2002	154	173746	0.9 (0.8-1.0)
Clarke et al,[42] 2012	74	107440	0.7 (0.5-0.9)
Studer et al,[43] 2016	**70**	114155	0.6 (0.5-0.8)
Gyurkovits et al,[37] 2019	**1**	1636	0.6 (0-3.4)
Paton et al,[50] 2005	19	34723	0.5 (0.3-0.9)
McAllister et al,[88] 2018	43	81387	0.5 (0.4-0.7)
Zenios et al,[47] 2000	7	14625	0.5 (0.2-1.0)
Woodacre et al,[48] 2016	16	37233	0.4 (0.2-0.7)
Giannakopoulou et al,[39] 2002	2	6140	0.3 (0-1.2)
Talbot et al,[6] 2017	18	64670	0.3 (0.2-0.4)
Tyagi et al,[79] 2016	1	3618	0.3 (0-1.5)
Moosa et al,[38] 2009	1	3786	0.3 (0-1.5)
Kural et al,[44] 2019	1	9758	0.1 (0-0.6)
Kishore Kumar et al,[52] 2016	2	**23295**	**0.1 (0-0.3)**
Random-effects model	NA	855109	0.5 (0.4-0.7)
I^2 = 86%			
Universal ultrasonographic screening			
Wirth et al,[62] 2004	32	12331	2.6 (1.8-3.7)
Treiber et al,[22] 2008	19	17393	1.1 (0.7-1.7)
Köse et al,[20] 2006	1	975	1.0 (0-5.7)
Biedermann et al,[80] 2018	25	27808	0.9 (0.6-1.3)
Sepúlveda et al,[89] 2021	961	1479308	0.6 (0.6-0.7)
Von Kries et al,[90] 2003	**1029**	3957692	0.3 (0.2-0.3)
Treiber et al,[23] 2021	5	21676	0.2 (0.1-0.5)
Riboni et al,[74] 2003	1	8896	0.1 (0-0.6)
Peled et al,[63] 2008	2	18067	0.1 (0-0.4)
Bache et al,[56] 2003	2	29323	0.1 (0-0.2)
Olsen et al,[8] 2018	0	4245	0 (0-0.9)
Random-effects model	NA	**5577714**	**0.4 (0.2-0.7)**
I^2 = 98%			



Operative treatment included closed and open reductions. A random-effects model was used to calculate pooled incidences per 1000 newborns in each screening group. NA indicates not applicable.

screening practices can be made and, for example, swaddling practices can be assessed and considered to be changed prior to implementation of more costly screening programs. The results of our study can be used in decision-making and comparison of acceptable rates of late diagnoses and operative treatments.

Randomized clinical trials are the gold standard in evidence-based medicine.[98] However, it is not feasible to conduct a randomized clinical trial to address the optimal screening method for DDH owing to the rare event rates. The main outcome measure used to justify universal ultrasonographic screening in the recent literature has been the rates of late-detected DDH and operative treatment. In this meta-analysis, we found that the rate of late-detected DDH was 0.02% among universally screened newborns and 0.05% among clinically or selectively screened newborns, which is not a statistically significant difference. Corresponding operative treatment rates were 0.04% in infants who received universal screening, 0.05% in neonates who received selective ultrasonographic screening, and 0.02% in neonates who received clinical screening, which was also not statistically significant. Therefore, a randomized clinical trial with a 1:1 design comparing a universal screening strategy with a selective strategy would require approximately 61 000 newborns per group to detect the anticipated absolute risk difference of 0.03% in late detection rate (standard α error = .05 and power of 0.80). Given this assumption, the number of screenings needed to avoid a single late-detected DDH would be 3333, and the question of whether this absolute difference should be considered a clinically relevant finding would have to be decided. Considering this, given the clearly underpowered sample sizes, it does not seem surprising that previous randomized clinical trials have not found statistically significant differences, although the late detection rate was high, especially in the study by Rosendahl et al.[11,12]

Limitations

This study has some limitations. We did not find any new randomized clinical trials since the most recent Cochrane review from 2013,[13] so we decided not to duplicate previous meta-analysis; therefore, we included only observational studies, which may create a risk of selection and reporting bias in the results. However, based on the risk of bias assessment, the included studies had no major issues. The study settings and reporting practices used in the included studies had high heterogeneity because some of the studies were single-institute prospective series and some of the studies were nationwide register-based retrospective analyses without precise information on the initial hip screening findings. We had to estimate the rates of patients having dysplastic hips because some of the studies reported only pathologic hips in their results, and this may have caused overestimation or underestimation in our results regarding the rates of early-detected DDH. Most studies were conducted in Europe or Asia, and there were no studies from Africa, which creates generalization bias in our global incidence estimations. Furthermore, there are different factors confounding the comparisons between countries; for example, swaddling, clothing for cold weather, and carrying habits have been found to be associated with the incidences of late-detected DDH.[99-102] Additionally, we did not search EMBASE database, as it was not available in our institutes, and we also excluded non-English–language reports. Therefore, it is possible that we might have missed some studies. However, our current report is by far the largest effort to gather estimation on the incidences of DDH, late-detected DDH, and non-operative and operative treatments, to our knowledge.

Conclusions

This systematic review and meta-analysis found that reported rates of early-detected DDH and initial nonoperative treatments are higher in settings with universal ultrasonographic screening compared with clinical screening and selective ultrasonographic screening programs. However, the incidences of late detected DDH and surgical treatment rates were not significantly different among different screening strategies.

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Obtained funding: Kuitunen.

Administrative, technical, or material support: Kuitunen, Haapanen.

Supervision: Sund, Helenius, Ponkilainen.

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SUPPLEMENT 1.

eFigure 1. Search Strategy and PRISMA Flowchart of the Review Process for Randomized Controlled Trials
eFigure 2. Overall Risk of Bias Summary Figure for Assessed Domains
eTable 1. Modified PRISMA Flowchart of the Screening Process
eTable 2. Background Characteristics of the Included Studies
eTable 3. Risk of Bias Assessment of the Individual Studies Based on the Joanna Briggs Institute Critical Appraisal Tool for Prevalence Studies

SUPPLEMENT 2.

Review Protocol