Health Information and Quality Authority

An tÚdarás Um Fhaisnéis agus Cáilíocht Sláinte

# Protocol for the evidence review of universal ultrasound screening for developmental dysplasia of the hip (DDH) in infants in Ireland.

Published 27 October 2023

## About the Health Information and Quality Authority (HIQA)

The Health Information and Quality Authority (HIQA) is an independent statutory authority established to promote safety and quality in the provision of health and social care services for the benefit of the health and welfare of the public.

HIQA's mandate to date extends across a wide range of public, private and voluntary sector services. Reporting to the Minister for Health and engaging with the Minister for Children, Equality, Disability, Integration and Youth, HIQA has responsibility for the following:

- **Setting standards for health and social care services** — Developing person-centred standards and guidance, based on evidence and international best practice, for health and social care services in Ireland.

- **Regulating social care services** — The Chief Inspector within HIQA is responsible for registering and inspecting residential services for older people and people with a disability, and children's special care units.

- **Regulating health services** — Regulating medical exposure to ionising radiation.

- **Monitoring services** — Monitoring the safety and quality of health services and children's social services, and investigating as necessary serious concerns about the health and welfare of people who use these services.

- **Health technology assessment** — Evaluating the clinical and cost-effectiveness of health programmes, policies, medicines, medical equipment, diagnostic and surgical techniques, health promotion and protection activities, and providing advice to enable the best use of resources and the best outcomes for people who use our health service.

- **Health information** — Advising on the efficient and secure collection and sharing of health information, setting standards, evaluating information resources and publishing information on the delivery and performance of Ireland's health and social care services.

- **National Care Experience Programme** — Carrying out national service-user experience surveys across a range of health services, in conjunction with the Department of Health and the HSE.

# Table of contents

# Background to the HIQA/NSAC work programme

The National Screening Advisory Committee (NSAC) was established in 2019 by the Minister for Health as an independent advisory committee to play a significant strategic role in developing and considering population-based screening programmes in Ireland. At the request of the Department of Health, the Health Technology Assessment (HTA) Directorate within the Health Information and Quality Authority (HIQA) undertakes evidence synthesis and provides evidence-based advice to NSAC on behalf of the Minister for Health.

# 1 Introduction

## 1.1 Background

Developmental dysplasia of the hip (DDH) is a congenital disorder of the musculoskeletal system, with a wide range of severity, in which there is abnormal development of the hip in infancy.[1, 2] In Ireland in 2017, the National Clinical Programme for Paediatrics and Neonatology and the DDH Subgroup of the National Child Health Review Steering Group recommended the implementation of a targeted ultrasound screening programme for infants at risk of DDH.[3] At-risk infants were defined as those with a first degree family history of DDH (that is, mother, father, or sibling), those who had a breech presentation, and infants with a positive clinical examination for DDH within 72 hours of birth.

In March 2023, at the request of NSAC, HIQA agreed to undertake an evidence review of universal ultrasound screening for developmental dysplasia of the hip (DDH) in infants in Ireland. This protocol describes the evidence synthesis approach that will be adopted.

## 1.2 Condition and screening technology

DDH is a congenital disorder of the musculoskeletal system in which there is abnormal development of the hip in infancy.[1, 2] The condition can have a wide range of severity, from mild dysplasia, where the socket of the hip (known as the acetabulum) is partially flattened, to a complete dislocation of the femoral head. The acetabulum, the femoral head, or both, can be deformed.[4] While the causes are unclear, there appears to be a codependence between development of the acetabulum and the femoral head, whereby the normal development of one stimulates the normal development and placement of the other, and vice versa. Additionally, the aetiology (that is, the cause of a disease) of DDH is multifactorial. Major risk factors include breech position in utero, female sex, family history of DDH, and being a firstborn child.[1, 2] Other potential risks include oligohydramnios (that is,

having low amniotic fluid during pregnancy), high birth weight, and post maturity birth (that is, being born overdue).[4]

The natural history of DDH depends on severity.[4] While DDH is usually asymptomatic at birth, the condition starts to become apparent in the first months of life and as the child reaches walking age.[2, 4] Symptoms may appear as an abnormal gait or limp, difference in length between the two legs, and or delayed walking age. In milder forms of the condition, the symptoms can disappear over time, but in more severe cases, the condition can lead to pain, early osteoarthritis, and a requirement for hip replacement.[2, 4, 5]

Treatment options depend on the severity of the condition. In mild cases, the infant may be observed for several weeks prior to a decision to treat, as some cases may resolve on their own.[6] If treatment is elected (either immediately in more severe cases, or after observation in milder cases), first-line, non-surgical management in infants usually involves abduction splinting (that is, holding the hips in the correct position). Early diagnosis of DDH may improve clinical outcomes and contribute to a reduction in hip osteoarthritis in young adults.[2]

Clinical examination for DDH is commonly performed as part of the routine physical examination of newborns.[6] Several commonly performed physical tests can be used as part of the examination, including the Ortolani, Barlow, and Galeazzi tests.[2, 6] The Galeazzi test evaluates the following: posture, leg length, symmetry of the lateral profile of the pelvis, and hip range of motion during abduction of the thighs. The Ortolani and Barlow manoeuvres involve assessing for a 'clunk' or 'click' sound in the hips, indicating an unstable and potentially dislocated hip. However, these manoeuvres may not detect a dislocated, irreducible (that is, the dislocation cannot be restored to normal location with manipulation) hip. These manoeuvres may become more difficult in older infants (above two to three months), as the hip laxity (that is, looseness of the limb) reduces and the hip may remain in the dislocated position. Therefore, some potential cases of DDH may be missed by clinical examination alone.

Ultrasounds of the hip may allow for detection of cases of DDH that would have otherwise been missed by screening involving clinical examination only.[2, 7] Ultrasound can allow for evaluation of the components of the hips, and any alterations from the first few days of life. Several main techniques for ultrasound evaluation of the hip exist, including: the Graf technique (mainly used in Europe), the Harcke technique (mainly used in the USA), and Morin-Terjesen technique (mainly used in Scandinavian countries).

Ultrasound screening programmes for DDH can either be selective (that is, only those with certain risk factors or clinical signs undergo ultrasound screening), or

universal (that is, all infants undergo ultrasound screening).[8] While a universal ultrasound screening programme may detect cases of DDH that may be missed using either a selective or no ultrasound screening programme, potentially reducing the rates of late presenting DDH and associated complications, there is also the concern of overtreatment.[7-11] The optimal method of a screening programme, whether it be selective or universal, remains unclear.[7-13]

## 1.3 Evidence synthesis approach

HTA is a multidisciplinary process that summarises information about the medical, social, economic and ethical issues related to the use of a health technology and does so in a systematic, transparent, unbiased, and comprehensive manner. HTAs are designed to inform safe and effective health policies that are both patient focused and achieve the best value.

The HTAs conducted by HIQA's HTA Directorate follow the HTA Core Model® proposed by the European Network for Health Technology Assessment (EUnetHTA).[14] The domains selected for this evidence review are consistent with a rapid relative effectiveness assessment synthesis approach, and include:

- description of the technology
- epidemiology
- clinical effectiveness and safety

HIQA will convene a multidisciplinary expert advisory group (EAG) to advise the Evaluation Team during the course of the review. The role of the HIQA EAG is to inform and guide the process, provide expert advice and information, and to provide access to data where appropriate.

## 1.4 Aims and objectives

In the subsequent sections of this protocol, the scope and methods of the review are described according to the domains that will be assessed. The approach taken will be that of an overview of the key pertinent evidence rather a comprehensive examination of the full extent of the literature. The objectives of this review are as follows:

- describe the existing and proposed ultrasound screening for DDH in infants in Ireland, including initial clinical assessment, ultrasound screening referral algorithm, diagnostics, and treatment
- describe the international practice in the use of ultrasound screening for DDH
- describe the epidemiology and burden of disease of DDH

- provide an overview of the clinical effectiveness of a universal ultrasound screening programme for DDH as compared to a selective ultrasound screening programme.

# 2 Description of technology

The purpose of this chapter is to provide an overview of a universal ultrasound screening programme for DDH. The specific aims of this chapter will be to describe:

- the current recommendations for a selective ultrasound screening programme for DDH in Ireland, including initial clinical assessment, referral for screening algorithm, diagnostics, and treatment for infants
- the method of ultrasound screening for DDH in children, including test accuracy
- international practice in the use of ultrasound screening for DDH in infants.

Further details on these aims are described in sections 2.1 to 2.2.

## 2.1 Screening, diagnostic and treatment pathways in Ireland

The current and proposed method of ultrasound screening for DDH in infants will be described alongside the process for referral and treatment. This description will be informed by review of international clinical guidelines, publicly available literature, consideration of current Irish pathways, and by expert clinical opinion.

## 2.2 International practice

An overview of international practice, describing the countries that currently have ultrasound screening programmes in place for DDH and the ultrasound screening methods used, will be provided. The overview will be informed by reviewing grey literature sources (for example, national public health organisations, and the websites of governmental departments and relevant agencies), and recent peer-reviewed literature. The overview will focus on the following countries deemed to be of most relevance to Ireland, based on a combination of geographical proximity to Ireland, population size, European Union membership and or availability of documents in English:

EU/EEA

- Austria
- Belgium
- Denmark
- Finland
- France
- Germany

Non-EU

- Australia
- Canada
- Israel
- New Zealand
- United Kingdom

- Italy
- Netherlands
- Norway
- Portugal
- Spain
- Sweden
- Switzerland.

# 3 Epidemiology

The purpose of this chapter is to provide an overview of the epidemiology of DDH. The specific aims of this chapter will be to describe:

- the aetiology, symptoms and natural history of DDH
- the burden (incidence, prevalence, and morbidity) of DDH.

Where available, national data and relevant international data on the burden of disease associated with DDH will be presented. An overview of the literature and clinical data on the aetiology, symptom presentation and natural history of DDH will be presented. A broad scoping exercise will be conducted to identify relevant literature and national and international data sources.

# 4 Clinical effectiveness of screening

As previously noted, in Ireland, there are currently recommendations for a selective ultrasound screening programme for DDH. Accordingly, this section of the review aims to describe the clinical effectiveness and safety of selective ultrasound screening for DDH compared to universal ultrasound screening for DDH.

Preliminary scoping exercises identified a number of recent systematic reviews assessing the effectiveness of universal ultrasound screening for DDH compared with selective ultrasound. Therefore, in the first instance, a summary of relevant systematic reviews will be undertaken and the underlying studies will be examined to identify comparative evidence for the effectiveness of universal ultrasound versus selective ultrasound screening. The proposed question and methods are outlined below.

## 4.1 Review question

### 4.1.1 Eligibility criteria

The review question was formulated according to the Population, Intervention, Comparator, Outcome (PICO) framework. The inclusion and exclusion criteria for

eligibility for the summary of systematic reviews are presented in Table 2. The summary seeks to answer the following question:

- What are the outcomes of universal ultrasound screening for DDH compared to selective screening ultrasound for DDH?

**Table 2. Review question for assessing clinical effectiveness of universal screening for DDH**

| | |
|---|---|
| **Population** | Infants |
| **Intervention** | Universal ultrasound screening for DDH (including pilots) |
| **Comparator** | Selective ultrasound screening for DDH† |
| **Outcomes** | ▪ Clinical outcomes (for example, morbidity)<br>▪ Pathway timings (for example, time to diagnosis, time to treatment)<br>▪ Harms |
| **Study design** | Include:<br>▪ Systematic reviews (with or without a meta-analysis) that:<br>○ clearly state a set of objectives with an explicit, reproducible methodology<br>○ contain a systematic search that attempts to identify all studies that would meet the eligibility criteria<br>○ have a systematic presentation, and synthesis, of the characteristics and findings of the included studies.<br>○ compares universal ultrasound screening with selective ultrasound screening for DDH<br>○ reports one or more of the outcomes of interest<br>Exclude:<br>▪ Literature reviews not completed using systematic review methodology<br>▪ Non-human studies; papers not available in English or for which an adequate English translation cannot be obtained; letters, editorials, commentaries, preprints, and conference abstracts<br>▪ Reviews in a neonatal population with diagnosed hip pathology |

| | |
|---|---|
| | ▪ Reviews in which the comparator is clinical examination alone (that is, without selective ultrasound screening) |

† Selective ultrasound screening was defined as only screening infants with risk factors for DDH (for example, family history or breech presentation) or where abnormal results of clinical examination are evaluated by ultrasound.

### 4.1.2 Search methods

A search for relevant literature, as per the above PICO, will be conducted in PubMed using the specialised PubMed Clinical Queries tool. The search terms will include combinations and variations of the following, in accordance with the PICO framework: "selective", "targeted", "universal", "population-based", "screening", "developmental dysplasia of the hip", and "congenital dislocation of the hip". The literature will be supplemented with a Google Scholar search and a targeted search of the websites of selected HTA agencies. Results of the Google Scholar search will be sorted by relevance with the review article filter applied. Forward citation searching and searching of the reference lists of included studies will also be undertaken.

For the review question, we will include systematic reviews which compare universal ultrasound screening to selective ultrasound screening as the primary source of evidence.

### 4.1.3 Data collection and analysis

**Selection of studies**

All potentially eligible studies will be screened independently by two reviewers as per the inclusion criteria, with disagreements resolved by discussion.

**Data extraction and management**

Data extraction will be performed by one reviewer using Microsoft Excel software. All data extracted will be reviewed by a second person, with disagreements resolved by discussion. A standardised data extraction template will be developed prior to undertaking the review.

The following data, where available, will be extracted from the systematic reviews:

- eligibility criteria for inclusion in the systematic review
- number of relevant studies identified (that is, studies comparing universal ultrasound screening to selective ultrasound screening)
- population characteristics (country, sample size)

- outcomes
    - morbidity (for example, requirement for surgery)
    - pathway timings, where reported (for example, age at diagnosis, age at treatment, time to diagnosis, time to treatment)
    - harms (for example, overtreatment, false positives)
    - screening acceptability measures (for example, uptake rate)

### Quality assessment

The key studies will be assessed by one reviewer, with the assessment cross-checked by a second reviewer. The quality will be assessed using the ROBIS tool, which is a tool for assessing the risk of bias in systematic reviews. Results will be summarised narratively.

### Data synthesis

The findings of the included studies will be narratively synthesised. The synthesis will include a discussion on the potential harms from the universal screening for DDH.

# 5 Anticipated timeline

It is expected that a draft report will be completed in October 2023. However, this timeline is dependent on available resources and the extent of the literature. The draft report will then be circulated and reviewed at a meeting of the HIQA EAG. Necessary amendments and revisions to the draft report will be made following the meeting of the HIQA EAG before the final report is circulated to NSAC for consideration (expected delivery November 2023).

# References

1. Yang S, Zusman N, Lieberman E, Goldstein RY. Developmental dysplasia of the hip. Pediatrics. 2019;143(1).
2. Agostiniani R, Atti G, Bonforte S, Casini C, Cirillo M, De Pellegrin M, et al. Recommendations for early diagnosis of Developmental Dysplasia of the Hip (DDH): working group intersociety consensus document. Italian Journal of Pediatrics. 2020;46(1):1-7.
3. National Targeted Ultrasound Screening for Developmental Dysplasia of the Hip Steering Group. National Selective Ultrasound Screening Programme for Developmental Dysplasia of the Hip in Infants: Implementation Pack. 2017.
4. Kilsdonk I, Witbreuk M, Van Der Woude H-J. Ultrasound of the neonatal hip as a screening tool for DDH: how to screen and differences in screening programs between European countries. Journal of Ultrasonography. 2021;21(85):147-53.
5. Zomar BO, Mulpuri K, Schaeffer EK. Examining the short-term natural history of developmental dysplasia of the hip in infancy: a systematic review. Indian Journal of Orthopaedics. 2021:1-16.
6. Agarwal A, Gupta N. Risk factors and diagnosis of developmental dysplasia of hip in children. Journal of clinical Orthopaedics and Trauma. 2012;3(1):10-4.
7. Kuitunen I, Uimonen MM, Haapanen M, Sund R, Helenius I, Ponkilainen VT. Incidence of neonatal developmental dysplasia of the hip and late detection rates based on screening strategy: a systematic review and meta-analysis. JAMA Network Open. 2022;5(8):e2227638-e.
8. Jung HW, Jang WY. Effectiveness of different types of ultrasonography screening for developmental dysplasia of the hip: a meta-analysis. Medicine. 2020;99(50).
9. Pandey RA, Johari AN. Screening of newborns and infants for developmental dysplasia of the hip: A systematic review. Indian Journal of Orthopaedics. 2021:1-14.
10. Shipman SA, Helfand M, Moyer VA, Yawn BP. Screening for developmental dysplasia of the hip: a systematic literature review for the US Preventive Services Task Force. Pediatrics. 2006;117(3):e557-e76.
11. Shorter D, Hong T, Osborn DA. Cochrane Review: Screening programmes for developmental dysplasia of the hip in newborn infants. Evidence - Based Child Health: A Cochrane Review Journal. 2013;8(1):11-54.
12. Laborie LB, Rosendahl K, Dhouib A, Simoni P, Tomà P, Offiah AC. The effect of selective ultrasound screening on the incidence of late presentation of developmental hip dysplasia—a meta-analysis. Pediatric Radiology. 2023:1-12.
13. Woolacott NF, Puhan MA, Steurer J, Kleijnen J. Ultrasonography in screening for developmental dysplasia of the hip in newborns: systematic review. BMJ. 2005;330(7505):1413.
14. European Network for Health Technology Assessment (EUnetHTA). HTA Core Model: [Available from: https://www.eunethta.eu/hta-core-model/.

**Published by the Health Information and Quality Authority (HIQA).**

**For further information please contact:**

**Health Information and Quality Authority**

**George's Court**

**George's Lane**

**Smithfield**

**Dublin 7**

**D07 E98Y**

**+353 (0)1 8147400**

**info@hiqa.ie**

**www.hiqa.ie**