Health Information and Quality Authority

An tÚdarás Um Fhaisnéis agus Cáilíocht Sláinte

Evidence review of universal ultrasound screening for developmental dysplasia of the hip (DDH) in infants in Ireland

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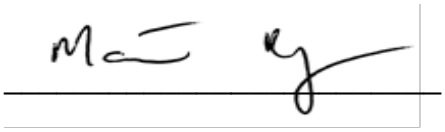
Table of contents

Foreword

The National Screening Advisory Committee (NSAC) was established in 2019 by the Minister for Health as an independent advisory committee to play a strategic role in the development and consideration of population-based screening programmes in Ireland. The role of the NSAC is to provide advice to the Minister for Health and the Department of Health on new screening proposals and proposed changes to existing screening programmes. 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.

Developmental dysplasia of the hip (DDH) is a congenital disease of the musculoskeletal system in which there is abnormal development of the hip in infancy. The condition can have a wide range of severity, which, along with the age of diagnosis, impacts the first-line treatment choice for the condition. Ultrasound screening for DDH is possible and has the potential to enable earlier identification and diagnosis, thereby facilitating earlier and less invasive treatment than if the condition is detected later.

Work on this evidence review was undertaken by an Evaluation Team from the HTA Directorate in HIQA. A multidisciplinary Expert Advisory Group was convened to advise the Evaluation Team during the course of the HTA. HIQA would like to thank the Evaluation Team, the members of the Expert Advisory Group and all who contributed to the preparation of this report.



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Deputy Chief Executive and Director of Health Technology Assessment

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HIQA further notes that the findings set out in the advice represent the interpretation by HIQA of the available evidence and do not necessarily reflect the opinion of all members of the EAG.

The membership of the EAG was as follows:

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Evaluation Team

The following individuals contributed to the management, technical writing or dissemination of this report:

Arielle Maher (project lead), Éanán Finnegan, Karen Jordan, Louise Larkin, Michelle O'Neill, Michelle O'Shea, Máirín Ryan, Debra Spillane, Susan Spillane.

Conflicts of interest

No conflicts of interest were declared.

Key findings and Advice to the NSAC

At the request of the National Screening Advisory Committee (NSAC), HIQA agreed to undertake an evidence review of universal ultrasound screening for developmental dysplasia of the hip (DDH) in infants in Ireland.

The approach taken was that of an overview of the key pertinent evidence rather than a comprehensive examination of the full extent of the literature. The domains of the HTA Core Model® selected for this evidence review are consistent with a rapid relative effectiveness assessment synthesis approach (that is, focusing on the clinical benefit of the intervention) and include: 1) description of the technology; 2) epidemiology; 3) clinical effectiveness and safety.

The key findings of this evidence review, which informed HIQA's advice to NSAC, were:

- DDH is a congenital disease of the musculoskeletal system in which there is abnormal development of the hip in infancy. The severity can range from mild dysplasia to complete hip dislocation.
    - There are several major risk factors for DDH, including breech position in utero, female sex, and family history of DDH. However, the majority of cases of DDH have no identifiable risk factor.
    - If DDH is not identified and treated in the early months of life, most of the significant cases of dysplasia will present symptomatically after walking age.
    - Hip instability is age dependent, and decreases in the first weeks of life with increases in muscle tone. Many cases of mild hip instability identified in newborns resolve without treatment within the first six to eight weeks of life; however, the true proportion is unclear.
    - Persistent dysplasia and dislocation can lead to wear of the cartilage of the hip and osteoarthritis in young adults. DDH is a common cause of total hip replacements in young adults.
- The incidence of DDH varies geographically due to genetic and cultural differences (such as post-natal swaddling practices). Reasons for variation in the reported incidence include differences in the age at examination, experience of the examiner, screening methods (for example, clinical examination only, selective ultrasound screening or universal ultrasound screening), screening protocols, and the definition of DDH applied.

  - There is no national centralised database of cases of DDH in Ireland. Therefore, there is considerable uncertainty regarding the current incidence of DDH in Ireland. Estimates of the incidence of DDH were available from a single centre and ranged between 7 per 1,000 and 31 per 1,000. However, these estimates may not be nationally representative of practice or case numbers in the rest of Ireland.

- Clinical examinations performed on newborns within the first few days of life, including investigation of hip stability, can serve as the first point of screening and identification of DDH. However, cases of DDH may go undetected by clinical examination alone.

- No internationally accepted treatment guidelines for DDH were identified. However, the following principles of treatment were apparent from the literature:

  - Consensus-based recommendations from two multidisciplinary expert groups, based primarily on expert opinion, support early diagnosis and treatment of DDH.
  - Treatment becomes more invasive, with an increase in the risk of treatment complications, as the age of infant at first treatment increases.
  - Cases who are less than six weeks of age may be monitored for resolution of hip instability over time. If DDH is diagnosed when the baby is less than six months old, treatment with a Pavlik harness may be advised. For older infants, more invasive treatments or surgery may be required.
  - Avascular necrosis of the femoral head is a potential major treatment-related complication which results from disruption in the blood supply to the bone; however, the risk of this complication is understood to be low. As a result, adequate identification must be balanced against the risk of unnecessary treatment.

- Ultrasound may be used as part of the diagnostic pathway for DDH as well as for screening for the condition. While several techniques for ultrasound examination for DDH exist, the Graf technique is currently used for screening and diagnosis of DDH in Ireland.

  - Ultrasound screening may detect DDH cases that may have been missed with clinical examination alone, potentially reducing the rates of late presenting DDH and associated complications. However, ultrasound screening can result in unnecessary treatment mostly due to the potential for spontaneous correction of the identified DDH. This risk of unnecessary treatment may be reduced by training in Graf technique and classification.

- Ultrasound screening programmes for DDH can be selective or universal. With selective screening, only those with certain risk factors and or clinical signs undergo ultrasound screening. Universal ultrasound screening means that all infants undergo ultrasound screening. The optimal ultrasound screening programme, whether it be selective or universal, remains unclear.
    - A review of ultrasound screening in 18 countries found that universal ultrasound screening for DDH was nationally implemented in two countries, while 14 have selective ultrasound screening based on clinical signs and or risk factors. No programme or guidelines on ultrasound screening for DDH were identified from the remaining two countries.

- Recommendations for a selective ultrasound screening programme for DDH in Ireland were published in 2017. As of November 2023, the recommendations are included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme. This programme sits within the governance of the Health Service Executive (HSE) Clinical Design and Innovation Division.
    - A national audit of centres performing ultrasound to assess for DDH in 2021 and 2022 found that 18.5% of all infants born were referred for ultrasound of the hips. It is important to note that the data underlying this statistic may not be complete as information was submitted voluntarily by individual hospitals.
    - As the recommendations have not been implemented under a formal national screening programme, the pathway does not have the governance and quality assurance that would be included in such a programme.
    - There is evidence to suggest that there may be variation in the implementation of this targeted clinical diagnostics programme.

- A summary of reviews was undertaken to synthesise and assess the clinical effectiveness and safety of universal ultrasound screening for DDH, relative to selective ultrasound screening.
    - Across eight systematic reviews, four primary studies were identified which compared universal with selective screening for DDH. These included two randomised controlled trials (RCTs) and two retrospective cohort studies.
    - For the outcome of late DDH, meta-analysis of the two RCTs found that universal ultrasound screening did not result in a statistically significant reduction in late DDH, compared to selective ultrasound screening.

Similarly, in one retrospective cohort study, there was no significant difference in the incidence of late DDH between the universal (0.50 per 1,000; n = 10,015 births) and selective (0.28 per 1,000 births; n = 18,053) ultrasound screening groups. Of note, cases of late DDH in the universal screening group were reported to be due to clinical appointments not being made or kept, rather than representing false negative ultrasound results. This underscores the importance of considering the overall effectiveness of the care pathway in providing end-to-end care.

- There is evidence of increased non-surgical intervention associated with universal ultrasound screening, relative to selective screening, without a corresponding reduction in the incidence of late DDH or requirements for surgical intervention. This suggests that the additional cases identified through universal screening are likely mild, and may resolve spontaneously in the absence of treatment.
- No data on functional complications were identified. Evidence of potential harms was limited to avascular necrosis of the femoral head, with no statistically significant differences found between the universal and selective ultrasound screening groups. However, studies were likely underpowered for this outcome (that is, the studies were not suitably designed to assess this).

- Overall, the relative benefit of a universal ultrasound screening programme compared with selective screening is unclear in the absence of high-quality comparative studies. Although limited, the available evidence suggests that screening all infants with ultrasound for the detection of DDH has the potential to lead to unnecessary treatment, with the risk of clinically significant consequences.

Arising from this evidence review, HIQA's advice to NSAC is as follows:

- Developmental dysplasia of the hip (DDH) is a congenital disease in which there is abnormal development of the hip in infancy. The severity can range from mild dysplasia to complete hip dislocation.
  - While many cases of mild hip instability identified in newborns resolve without treatment within the first six to eight weeks of life, persistent dysplasia and dislocation can lead to wear of the cartilage of the hip and osteoarthritis in young adults.
- Some cases of DDH may be missed by clinical examination alone. Ultrasound screening can be used to support identification of a greater number of cases. However, it is unclear whether or not all cases identified would be clinically

significant (that is, requiring intervention). The risk of unnecessary treatment in cases that would resolve without treatment may be reduced by training in Graf technique and classification.
    - Studies of universal ultrasound screening relative to selective ultrasound screening identified within this review were underpowered to detect potential harms. Therefore, there is insufficient evidence to assess the benefit-harm balance.
    - The optimal ultrasound screening programme, whether it be selective or universal, remains unclear.
- Recommendations for a selective ultrasound screening programme for DDH in Ireland were published in 2017. As of November 2023, these recommendations are included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme.
    - There is evidence to suggest that there may be variation in the implementation of the current recommendations for ultrasound screening in Ireland. Further understanding of current practice and barriers to uptake of the existing recommendations across Ireland would facilitate successful implementation of a formal screening programme.
    - Consideration could be given to implementing the current selective ultrasound screening recommendations as a formal screening programme with appropriate governance, end-to-end care, quality assurance and monitoring of outcomes.

Executive Summary

This report summarises a review of the evidence surrounding universal ultrasound screening for developmental dysplasia of the hip (DDH).

Background

DDH is a congenital disease of the musculoskeletal system in which there is abnormal development of the hip in infancy. The severity can range from mild dysplasia to complete hip dislocation.

In March 2023, at the request of National Screening Advisory Committee (NSAC), HIQA agreed to undertake an evidence review of universal ultrasound screening for developmental DDH in infants in Ireland.

Methods

This research was carried out in accordance with HIQA's guidelines for the conduct of HTAs. In summary, the following took place:

- The Terms of Reference of the evidence review were agreed between HIQA and the Chair of the NSAC, on behalf of the NSAC.
- An Expert Advisory Group (EAG) was convened by HIQA comprising representation from relevant stakeholders. These included: the Department of Health, the Health Service Executive (HSE), the Irish Society of Chartered Physiotherapists, the Irish Institute of Radiography and Radiation Therapy, parent representatives (Cuidiú), and clinicians with specialist expertise in public health, neonatology, paediatrics, radiology and orthopaedics.
- Potential diagnostic and treatment modalities as well as current Irish screening recommendations were described.
- The epidemiology of DDH in Ireland and internationally was described.
- A brief review of the clinical effectiveness of universal ultrasound screening compared to selective ultrasound screening was carried out.

Description of technology

DDH is a congenital disease of the musculoskeletal system in which there is abnormal development of the hip in infancy, with a wide range of severity. In mild cases, symptoms may resolve without treatment. However, in more severe cases, the condition can lead to pain, early osteoarthritis, and a requirement for hip replacement in the longer term.

Clinical examinations performed on newborns within the first few days of life can serve as the first point of screening and identification of DDH. However, some cases of DDH may be missed using clinical examination alone. It is also important to note that hip instability is age dependent, and decreases in the first weeks of life with increases in muscle tone. Ultrasound screening can detect DDH cases that may have gone undetected with clinical examination alone. However, while ultrasound screening would detect most cases of DDH, it cannot distinguish between those that may resolve spontaneously and those that would require intervention. The risk of unnecessary treatment for those cases which would resolve spontaneously may be reduced by training in Graf technique and classification.

Ultrasound screening programmes for DDH can be selective or universal. With selective screening, only those with certain risk factors or clinical signs undergo ultrasound screening. In universal ultrasound screening all infants undergo ultrasound screening. The optimal method of an ultrasound screening programme, whether it be selective or universal, remains unclear. Recommendations for a selective ultrasound screening programme for DDH in Ireland were published in 2017. As of November 2023, the recommendations have not been implemented as a screening programme in Ireland. Rather, the recommendations are included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme. This programme sits within the governance of the Health Service Executive (HSE) Clinical Design and Innovation Division. As the recommendations have not been implemented under a formal national screening programme, the pathway does not have the governance and quality assurance that would be included in such a programme. There is evidence to suggest that there may be variation in the implementation of this targeted clinical diagnostics programme.

No internationally accepted guidelines for the treatment of DDH were identified as part of this review. Treatment recommendations for DDH, for example, use of Pavlik harness or reduction surgery, generally depend on the age of the patient; as the age at which the infant starts treatment increases, treatment becomes more invasive (with an increase in the risk of treatment complications). A large proportion of cases of DDH will experience resolution of hip instability without treatment; therefore, in some cases, the approach taken will be to monitor the condition. Consensus-based recommendations from two multidisciplinary expert groups, based primarily on expert opinion, supported early diagnosis and treatment of DDH.

A review of ultrasound screening in 18 countries found that universal ultrasound screening for DDH was nationally implemented in two countries, while 14 have selective ultrasound screening based on clinical signs and or risk factors. No programme or guidelines on ultrasound screening for DDH were identified for the two remaining countries included in the review.

Epidemiology and burden of disease

DDH occurs when there is an abnormality in the development of the hip. The severity can range from mild dysplasia to complete hip dislocation. There are several major risk factors for DDH, including breech position in utero, female sex, and family history of DDH. Other factors associated with risk of DDH include being a firstborn child, foot abnormalities, tilted neck position, low amniotic fluid during pregnancy, high birth weight, being born overdue, and postnatal swaddling of the infant; however, limited evidence exists for the strength of their association or ability to predict the occurrence DDH.

The natural history of DDH is poorly understood. If the condition is not identified and treated in the early months of life, most of the significant cases of dysplasia will present symptomatically after walking age. Many cases of mild hip instability identified in newborns resolve without treatment within the first six to eight weeks of life; however, the true proportion is unclear. Persistent dysplasia and dislocation can lead to wear of the cartilage of the hip and osteoarthritis in young adults. Studies in Denmark and Norway have found that DDH is a common cause of total hip replacements in young adults, representing the cause of 2.6% to 9.1% of total hip replacements overall. Irish data similarly showed that in patients under 50 years of age who were undergoing total hip replacement, over 40% had evidence of dysplasia. However, it was not reported whether this dysplasia was present when the patients were infants or developed later in life. Considering patients with DDH, one systematic review, with more than 40-years follow-up, assessed long-term outcomes of late detected DDH hips treated after walking age; this study found that up to 35% of such patients required total hip replacement.

The incidence of DDH varies geographically due to factors such as genetic and cultural differences (such as post-natal swaddling practices). Reasons for variation in the reported incidence include differences in the experience of the examiner, screening methods (for example, clinical examination only, selective ultrasound screening or universal ultrasound screening), screening protocols (for example, age at examination or timing of follow-up examination) and definitions of DDH applied.

There is no national centralised database of cases of DDH in Ireland. Therefore, it is challenging to estimate the incidence of the condition. Available incidence estimates are from a single centre. The estimates of the incidence of DDH at this centre ranged between 7 per 1,000 (based on children born in 2009, prior to the publication of the 2017 national recommendations for selective ultrasound screening) and 31 per 1,000 (based on children diagnosed in 2018 and 2019, after the publication of the recommendations). Of note, these estimates may not be representative of practice and case numbers in the rest of Ireland.

Clinical effectiveness of screening

A summary of reviews was undertaken to synthesise and assess the clinical effectiveness and safety of universal ultrasound screening for DDH, relative to selective ultrasound screening.

Eight relevant systematic reviews were identified. Outcomes considered by the systematic reviews included incidence of late DDH, rates of non-surgical and surgical treatment, functional complications (for example, delayed walking, gait disturbances or pain), and harms (for example, unnecessary treatment or psychological distress).

Concerning late DDH, meta-analysis of the two randomised controlled trials (RCTs) found that universal ultrasound screening did not result in a statistically significant reduction in this outcome, compared to selective ultrasound screening (relative risk (RR) 0.49, 95% confidence interval (CI): 0.19 to 1.26). In one retrospective cohort study, there was no significant difference in the incidence of late DDH between the universal (0.50 per 1,000; n = 10,015 births) and selective (0.28 per 1,000 births; n = 18,053) ultrasound screening groups. Of note, cases of late DDH in the universal screening group were reported to be due to clinical appointments not being made or kept, rather than representing false negative ultrasound results.

For non-surgical treatment rates, universal ultrasound screening for DDH may be associated with an increase in the rate of this outcome, relative to selective screening, although the magnitude of this effect is subject to uncertainty. For surgical intervention, meta-analysis of two RCTs found no statistically significant difference in requirements for surgical intervention with universal screening relative to selective screening (RR 0.36, 95% CI: 0.04 to 3.48). Consistent with these findings, one retrospective cohort study identified reported no significant difference in the incidence of surgical intervention between the universal and selective screening cohorts. The findings of the second retrospective cohort study were unclear.

Taken together, there is evidence of increased non-surgical intervention associated with universal ultrasound screening, relative to selective screening, without a corresponding reduction in the incidence of late DDH or requirements for surgical intervention. This suggests that the additional cases identified through universal screening are likely mild, and may resolve spontaneously in the absence of treatment.

No data on outcomes of functional complications were identified. In included RCTs (n = 2), evidence of potential harms was limited to avascular necrosis of the femoral head, with no significant differences found between the universal and selective ultrasound screening groups. However, studies were likely underpowered for this outcome. Outcomes of the universal ultrasound DDH screening programme in Austria demonstrate a reduction in hospital admissions and the number of patients requiring surgery since the introduction of the programme in 1992. These findings

are unlikely to be directly attributable to ultrasound screening alone; rather, they may have represented the beneficial combined effects of a structured screening programme and changes in clinical practice over time. It is important to note, however, that the non-comparative nature of the evidence does not provide insight into the relative benefit of a universal screening compared with selective screening.

The relative benefit of universal ultrasound screening, compared with selective screening, is unclear in the absence of high-quality comparative studies. Although limited, the available evidence suggests that screening all infants with ultrasound for the detection of DDH has the potential to lead to unnecessary treatment, with the risk of clinically significant consequences.

Conclusion

The purpose of this report was to provide an evidence review of universal ultrasound screening for developmental dysplasia of the hip in infants in Ireland. Overall, the information included in this evidence review was limited for several reasons; these included the lack of international guidelines for screening for DDH and the limited evidence available for the natural history, aetiology and epidemiology of the condition. There was limited evidence found to determine whether universal ultrasound screening for DDH compared to selective ultrasound screening leads to improved functional outcomes, decreased need for surgical interventions and reduced harms. These knowledge gaps combine to produce significant uncertainty regarding the benefit of introducing a universal ultrasound screening programme over the current recommendations for selective ultrasound screening in Ireland.

Given the variable natural history of DDH, with a high rate of spontaneous resolution of hip instability, and the potential risk of serious complications from treatment, the potential benefits of earlier diagnosis which may be achieved through widespread screening need to be weighed against the potential harms of unnecessary treatment. In the absence of nationally representative data regarding outcomes of current selective ultrasound practices in Ireland, the relative benefit of a universal ultrasound screening programme is uncertain.

The 2017 recommendations for selective ultrasound screening in Ireland have not been implemented as part of a formal national screening programme, and current practice is therefore not supported by the governance, monitoring and evaluation that would be associated with such a programme. Further understanding of current practice and barriers to following the recommendations across Ireland may facilitate the successful implementation of a formal screening programme to ensure standardised practice across the country.

Plain Language Summary

Developmental dysplasia of the hip (DDH) is a condition in which the 'ball and socket' joint of the hip does not form correctly in babies and young children. The top of the bone of the thigh (or 'ball') sits inside a 'socket' that is part of the pelvis. In a normal hip joint, the ball sits firmly in the socket. This helps the hips to remain stable during movement and weight-bearing activities, such as standing and walking.

DDH includes a wide range of hip problems. Some babies have a mild looseness in one or both of their hips. For other babies, the ball easily comes completely out of the socket, which can mean the bone can be pulled out of its joint completely, known as a dislocation. If DDH is not treated, the hip joint may not form correctly, which could lead to pain while walking or arthritis of the hip at a young age. This may then lead to the need for a hip replacement in later life. The cause of DDH is unknown, and there are likely a number of reasons why people have DDH. There are several risk factors for DDH. These include breech position during late pregnancy (where the baby is lying bottom-down in the womb), being born female and having a family member with DDH.

It is important to note that a baby's hips continue to develop during their first weeks of life. As a result, many cases of DDH identified shortly after birth may resolve without treatment; however, the exact amount is hard to know. It is difficult to estimate how many babies are born each year with DDH. In Ireland, a small number of studies estimate that there are between 400 and 1,800 babies born with DDH each year. However, not all of these children would require treatment.

To look for DDH and for other health problems, newborns have a physical examination after birth and regularly during early life. This is carried out by a trained healthcare professional. When looking for DDH, the examination aims to find out if the baby's hips are stable, unstable or dislocated. However, this physical examination cannot find all cases of DDH. If cases are missed, this can lead to a later diagnosis and older age at starting treatment. Ultrasound screening is an imaging test that can be used to produce pictures of the inside of the body. It can be used in addition to physical examination to help find cases that may be missed. Ultrasound screening can be 'selective' or 'universal'. With selective ultrasound screening, only babies with risk factors or clinical signs found during the physical examination undergo ultrasound screening. With universal screening, every baby undergoes ultrasound screening. Ultrasound screening can identify cases of DDH, but it cannot tell which babies have mild DDH that would resolve over time without treatment, and which babies have DDH that will require treatment. However, this may be improved if those doing and reviewing the ultrasound are closely following certain methods used with ultrasound. In Ireland, recommendations for a selective ultrasound screening programme which would look for or 'screen' for DDH were

published in 2017. However, a formal screening programme has not been implemented. A formal screening programme ideally includes the complete care pathway from screening and diagnosis through to long-term follow-up. Formal screening programmes also have processes for monitoring how well the programme is performing. As a formal screening programme is not in place, it is unclear if the recommendations for selective ultrasound screening are followed exactly in all hospitals.

The goal of treatment is to put the ball of the hip in the socket and to keep it there to allow the joint to develop normally. The type of treatment depends on a number of factors including the baby's age and the severity of their condition (how much of the ball is outside the hip socket). At an older age, treatment involves a more complicated surgery with a higher risk of problems, such as damage to the bone. If DDH is diagnosed when the baby is less than six months old, the baby may need to wear an abduction brace called a Pavlik harness to hold their hips in place. This helps to encourage the hip joint to form correctly. The Pavlik harness allows some movement in the legs, and so is less likely to result in complications. The harness usually works well at keeping the hips in the correct position. If the baby is more than six months old at the time of diagnosis, or if one or both of the baby's hips are still partly or completely dislocated even with the use of a harness, the baby may need more invasive procedures, or surgery. If older than six months, the baby may need to wear a solid cast to hold its hips in place. In some cases, typically in toddlers, surgery may be required to place the ball of the joint back into the socket. No internationally accepted guidelines for diagnosis and treatment of DDH were identified and treatment pathways appear to vary between countries.

HIQA performed a review of published evidence on the clinical effectiveness and safety of universal ultrasound screening versus selective ultrasound screening. This found that universal ultrasound screening resulted in greater use of treatments like the Pavlik harness, as compared with when selective ultrasound screening is used. However, universal ultrasound screening did not result in a fewer number of late diagnoses of DDH or requirements for surgery. Only two clinical trials reported on the potential harms of universal ultrasound screening compared with selective ultrasound screening. Based on evidence from these two clinical trials, there was no difference found in the risk of damage to the hip when comparing the universal and selective screening groups. However, the identified studies were not designed to investigate this, so an increased risk of these harms cannot be ruled out. A larger study would be needed to assess the potential harms of universal relative to selective ultrasound screening.

In summary, weighing up the possible benefits and harms, it is not known if universal ultrasound screening for DDH is better than selective ultrasound screening. This is due to weaknesses in the information and evidence available. The 2017

recommendations for selective ultrasound screening in Ireland have not been applied as part of a formal screening programme. It would be helpful to have a better understanding of the current ways that DDH is tested across Ireland. This would be useful information for planning how a screening programme should be organised, if a decision is made to introduce a screening programme.

List of abbreviations used in this report

AAOS	American Academy of Orthopaedic Surgeons
AVN	avascular necrosis
DDH	developmental dysplasia of the hip
CI	confidence interval
EAG	Expert Advisory Group
HIQA	Health Information and Quality Authority
HTA	Health Technology Assessment
NSAC	National Screening Advisory Committee
NPV	negative predictive value
PPV	positive predictive value
RCT	randomised controlled trial
RR	relative risk or risk ratio
USPSTF	US Preventive Services Task Force
UHW	University Hospital Waterford

1 Introduction

1.1 Background to the request

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] Early diagnosis of DDH may improve clinical outcomes and contribute to a decrease in hip osteoarthritis in young adults.[2] Treatment effectiveness may be maximised when initiated within the first few days to months of life. Additionally, earlier detection may allow for nonsurgical treatment with abduction splinting (that is, holding the hips in the correct position).

Clinical examination for DDH is commonly performed as part of the routine physical examination of newborns.[3] However, some potential cases of DDH may be missed by clinical examination alone.[2-4] Ultrasound may be used to screen for DDH in infancy.[2, 5] Ultrasound screening may allow for the detection of cases of DDH that may have been missed with clinical examination alone, potentially reducing the rates of late presenting DDH and associated complications. However, ultrasound screening can result in unnecessary treatment, which is mostly due to the potential for spontaneous correction of DDH.[5-10] This risk of unnecessary treatment may be reduced by training in Graf technique and classification.[11, 12] Ultrasound screening programmes for DDH can either be selective or universal. With selective ultrasound screening, only infants with one or more risk factors (for example, family history or breech presentation) or clinical signs undergo ultrasound screening. In a universal ultrasound screening programme, all infants undergo ultrasound, irrespective of their risk factors.[7] The optimal method of an ultrasound screening programme, whether it be selective or universal is unclear.[5-9, 13, 14]

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 selective ultrasound screening programme for infants at risk of DDH.[15] 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. Of note, as of November 2023, the recommendations have not been implemented as a screening programme in Ireland.[16] Rather, the recommendations are included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme. This programme sits within the governance of the Health Service Executive (HSE) Clinical Design and Innovation Division, and does not have the governance of a screening programme.

1.2 Terms of reference

The terms of reference for this evidence review, as agreed between HIQA and the Chair of the NSAC, on behalf of the NSAC, are to:

- Describe the existing and proposed ultrasound screening for developmental dysplasia of the hip (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.
- Produce a report summarising the above pieces of work.
- Convene meetings of the HIQA Expert Advisory Group (EAG), and present the above findings to the EAG for their interpretation and input.
- Subject to review and approval by the CEO, provide a final report summarising the overall findings of the assessment and HIQA's advice to NSAC.

1.3 Overall approach

The approach taken was that of an overview of the key pertinent evidence rather than a comprehensive examination of the full extent of the literature. The domains of the HTA Core Model® selected for this evidence review are consistent with a rapid relative effectiveness assessment synthesis approach (that is, focusing on the clinical benefit of the intervention), and include: 1) description of the technology; 2) epidemiology; 3) clinical effectiveness and safety.

A multidisciplinary Expert Advisory Group (EAG) was convened by HIQA comprising representation from relevant stakeholders including the Department of Health, the HSE, the Irish Society of Chartered Physiotherapists, the Irish Institute of Radiography and Radiation Therapy, parent representatives (Cuidiú), and clinicians with specialist expertise in public health, neonatology, paediatrics, radiology, and orthopaedics. The role of the EAG is to inform and guide the process, provide expert advice and information, and to provide access to data where appropriate. A full list of the membership of the EAG is available in the acknowledgements section of this report.

The terms of reference of the EAG are to:

- Contribute to the provision of high quality research and considered advice by HIQA to NSAC on behalf of the Minister for Health.
- Contribute to the work of the group by providing expert guidance, as appropriate.
- Be prepared to provide expert advice on relevant issues outside of group meetings, as requested.
- Provide advice to HIQA regarding the scope of the assessment.
- Review the project plan outline and advise on priorities, as required.
- Support the Evaluation Team during the assessment process by providing expert opinion and access to pertinent data, as appropriate.
- Review the draft report from the Evaluation Team and recommend amendments, as appropriate.
- Contribute to HIQA's development of its approach to HTA by participating in an evaluation of the process on the conclusion of the assessment.
- Notify the project lead if a nominee can no longer participate or contribute to the process, as non-participation may require alternative EAG membership to be sought.

At the meeting of the Expert Advisory Group, the Terms of Reference of the evidence review were reviewed and the draft chapters of the report discussed. Draft versions of this report were circulated for review by the EAG and were amended as appropriate. Consistent with standard HIQA governance, the final draft of the evidence review was submitted to the CEO for review and approval. Following approval, the finalised report will be submitted to NSAC for consideration and published on the HIQA website.

2 Description of the technology

Key points

- Developmental dysplasia of the hip (DDH) is a congenital disease of the musculoskeletal system in which there is abnormal development of the hip in infancy, with a wide range of severity.
    - In mild cases, spontaneous resolution may occur without treatment. However, in more severe cases, the condition can lead to pain, early osteoarthritis, and a requirement for hip replacement in the longer term.
- Clinical examinations performed on newborns within the first few days of life can serve as the first point of screening and identification of DDH. However, this has low accuracy for detection of DDH, and cases of DDH may go undetected by clinical examination alone. It is not clear how many of these cases would be clinically significant (that is, require intervention). It is also important to note that hip instability is age dependent, and decreases in the first weeks of life with increases in muscle tone.
- Ultrasound may be used as part of the diagnostic pathway for DDH as well as for screening for the condition. Anatomical alteration of the hip can be detected with ultrasound within the first few days of life. While several techniques for ultrasound examination for DDH exist, the Graf technique is currently used in Ireland.
    - Ultrasound screening may detect DDH cases that may have been missed with clinical examination alone, potentially reducing the rates of late presenting DDH and associated complications. However, ultrasound screening can result in unnecessary treatment mostly due to the potential for spontaneous correction of the identified DDH. This risk of unnecessary treatment may be reduced by training in Graf technique and classification.
- Ultrasound screening programmes for DDH can be selective or universal. With selective ultrasound screening, only those with certain risk factors or clinical signs undergo ultrasound screening. Universal ultrasound screening means that all infants undergo ultrasound screening. The optimal method of an ultrasound screening programme, whether it be selective or universal, remains unclear.
- Recommendations for a selective ultrasound screening programme for DDH in Ireland were published in 2017. As of November 2023, the recommendations have not been implemented as a screening programme in Ireland. Rather, the

recommendations are included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme. As the recommendations have not been implemented under a formal national screening programme the pathway does not have the governance, end-to-end care, and quality assurance that would be included in such a programme.

- No internationally accepted guidelines for the treatment of DDH were identified. Treatment recommendations for DDH generally depend on the age of the patient. As the age at which the infant starts treatment increases, treatment becomes more invasive, with an increase in the risk of treatment complications.
    - Consensus-based recommendations from two international multidisciplinary expert groups, based primarily on expert opinion, supported early diagnosis and treatment of DDH.
- A review of ultrasound screening in 18 countries found that universal ultrasound screening for DDH was nationally implemented in two countries, while 14 have selective ultrasound screening based on clinical signs and or risk factors. No programme or guidelines on ultrasound screening for DDH were identified for the remaining two countries.

2.1 Introduction

The purpose of this chapter is to describe key elements of the technology under consideration, namely the ultrasound screening programme for developmental dysplasia of the hip (DDH). The diagnosis and treatment of DDH are briefly described, followed by a description of the current selective ultrasound screening recommendations for DDH in infants in Ireland and a proposed universal ultrasound screening programme. An overview of the status of universal and selective ultrasound screening programmes internationally is then presented.

2.2 Developmental dysplasia of the hip

DDH, also known as congenital dislocation of the hip or hip dysplasia, is a congenital disease of the musculoskeletal system in which there is abnormal development of the hip in infancy.[1, 2, 17] In normal development of the hip joint, the head of the femur (that is, the thigh bone) should be rounded and sit within the cup-shaped acetabulum (that is, the hip socket). However, in those with DDH, the acetabulum is too shallow, and the head of the femur is not properly held in place. Therefore, the hip joint is loose. In severe cases, the femur can come out of the acetabulum, known as a dislocation.

A precise definition of DDH varies, but includes a spectrum of hip abnormalities from mild dysplasia (that is, the femur not sitting properly within the acetabulum), subluxated (that is, a partial dislocation), dislocatable (that is, has the potential to dislocate), and dislocated. In those with DDH, the acetabulum, the femoral head, or both can be deformed.[18] 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. Hip instability is also noted to be age dependent, and decreases in the first weeks of life with increases in muscle tone.[19] The aetiology of DDH is multifactorial. Risk factors are described in Chapter 3: epidemiology.

The natural history of DDH depends on severity.[18] DDH is usually asymptomatic at birth. If the condition is not identified and treated in the early months of life, most of the significant cases of dysplasia will present symptomatically after walking age.[2, 18] 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, spontaneous resolution can occur without treatment, but in more severe cases, the condition can lead to pain, early osteoarthritis, and a requirement for hip replacement in the longer term.[2, 18, 20] Residual complications from DDH are the leading cause of early hip osteoarthritis in adults.[1] Further details of the natural history are described in Chapter 3: epidemiology.

2.2.1 Screening and diagnosis of DDH

Clinical examination

Clinical examinations are usually performed for newborns in the first few days of life, with testing for DDH commonly performed as part of the newborn physical examination.[3] These examinations represent a form of screening for DDH, based on physical examination alone. Several commonly performed physical tests are often performed as part of the examination, including the Galeazzi test and the Ortolani and Barlow manoeuvres.[2-4, 19] 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 manoeuvre tests the hip joint's reducibility (that is, ability to position correctly in the socket). Forward pressure is applied to each femoral head, in an attempt to move a potentially dislocated femoral head forward into the acetabulum. Movement and a 'clunk' sound in the hips suggests an unstable and potentially dislocated hip. The Barlow manoeuvre tests for subluxation (that is, partial dislocation) of the hip joints. Backwards pressure is applied to each femoral head, to check for subluxation of the femoral head, which indicates a partial or complete dislocation. The Ortolani and Barlow manoeuvres may not detect a dislocated, irreducible (that is, the dislocation cannot be restored to

normal location with manipulation) hip, and the 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. Additionally, as these manoeuvres aim to detect unstable joints, they may not detect milder forms of dysplasia, which may not have clinical symptoms.

Cases of DDH may be missed by clinical examination alone.[2-4] A 2001 systematic review by the Canadian Task Force on Preventive Health Care aimed to explore the effectiveness of serial clinical examination (that is, multiple clinical examinations at various time intervals) for DDH.[21] The review found that the Barlow manoeuvre alone had a high negative predictive value (NPV) (that is, the probability that when a test result is negative, the person truly does not have the condition), at 99%, but a low positive predictive value (PPV) (that is the probability that when a test result is positive, the person truly has the condition), at 22%. Additionally, the Ortolani manoeuvre is reported to have a sensitivity (that is, the proportion of those with the condition that are correctly classified as positive by a test) of only 60%.[22] However, when the Ortolani and Barlow manoeuvres were used in combination, the specificity (that is, the proportion of those without the condition that are correctly classified as negative by a test) ranged from 98% to 99%. Another systematic review, published in 2022, which aimed to compare clinical examination versus ultrasound for detection of DDH, found that the Ortolani-Barlow tests in combination have a sensitivity of 36% (95% confidence interval (CI): 25% to 48%) and specificity of 98% (95% CI: 0.93 to 0.99).[23] When these manoeuvres were used in combination with tests of hip abduction, the sensitivity was found to be 57% (95% CI: 30% to 82%) and specificity was 95% (95% CI: 68% to 99%). These reviews noted that low and varied sensitivity may be due to the skill of the clinical examiner, the number of examinations performed, and or the timing of the examinations.[21, 23] Based on these results, the evidence suggests that clinical examination is associated with a low risk of false positive results, but many cases of DDH would be missed by physical examination alone.

Ultrasound

Ultrasound may be used as part of the diagnosis of DDH as well as for screening.[2, 5] Ultrasound can allow for evaluation of the components of the hips and any anatomical alterations from the first few days of life. Several main techniques for ultrasound evaluation of the hip exist. The various techniques have evolved primarily due to historical use in different regions of the world. The main techniques include: the Graf technique (mainly used in Europe), the Harcke technique (mainly used in the USA), and Morin-Terjesen technique (mainly used in Scandinavian countries). A 2019 publication from an international interdisciplinary consensus meeting on DDH, composed of representatives from various European countries, including Ireland, came to a strong agreement (that is, over 90% supported the recommendation) that

the preferred ultrasound examination technique for early detection and treatment of DDH is the Graf technique.[12] The publication also noted that comparisons of the three techniques have found good correlation in results of hip assessment between the techniques. As the Graf technique is the ultrasound method used in Ireland and most of Europe, the other techniques will not be further discussed.

The Graf technique examines the morphology of all the joint components and measures the angles of bony and cartilaginous components of the acetabulum.[2, 24, 25] The severity of DDH observed is rated using the Graf classification.[18] The classification ranges from type Ia (normal hips) to type IV (most severe form) and is dependent on several factors:

- Graf-α angle. This is the angle between a horizontal line through the lateral side of the iliac bone, a line through the bony acetabular rim, and the triradiate cartilage (that is, where three parts of the acetabulum join)
- Graf-β angle. This is the angle between the bony acetabular rim and the cartilaginous acetabular labrum
- the appearance and coverage of the acetabulum and the femoral head
- age.

A systematic review and meta-analysis was identified which explored the performance of ultrasound using the Graf method for detecting DDH in infants less than 12 weeks of age.[26] Diagnosis must have been confirmed by clinical, ultrasound, or radiological follow-up examinations. Overall, the review found that the pooled sensitivity from the six included articles was 93% (95% CI: 57% to 99%), and the pooled specificity was 97% (95% CI: 86% to 99%). Therefore, some potential cases of DDH may go undetected, even with ultrasound. The review noted that the test performance may be impacted by the training level of the operator.

Ultrasound screening may allow for the detection of cases of DDH that may have been missed with clinical examination alone, potentially reducing the rates of late presenting DDH and associated complications. However, ultrasound screening can result in unnecessary treatment mostly due to the potential for spontaneous correction of the identified DDH.[5-10] This risk of unnecessary treatment may be reduced by training in Graf technique and classification.[11, 12]

Radiography

Radiography can also be used in the diagnosis of DDH.[2, 27] Due to the risks associated with radiological exposures, radiography is used primarily in diagnostic investigations to confirm a clinical or ultrasound suspicion of DDH, and not as a first-line method of screening. Radiological examinations are only useful from six months

of age and onwards. At birth, the proximal region of the femur is completely made of cartilage.[27] This structure starts to ossify (that is, to form bone) after birth; by three to four months of age the skeletal structures of the infant have sufficient mineralisation to be visualised on X-ray.[2, 26, 27] However, portions of the hip joint remain as cartilage until around six months of age, and therefore are not able to be visualised on X-ray during this time.

Ultrasound screening programmes

Ultrasound screening programmes for DDH can either be selective or universal. With selective screening only infants with one or more risk factors (for example, family history or breech presentation) or clinical signs undergo ultrasound screening. In a universal screening programme, all infants undergo screening, irrespective of their risk factors.[7]

Cases of late diagnosed DDH can still occur following ultrasound screening.[13] The optimal method of an ultrasound screening programme, whether it be selective or universal, remains unclear.[5-8, 13, 14, 28] It is not clear whether a universal screening programme, in which all infants are screened for DDH, leads to a reduction in late presentation and better outcomes. The clinical effectiveness of universal ultrasound screening compared to selective ultrasound screening is described in Chapter 4.

Examinations for DDH routinely performed in Ireland

In Ireland, newborns receive a routine full clinical examination within 72 hours of birth performed by either a physician or a specialist midwife.[29, 30] This examination includes clinical history and assessment of the hips to check for signs of DDH. The physical assessment for DDH includes the Ortolani and Barlow manoeuvres as well as assessment of leg length, gluteal fold symmetry and symmetry of hip abduction. The algorithm for referral for further DDH assessment (based primarily on ultrasound testing) if there is clinical suspicion or risk factors for DDH is outlined in section 2.3.1. If DDH is not suspected, the infant receives another assessment at approximately six weeks of age, generally with a general practitioner (GP).[29, 30] The infant will then receive developmental checks by a public health nurse at three months, and at seven to nine months, including assessment of gross and fine motor movements.

2.2.2 Treatment

Treatment options for DDH depend on the severity of the condition and the age of the patient.[1, 2] The aim of treatment is to achieve and maintain concentric reduction of the femoral head in the acetabulum (that is, the femoral head is aligned properly in the hip socket), therefore allowing for normal continued hip development in infancy and childhood and preventing subsequent impairment.[1]

No internationally accepted guidelines for the treatment of DDH were identified in the course of this evidence review; however, treatment recommendations are generally similar across the recently published literature.[1, 3, 27] Variation in treatment is based on individual patient characteristics; however, general algorithms exist (see Chapter 3: epidemiology).[3] In some cases, treating physicians will choose to monitor infants with hip classification up to Graf IIa ultrasound, and treat those with more severe classifications.[19]

If the decision is made to treat, in infants up to six months of age with hip instability, dysplasia, or dislocation, the condition is generally treated with abduction splinting.[1, 3, 27] The Pavlik harness is the most commonly used brace for the splinting.[1, 3, 27, 31] In patients aged six to 18 months, the condition is often treated with closed reduction (that is, the femoral head is placed in the hip socket without surgery) along with a hip spica cast (that is, a cast that keeps the hips in the required position while healing).[1, 3, 27] For older children (generally older than two years), open reduction (that is, the femoral head is placed in the hip socket with surgery) is usually required, with or without osteotomy (that is, a surgical procedure to reshape and realign a bone). Femoral shortening osteotomy may be required if there is the need to reduce tension on the hip reduction, and pelvic osteotomy may be required if there is residual shallow dysplastic acetabulum (that is, a shallow hip socket). Finally, for children eight years and older it is unclear whether to perform a reduction surgery, or whether to observe for the need for eventual arthroplasty (that is, joint replacement, remodelling, or realignment) when required later in life.

In 2020, a multidisciplinary group of experts representing various Italian clinical societies, which included paediatricians, radiologists and paediatric orthopaedic surgeons, published a consensus document of recommendations for early diagnosis and treatment of DDH.[2] The recommendations were prepared through a review of the available scientific literature and expert opinions and are set to be reviewed every three years. The recommendations noted that early diagnosis of DDH may improve clinical outcomes and contribute to a decrease in hip osteoarthritis in young adults. Early treatment was also strongly supported in the 2019 publication from an international interdisciplinary consensus meeting on DDH, composed of representatives from various European countries. The publication from this group noted strong agreement that the principle of treatment was the application of a device that holds the hips in the appropriate flexion and abduction, but that the type of device is less important than early and accurate diagnosis and treatment initiation.[12]

Treatment effectiveness may be maximised when initiated within the first few days to months of life.[2] Earlier detection may also allow for nonsurgical treatment, such as harnessing or closed reductions. Notably, if DDH is present at birth, the anatomical changes associated with the dislocation may not be solidified (that is,

becoming more permanent), which would occur at age two to three months. This highlights the potential importance of early treatment to correct the DDH before the changes are permanent. Permanent changes of the anatomical structure should be avoided to reduce the incidence of hip osteoarthritis in adulthood.[2, 32] Additionally, while clinical symptoms of DDH may disappear over time, some evidence suggests that the anatomical changes associated with the condition can remain, potentially leading to the requirement for surgery later in life.[2, 32, 33]

Conversely, while evidence shows that earlier treatment generally leads to better outcomes for the patient, there is a risk of complications.[2, 4, 34] While harnessing and closed reduction techniques may be considered minimally invasive, there can be complications associated with these procedures, most commonly avascular necrosis (AVN) and femoral nerve palsy. AVN of the femoral head is caused by an interruption in blood supply to the femoral head, resulting in the death of bone tissue. This may occur as a result of excessive pressure applied to the femoral head during treatment, resulting in decreased blood supply. A 2016 systematic review found that, with a mean follow-up length of 7.7 years (range 5.0 to 18.8 years), the rate of AVN for children who underwent closed reduction for DDH at younger than two years of age (n = 538 hips) was 10%.[34] It is important to note, however, that the method of detection of the DDH (for example, Graf method) was not reported in this review, as this may have impacted findings. AVN is thought to be more common in closed reduction which involves a hip spica cast, as the positioning of the femoral head is more static, compared to harnessing procedures such as the Pavlik harness, where the positioning is more dynamic, and therefore the risk of continuous interruption to blood supply is reduced.[35]

Furthermore, a 2007 review of DDH found that early treatment does not completely avoid the need for subsequent surgery.[19] In one study within the 2007 review, it was noted that up to 5% of infants who were treated with abduction splinting required surgery at some point later in life, indicating that the abduction splinting was unsuccessful in fully treating the DDH.[10, 19] Additionally, a 1998 study from the UK Medical Research Council found that one fifth of children who needed surgery for DDH by the age of five years had been treated previously with abduction splinting, again indicating unsuccessful treatment.[36] The 2007 review noted that it was unclear whether age at abduction splinting is predictive of later need for surgery. This lack of clarity was due to confounding by severity and mode of detection, and the variation in adherence to the splinting by parents.

2.3 Screening for DDH in Ireland

2.3.1 Current ultrasound screening recommendations in Ireland

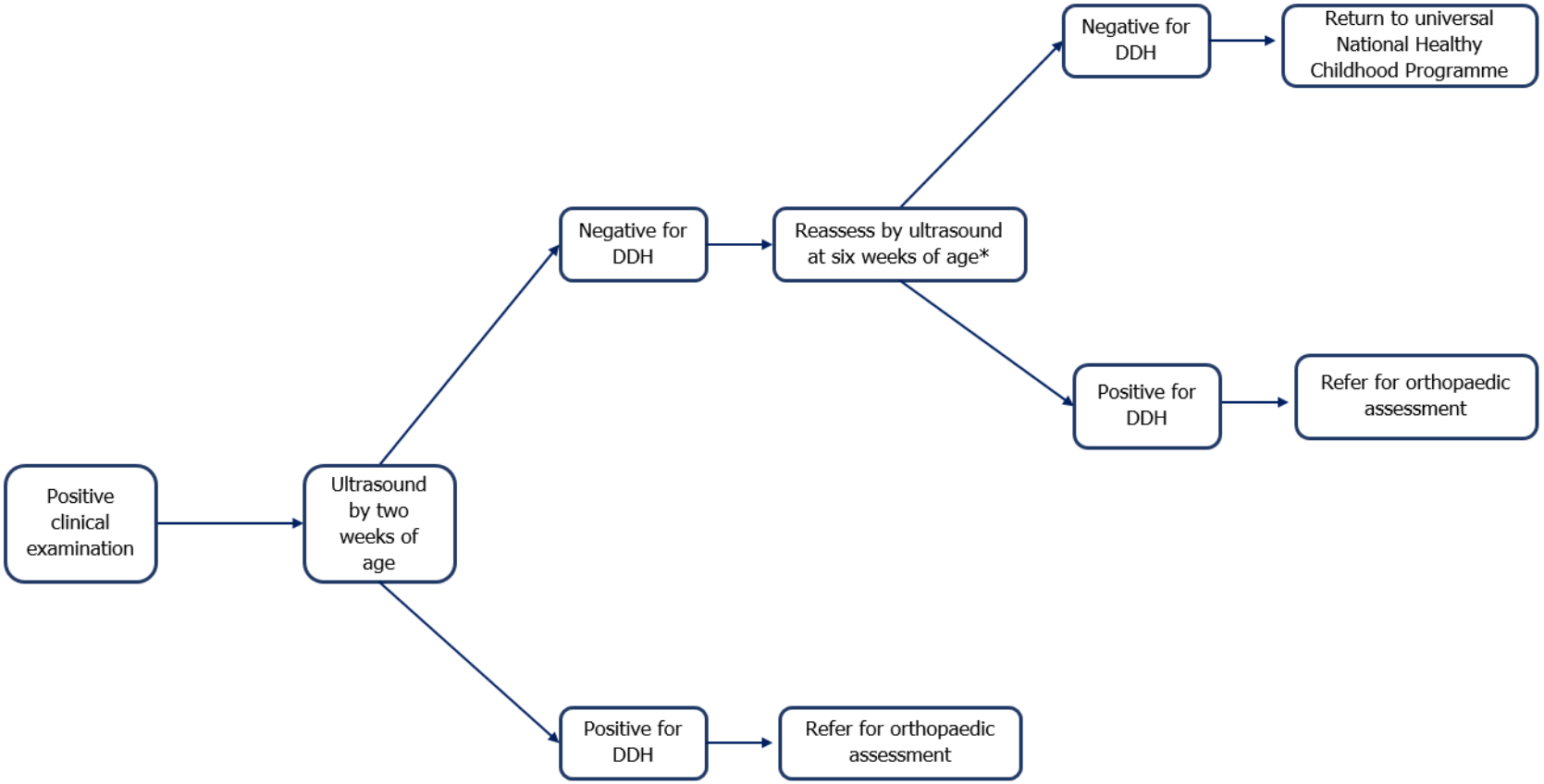
In Ireland in 2017, the National Clinical Programme for Paediatrics and Neonatology and the DDH Subgroup of the National Child Health Review Steering Group published an implementation pack which recommended a selective ultrasound screening programme for infants at risk of DDH.[15] The pack, further outlined in the following paragraphs of this report, described the standards required for a selective ultrasound screening programme for infants in Ireland, outlined which infants would receive ultrasound under this programme, when and how this should occur, and provided guidance for what steps to take following the ultrasound examination. The pack also described what data should be collected and the reporting format. Of note, as of 2023, the recommendations have not been implemented as a screening programme in Ireland.[16] Rather, the recommendations are included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme. This programme sits within the governance of the Health Service Executive (HSE) Clinical Design and Innovation Division. As the recommendations have not been implemented under a formal national screening programme, the pathway does not have the governance, end-to-end care (that is, the complete care pathway from diagnosis of the patient through to long-term follow-up), and quality assurance that would be included in such a programme.

It was noted that the Graf method of ultrasound should be used.[15] The screening algorithm is outlined below, and is dependent on findings from the standard development check performed within 72 hours of birth[30] and or certain risk factors for DDH; these included having a first degree family history of DDH (that is, mother, father, or sibling) or having a breech presentation.

Recommendations for infants with a positive clinical examination at the neonatal check

The pack recommends that in infants who have a positive clinical examination for suspected DDH at the standard developmental check performed within 72 hours of birth (Figure 2.1), they should receive an ultrasound by two weeks of age and a consultation with an expert (paediatrician or orthopaedic consultant).[15] Ideally, the ultrasound should take place within three days. If DDH is confirmed on the ultrasound (defined as Graf IIb to Graf IV ultrasound), immediate treatment with a Pavlik harness is advised. Further management then falls under the care of orthopaedic services. If the initial ultrasound is negative (defined as Graf I ultrasound) for DDH, the infant should receive another ultrasound by six weeks of age (see Figure 2.2). If this follow-up ultrasound is again negative for DDH, the infant is returned to the universal National Healthy Childhood Programme.

Figure 2.1 Recommended screening algorithm in infants with a positive clinical examination



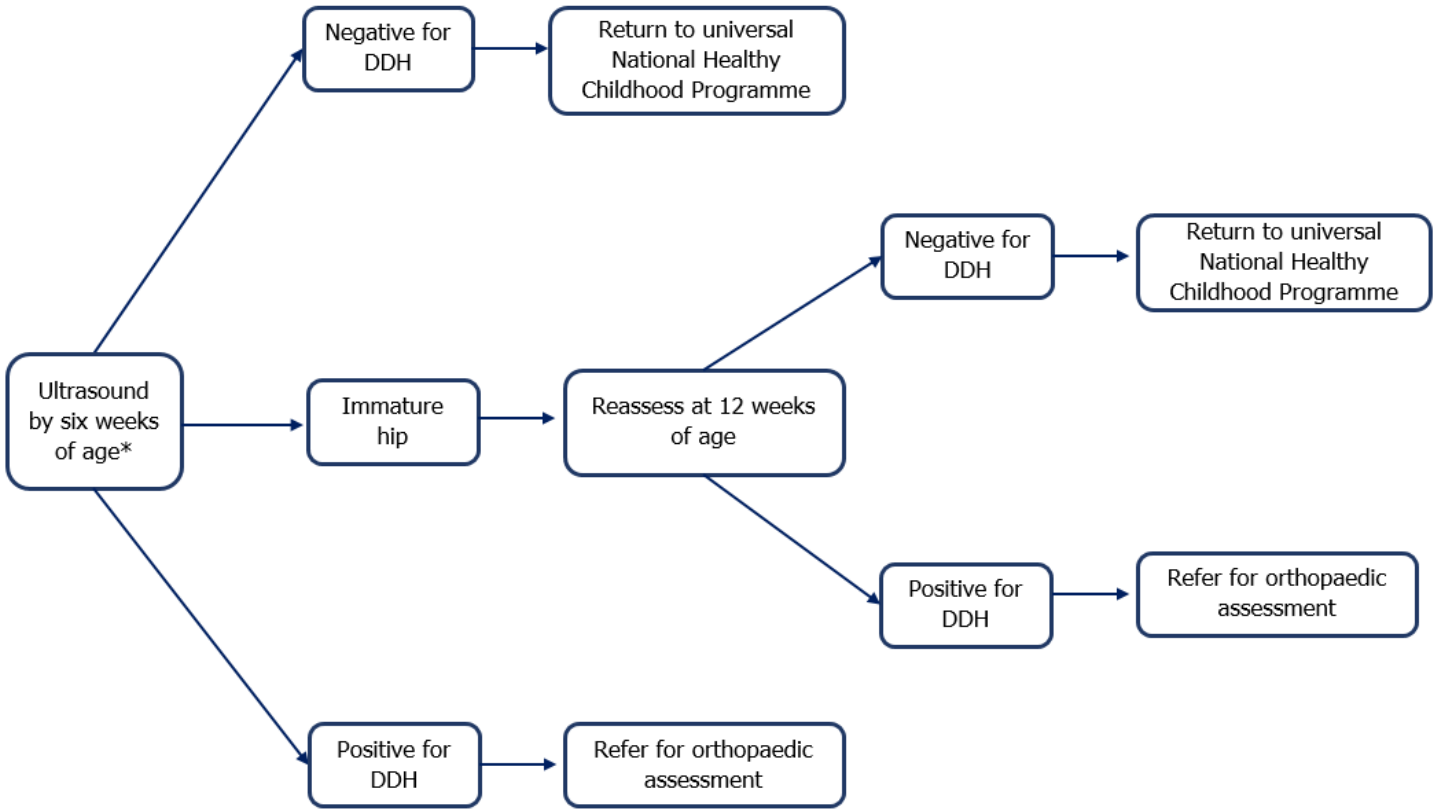
Key: DDH – developmental dysplasia of the hip.
* See Figure 2.2 for algorithm for the six week ultrasound.

Recommendations for infants with a negative clinical examination at the neonatal check and one or more risk factors

Infants with a negative clinical examination for suspicion of DDH, but either a first degree family history of DDH (that is, mother, father, or sibling) or those who had a breech presentation, are recommended to have an ultrasound examination by six weeks of age (adjusted for prematurity) (Figure 2.2).[15] Breech presentation includes babies who are breech presentation at or after 36 weeks gestation (adjusted for prematurity), regardless of presentation at birth or final mode of delivery. In the case of multiple births, all babies should be screened even if only one baby was breech. Infants requiring an ultrasound should be referred to the radiology department in the hospital of birth, unless there are local protocols that define the pathway service for ultrasound.

If the six week of age ultrasound is negative (defined as Graf I ultrasound), the infant is returned to the universal National Healthy Childhood Programme. If the ultrasound is positive for DDH (defined as Graf IIb to Graf IV ultrasound), it is recommended that the infant receive an urgent specialist opinion and be referred for orthopaedic assessment in line with local protocols. If the ultrasound reveals a Graf IIa hip, the hip is classified as 'immature' (that is, hips that have not fully developed and may continue to develop normally). These infants receive another ultrasound at 12 weeks of age, and follow the algorithm for positive or negative for DDH as outlined for the ultrasound by six weeks of age. If the ultrasound still reveals a Graf IIa hip, the hip is classified as positive for DDH, and the child follows the referral pathways for Graf IIb to Graf IV hips.

Figure 2.2 Recommended screening algorithm for the screening ultrasound by six weeks of age*



Key: DDH – developmental dysplasia of the hip.
* Screening by six weeks of age either following a negative clinical examination at the neonatal check and one or more risk factors or if, following a positive clinical examination at the neonatal examination, the ultrasound by two weeks of age is negative for DDH.

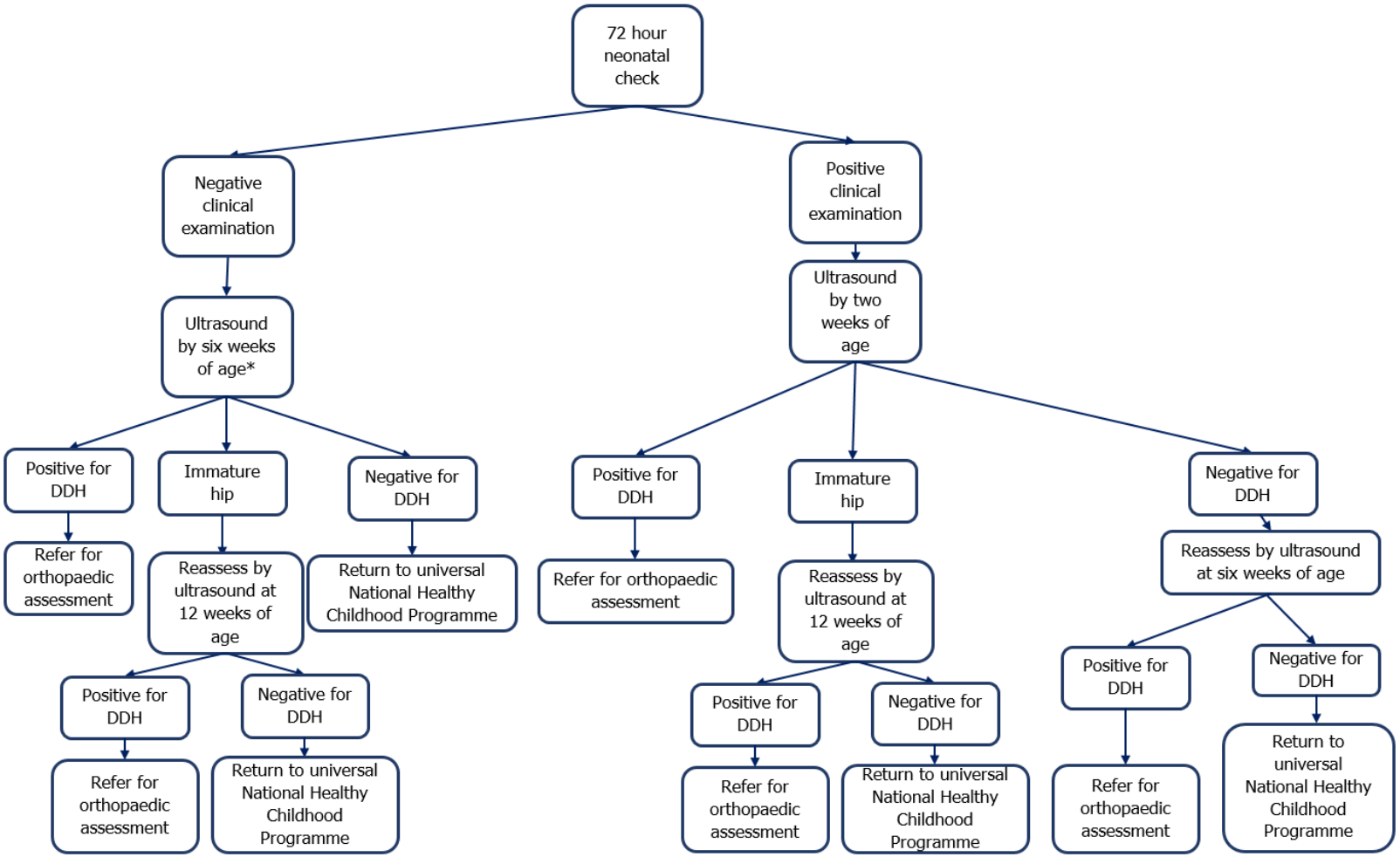
Recommendations for infants at the six-week clinical visit

Infants who have a positive clinical examination for suspected DDH at the standard six-week clinical visit should receive urgent referrals as per local protocols.[30] Additionally, if an infant is identified as having a risk factor for DDH (that is, first degree family history of DDH or breech presentation) that was not identified at the 72 hour clinical examination, the infant should be referred for an urgent ultrasound, irrespective of if their clinical examination is negative for suspected DDH. In the absence of local protocols, the infant should be referred to their hospital of birth and should be seen within two weeks.

2.3.2 Possible universal ultrasound screening programme in Ireland

A universal ultrasound screening programme in Ireland would allow for all infants, regardless of identified risk factors, to receive an ultrasound of the hip. A possible screening algorithm for a universal ultrasound screening programme, based on the pathway outlined in the current recommendations for selective ultrasound screening, is outlined in Figure 2.3. Briefly, if an infant were identified by clinical examination within 72 hours of birth as a suspected case of DDH, the infant would receive an ultrasound of the hip by two weeks of age. If the 72 hour clinical examination is negative for suspected DDH, the infant would follow the screening algorithm under the current recommendations for an infant receiving an ultrasound by six weeks due to an identified risk factor. This six week ultrasound would occur irrespective of risk factors (that is, all infants would receive ultrasound).

Figure 2.3 Possible screening algorithm for a universal ultrasound screening programme in Ireland†



Key: DDH – developmental dysplasia of the hip.

† Based on the pathway outlined in the current recommendations for selective ultrasound screening.

* Screening by six weeks of age either following a negative clinical examination at the neonatal check or, if, following a positive clinical examination at the neonatal examination, the ultrasound by two weeks of age is negative for DDH.

2.3.3 Outcomes reported with screening practices for DDH in Ireland

Three studies were identified reporting on screening practice for DDH in Ireland.[37-39] Two of these were undertaken prior to the publication of the selective screening ultrasound implementation pack in 2017.[38, 39] The reporting was limited to the same single centre in two of the studies and may not be representative of processes in use in other hospitals.[37, 38]

In 2006, a survey of consultant paediatricians and neonatologists linked to 19 maternity units in Ireland was conducted to ascertain the approaches to screening for DDH in use in Ireland at the time.[39] Of note, this study was performed prior to the publication of the selective screening ultrasound implementation pack in 2017. The study found that eight (42%) of the hospitals had a formal DDH screening protocol or algorithm. Responses from clinicians working in the same hospital were contradictory in three (16%) hospitals, suggesting variation in practice within these hospitals depending on the treating clinician. A total of 16 (84%) hospitals used radiography as the primary method of screening, two (11%) used ultrasound, and one (6%) used both methods equally. Radiography was performed at four months, five months, and six months in three (18%), eight (47%), and six (36%) of the 17 hospitals completing this survey question, respectively. In the three hospitals using ultrasound, the ultrasounds were performed between six and eight weeks of age. A consultant paediatrician examined the hips of all newborns in six (32%) centres. Of all births in Ireland at the time, 29.1% of infants overall had hip examination performed by an experienced examiner. Overall, the study highlighted the variation in screening protocols across the country, with a majority reliant on radiography at the time of the study.

A retrospective review of patients was identified which presented data from a single hospital network in Ireland in 2015.[38] This study was performed prior to the publication of the selective ultrasound screening implementation pack in 2017. The study aimed to evaluate the outcomes from radiography at six months of age in infants with normal physical examination and ultrasound at six weeks. The report stated that selective ultrasound screening was in place in the hospital, but did not define the selection criteria. While not explicitly stated, routine practice at this centre appeared to involve a re-check with radiography at six months for infants who were referred for selective ultrasound and had normal ultrasound findings. Consistent with the previously mentioned study, there was a lack of clarity regarding the selective screening pathway. In particular, the selection criteria, and follow-up processes were unclear. Of 829 included patients, 63 (8%) patients had abnormal hip radiographs at six months. The authors concluded that radiographic follow-up should be recommended for infants who present for the selective ultrasound screening, even when the ultrasound is normal.

An audit of all referrals made to the University Hospital Waterford (UHW) in 2020 aimed to examine referrals for ultrasound diagnostics for DDH at the hospital.[37] This hospital provides the selective ultrasound screening service for all infants born in Waterford, Kilkenny, Wexford and South Tipperary hospitals. The hospital adapted the recommendations from the 2017 implementation pack for selective ultrasound screening of referrals. According to the implementation pack, infants with normal ultrasounds can be discharged. However, the Southeast diagnostic protocol differs from the implementation pack recommendations in that all at-risk babies who have a normal ultrasound are followed up with radiograph at six months. This deviation was informed by the conclusions of the previously described publication which suggested that some cases of DDH may not be identified with early screening.[38]

A total of 1,031 infants were referred for ultrasound diagnostics. While the total number of infants examined clinically was not reported in the study, a total of 7,213 births were reported in the catchment area served by the selective ultrasound diagnostics service for DDH at UHW in 2018. This suggests that approximately 14% of children born in UHW were referred for selective ultrasound diagnostics. Of the 1,031 infants referred, 992 (96%) were scanned, of whom 641 (65%) were scanned within the recommended six weeks of age. The authors noted delays due to the COVID-19 pandemic. In total, 255 (26%) of the patients scanned were referred onwards to orthopaedics, 161 (63%) of whom were referred onwards due to abnormalities noted in the ultrasound, and 94 (37%) of whom were referred due to continued clinical or radiographic suspicion of DDH, even though the ultrasound was normal. A total of 198 infants received treatment for DDH, 122 (62%) of whom were initially treated with a harness. All of these 122 patients had received their initial ultrasound within the recommended six weeks of age, and at the time of publication, harnessing was successful in treating the DDH, with no operative management required. Of 864 infants with normal ultrasound, 71 (8%) were subsequently referred to the orthopaedic clinic for radiographic examination. Overall, the sensitivity of ultrasound for the diagnosis of DDH was calculated at 64.1%, based on the number of patients identified through ultrasound and the number with normal ultrasound findings who were subsequently treated in the orthopaedic clinic. The sensitivity reported in this study is lower than that reported from the systematic review[26] in section 2.2.1 (at 93%). Assuming all treated infants truly had DDH, based on imaging and examination findings, specificity and PPV were assumed to be 100%. The NPV was calculated to be 91.8%. Of note, of all treated infants, there were no cases of AVN reported.

2.4 International practice in screening for DDH in infants

To provide an overview of current international practice regarding ultrasound screening for DDH, a scoping search was performed up to September 2023, which examined countries deemed to be of most relevance to Ireland. These were chosen

based on a combination of geographical proximity to Ireland, population size, European Union (EU) membership and or availability of documents in English. A targeted grey literature search (for example, searching national public health organisations, and the websites of governmental departments and relevant agencies), supplemented by a search of primary literature, was performed. The below countries were included in this review:

EU/EEA

- Austria
- Belgium
- Denmark
- Finland
- France
- Germany
- Italy
- Netherlands
- Norway
- Portugal
- Spain
- Sweden
- Switzerland

Non-EU

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

A summary of the findings of this review is presented in Table 2.1, followed by detailed findings for each specific country employing universal ultrasound screening. Of the 18 countries that were examined, universal ultrasound screening for DDH was nationally implemented in two countries, while 14 have selective ultrasound screening based on clinical signs and or risk factors. For two countries, no programme or guidelines of an ultrasound screening programme were identified. Of note, screening for DDH is not clearly outlined as part of an official national screening programme in some countries and is considered an element of an infant's general health examination or 'check-up'. In the countries with selective ultrasound screening, there was variation in aspects of the programme or guidelines, such as timing and frequency of clinical examination, and criteria for referral to ultrasound. Selection to receive ultrasound screening was most commonly due to the presence of clinical signs during routine examinations and risk factors. Common risk factors included family history and breech position.

Specific criteria and guidelines for the selective ultrasound screening programme were limited, with few countries providing specific details of the care pathway. While no programme or guidelines of an ultrasound screening programme were identified for two of the countries (Finland and Portugal) in this review, it is likely that some manner of screening for DDH is in place in these countries. Additionally, there may

be local implementation or recommendations from professional societies in some countries that were not captured at a national level review.

Table 2.1 Overview of international practice in DDH screening

Country/Provence	Screening
Austria	Universal ultrasound[40]
Australia	Selective ultrasound[41]
Belgium	Selective ultrasound[42]
Canada	Selective ultrasound[43]
Denmark	Selective screening[44]
Finland	*No programme or guidelines identified*
France	Selective ultrasound[45]
Germany	Universal ultrasound[46]
Israel	Selective screening[47]
Italy	Selective ultrasound[48]
The Netherlands	Selective ultrasound[49]
New Zealand	Selective ultrasound[50]
Norway	Selective ultrasound[51]
Portugal	*No programme or guidelines identified*
Spain	Selective ultrasound[52]
Sweden	Selective ultrasound[53]
Switzerland	Selective ultrasound[22]
United Kingdom	Selective ultrasound[54]

The following three countries were identified as having or have had a universal ultrasound screening programme for DDH:

Austria

In 1992, a nationwide general ultrasound screening program using Graf technique was introduced to detect DDH in Austria.[40]

Germany

In Germany, a screening programme for developmental hip dysplasia that included universal ultrasound imaging for all children started in January 1996.[46]

Switzerland

Of note, universal ultrasound screening was active in Switzerland until 2004. The programme was then discontinued as the evidence was considered insufficient to support its continuation.[22]

2.5 Discussion

This chapter aimed to describe the key elements of the technology under consideration (that is, universal ultrasound screening for DDH in infants). It is important to note that in Ireland, there are published recommendations for a selective ultrasound screening programme of infants, with referrals for ultrasound based on clinical findings at the newborn examination and or specific risk factors. As of November 2023, the recommendations are not implemented as part of a formal screening programme. Rather, there is a targeted clinical diagnostics programme in place for DDH in Ireland. This programme sits within the governance of the Health Service Executive Clinical Design and Innovation Division. As the recommendations have not been implemented under a formal national screening programme, the pathway is not subject to the governance, end-to-end care, and quality assurance that would be included in such a programme; this is discussed further in Chapter 5 (Discussion – 'The Screening Programme').[55, 56]

Treatment options for DDH differ depending on the age of the patient, with younger patients generally able to receive less invasive intervention. Ideally, the patient would receive the least invasive treatment option available, supporting earlier identification and treatment. It is important to note that, regardless of treatment selection, there is the possibility of complications arising from the treatment, most seriously AVN of the femoral head. However, the risk of complications is thought to be lower in infants who receive abduction bracing rather than a solid cast, as there is still some movement in the hips. This would again support earlier detection and treatment of DDH.

As there may be undetected cases with clinical examination for DDH alone, due to the low sensitivity and specificity of the physical examination, ultrasound screening, either selective or universal, for DDH can be used to improve early detection and treatment. As noted from the overview of current international practice, ultrasound is widely implemented as a selective or universal screening tool for DDH. However, no international guidelines were found outlining a standardised protocol for screening, including risk factors for selective screening for DDH or factors such as the timing of screening or protocols for follow-up. The absence of an internationally agreed best practice approach is a notable challenge. There is also variation in recommendations for the timing of the ultrasound screening between different organisations. The optimal timing of ultrasound examination is important to reduce the potential for unnecessary treatment; this is further discussed in Chapter 3.

Two publications of international experts support the early identification and treatment of DDH.[2, 12] Of note, these publications were based primarily on expert opinion, though one (representing Italian clinical societies) was based on a review of the literature, followed by analysis of the results within a consensus conference with formulation of recommendations based on the experts' advice.[2] The 2019 publication from an international interdisciplinary consensus meeting on DDH, composed of representatives from various European countries, concluded that there was a strong agreement among the experts that clinical examination alone is insufficient for assessment for DDH, and that ultrasound is essential.[12] They further agreed that ultrasound should be carried out as soon as possible, and no later than six weeks of age. There was also strong agreement in favour of a universal ultrasound screening programme using the Graf technique, which would not result in overtreatment and would result in a reduction of later complications of DDH. Furthermore, this publication noted that there was strong agreement that the principle of treatment was the application of a device that holds the hips in the appropriate flexion and abduction, but that the type of device is less important than early and accurate diagnosis and treatment initiation. Early diagnosis and treatment of DDH was also supported in the 2020 publication recommendations for early diagnosis and treatment of DDH emerging from a consensus meeting.[2] This publication concluded that all newborns must undergo clinical examination by a neonatologist or paediatrician at birth, and that if there are any signs of DDH, an ultrasound is required before discharge or within the first six weeks of life. Additionally, they noted that a DDH screening programme is needed, with all newborns requiring ultrasound by four to six weeks of age.

In the absence of international guidelines, an Irish-specific pathway for selective ultrasound screening has been developed. These recommendations were however informed by the selective ultrasound screening practice in the UK.[57] In 2017, these recommendations for a selective ultrasound screening programme were published, with the aim of providing guidance for DDH screening across the country. A national audit of centres performing ultrasound to assess for DDH in 2021 and 2022 found that 18.5% of all infants born were referred for ultrasound of the hips in these years.[58] However, as noted above, there is currently no formal screening programme in place and the recommendations are instead included as part of the diagnostic pathway for DDH, referred to as a targeted clinical diagnostics programme. Central to the concept of a screening programme is the need for appropriate governance and performance metrics against which outcomes are measured in relation to the management of individuals who present for screening and follow-up of those with a positive screening test result. As noted by the WHO, a screening programme is not just a single test; reporting of outcomes and ongoing evaluation of a screening programme are important as part of quality assurance processes at a programme level in order to achieve the greatest benefit with the

least harm.[59] Such a structured approach is not currently in place for the detection of DDH in infants in Ireland. Therefore, it is challenging to comprehensively understand current practice across the country, with the ultrasound programme managed locally at each unit.

It is unclear to what degree the recommendations are implemented and followed as specified throughout Ireland, and there seems to be variation in the implementation of the recommendations across hospitals. Highlighting the potential for inconsistency in practice between different hospitals, a study of referrals made to the UHW in 2022 outlined that radiograph follow-up at six months of age was performed for all infants originally referred for selective ultrasound but who had normal hips as determined by ultrasonography. This was inconsistent with the 2017 recommendations, in which children with normal ultrasounds were recommended to be returned to normal clinical care. The authors highlighted that this additional examination by radiography was performed following the results of early publications which showed that some infants with DDH are potentially missed with the current selective ultrasound screening programme, and may present later in life. However, it is important to highlight that the study that informed the recommendation at UHW was purely descriptive and did not include a comparator arm of infants in the six month group who did not have X-ray. Therefore, it is not possible to determine the outcomes of these infants were they not to have received the additional X-ray.

It is unclear whether practice in other countries includes follow-up radiography in those with normal hips at ultrasound. While this practice may identify late presenting cases of DDH, it is important to consider the risks associated with radiography in the pelvic region, especially at the young age of the infants. Additionally, even at the newborn clinical examination, there is likely discrepancy in terms of level of expertise in those performing the examinations, and therefore in which infants are referred onwards for ultrasound screening.

Access to ultrasound testing for the diagnosis of DDH was noted to be implemented across a number of the countries examined in this review on a selective or universal basis. It is unclear if selective and ultrasound screening in these contexts have been implemented as part of a formal screening programme, including appropriate governance structures. While many countries reported that there was a form of selective ultrasound screening for DDH, few reported on the criteria for referral for ultrasound, and of those that did, there was a lack of uniformity in referral algorithms, with some noting referral was based on clinical suspicion alone, and others nothing clinical examination findings and or various risk factors. Information regarding the timing of screening was not readily available. It was unclear whether this was due to variation in clinical practice at a local level within countries included in the review, or the absence of clear guidelines. Additionally, while there were no programme or guidelines identified for a selective ultrasound screening programme

for two countries, it is likely that newborns in these countries do receive some form of clinical examination for hip abnormalities. It is also important to note that only two countries were identified which have a universal ultrasound screening programme for DDH. The programmes in these two countries have been in place since the 1990s.[40, 46] An additional country (Switzerland) opted to not continue with universal ultrasound screening after a temporary programme was implemented with a view to evaluating the outcomes of the practice.[22] There was insufficient evidence to support its continuation. This review focused on a limited number of countries considered most applicable to the Irish context, and did not include an exhaustive search of all countries.

Ultrasound screening for DDH in infants can be used as part of a selective or a universal screening programme. At present in Ireland, there are recommendations for a selective ultrasound screening programme in place, however these have not been implemented as a formal screening programme. Uptake of the recommendations published in 2017 is not well documented; however, there is some evidence to suggest inconsistencies in practice. As noted, the outcomes of a universal programme compared to a selective programme are discussed in Chapter 4.

3 Epidemiology and burden of disease

Key points

- Developmental dysplasia of the hip (DDH) occurs when there is an abnormality in the development of the hip. The severity can range from mild dysplasia to complete hip dislocation.
- If DDH is not identified and treated in the early months of life, most of the significant cases of dysplasia will present symptomatically after walking age. Many cases of mild hip instability identified in newborns resolve without treatment within the first six to eight weeks of life. However, the natural history of DDH is poorly understood, and the true proportion of cases that will resolve spontaneously remains unclear.
- There are several major risk factors for DDH, including breech position in utero, female sex, and family history of DDH. Other potential risk factors include being a firstborn child, foot abnormalities, tilted neck position, low amniotic fluid during pregnancy, high birth weight, being born overdue and postnatal swaddling of the infant. However, the majority of cases of DDH have no identifiable risk factor.
- Persistent dysplasia and dislocation can lead to wear of the cartilage of the hip and osteoarthritis in young adults. DDH is a common cause of total hip replacements in young adults.
- The incidence of DDH varies geographically due to factors such as genetic and cultural differences (such as post-natal swaddling practices). Reasons for variation in the reported incidence include differences in the age at examination, experience of the examiner, screening methods (for example, clinical examination only, selective ultrasound screening or universal ultrasound screening), screening protocols and definitions of DDH applied.
    - An analysis of infants born consecutively in Austria, where a universal ultrasound screening programme for DDH has been implemented since 1992, reported 9.8% of infants had a hip higher than Graf type I (normal hip). Most of these cases were classified as Graf type IIa (immature hips).
- There is no national centralised database of cases of DDH in Ireland. Therefore, it is challenging to estimate the incidence of the condition.
    - There is considerable uncertainty regarding the current incidence of DDH in Ireland; available incidence estimates are from a single centre.

The estimates of the incidence of DDH at this centre ranged between 7 per 1,000 (based on children born in 2009, prior to the publication of the 2017 national recommendations for selective ultrasound screening) and 31 per 1,000 (based on children diagnosed in 2018 and 2019, after the publication of the recommendations). These estimates may not be representative of practice and case numbers in the rest of Ireland.

- Under the current targeted clinical diagnostics programme, a national audit of centres performing ultrasound to assess for DDH in 2021 and 2022 found that 18.5% of all infants born were referred for ultrasound of the hips.

3.1 Introduction

The purpose of this chapter is to describe the epidemiology and burden of disease associated with developmental dysplasia of the hip (DDH). The chapter outlines the aetiology of the condition, followed by the natural history and burden of disease, and the incidence. It is important to note, as outlined in Chapter 2, that there is no standardised definition of DDH. The definition varies, but can include a wide spectrum of hip abnormalities from mild dysplasia to complete hip dislocation.

3.2 Aetiology

The causes and aetiology of DDH are multifactorial, but are often said to be poorly understood.[1, 18] 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.[18, 27] Several risk factors for DDH have been identified, however it is important to highlight that the majority of cases of DDH have no identifiable risk factor.[60, 61] Major risk factors include breech position in utero, female sex, and family history of DDH.[1, 2] Other potential risk factors include being a firstborn child, foot abnormalities (such as, talipes or asymmetrical calcaneovalgus foot deformities), torticollis (that is, tilted neck position), oligohydramnios (that is, having low amniotic fluid during pregnancy), high birth weight, being born overdue, and postnatal swaddling of the infant. However, limited evidence exists to the degree of association or ability to predict DDH with these factors.[1, 18] Furthermore, there is some level of familial risk for DDH.[19] Some studies have suggested that acetabular dysplasia and joint laxity are heritable, both of which are associated with DDH. In one study using the Norwegian Twin Registry, the odds of DDH in relatives of twins with DDH were compared with the odds of DDH in relatives of twins without DDH. This comparison was performed for each of fathers, mothers, siblings and offspring,

where the odds ratio for mothers was 35.8, more than four times that in fathers, suggesting a maternal effect.[62]

3.3 Natural history and burden of disease

The natural history of DDH is often said to be unclear, mostly due to inconsistencies in terminology and the lack of long-term studies which follow infant hip instability of all severities to maturity.[10, 20, 21, 27]

Mild dysplasia may never result in clinical symptoms, or, conversely, it may not become apparent until adult life.[19] The longer term natural history of hip dysplasia, in the absence of a true dislocation or subluxation, is largely unknown because it often goes undiagnosed. Several observational studies have shown that mild instability in the hip resolves on its own without intervention, with these studies reporting that up to 90% resolved spontaneously within the first six to eight weeks of life.[20, 21, 27, 63]

Conversely, some studies suggest that there are cases of DDH without physical symptoms in infancy that become symptomatic later in life.[10] In those with undetected persistent dysplasia (that is, the dysplasia does not resolve on its own) that is not identified and treated in the early months of life, the condition will most likely present clinically in infancy or early childhood, often after walking age.[19] The persistent dysplasia alters the hip and can increase load on the cartilage that covers the femur and acetabulum, leading to wear of the cartilage, and osteoarthritis. In those with subluxation or dislocation, osteoarthritis usually occurs.

As noted, long-term complications of untreated DDH include hip, knee, and lower back pain, gait disturbance, and degeneration of the hip joint.[19] However, the risk and incidence of these complications has not been well reported. After an average of 50-years follow-up in several studies, it was reported that 11% to 41% of those with untreated dislocations remained pain free.[27, 63] However, it was also estimated from studies in Denmark and Norway that DDH represents the cause of 2.6% to 9.1% of total hip replacements, and is a common cause of total hip replacements in young adults.[27, 63] Irish data showed that in patients under 50 years of age who were undergoing total hip replacement, over 40% had evidence of dysplasia. However, it was not reported whether this dysplasia was present when the patients were infants or developed later in life, and whether or not the patient had received treatment for the dysplasia prior to the hip replacement.[64] Considering patients with DDH, one - systematic review, with more than 40-years follow-up, assessing long-term outcomes of late detected DDH hips treated after walking age found that up to 35% of patients required total hip replacement.[65] Also, the risk of total hip replacement was estimated to be more than twice greater (relative risk 2.6) when there is hip instability at birth compared to a normal hip.[27, 63]

3.4 Incidence of DDH

The incidence of DDH varies geographically.[4, 19] This variation is likely due to genetic and cultural factors (such as post-natal swaddling practices). The reported incidence of DDH may also vary due to the age at examination, experience of the examiner, screening methods, and definitions of DDH applied. There are inconsistencies noted in terms of the specific condition and criteria reported. For example, some reported incidence estimates include: irreducible hip dislocation, clinical hip instability, DDH in an unscreened population, or DDH in a screened population (either clinical screening or ultrasound screening). These are detailed in the next paragraphs.

The reported incidence figures included in this review are derived from studies deemed most relevant to this report. This includes results from:

1) a report from Austria, where a universal ultrasound screening programme has been in place since 1992, and therefore may represent the longest follow-up for the highest number of patients with DDH that will be identified in a population;[66]

2) a report from the UK, where there are similar screening practices in place to Ireland;[4]

3) a report from an unscreened population.[19]

Additionally, results from a study on the prevalence of DDH, based on an international review of unscreened populations, is presented.[19] While incidence (that is, the number of newly detected cases) is most relevant to this report, information on prevalent cases can help inform how the number of cases change over time, potentially informing the rate of spontaneous resolution of DDH.

Results from an analysis of infants born consecutively in Austria, where a universal ultrasound screening programme for DDH has been implemented since 1992, summarised incidence of DDH in the country.[66] Of the 27,808 patients eligible for the analysis, 2,715 (9.8%), had a hip higher than Graf type I (normal hip). A total of 2,497 (9.0%) had hips that were classified as Graf type IIa (immature hips). The remaining patients with hips higher than Graf type I (0.8%) were classified as the following: type IIb – 9 (<0.1%), type IIc – 182 (0.7%), type III – 19 (0.1%), and type IV – 8 (<0.1%).

In the UK, the incidence of irreducible hip dislocation has been reported as between 0.5 and 0.8 per 1,000 live births in a population with a selective ultrasound screening programme (referral based on clinical symptoms and risk factors). However, the incidence may be higher due to the potential for undetected cases during screening.[4] Several observational studies have shown that mild instability in

the hip is present in 1.0% to 1.5% of infants (an incidence of 5 per 1,000 in males and 13 per 1,000 in females).[19, 21, 27, 63]

A 2007 review aimed to report the incidence of ultrasound-detected hip abnormalities from universal screening programmes in newborn populations.[19] Five studies were included which used the Graf technique. These were published between 1986 and 1997 and the studies included between 615 and 4,648 infants screened. The results are summarised as follows:

- Type I (normal hips) – 40.1% to 84.7%
- Type IIa (immature hips) – 2.3% to 56.6%
- Type IIc or type D – 1.2% to 4.5%
- Type III or type IV – 0.2% to 0.7%.

These reported values come from a universally screened population, which may represent the incidence in the overall infant population. However, it is important to note there was large variation in the frequencies reported, and that the age at screening was not reported in the review. Therefore, the reported incidence should be interpreted with regards to these limitations.

This 2007 review also reported that in an unscreened population, the median prevalence of DDH was 1.3 per 1,000 (range 0.8 to 1.5 per 1,000).[19] The reported prevalence was based on a review of 44 unscreened populations, predominantly from the USA, Canada, Scandinavia, and the UK. It was not clear whether the 'unscreened population' only included those who did not receive ultrasound screening, or if there was no clinical examination either. The same publication reported the prevalence of clinical hip instability detected using the Ortolani and Barlow manoeuvres; this ranged from 1.6 to 28.5 per 1,000 in neonates.[19] Furthermore, the prevalence of ultrasound-detected hip DDH in newborns, when ultrasound was used as the primary screening method, ranged from between 34.0 and 60.3 per 1,000. It was not clear whether the reported prevalence included studies of both universal and selective screening programmes. The authors noted that the higher prevalence of clinical hip instability, based on the clinical screening programmes, than clinically diagnosed DDH, is consistent with findings that hip instability often resolves in the first few weeks of life.

3.5 DDH in Ireland

There is no national database of cases of DDH in Ireland. Therefore, estimations of the incidence of the condition in the country are challenging. Two publications from the University Hospital Waterford (UHW) were identified which reported on the incidence of DDH in Ireland.[29, 67] One of these was from 2009, prior to the

publication of the implementation pack for the selective ultrasound screening recommendations in 2017, and may not be applicable to the current context in Ireland.[67] The second publication reported on all infants at a single centre diagnosed with DDH, including those referred for selective ultrasound diagnostics, and those identified later, without being initially identified at the newborn examination with clinical signs of DDH or risk factors.[29] Additionally, one publication, also from UHW, was identified which included the Graf classification from infants referred to the selective ultrasound diagnostics programme.[37] Of note, as these infants were referred for selective ultrasound diagnostics due to clinical suspicion and or risk factors, the DDH severity by Graf classification is not representative of the general infant population. However, the results are illustrative of the population that are referred under the current selective ultrasound diagnostic practice at that centre.

The first study was retrospective and aimed to estimate the incidence and treatment outcomes in the Southeast of Ireland (where all cases are referred to the same centre, UHW) from a chart review of children born in 2009.[67] The authors noted that at the time of the study, standard practice was to perform examination of the hips of infants prior to discharge from the hospital. Most examinations were performed by an experienced paediatrician, with some examinations performed by paediatric trainees. All home births were examined by the area medical officer or general practitioner. All newborns were examined again at approximately six weeks of age by the area medical officer or general practitioner. The Barlow and Ortolani manoeuvres were used to detect hip instability. Infants with abnormal clinical examinations and or risk factors for DDH were referred to the DDH assessment clinic. The risk factors were not defined in the publication, and the authors noted that at the time of the study, there was no standard protocol for referring patients based on positive risk factors alone. Overall, the incidence of DDH in the Southeast of Ireland was estimated to be 6.73 cases per 1,000 live births. The incidence of early diagnosis (defined as those identified and treated before three months of age) was 3.97 per 1,000 live births, and the incidence of late diagnosis (defined as those identified after three months of age) was 3.77 per 1,000 live births. In the early diagnosis group, the incidence of operative management was 0.24 per 1,000 live births, compared to 0.84 per 1,000 live births in the late diagnosis group, with a significant difference in the requirement for surgical management between the two groups ($p = 0.024$). The authors reported that at the time of the study, there was no universal ultrasound screening programme. To note, current recommendations in Ireland for selective ultrasound screening may not reflect the practices described at the time of the study.

A review was published of children diagnosed with DDH in Southeast Ireland (again, all referred to UHW) over an 18-month period from January 2018 to June 2019.[29]

All infants diagnosed with DDH clinically or with imaging who were treated locally with abduction bracing or referred onwards for tertiary care were included in the calculation of incidence. The incidence was found to be 31.1 cases per 1,000 live births. Of these, 7.9 per 1,000 live births were considered as late-diagnosed DDH (defined as those diagnosed at or after three months of age).

Additionally, the previously described publication (in Chapter 2, section 2.3) of an audit of all referrals made to the UHW in 2020 described the Graf classification findings from the 992 infants referred for selective ultrasound diagnostics at the hospital.[37] Almost all (88.2%) of the hips were Graf type I (normal hips), or Graf type IIa (immature hips, 6.75%).

Finally, unpublished data from an audit of all hospitals in Ireland that carry out DDH diagnostics was performed for the years 2021 and 2022.[58] Data were requested from all hospitals and the information was submitted voluntarily (Table 3.1). Most hospitals provided the requested data. In both years, 18.5% of infants born received an ultrasound for DDH assessment. Of those included in the audit (69.8% and 77.8% of those who received ultrasound in 2021 and 2022, respectively), 9.6% and 11.8%, in 2021 and 2022, respectively, had abnormal hips on ultrasound. As outcome data were not available for a large percentage of infants who received an ultrasound assessment for DDH, the national incidence of DDH cannot be calculated from this data.

Table 3.1 Results from national audit of centres carrying out ultrasound screening for DDH in Ireland

	2021	2022
Births in Ireland	58,443	57,540
Infants who received ultrasound for DDH assessment, n (%)	10,796 (18.5%)	10,659 (18.5%)
Infants included in national audit, n	7,533	8,292
Infants scanned by ultrasound but outcomes not available for this audit, n	3,263	2,367
Infants treated with harnessing, n (%)*	317 (4.2%)	196 (2.4%)
Infants received follow-up imaging for immature hips, n (%)*	407 (5.4%)	780 (9.4%)
Infants with abnormal hips on ultrasound, n (%)*	724 (9.6%)	976 (11.8%)

Key: DDH – developmental dysplasia of the hip.
* Percentage of those included in the national audit.

3.6 Discussion

The purpose of this chapter was to outline the epidemiology of DDH, including the aetiology, natural history, and burden of the disease. The chapter was informed by

international and national data, where available. The cause of DDH is multifactorial and said to be poorly understood. There are several known risk factors which may contribute to the development of DDH. As previously noted, the definition of DDH varies. For this chapter, a strict definition of DDH was not adopted, given the limited data available, and uncertainty regarding the clinical course of disease.

The natural history of DDH is poorly understood, mostly due to inconsistencies in terminology, the lack of long-term studies which follow all severities of infant hip instability to maturity, and the scarcity of prospective studies. Some publications of the natural history of DDH report from the stage of hip instability, while others report from the stage of a diagnosis of DDH. It is not clear from all studies at which point the cases are followed, leading to discrepancies in reporting of outcomes. Additionally, it was noted that there is a large percentage of cases of mild hip instability that resolves spontaneously, without treatment, in the first few weeks of life. Therefore studies of natural history and outcomes that include all cases of mild hip instability identified from soon after birth may report better outcomes, in the absence of treatment, that those that include only cases that are identified at slightly older ages. This would likely be due to the cases identified older only being the cases that did not resolve on their own, and therefore may be more severe. Conversely, some studies suggest that there are cases of DDH without physical symptoms in infancy that become symptomatic later in life. Overall, the natural history from infancy of DDH remains unclear.

Variation in reporting of the incidence of DDH was also noted, with limited large-scale studies aiming to identifying the incidence. There was variation in terms of the definition of DDH applied, with some studies reporting on subtypes across the spectrum of disease, such as irreducible hip dislocation, or clinical hip instability. In studies that specifically defined the overall incidence of DDH, studies reported on unscreened populations, clinically screened populations, and or populations screened with imaging. Most studies were not clear on how the cases were identified, and in those that reported on screened or unscreened populations, it was not clear which form of screening (that is, clinical, selective ultrasound, or universal ultrasound) was applied, and whether clinical examinations were considered to be screening. Furthermore, there is a noted variation geographically in the incidence of DDH, partially due to genetic factors and swaddling practices. Overall, variation in reporting between identified studies (such as the definition of DDH or the timing of ultrasound) makes the true burden of DDH difficult to estimate.

Results from an audit of all referrals made to the UHW centre in 2020 provided percentages of different Graf classifications of hips determined from imaging.[37] Incidence of DDH at this centre reported from 2018 and 2019 was found to be 31.1 cases per 1,000 live births.[29] All infants diagnosed with DDH clinically or with imaging who were treated locally with abduction bracing or referred onwards for

tertiary care were included in the calculation of incidence. The calculation of incidence (31.1 cases per 1,000 live births) included: 1) all those identified as DDH with the initial ultrasound for those referred for selective ultrasound diagnostics; 2) those with a Graf type IIa hip who are identified as positive for DDH at the follow-up; and 3) those identified at the six month radiograph. It is, however, challenging to compare the incidence of DDH reported in Ireland to incidence rates reported internationally. It would be expected that the incidence from countries with universal ultrasound screening for DDH would represent the highest plausible incidence of DDH.

An important consideration is the impact of screening on the incidence of observed cases. The reported incidence is likely to be higher in regions with screening for DDH. This relative increase would likely be larger for countries with ultrasound screening programmes, and specifically those with universal ultrasound screening programmes. If screening is in place, cases of hip instability and DDH that may not have been detected in the absence of such screening would be included in estimates of incidence. It is important to note however that it is not clear how many of these cases would truly be clinically significant (that is, require intervention). As previously noted, there can be resolution of hip instability without treatment. These cases may have never contributed to long term burden of the disease. The timing of the screening may play an important factor in the estimation of the incidence of the condition.

As part of the universal ultrasound screening programme in Austria, between 1998 and 2014, the incidence of Graf type higher than IIa hips was reported to be 8 per 1,000 in a consecutive sample of 28,092 neonates in a single hospital.[66] Similarly, the study based on Irish data prior to the publication of the recommendations for the selective ultrasound screening programme reported an incidence of DDH of 7 per 1,000 births. However, available Irish data reported from a single centre with selective ultrasound screening from a period after the publication of the recommendations indicate a higher estimate of 31 per 1,000.[29] This incidence reported at the single centre may not be representative of the whole country, and is based on a small sample size and had a very short study period (18 months) compared with the Austrian study. Therefore, it is difficult to determine whether the results suggest a high incidence of DDH in Ireland, if the current selective ultrasound recommendations in Ireland are highly sensitive, or if the data from this centre are not nationally representative.

In terms of Irish data, it is important to note that there is no central register of cases of DDH. Therefore, estimates of incidence in the country are largely based on reporting from individual hospitals. Three studies from the same hospital were identified which reported on the incidence and selected outcomes for cases of DDH. It is not clear how representative this hospital is of practice and case numbers

across the rest of the country; for example, this centre may have a particular focus and interest in DDH screening, and therefore there may be more cases identified due to awareness of the condition and appropriate diagnostics methodology. The incidence reported in a later publication, after the publication of the selective ultrasound screening programme recommendations, showed a higher reported incidence of DDH than reported in the earlier publication, prior to the use of selective ultrasound screening. This suggests increased identification of cases when the criteria for referral for selective ultrasound are applied. It is also important to highlight however that the centre also includes follow-up radiography at six months for all infants referred for selective ultrasound diagnostics, for whom no hip abnormalities were detected. As noted in the publication, some additional cases of DDH are identified at this follow-up radiography. Therefore, the authors suggested that the current recommendations may be insufficient in identifying all cases of DDH.

In practice, there is no diagnostic test which can confirm whether or not a case of hip instability detected in infancy would require treatment, or would resolve on its own. Given the changing natural history of the condition in early life (that is, some cases may resolve spontaneously and some cases that may have been detected in infancy will not present later in life), it is unlikely that any test can be highly sensitive and specific.

4 Clinical effectiveness of universal ultrasound screening compared to selective ultrasound screening

Key points

- A summary of reviews was undertaken to synthesise and assess the clinical effectiveness and safety of universal ultrasound screening for developmental dysplasia of the hip (DDH), relative to selective ultrasound screening.
- Eight relevant systematic reviews were identified. Outcomes considered by the systematic reviews included incidence of late DDH, rates of non-surgical and surgical treatment, functional complications (for example, delayed walking, gait disturbances or pain), and harms (for example, unnecessary treatment or psychological distress).
- Four primary studies were identified from these systematic reviews which: 1) compared universal with selective ultrasound screening for DDH; and, 2) reported on at least one relevant outcome. These included two randomised controlled trials (RCTs) and two retrospective cohort studies. One of the systematic reviews included meta-analyses of the RCT evidence.
- For the outcome of late DDH, meta-analysis of the two RCTs found that universal ultrasound screening did not result in a statistically significant reduction in this outcome, compared to selective ultrasound screening (relative risk (RR) 0.49, 95% confidence interval (CI): 0.19 to 1.26). In one retrospective cohort study, there was no statistically significant difference found in the incidence of late DDH between the universal (0.50 per 1,000; n = 10,015 births) and selective (0.28 per 1,000 births; n = 18,053) ultrasound screening groups. Of note, cases of late DDH in the universal screening group were reported to be due to clinical appointments not being made or kept, rather than representing false negative ultrasound results.
- Universal ultrasound screening for DDH may be associated with an increase in non-surgical treatment rates, relative to selective screening, although the magnitude of this effect is subject to uncertainty.
- Considering surgical intervention, meta-analysis of two RCTs found no statistically significant difference in requirements for surgical intervention with universal screening relative to selective screening (RR 0.36, 95% CI: 0.04 to 3.48). Consistent with these findings, one of the two retrospective cohort studies identified reported no significant difference in the incidence of surgical

intervention between the universal and selective screening cohorts. The findings of the second retrospective cohort study were unclear.

- Taken together, there is evidence of increased non-surgical intervention associated with universal ultrasound screening, relative to selective screening, without a corresponding reduction in the incidence of late DDH or requirements for surgical intervention. This suggests that the additional cases identified through universal screening are likely mild, and may resolve spontaneously in the absence of treatment.
- No data on outcomes of functional complications were identified. In included RCTs (n = 2), evidence of potential harms was limited to avascular necrosis of the femoral head, with no significant differences found between the universal and selective ultrasound screening groups. However, studies were likely underpowered for this outcome.
- The relative benefit of universal ultrasound screening, compared with selective screening, is unclear in the absence of high-quality comparative studies. Although limited, the available evidence suggests that screening all infants with ultrasound for the detection of DDH has the potential to lead to unnecessary treatment, with the risk of clinically significant consequences.

4.1 Introduction

Early detection of developmental dysplasia of the hip (DDH) may enable less invasive and potentially more effective treatment options. The aim of this chapter is to describe the clinical effectiveness and safety of universal ultrasound screening for DDH relative to selective ultrasound screening. As noted in Chapter 2, there are recommendations for selective ultrasound screening for DDH in place in Ireland.

A number of systematic reviews comparing the use of universal versus selective ultrasound screening for DDH have been published. In order to assess the clinical efficacy and effectiveness of these two ultrasound screening strategies, a summary of systematic reviews was undertaken to produce an overview of the relevant evidence.

4.2 Methods

Detailed methods are available in the accompanying evidence review protocol (link). Briefly, this review sought to answer the following question through identification, in the first instance, of systematic review literature:

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

Table 4.1 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) ▪ Pathway timings (for example, time to diagnosis, time to treatment) ▪ Harms
Study design	Include: ▪ Systematic reviews (with or without a meta-analysis) that: ○ clearly state a set of objectives with an explicit, reproducible methodology ○ contain a systematic search that attempts to identify all studies that would meet the eligibility criteria ○ have a systematic presentation, and synthesis, of the characteristics and findings of the included studies ○ compare universal ultrasound screening with selective ultrasound screening for DDH

	○ report one or more of the outcomes of interest Exclude: ▪ Literature reviews not completed using systematic review methodology ▪ 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 ▪ Reviews in a neonatal population with diagnosed hip pathology ▪ Reviews in which the comparator is clinical examination alone (that is, not selective ultrasound screening)

Key: DDH - developmental dysplasia of the hip.
† Selective ultrasound screening was defined as screening only infants with risk factors for DDH (for example, family history or breech presentation) or abnormal results of clinical examination.

4.2.1 Search methods

A search for relevant literature was conducted in PubMed using the specialised PubMed Clinical Queries tool. Details of the search strategy are available in the protocol.

4.2.2 Review selection, data extraction and management

Selection of studies

All potentially eligible systematic reviews identified through the search methodology were screened independently by two reviewers, with disagreements resolved by discussion. Reviews reporting on at least one outcome of interest were considered eligible for inclusion.

Once included, the potential for overlap of primary studies across identified systematic reviews was investigated by developing an evidence matrix. The aim of the matrix was to avoid duplicating the reporting of information from primary studies.[68] Where there was substantial overlap in the studies included, the reviews which included the greatest number of relevant studies were prioritised for data extraction of individual study findings and for quality assessment.

Data extraction and management

Systematic review characteristics (for example, study designs included, outcomes assessed and author conclusions) were reported for all relevant reviews identified. As noted, data extraction of individual study findings was performed for reviews prioritised based on the evidence matrix.

Data extraction was performed by one reviewer using Microsoft Excel software. All data extracted were checked by a second reviewer, with disagreements resolved by

discussion. A standardised, pre-piloted data extraction template was developed prior to undertaking the review. Relevant clinical outcomes and harms were identified with consideration to the clinical outcomes that the systematic reviews sought to assess (see Table 4.2). The clinical outcomes identified and extracted (where results were available) were as follows:

- incidence of late DDH (as defined by the systematic review authors)
- treatment (surgical and non-surgical)
- time to surgical treatment
- functional outcomes (for example, crawling difficulties, delayed walking, gait disturbances, chronic pain, osteoarthritis of the hip, or osteoarthritis requiring joint replacement)
- harms (for example, unnecessary treatment, psychological distress).

Data synthesis

The findings of the included systematic reviews, including results of meta-analyses performed, were narratively synthesised.

Quality assessment

Systematic review quality was assessed using the ROBIS quality assessment tool.[69] Only systematic reviews prioritised for data extraction of study findings were quality appraised (see section 4.3.1, Evidence Matrix). The ROBIS tool evaluates the quality of systematic reviews over three phases; assessing relevance, identifying concerns, and judging risk of bias. Results for phase one (assessing relevance) are not shown, as reviews that did not meet the pre-specified inclusion criteria were excluded during study selection.

4.3 Results

4.3.1 Characteristics of included studies

Eight systematic reviews were identified in which all or a proportion of the included primary studies presented data comparing universal ultrasound screening with selective ultrasound screening for DDH (Table 4.2). The publication dates of these reviews ranged between 2005 and 2023.

Study design

Of the identified systematic reviews, five reviews included clinical trials (that is, randomised, quasi-randomised or cluster-randomised clinical trials) and observational evidence,[6, 7, 13, 14, 70] one review included clinical trials only,[28] one

review included observational evidence only,[5] and one review set out to include the best available evidence in line with the hierarchy of evidence (ranging from systematic reviews, representing the top tier of evidence, to case series, representing the lowest) for a given outcome.[8] In some cases, the systematic reviews presented data for single-arm observational studies (that is, non-comparative data); these were not considered relevant to the research question outlined in Table 4.1.

Evidence matrix

In order to determine the overlap between the primary studies included in the identified systematic reviews, an evidence matrix was completed (Table 4.3). In examining the evidence from the eight systematic reviews,[5-9, 13, 14, 70] four primary studies were identified which compared universal and selective ultrasound screening for DDH, and included outcomes of interest to this review.[71-74]

As demonstrated in Table 4.3, three of the four relevant primary comparative studies are captured in the systematic review undertaken by Pandey et al. 2021,[6] while the remaining study was captured in both Laborie et al. 2023,[13] and Cheok et al. 2023.[70] The remaining systematic reviews captured fewer than three of these studies. Therefore, the Pandey et al. 2021, Laborie et al. 2023, and Cheok et al. 2023 systematic reviews were used to outline the clinical effectiveness of selective versus universal screening for DDH. The additional systematic reviews presented in Table 4.2 were used to supplement and corroborate the findings of these three reviews, where appropriate.

Characteristics of primary studies

Four primary studies, comprising two randomised controlled trials (RCTs) and two retrospective cohort studies, were identified which compared universal and selective ultrasound screening for DDH.[71-74] The mean duration of follow-up in the RCTs ranged from 42.4 months[71] to 8.5 years.[73] The Graf technique of ultrasound was used in two of the primary studies (one RCT and one retrospective cohort study),[71, 74], while the Harcke technique[72] and the Morin-Terjesen technique[73] were used in the remaining retrospective cohort study and the remaining RCT, respectively.

In one retrospective cohort study all infants born between June 2005 and May 2008 formed the universal screening cohort[74], while infants born between January 2009 and December 2011 formed the selective screening cohort; outcomes of screening were compared between these two time periods. The minimum duration of follow-up in the selective screening cohort was two years. A second retrospective cohort study adopted a similar study design.[72] From 1986 to May 1989, all newborns underwent selective ultrasound screening. In June 1989 to 1996, universal ultrasound screening

was implemented. The minimum duration of follow-up for this study was not reported.

In addition, follow-up data from one of the RCTs included in Table 4.3 was identified through forward citation searching.[75] The follow-up study aimed to assess radiographic and clinical features related to acetabular dysplasia and early degenerative change at skeletal maturity (mean age: 18.6 years, range 17.2 to 20.1), which may be associated with early onset hip osteoarthritis. From the original trial including 11,925 newborns,[71] a subset of 3,935 adolescents were invited for follow-up at 18 to 20 years of age. A final sample of 2,011 participated in the follow-up study.[75]

Outcomes reported

Outcomes reported in the available evidence varied according to the primary studies contained within the systematic reviews. The incidence of each of late DDH and non-surgical treatment was reported in three primary studies.[71, 73, 74] All four primary studies reported on the rate of surgical intervention in infants with universal ultrasound screening compared to those with targeted ultrasound.[71-74] Functional outcomes were not addressed in any of the studies. One systematic review presented data on clinical harms as reported in two primary studies.[28]

Table 4.2 Characteristics of identified systematic reviews

Review (search date)	Population	Screening strategies	Length of follow -up	Outcomes assessed	Definition of late DDH	Review author's conclusion	Quality appraisal of primary studies as reported in the systematic review
Cheok 2023[70] (27 March 2022)	Newborns	▪ Universal ultrasound screening ▪ Selective ultrasound screening	NR	▪ Prevalence of late diagnosis of DDH ▪ Prevalence of abduction bracing treatment ▪ Prevalence of surgical treatment	Established after 3 months of life. Range 3 to 12 months in included studies.	Universal screening showed a trend towards lower prevalence of late DDH compared to selective screening. However, it was also associated with a significant increase in the prevalence of abduction bracing without a significant reduction in the prevalence of surgical procedures in childhood for dysplasia being performed. High-quality studies comparing both treatment methods are required, in addition to studies into the natural history of missed DDH.	Of the 31 studies included, 29 were considered at low risk of bias. In most studies, there was a lack of a reliable measure to assess the incidence of late DDH.
Laborie 2023[13] (February 2021)	Newborns up to 6 weeks without underlying congenital disorders, including cerebral palsy and other neurological conditions	▪ Universal ultrasound screening ▪ Selective ultrasound screening	22 weeks to 5.5 years.	▪ Incidence/ prevalence of late presentation of DDH ▪ Incidence/ prevalence of sequelae of DDH (e.g., avascular necrosis, limp)	The definition differed between individual studies. For each study, the age at which 'late presentation' was defined, from 4 weeks of age and beyond, was always after the initial newborn screening period.	Selective ultrasound screening appears to slightly increase the rate of late presentation compared to universal ultrasound screening for DDH, although universal screening does not eliminate the risk of late presentation. However, uniformity in design and in reporting of DDH studies is required particularly in relation to age at clinical and ultrasound screening, to method of clinical and ultrasound screening, to duration of follow-up and to the definition of late presentation. Furthermore, a cost-effectiveness analysis is needed to update current recommendations.	All studies used appropriate methodology to address the research question.
Kuitenen 2022[5] (25 and 27	Newborns	▪ Clinical screening ▪ Selective ultrasonogr	NR	▪ Country and screening stratified incidences of early-detected DDH (age <12 weeks)	Late detected cases (≥ 12 weeks)	This meta-analysis found that early detection rates and non-operative treatments were higher with universal screening. The late detection and operative treatment rates with universal	Included studies had no major issues. However, only observational studies were

Review (search date)	Population	Screening strategies	Length of follow-up	Outcomes assessed	Definition of late DDH	Review author's conclusion	Quality appraisal of primary studies as reported in the systematic review
November 2021)		aphy screening ▪ Universal ultrasonography screening		▪ Initial non-operative treatment rate ▪ Incidence of late-detected DDH (age ≥12 weeks) ▪ Operative treatment rate.		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.	included, which may create a risk of selection and reporting bias.
Pandey 2021[6] (search date not reported)	Newborns up to 4 weeks	▪ No screening ▪ Universal clinical screening ▪ Selective ultrasonography screening ▪ Universal ultrasonography screening	1 to 40 years	▪ Sensitivity ▪ Specificity ▪ Treatment rate ▪ Mean age at treatment ▪ Mean age at first operation ▪ Cost-effectiveness of each method ▪ Challenges encountered, in implementation of screening strategy	Variable - range 1 to 12 months. The majority of studies considered cases presenting after 3 months as late presentation.	The literature supports universal ultrasound screening and has proved its cost-effectiveness. However, considering the logistic and financial challenges in our country, immediate implementation of universal ultrasound screening seems impractical. In the absence of any current guidelines for screening for DDH in India, we suggest professional organisations involved in the care of children and public health policy-makers to come together to develop national screening guidelines for DDH.	NR
Jung 2020[7] (January 2020)	Infants	▪ Universal hip ultrasonography screening ▪ selective hip ultrasonography screening	NR	▪ Incidence of late-diagnosed DDH	> 1 month to > 6 months	This meta-analysis suggests that a statistically significant decrease in the incidence of late-diagnosed DDH is possible when universal hip ultrasonography screening is adopted compared with selective hip ultrasonography screening. However, the strategy of infant hip screening that is appropriate should be considered individually, by each country, in the context of socioeconomic factors and healthcare policies, including insurance.	The risk of bias in the RCTs was poor; however, the cohort studies were of high quality.
Shorter 2011[28]	Newborns up to 6 weeks	▪ No screening	Within 2 years	▪ Incidence of late diagnosed DDH ▪ Any treatment	After 8 weeks of age	There is insufficient evidence to give clear recommendations for practice. There is inconsistent evidence that	There were substantial methodological

Review (search date)	Population	Screening strategies	Length of follow-up	Outcomes assessed	Definition of late DDH	Review author's conclusion	Quality appraisal of primary studies as reported in the systematic review
(February 2010, updated January 2011)		▪ Clinical screening ▪ Ultrasound screening (universal or targeted) alone or in combination		▪ Delayed abduction splinting >8 weeks of age. ▪ Open surgery for correction of hip dysplasia. ▪ Avascular necrosis or osteoarthritis of the hip, at any age. ▪ Delayed walking >18 months of age ▪ Limb length discrepancy at any age. ▪ Gait abnormality at any age. ▪ Chronic hip pain, at any age ▪ Hip replacement		universal ultrasound results in a significant increase in treatment compared to the use of targeted ultrasound or clinical examination alone. Neither of the ultrasound strategies have been demonstrated to improve clinical outcomes including late diagnosed DDH and surgery. The studies are substantially underpowered to detect significant differences in the uncommon event of late detected DDH or surgery. For infants with unstable hips or mildly dysplastic hips, use of delayed ultrasound and targeted splinting reduces treatment without significantly increasing the rate of late diagnosed DDH or surgery.	concerns for both studies.
Shipman 2006[8] (January 2005)	Infants from birth to 6 months	▪ Clinical examination and universal ultrasound screening ▪ Clinical examination and selective ultrasound ▪ Clinical examination only	NR	▪ Reduced need for surgery ▪ Improved functional outcomes such as gait, physical functioning activity level, peer relations, family relations, and school and occupational performance ▪ In the absence of data on functional outcomes, non-functional outcomes (the need for surgical treatment, age at surgical treatment, mean number of visits	NR	Screening with clinical examination or ultrasound can identify newborns at increased risk for DDH, but because of 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.	NR

Review (search date)	Population	Screening strategies	Length of follow-up	Outcomes assessed	Definition of late DDH	Review author's conclusion	Quality appraisal of primary studies as reported in the systematic review
				at outpatient clinics, total hip-related hospitalisations, and the occurrence of definite or suspected avascular necrosis)			
Woolacott 2005[14] (March 2004)	Unselected newborns	▪ General clinical screening ▪ General clinical screening plus ultrasound screening ▪ General clinical screening plus selective ultrasonography	NR	▪ Diagnostic accuracy ▪ Overall treatment rates ▪ Rates of operative intervention ▪ Rates of abduction splinting ▪ Rate of delayed diagnosis ▪ Time to treatment ▪ Duration of treatment ▪ Rate of treatment complications ▪ False diagnostic labelling ▪ Any long term functional outcomes	Variable - range 1 to 8 months	Clear evidence is lacking either for or against general ultrasound screening of newborn infants for DDH. Studies that investigate the natural course of the disorder, the optimal treatment for DDH, and the best strategy for ultrasound screening are needed.	Methodological concerns related to blinding (n=2), assessment bias (n=1), and randomisation procedures (n=1)

Key: DDH – developmental dysplasia of the hip, NR – not reported.

Table 4.3 Relevant primary studies included in each systematic review

			Systematic reviews							
			Cheok 2023[70]	Laborie 2023[13]	Kuitunen 2022[5] †	Pandey 2021[6]	Jung 2020[7]	Shorter 2011[28]	Shipman 2006[8]	Woolacott 2005[14]
Primary studies included	Rosendahl 1994[71]	RCT		✓		✓	✓	✓	✓	✓
	Clegg 1999[72]	Retrospective cohort				✓	✓			
	Holen 2002[73]	RCT	✓	✓		✓	✓	✓	✓	✓
	Westacott 2018[74]	Retrospective cohort	✓	✓						

Key: RCT – randomised controlled trial.

† The systematic literature search did not identify any new RCTs since the Cochrane review in 2011 (Shorter et al. 2011). An update of the Cochrane review was therefore not considered necessary by the study authors. Kuitunen et al. 2023 included non-comparative observational evidence only.

4.3.2 Late DDH

As noted in Chapter 2, late presentation of DDH (here 'late DDH') in infancy is associated with poor clinical outcomes. Three studies, comprising two RCTs and one retrospective cohort study,[71, 73, 74] reported across two systematic reviews, compared the incidence of late DDH between universal and selective ultrasound screening.[6, 13] Late DDH was defined differently in the underlying studies included in identified systematic reviews. Of the two RCTs included in Pandey et al. 2021,[6] both RCTs defined late DDH as those diagnosed after one month of age[73].[71] The additional retrospective cohort included in Laborie et al. 2023[13] defined late DDH as identification later than three months of age.[74]

Meta-analysis of the two RCTs, including 23,530 infants, demonstrated that the difference in the incidence of late DDH between the universal and selective ultrasound screening groups was not statistically significant (relative risk (RR) 0.49, 95% confidence interval (CI): 0.19 to 1.26).[28] The additional comparative study included in Laborie et al. 2023[13] reported a higher incidence of late DDH in the universal ultrasound screening group (0.50 per 1,000; n = 10,015 births) than in the selective screening group (0.28 per 1,000 births; n = 18,053), although the difference was not statistically significant.[13, 74] It is important to note that the authors of this retrospective cohort study reported that cases of late DDH in the universal ultrasound screening group were not due to the screening test, but rather were attributable to challenges associated with the overall care pathway (that is, failure to attend the neonatal scan, and failure to make follow-up appointments in part due to clinical appointments not being made or kept).[74]

4.3.3 Treatment

Non-surgical intervention

Three studies compared non-surgical intervention rates between universal and selective ultrasound screening groups.[71, 73, 74] Two RCTs included in Pandey et al. 2021 reported on abduction splinting treatment rates.[6] One RCT reported a rate of 9.6 per 1,000 in the universal ultrasound screening group and 8.6 per 1,000 in the selective ultrasound screening group.[73] While this suggests a higher treatment rate in the universal ultrasound screening group, the difference between groups was not statistically significant. The second RCT reported a statistically significant increase in the rate of treatment with abduction splinting among those in the universal ultrasound screening group (34 per 1,000) compared to the selective ultrasound screening group (20 per 1,000).[71] Of note, 60% of infants with dysplasia in the universal ultrasound screening group were considered low risk (that is, did not have known risk factors or evidence of hip instability during clinical examination). Shorter et al. 2011 noted that meta-analysis of these results was not considered appropriate due to evidence of substantial statistical heterogeneity (I^2 = 77%).[28] Differences in

treatment thresholds and in the experience of the clinical hip examiners were reported as potential reasons for evidence of substantial statistical heterogeneity.[6]

The retrospective cohort study included in Laborie et al. 2023 also reported a statistically significantly higher abduction rate in the universal ultrasound screening group (7.90 per 1,000, 95% CI: 6.24 to 9.72) compared with the selective ultrasound screening group (2.30 per 1,000, 95% CI: 1.67 to 3.09).[13, 70, 74]

Surgical intervention

Four primary studies reported across two systematic reviews reported on the rate of surgical intervention in infants with universal ultrasound screening compared to those with targeted ultrasound.[71-74] Fixed effect meta-analysis of two RCTs[71, 73] carried out by Shorter et al. 2011, reporting on outcomes of 23,530 infants, did not find a significant difference in surgery (RR 0.36, 95% CI: 0.04 to 3.48) in infants screened using universal ultrasound screening compared to infants screened using selective ultrasound screening.[28] There was no evidence of statistical heterogeneity (I^2 = 0.0%).

The retrospective cohort study within Pandey et al. 2021[6] also compared the rate of surgical interventions between a universal screening cohort (June 1989 to 1996) and selective screening cohort (1986 to May 1989).[72] The study reported an average surgical intervention rate of 2.5 per year in the universal ultrasound screening group, compared to a rate of 5.4 per year in the selective ultrasound screening group. The absolute number of cases during the study period was not reported. The retrospective cohort study included in Laborie et al. 2023[13] outlined a surgical intervention rate of 0.90 per 1,000 live births in those that underwent universal ultrasound screening (n = 10,015, June 2005 to May 2008), and 0.60 per 1,000 live births in those that underwent selective ultrasound screening (n = 18,053, January 2009 to December 2011); however, this difference was not statistically significant.[74] As noted in the preceding section, this study reported a significantly higher rate of non-surgical treatment in the universal screening cohort, compared with the selective screening cohort (0.79% versus 0.23% of cases screened, respectively). This suggests that despite earlier identification and treatment of a greater number of cases, this did not translate into a significant reduction in requirements for surgical intervention.

4.3.4 Pathway timings

Time to surgical treatment

The age at first operation was only described by one retrospective cohort study; this was reported in Pandey et al.[6, 72] The mean age at first operation was 6.7 months and 14.2 months for those in the universal and selective ultrasound screening

groups, respectively. While age at first operation was younger in the universal ultrasound screening group, given that outcomes were compared across different time periods, it is not possible to determine if this was due to differences in the intervention, or changes in clinical practice over time.

4.3.5 Functional outcomes

No data were identified comparing functional outcomes (for example, crawling difficulties, delayed walking, gait disturbances, chronic pain, osteoarthritis of the hip, or osteoarthritis requiring joint replacement) of universal compared to selective ultrasound screening for DDH.

4.3.6 Harms

Overall, evidence of potential harms of screening was limited in identified systematic reviews. Meta-analysis by Shorter et al. 2011 of two studies did not show a significant difference in avascular necrosis or osteoarthritis in a fixed-effect model (RR 0.33, 0.01 to 8.02); however, it is likely that these studies were underpowered for this outcome.[28]

The 2013 follow-up study of young adults (mean age: 18.6 years, range 17.2 to 20.1) enrolled in an RCT assessing the efficacy of screening for DDH in newborns (1988 to 1990), identified through forward citation searching, found that there were no statistically significant differences between radiographic findings associated with acetabular dysplasia or early degenerative change at skeletal maturity between the universal and selective ultrasound screening groups.[71, 75] Furthermore, the authors suggested that a universal ultrasound screening strategy did not appear to result in higher rates of avascular necrosis (AVN) (no instances of AVN reported), relative to a selective ultrasound screening strategy,[75] despite the higher treatment rates reported in the universal screening group at a minimum of 27 months follow-up in the original RCT.[71] However, the authors acknowledge that the original trial was not designed to detect such differences between the groups at the time of follow-up. No identified studies reported on differences between ultrasound and selective ultrasound screening groups for outcomes such as unnecessary treatment, psychological harms for parents/guardians and affected cases, or additional treatment-related harms.

4.3.7 Quality assessment

The ROBIS tool was used to assess Pandey et al. 2021, Laborie et al. 2023, and Cheok et al. 2023.[6, 13, 70] For each of the three systematic reviews evaluated using the tool, no concerns were raised in phase one of the assessment. For phase two, areas such as study eligibility criteria, identification and selection of studies, and data collection and study appraisal, did not raise significant issues for any of the included systematic reviews. However, concerns were present for all three reviews

with regards to synthesis and findings. All three studies failed to adequately address the large amount of clinical heterogeneity (for example, differences in the definition of late DDH, the experience of examiners, treatment thresholds, and care pathways) that existed between the primary studies included in each of their respective reviews. It is plausible that this clinical heterogeneity contributed to uncertainty observed in the treatment effect. In phase three, which assesses the overall risk of bias in the review, the risk in each of the systematic reviews was judged to be unclear. Results of the quality assessment are presented in Table 4.4.

Table 4.4 Summary of quality assessment using ROBIS tool†

Review	ROBIS phase 2				ROBIS phase 3
	Study eligibility criteria	Identification and selection of studies	Data collection and quality appraisal	Synthesis and findings	Risk of bias in the review
Cheok et al. 2023[70]	Low risk	Low risk	Low risk	High risk	Unclear
Laborie et al. 2023[13]	Low risk	Low risk	Low risk	High risk	Unclear
Pandey et al. 2021[6]	Low risk	Low risk	Low risk	High risk	Unclear

† Results for phase one (assessing relevance) are not shown, as reviews that did not meet the pre-specified inclusion criteria were excluded during study selection.

4.4 Discussion

The aim of this chapter was to investigate the clinical effectiveness of universal ultrasound screening relative to selective ultrasound screening. The evidence base was limited, with four studies identified that directly compared universal ultrasound screening to selective ultrasound screening. While eight systematic reviews were identified, most of the primary literature included in these systematic reviews consisted of single arm studies (either universal ultrasound screening, selective ultrasound screening or screening using clinical examination only) and were therefore not comparative in nature.

While each of the systematic reviews identified aimed to answer a unique question or set of questions, and included different studies, in general, the majority of systematic reviews reported that there was insufficient evidence to suggest that universal ultrasound screening would result in additional clinical benefits when compared to selective ultrasound screening.[5, 8, 14, 28, 70] Two systematic reviews specifically highlighted the potential association between universal ultrasound screening and unnecessary treatment given the potential for spontaneous resolution of DDH.[5, 8] Only one of the eight systematic reviews identified concluded that there was evidence to support universal ultrasound screening. However, the authors also noted that its introduction may be impractical as a result of challenges associated with implementation.[6] Similarly, another systematic review noted that, as a result of potential logistical challenges, the optimal screening strategy should be determined at a country level.[7]

As outlined in the National Screening Advisory Committee (NSAC) criteria for appraising the viability, effectiveness and appropriateness of a screening programme, to be effective screening for DDH should identify cases earlier than they would have been identified in the absence of a universal screening programme.[76] Furthermore, early identification should lead to earlier treatment, and earlier treatment should be associated with better outcomes when compared with later treatment. As described in the following text, limited direct evidence was identified to suggest that earlier identification of DDH through universal ultrasound screening is associated with improved clinical outcomes, when compared with a scenario where selective ultrasound screening is already in place.

Evidence from three primary studies[71, 73, 74] reported across two systematic reviews[6, 13] suggested that the incidence of DDH cases undergoing non-surgical treatment was higher in the universal ultrasound screening group than the selective ultrasound screening group. As noted in Chapter 3, there is substantial uncertainty regarding the natural history of DDH. The proportion of DDH cases identified by universal ultrasound screening which may otherwise have resolved spontaneously in the absence of treatment is unclear. Thus, while it may be that more cases of DDH

are being treated with non-surgical interventions in a universally screened cohort compared to a selectively screened cohort, there may be some cases that are being treated unnecessarily.

It may be argued that early detection of DDH could enable less invasive, and potentially more effective, corrective procedures. However, based on the findings of the present review, universal ultrasound screening was not associated with a significant reduction in surgical treatment of DDH when compared with selective ultrasound screening.[71, 73] Consistent with the findings presented in this summary, a meta-analysis of non-comparative studies found that the universal and selective screening groups had a similar rate of surgical procedures in childhood for dysplasia (0.48 (95% CI: 0.07 to 1.13) and 0.49 (95% CI: 0.31 to 0.71) per 1,000 live births, respectively).[70] While direct comparison between single-arm studies may not be appropriate due to the likely presence of substantial clinical heterogeneity, the available evidence suggests that universal screening is not associated with a reduction in requirements for surgical intervention.

Furthermore, there is no internationally recognised 'gold standard' test which can distinguish, in early life, between clinically significant DDH (that is, DDH which will require intervention), and cases that may resolve spontaneously. Diagnosis of DDH is complicated by the evolving anatomy of the hips in early life, which may reduce the clinical utility of universal ultrasound screening in the early life. The authors of one identified systematic review concluded that while the universal ultrasound screening cohort may have had a significantly higher treatment rate overall, the rate among children at high risk of DDH (that is, at least one risk factor and or evidence of clinical hip instability) was not increased.[8] This is an important point to consider, as there is evidence to suggest that additional cases identified through universal ultrasound screening, compared to selective ultrasound screening, may have DDH that is mild in nature and therefore more likely to resolve spontaneously. In one included RCT, more than half of infants identified in the universal screening group were considered at low risk of DDH (that is, no risk factors or evidence of clinical hip instability in the first clinical examination).[71] Identification of these cases may not lead to clinical benefit for the patient, and has the potential to result in unnecessary treatment, as well as exposure to potential harms of treatment. Whether or not universal screening would lead to earlier detection of clinically significant DDH is not known due to the lack of high-quality evidence.

Evidence from three primary studies, reported across two systematic reviews, suggested that there was no statistically significant difference in the incidence of late DDH between universal and selective ultrasound screening strategies. However, interpretation of the evidence is likely highly dependent on the definition of late DDH applied. If ultrasound is performed too early, some infants with transient immature and physiologically unstable hip joints may be diagnosed as positive cases. It is

important to note that in some screening recommendations, including the Irish recommendations, hips that are classified as immature can be followed and reassessed before recommending treatment in order to minimise the risk of unnecessary intervention.[15, 57] Most studies evaluating this outcome rely on the assumption that additional cases identified through universal ultrasound screening would eventually have presented with clinically significant disease requiring treatment, without taking into consideration the potential for spontaneous resolution of DDH in the absence of treatment, as described in Chapter 3.

Concerning the definition of late DDH as described in the systematic reviews as well as the primary literature, the 2023 systematic review carried out by Laborie et al. investigated the potential effect of the varying definitions of late DDH.[13] This analysis showed no significant difference between the rate of late DDH diagnosis when considering a definition of later than three months (0.35 per 1,000 births), versus less than three months (0.62 per 1,000 births).[13] While the results of this investigation are not statistically significant, the direction of the effect may suggest that earlier screening could result in overdiagnosis of DDH. This would be consistent the epidemiological literature described in Chapter 3 which reports a high resolution of cases of DDH over time. It is important to note that the systematic review authors did not distinguish between screening strategies in this analysis (that is, universal and selective screening), and the results were derived from indirect comparisons of universal and selective ultrasound screening groups.

Despite the paucity of high-quality studies comparing universal and selective ultrasound screening, two systematic reviews noted that further good quality research in the area was unlikely, due to the early adoption of ultrasound screening for DDH.[14, 28] Shorter et al. 2011 suggested that, despite concerns regarding harms of excess treatment, given widespread acceptance of ultrasound as a screening tool, the focus of research would likely be on management protocols, rather than diagnosis.[28] Where a screening modality has been implemented at a local or national level there may be reluctance to alter an established programme in the absence of high-quality evidence supporting an alternative screening method. This in turn may have led to a lack of demand and or funding for further research in this area, resulting in unresolved uncertainty. Furthermore, detection of clinically relevant outcomes associated with ultrasound screening (universal or selective) would require long-term follow-up, as poor functional outcomes arising from hip pathology may not manifest for decades. However, studies with very long-term follow-up would be extremely challenging to conduct due to the sample sizes needed to detect low probability events (estimated to be in excess of 100,000 infants),[8] and the high risk of loss to follow-up in the long term. The financial and logistical challenges associated with a long-term, large scale study such as this likely represent a barrier to research. A search of ClinicalTrials.gov did not identify any ongoing RCTs that

have the potential to address uncertainty regarding the relative benefit of a universal ultrasound screening programme.

The harms of screening were not clearly reported in any of the included systematic reviews, owing to a dearth of primary studies reporting on this outcome. In included studies, evidence of potential harms was limited to AVN of the femoral head, with no significant differences found between the universal ultrasound and selective ultrasound screening groups.[28] However, studies were likely underpowered to detect this outcome given the low number of events. Although not quantified in the identified studies, Pandey et al. 2021 noted that potential false positive cases and subsequent unnecessary treatment could translate into an increase in complications related to abduction splinting, such as AVN, femoral nerve palsy, pressure sores, and parental anxiety.[6] Cheok et al. 2023 noted that the risk of such treatment-related harms may be increased in settings where there are limited resources for education and parental support.[70] It is acknowledged that in the context of screening for DDH; however, outcomes such as false positives or overtreatment are difficult to accurately quantify, given that once screen-detected DDH is treated, it cannot be determined if the condition would have resolved spontaneously.

In order to estimate the relative benefit of universal screening compared with selective screening, only studies with a comparator group were considered eligible for inclusion in this review. The relative benefit cannot be deduced from single-arm studies. However, it is acknowledged that identified clinical trials undertaken in the 1990s and early 2000s may not be applicable to the current clinical context. Real-world evidence from national population-based programmes of universal screening can provide valuable descriptive information. While this information cannot be used to ascertain the relative benefits of universal screening versus selective screening, it serves as hypothesis-generating data and enables consideration of what could potentially be observed if a programme were to be introduced. As noted in Chapter 2, universal screening for DDH was implemented in Austria in 1992. A retrospective registry-based study was identified which documented all cases of DDH in Austrian hospitals between 1992 and 2008.[40] This descriptive study reported a statistically significant decrease in hospital admissions for the treatment of DDH (from 9.5 to 3.6 per 1,000 live births, or a 62% decrease) over this time period.[40] Additionally, the number of patients requiring surgery to treat DDH (that is, acetabuloplasty, pelvic osteotomy, triple osteotomy, or periacetabular osteotomy) decreased by 46%, from 1.3 to 0.7 per 1,000 live births. There was no change in the number of open reductions (22%) were performed in children born in countries without ultrasound services for screening for DDH. When those born outside Austria were excluded, there was a decrease in the rate of open reductions. It is important to note that such observed reductions in admissions and requirements for surgery for children with DDH are unlikely to be directly attributable to ultrasound screening alone; rather,

they may have represented the beneficial combined effects of a structured screening programme, including increased awareness and training among healthcare professionals, appropriate resourcing of coordinated care pathways, and mechanisms for reporting and evaluation. Importantly, given the non-randomised, non-comparative nature of such studies, background effects (for example, epidemiological trends and evolution of approaches to identification and management) also cannot be ruled out.

As with all screening interventions, guidance from the American Academy of Orthopaedic Surgeons (AAOS) states that in the case of screening and early intervention programs, potential harms relate to the balance between under and overdiagnosis.[77] Specifically, the authors noted that potential overdiagnosis would lead to increased rates of further evaluation and unnecessary treatment.[77] However, in circumstances where clinical examination only, or a selective screening programme are in place, cases of DDH with normal clinical examination and no risk factors would not be identified, resulting in delayed clinical presentation, with potential progression to deformity.[77] The guidelines further reported deficits in the evidence base relating to the potential emotional impact of detecting a non-apparent musculoskeletal problem in a newborn through screening, as also noted in Shipman et al. 2006.[77] Furthermore, Shorter et al. 2011 noted that the negative psychological impacts are not limited to parents or guardians; late DDH diagnosis and the associated potential complications, including functional impairment and arthritis, have the potential to negatively impact the psychological health of affected patients in the longer term.[28]

Strengths and Limitations

Methodological studies across different disciplines have reported that authors of overviews rarely assess the overlap of primary studies, which has the potential to introduce bias.[68] A strength of the analysis in this report is the exploration of overlap across identified systematic reviews. This exploration allows a concise summary of key evidence without duplication of results from primary studies contained within systematic reviews. In addition, the quality of key systematic reviews was assessed to identify potential sources of bias that may influence the interpretation of the results or conclusions of identified reviews.

However, limitations associated with the evidence synthesis process and underlying evidence warrant careful consideration. For the purposes of this evidence review, only evidence considered in published systematic reviews was considered. Consistent with best practice, the unit of analysis was the systematic review; data extraction and quality appraisal were not undertaken at an individual study level.[78] Our results are therefore dependent on the accuracy and completeness of reporting of the study results by the authors of identified systematic reviews. Given the emphasis on

secondary research, this summary could not capture any relevant primary studies published since the included systematic reviews were undertaken. However, it is unlikely that additional primary studies were not identified by our search method, given that the most recent systematic search in identified reviews was conducted in March 2022. A search for relevant studies published since identified systematic reviews undertaken using scoping methodology did not identify any additional studies relevant to our specific research question; however, a systematic search would be necessary to confirm the absence of new studies with certainty. In addition, given this summary included only evidence comparing universal ultrasound screening and selective ultrasound screening, it comprises only a subset of the available evidence on the overall topic. Nonetheless, this subset represents the best available evidence currently.

Two of the four studies identified in included systematic reviews were retrospective cohort studies. There is potential for bias in these studies, as factors other than the intervention may differ across cohorts, limiting the ability to draw definitive conclusions. For example, where the groups compared were studied during different time periods, it is difficult to determine whether the results obtained were attributable to differences in the mode of screening (that is universal ultrasound or selective ultrasound), or if they are as a result of factors such as data quality or changes in clinical practice over time (for example, increased awareness or adoption of an observation period prior to intervention).

Variation in elements of the screening pathways, such as the timing of ultrasound, the ultrasound technique used, experience of the examiners, thresholds for treatment, and the care pathway, makes comparison of results reported across studies difficult. Such clinical heterogeneity presents challenges for comparison of results across studies. For example, while both of the identified RCTs aimed to carry out ultrasound examination in the first 72 hours of life, one of the identified observational studies reported that participants were invited to attend for ultrasound within 'first 6 weeks of life'. Additionally, the ultrasound technique in the studies varied; two studies used the Graf technique,[71, 74] while the Harcke technique[72] and the Morin-Terjesen technique[73] were used in the remaining two studies. It is likely that differences in techniques used had minimal impact on the outcomes, given the previously noted correlation in results of hip assessment between the techniques (Chapter 2).[12] The definition of late DDH also varied between studies, and therefore the reported results for this outcome are subject to a high degree of uncertainty. These limitations are intrinsically linked as variability in the time to screen and the definition of late DDH, may directly contribute to the rate of late DDH being reported in each study. Therefore, the results of this outcome are subject to a large degree of uncertainty.

Conclusion

Variability in study design and reporting, coupled with a limited evidence base, present challenges for interpretation of the evidence. There is considerable uncertainty surrounding the potential benefit of universal, relative to selective ultrasound screening for DDH. However, overall, the results suggest that compared with selective ultrasound screening, universal ultrasound screening may be associated with an increase in non-surgical treatment of DDH, without evidence of a subsequent reduction in the incidence of late DDH, or a reduction in requirements for surgical intervention. No evidence was identified comparing universal and selective screening strategies on long-term clinical outcomes. In the Irish context, the relative benefit of implementing a universal ultrasound screening programme would be highly dependent on the effectiveness of the current selective screening strategy in identifying cases with clinically significant disease.

5 Discussion

5.1 Introduction

In March 2023, at the request of the National Screening Advisory Committee (NSAC), HIQA agreed to undertake an evidence review of universal ultrasound screening for developmental dysplasia of the hip (DDH) in infants in Ireland. The purpose of this discussion chapter is to summarise the key findings of the evidence review, contextualise these findings relative to other reviews completed internationally, and present the strengths and limitations of the review overall.

5.2 Summary of key findings

The NSAC have produced a list of 20 criteria for appraising the viability, effectiveness and appropriateness of a screening programme (Appendix 1). These are grouped under five headings: the condition, the screening method, the intervention, the screening programme, and implementation criteria.[76] This review explored evidence for four of the five headings, namely: the condition, the screening method, the intervention and the screening programme. Implementation criteria were outside of the scope of the evidence review.

5.2.1 The condition

As set out in the NSAC criteria, the condition should be an important health problem.[76] Additionally, the criteria note that the epidemiology, incidence, prevalence and natural history of the condition should be understood. As highlighted throughout this report, there is no consensus on a precise definition of DDH, which can vary from mild hip instability to full hip dislocation.[60] This wide range makes understanding the epidemiology of the condition challenging. Additionally, a limited amount of evidence was identified that reported on the aetiology, natural history, and incidence of the condition (see Chapter 3). While there are several known risk factors for DDH, most identified DDH cases have no known risk factors, making identification based on risk factors challenging. It is however important to note that in most countries (see Chapter 2), all newborns receive a physical examination to check for signs of DDH, which represents a form of universal screening. Nonetheless, potential cases of DDH may be missed by clinical examination alone (Chapter 2), which underlies the rationale for ultrasound-based screening.[2-4]

Reports on the natural history of DDH are variable. As the condition is broad, it is unclear how different morphologies of the condition develop over time. Specifically, hip instability can be age dependent, and decreases in the first weeks of life with increases in muscle tone.[19] Up to 90% of mild forms of dysplasia detected in infants resolve on their own in the first six to eight weeks of life, without the need for treatment.[19-21, 27, 63] Clinical examination of the hips in the first few weeks of life

may therefore identify hip instability that would resolve, without the need for intervention. Conversely, some cases of DDH will present later in life, with no abnormalities detected in infancy either by clinical or ultrasound screening.[10]

While many cases of infant hip instability will resolve on their own, there are cases of DDH that do not resolve and can lead to long-term complications (see Chapter 3). DDH can lead to osteoarthritis; it is a significant source of overall requirement for total hip replacements, and is a common cause of total hip replacements in young adults.[27, 63, 65] Therefore, these untreated cases can be an important health problem. A screening programme using ultrasound may reduce the number of late identified cases of DDH, and help improve outcomes with early treatment.

The incidence of DDH is not well known or reported; where reported, the incidence varies (see Chapter 3). This variation may be true variation due to geographic differences but may also be due to differences in how incidence is measured and identified. For instance, there is variation in terms of what is classified as DDH, with some reports of DDH incidence including a broad definition, such as minor hip instability identified in the neonatal examination, while others included only those identified with subluxated hips or more severe dislocations. Studies using a broader definition of DDH are expected to have higher incidence estimates than those that applied narrower definitions. Additionally, the reported incidence differs depending on how the cases of DDH were identified. For example, in regions with universal ultrasound screening for DDH, the reported incidence is expected to be much higher than incidence reported in regions without screening, where only DDH cases that present clinically are identified. However, it is important to note that the higher incidence from the screening programmes may include more minor cases of DDH that would never have presented clinically, and would have potentially resolved without treatment.

It is challenging to compare the incidence of DDH reported in Ireland to internationally reported incidence, given the differences in screening methods, DDH definitions, and the level of implementation (for example, local implementation or national screening programme). Available Irish data reported from a single centre with selective ultrasound diagnostics, from a period after the publication of the 2017 selective ultrasound screening recommendations, indicate an estimated incidence of 31 per 1,000 births (see Chapter 3). This is higher than the incidence reported from Austria (8 per 1,000), which is based on several years of data from a universal ultrasound screening programme.[66] The incidence reported from the Irish centre may; however, not be representative of the rest of the country.

5.2.2 The screening method

NSAC criteria for 'the screening method' state that the method should be simple, safe, precise, reliable, and validated.[76] In this review, the screening method is

ultrasound screening of the hip for DDH in infants. While the Graf technique is the ultrasound method used in Ireland and most of Europe, and was the primary method of ultrasound reported in this review, other ultrasound methods are used internationally. There is evidence that supports good correlation between the results obtained from hip assessment based on other techniques compared to the Graf technique.[12] Therefore the inclusion of results based on techniques other than the Graf technique is not expected to majorly impact results in this report.

Results from analyses of Graf ultrasound for DDH demonstrate that the sensitivity and specificity are high (93% and 97% respectively). With regard to sensitivity, the possibility of some cases of DDH going undetected can be due to the training and experience of the operator, or due to the condition itself (that is, there are no signs at the time of the ultrasound) (see Chapter 2).[26] Training in Graf technique is likely to be required as part of an ultrasound screening programme for DDH to ensure reproducibility and reliability of the results and to reduce the risk of undetected cases or unnecessary treatment. With regard to specificity, it is important to highlight that while high values of specificity have been reported, these are likely calculated based on all cases of DDH, without knowledge of whether the cases would be clinically significant (that is, requiring intervention). There is no tool that can accurately predict whether identified cases of DDH would resolve spontaneously or require treatment.

Timing of screening

The timing of screening needs to be considered in any ultrasound screening programme given that many cases of minor hip instability in newborns resolve spontaneously (see Chapter 3). If the ultrasound is done too early, minor instability that would have resolved within a few weeks would be detected, and subsequently treated. It is unclear whether these cases should be labelled as 'false positives' as at the time of screening the hip would have been identified as having an abnormality. However, they would not have subsequently required treatment. There are no international guidelines outlining whether or not there should be an observation period to monitor for improvement prior to treatment initiation. As noted previously, the Irish recommendations for a selective ultrasound screening programme recommend that hips classified as immature should be followed and reassessed before recommending treatment in order to minimise the risk of unnecessary intervention.

Use of radiography following ultrasound

Data from one study at the University Hospital Waterford (UHW) suggest that some cases of DDH may be missed on early ultrasound. At this centre, infants referred for ultrasound of the hip who have normal hips on imaging are examined again and receive radiography at six months of age. UHW found that 8% of infants who

received radiography at six months had abnormal hips, which were not detected at the time of the selective ultrasound screening performed by six weeks of age.[38] This finding has led to the implementation of the six-month radiography follow-up as standard practice at this centre. It is important to note the limitations of the study that informed this practice, and to highlight that this is unlikely the standard of care in the rest of the country. The study that informed practice at UHW was descriptive and did not include a comparator arm; it is therefore not possible to determine the outcomes of infants who received the scan at six months of age had they not received the additional X-ray. Additionally, it was unclear whether the cases that were not detected at the ultrasound were due to the ultrasound technique itself, or if there truly were no signs at the time.

An unpublished review, performed by members of the group that published the recommendations for selective ultrasound screening programme in Ireland, aimed to explore whether pelvic X-ray is necessary for follow-up of infants with risk factors for DDH that had normal ultrasound at six weeks of age.[16] Of the 13 studies identified, nine recommended discontinuing routine X-rays at four to six months for all infants with risk factors who have a normal ultrasound at six to 10 weeks. The authors of this review noted that there was no international consensus on whether or not to perform X-ray. It is important to acknowledge the risks associated with radiography of the pelvic area in infants. In the context of a selective screening programme, consideration may be required for a screening algorithm that includes follow-up of those with risk factors for DDH but with normal findings at the initial ultrasound. It is important also to note that use of medical radiological procedures in the context of screening is subject to regulations as outlined in SI.256 of 2018.[79] HIQA is the competent authority with responsibility for inspecting and enforcing these regulations. These regulations indicate that if X-ray examination is being performed on an asymptomatic individual for the early detection of disease, this X-ray exposure should be as part of a screening programme or the individual justification for that exposure needs to be documented, in consultation with the referrer.

It is worth noting that, overall, there is no gold standard for screening of the hips for DDH. In practice, and given the changing natural history of the condition especially in early life, there is no diagnostic test that can confirm whether or not a case of hip instability detected in infancy would require treatment, or would resolve on its own.

5.2.3 The intervention

NSAC criteria relating to the intervention state that there should be an effective intervention for patients identified through screening, with evidence that intervention at a pre-symptomatic phase leads to better outcomes for the screened individual compared with usual care.[76] Another NSAC criterion states that there should be agreed evidence-based policies covering which individuals should be offered

interventions and the appropriate intervention to be offered. No internationally accepted guidelines for the treatment of DDH were identified in this evidence review. The treatment options for DDH depend primarily on the age of the patient at treatment initiation (see Chapter 3).[1, 2] Overall, treatment is less invasive when initiated earlier, with the lowest risk of complications in those treated with abduction splinting, such as the Pavlik harness. Complication risks, most frequently avascular necrosis (AVN) of the femoral head, increase with the use of a solid cast used with closed reductions. Finally, if the condition is treated with an open reduction, there may be several surgical risks, such as risk of infections or complications of anaesthesia. Therefore, there would be perceived benefit in identification of DDH earlier, where the least invasive treatment, and also the treatment less likely to result in complications, can be used. This aligns with recommendations for early diagnosis and treatment of DDH published in 2020 by a multidisciplinary group of experts, which included paediatricians, radiologists and paediatric orthopaedics.[2] It is unclear; however, whether universal screening would result in a clinical benefit to patients, compared with selective screening, as evidence from Chapter 4 suggests that the incidence of late DDH and surgical intervention did not differ between these groups.

Benefits achieved through screening must be balanced with the risk of harms. The true rate of spontaneous resolution of the hip instability is difficult to discern, given that many of the cases identified by a screening programme for DDH (clinical screening and or ultrasound screening) may have started treatment before the hip instability has the opportunity to resolve. While early initiated treatment is generally minimally invasive, there is still a risk of potential harms such as AVN of the femoral head, femoral nerve palsy, pressure sores, and parental anxiety. One systematic review found that 10% of children who underwent closed reduction experienced AVN.[34] While AVN is thought to be less common with harnessing compared to closed reduction, the risk of occurrence cannot be ignored, given the severity of the complications. Therefore some cases of DDH may be at risk of complications when they may not have truly received any benefit from treatment. It is important to note that in identified studies, evidence was retrieved for AVN of the femoral head only. In the absence of studies powered to assess the potential harms of universal versus selective ultrasound screening, the risk-benefit balance is unclear.

5.2.4 The screening programme

Ultrasound screening programmes for DDH can either be selective or universal (see Chapter 2). A universal ultrasound screening programme would lead to all infants being offered ultrasound of the hips, compared to a selective screening programme, in which only those that meet predefined clinical findings and or risk factors are referred for ultrasound. As there are already recommendations for a selective ultrasound screening programme for Ireland, this evidence review aimed to provide

an overview of the clinical effectiveness of universal ultrasound screening for DDH relative to selective ultrasound screening.

Clinical effectiveness

The rationale behind a universal screening programme is that it may allow for the detection of DDH cases that would have otherwise been missed with selective ultrasound or clinical examination alone (that is, those without physical symptoms noted on examination and or risk factors), potentially reducing the rates of late presenting DDH and associated complications (see Chapter 2). There are concerns that the clinical examination screening for DDH, which may lead to referral for selective ultrasound screening, is not sensitive enough, leading to potential undetected cases of DDH.[2-4] Additionally, as many infants who do develop DDH do not have known risk factors, there is the concern that using a selective ultrasound screening programme which refers for screening based on certain risk factors will miss many cases of DDH. However, it is important to note that it is unclear how many of the undetected cases would be clinically significant (that is, need treatment).

NSAC criteria for a screening programme note that there should be evidence from high-quality randomised controlled trials (RCTs) that the screening programme is effective in reducing mortality or morbidity.[76] Limited direct evidence was identified that would suggest that earlier identification of DDH through universal ultrasound screening, when compared with selective ultrasound screening, is associated with improved clinical outcomes (see Chapter 4). While evidence identified in the literature suggests that there may be more infants treated with abduction splinting where there is universal ultrasound screening, it is not clear whether this increased treatment rate is truly beneficial. If all infants are being screened by ultrasound, it is likely that many cases of hip instability that would have otherwise never been detected will end up being treated, and potential complications from treatment cannot be ignored. However, this must be balanced against the potential benefit of early detection and treatment of cases that are clinically significant. It is important to note that despite the higher treatment rate for universal ultrasound compared to selective, there was no increase in the rate of AVN in one RCT.[71, 75] However, this study was underpowered for this outcome, limiting the ability to draw definitive conclusions. Overall, it was unclear from the literature identified in this report whether there was a benefit of universal screening compared to selective screening in terms of a reduction of late detected DDH, surgical treatment rates, or long-term outcomes.[6, 13, 28, 70]

Ideally, evidence comparing universal ultrasound screening to selective ultrasound screening would come from studies of population-based screening programmes (in which there would be appropriate governance in place), rather than smaller scale

studies that may be underpowered to detect differences in outcomes and which may not represent the full features (such as appropriate governance) of a dedicated programme. While two RCTs were identified in this evidence review which compared universal ultrasound screening to selective ultrasound screening, both of these studies are potentially outdated (1994 and 2002) (see Chapter 4). The feasibility of conducting further RCTs has been questioned, largely due to concerns regarding the large sample sizes required for sufficient power.[8] Additionally, conducting sufficiently large RCTs may be unlikely due to the potential for legal and ethical concerns of missing cases of DDH, as well as the lack of feasibility of detecting long-term outcomes (such as need for hip replacement in later life).

The other two primary studies which informed the results of the clinical effectiveness of universal ultrasound screening relative to selective ultrasound screening were retrospective cohort studies conducted across different time periods; this limits the comparability of the universal and selective ultrasound screening groups. In the event that selective ultrasound screening is in place prior to the implementation of universal ultrasound screening, it is likely that awareness of the condition will have improved over time (by nature of being aware of the selective screening, and also changes in clinical practice). Therefore, the potential benefits of introducing universal ultrasound screening, when selective ultrasound screening is already in place, are unclear. A comparison to a situation with no ultrasound screening was not addressed in this evidence review, as this is not relevant to the Irish context. While there is no formal screening programme in the country, all infants access physical examination, including investigation of hip instability, as part of developmental checks. In addition, some infants undergo ultrasound depending on the outcome of physical examination or the presence of risk factors (that is, selective screening); a national audit of centres performing ultrasound to assess for DDH in 2021 and 2022 found that 18.5% of all infants born are referred for ultrasound of the hips. However, implementation of selective screening is not necessarily standardised across the country due to the absence of a formal screening programme.

International practice and experience

The absence of internationally agreed best practice is a notable challenge for ultrasound screening programmes for DDH. In most countries identified in the scoping search for international practice (Chapter 2), there is selective ultrasound screening for DDH in place. However, the referral criteria (suspicion or certain risk factors) for ultrasound were not clearly reported. For countries that did report the risk factors, there is variability in practice between countries. In one country, Switzerland, the universal screening programme for DDH was replaced by a selective screening programme due to insufficient evidence to support its continuation.[22] Additionally, no screening programme was found for two of the countries, Finland and Portugal, in the scoping search. It is likely that these countries do in fact have

some sort of screening, whether it be clinical or by risk factors. However, as previously highlighted, there is a lack of reporting and formalisation of screening for DDH internationally.

Universal ultrasound has, however, been in place in Austria since 1992 and in Germany since 1996.[40] It is unclear whether an evidence assessment was undertaken to inform the implementation of these programmes.[40, 46] While studies from countries with universal screening can help in understanding of the epidemiology of DDH in a universally ultrasound screened population and serve to describe changes in outcomes for DDH following the introduction of universal ultrasound screening, these are descriptive studies and therefore do not allow conclusions to be made regarding whether the introduction of universal screening, versus selective, has a beneficial effect on patient outcomes.

Acceptability and feasibility

A further NSAC criterion about the screening programme notes that there should be evidence that the complete screening programme is acceptable and can be implemented.[80] In Ireland, while there are recommendations for a selective ultrasound screening programme, the recommendations have not been implemented as a screening programme to date.[16] Rather, the recommendations are included as part of the diagnostic pathways for DDH, referred to as a targeted clinical diagnostics programme. At the time these recommendations were developed, implementation of a universal programme was not considered feasible. A selective programme was recommended with a view to building capacity for ultrasound across the country, training radiographers and radiologists in the Graf ultrasound technique, and bringing awareness of DDH in general to healthcare practitioners.

Within the current practice across the country, there is evidence to suggest that there may be variation in the implementation of the proposed care pathway (see Chapter 2). For example, publications from the UHW note that infants referred for selective ultrasound screening whose imaging does not show signs of DDH, receive follow-up radiography at six months of age. This differs from the recommendations in that infants with negative ultrasound are recommended to be returned to routine care, with no follow-up required. This deviation from the recommendations may demonstrate that some clinicians do not perceive the current recommendations to be acceptable or sufficient, and therefore feel the need to adapt them. Of note, for this evidence review, the screening algorithm in place for only one of the hospitals in Ireland was identified from publications. The practices in place in other hospitals across the country are unclear.

Organisational and programme factors

Various organisational factors may impact the effectiveness of screening practices. The authors of one of the studies included in the evidence comparing universal ultrasound to selective ultrasound noted that the cases of late DDH identified in the universal ultrasound screening group were not due to missed cases from the screening test itself, but rather from issues with the care pathways, such as failure to attend appointments.[74] This may demonstrate shortcomings of the programme itself, rather than the screening method. When screening happens several weeks after birth, the baby is seemingly healthy, and families have entered into a routine, engagement with follow-up appointments might be more challenging compared to screening that happens shortly after birth (for example, newborn bloodspot screening). While it would be likely that these issues could be common to both a universal and selective ultrasound screening programme, it may be amplified in the universal programme, as there would be more infants entering the pathways, and parents may not perceive the benefit of attending the appointments when no risk factors or symptoms have been identified. This underlines the importance of a programme incorporating follow-up, data collection, and communication strategies, to ensure the programme is delivered as specified. Appropriate governance is important for any screening programme, including monitoring of programme performance. An example of performance measures within DDH screening may be found in the National Health Service (NHS), where key performance indicators for the timeline of the ultrasound scan of the hips for DDH have been introduced.[81]

As noted by the World Health Organization (WHO), a screening programme is not just a single test but a process and a pathway.[59] This process begins by identifying those eligible for screening, includes diagnosis and treatment, and concludes with reporting of outcomes and evaluation of the screening programme. Similarly, the UK National Screening Committee notes that a nationally managed screening programme is most safely undertaken with the support of an information system that identifies participants and manages individuals through the pathway, and that effectiveness should be assessed through recording of health outcomes.[55] The Australian Population Based Screening Framework, devised by the Department of Health, notes key principles for the implementation and management of screening programmes.[56] These include consideration towards an agreed quality management plan and a continuous quality improvement framework, clearly defined governance and management structures, and appropriate monitoring, evaluation and review.

Cost effectiveness and resourcing factors

It is also important to note there may be logistical and resourcing challenges associated with implementing any screening programme. Organisational issues around delivery of screening represent an important consideration, albeit not explored in depth within the current review. Furthermore, it is necessary to understand whether the programme would be a good use of resources; cost

effectiveness and budget impact represent important factors in decision-making regarding introduction of a screening programme, selective or universal. While not examined within the present review, there may be challenges associated with ultrasound capacity and resources available to finance the programme.[57]

5.3 Findings relative to international assessments

A limited number of regional and national organisations have completed assessments regarding screening for DDH in infants; these include one assessment conducted in Canada (2001),[43] two from the US (the US Preventive Services Task Force, USPSTF, 2006; the American Academy of Orthopaedic Surgeons, AAOS, 2022),[60, 77, 82] and assessments in France (2013)[83] and Spain (2016).[84] Authorities from France, Spain, and the AAOS in the US all recommended against a universal ultrasound screening programme, and supported a selective ultrasound screening programme. The recommendations from the USPSTF indicated that there was insufficient evidence to provide a recommendation for screening for DDH.[60, 82] The authority from Canada recommended against either universal ultrasound, or selective ultrasound based on risk factors, screening for DDH.[43]

The reports from France, Spain and the AAOS all recommended against a universal ultrasound screening programme, and supported a selective ultrasound screening programme.[77, 83, 84] Consistent with most of the evidence identified in this report, the reports noted that while an ultrasound screening programme would increase the number of detected cases of DDH, it is not clear how many of these cases would have resolved without treatment. Consistent with the evidence presented in Chapter 4, the reports from Spain and the AAOS noted that treatment rates increase with universal ultrasound screening; however, there is no evidence to indicate a decrease in the number of late-detected cases of DDH or improvement in outcomes for treated cases. The three reports highlighted the risk of unnecessary treatments with a universal ultrasound screening programme. However, the report from France highlighted that for cases that are detected early, and which would have required treatment, treatment options are less invasive than if the condition is detected later. Due to the limitations noted in a universal ultrasound screening programme, and the potential for missing clinically relevant cases from a clinical examination alone, the three reports recommended a selective screening programme for infants with risk factors for DDH. Specifically, the AAOS recommended that in infants with risk factors for DDH but with a normal clinical examination, an ultrasound can be performed between two to six weeks of age, or radiography can be performed from four months of age. The AAOS also highlighted that there was limited evidence to support using ultrasound to guide the decision to initiate treatment with a brace, rather than wait and watch for spontaneous resolution in those less than six weeks of age with hip instability on clinical examination.

The recommendations from the USPSTF, published in 2006, indicated that there was insufficient evidence to provide a recommendation for screening for DDH.[60, 82] The recommendations were based on a 2006 systematic review.[8] Similar to the present evidence review, the report noted that the pathophysiology and natural history of DDH are poorly understood. Overall, the report concluded that it was not possible to assess the balance of benefits and harms of screening for DDH. As of October 2023, the USPSTF website indicates a decision not to review the evidence and update its recommendations for this topic, though acknowledges that the previous evidence review and recommendation may contain information that is outdated.[85]

The report from Canada, published in 2001, found broadly similar findings to the present evidence review.[43] Overall, the report recommended serial clinical examination by a trained clinician. The report recommended against either universal or selective ultrasound screening based on risk factors. The recommendation against universal ultrasound screening was made on the basis of universal screening increasing the intervention rate, the potential for false positives, and given no decrease in the rates of late DDH or requirement for surgery. The recommendation against selective ultrasound screening was made on the basis that most infants with DDH do not have risk factors, and selective ultrasound screening does not lead to a decrease in the rate of requirement for surgery.

5.4 Strengths and limitations

The findings of this evidence review should be considered in light of its overall strengths and limitations. In terms of strengths, a robust approach to the evidence review was employed with publication of a protocol for the review, and the establishment of an Expert Advisory Group (EAG) comprising a broad range of national key stakeholders to provide expert input and advice. Although a de novo systematic review was not conducted to identify evidence for universal ultrasound for DDH compared to selective ultrasound, a comprehensive summary of systematic reviews was undertaken.

However, some important limitations exist in relation to the currently available evidence and the methodological approaches applied in this evidence review, which must be considered in the context of the overall findings.

The aim of this report was to provide an overview of the key pertinent evidence rather than a comprehensive examination of the full extent of the literature. The majority of the evidence described was derived from international literature and there was limited national evidence available for this report. Scarce data were available in relation to selective screening as is currently in place in Ireland and nationally centralised data were unavailable. Therefore, it is challenging to estimate the incidence, referral rates, and outcomes of DDH across the country. However, the

review focused on a number of studies and countries considered most applicable to the Irish context, potentially mitigating these limitations. Furthermore, while the algorithm for the selective ultrasound screening practice in one hospital in Ireland was outlined in a recent publication, it is not clear what practice has been implemented in other hospitals across the country. Therefore, the applicability to the Irish context is unclear.

A summary of reviews was undertaken, rather than a de novo systematic review or a structured overview of reviews to explore the outcomes of universal ultrasound screening for DDH relative to selective ultrasound screening. As this summary only included evidence that compared universal ultrasound screening to selective ultrasound screening, it presents only a subset of the available evidence, and does not compare to a situation of no ultrasound screening or clinical screening only. There are further limitations associated with this evidence synthesis approach as it may not capture all relevant primary studies, and it would not capture primary studies published since the search dates of the included systematic reviews. However, given how recently the identified systematic reviews were published (literature searches performed up to March 2022), and the consistency in conclusions across all identified reviews, it is highly likely that a de novo systematic review or overview of reviews would have come to the same conclusions.

5.5 Conclusion

The purpose of this report was to provide an evidence review of universal ultrasound screening for developmental dysplasia of the hip in infants in Ireland. Overall, the information included in this evidence review was limited for several reasons. These included the lack of international guidelines for screening for DDH and the limited evidence available for the natural history, aetiology and epidemiology of the condition. Limited evidence was found to determine whether universal ultrasound screening for DDH compared to selective ultrasound screening leads to improved functional outcomes, decreased need for surgical interventions, and reduced harms. These knowledge gaps combine to produce significant uncertainty regarding the benefit of introducing a universal ultrasound screening programme over the current recommendations for selective ultrasound screening in Ireland.

Given the variable natural history of DDH, with a high rate of spontaneous resolution of hip instability, and the potential risk of serious complications from treatment, the potential benefits of earlier diagnosis, as may be achieved through widespread screening, need to be weighed against the potential harms of unnecessary treatment. In the absence of nationally representative data regarding outcomes of current selective ultrasound practices in Ireland, the relative benefit of a universal ultrasound screening programme is uncertain.

The 2017 recommendations for selective ultrasound screening in Ireland have not been implemented as part of a formal national screening programme, and current practice is therefore not supported by the governance, end-to-end care, quality assurance, and monitoring of outcomes that would be associated with such a programme. Further understanding of current practice and barriers to following the recommendations across Ireland may facilitate the successful implementation of a formal screening programme.

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Appendices

Appendix 1 NSAC criteria for appraising the viability, effectiveness and appropriateness of a screening programme

Criterion No.	NSAC Grouping	Criterion
1	The Condition	The condition should be an important health problem. The epidemiology, incidence, prevalence and natural history of the condition should be understood, including development from latent to declared disease and or there should be robust evidence about the association between the risk or disease marker and serious or treatable disease.
2		All the cost-effective primary prevention interventions should have been implemented as far as practicable.
3		If the carriers of a mutation are identified as a result of screening, the natural history of people with this status should be understood. The psychological implications should be considered, and the necessary psychological supports should be in place.
4	The Screening Method	The screening method should be, as far as is practicable: a) simple b) safe c) precise d) reliable e) validated.
5		The distribution of screening values in the target population should be assessed and suitable cut-off levels/measurements defined and agreed by the applicant.
6		The screening process should be acceptable to the target population.
7		There should be an agreed policy on the further diagnostic investigation of individuals with a positive screening result and on the choices available to those individuals.
8		If screening is for a particular mutation(s) or set of genetic variants, the method for their selection should be kept under review.
9	The Intervention	There should be an effective intervention for patients identified through screening, with evidence that intervention at a pre-symptomatic phase leads to better outcomes for the screened individual compared with usual care.
10		There should be agreed evidence-based policies covering which individuals should be offered interventions and the appropriate intervention to be offered.
11	The Screening Programme	Ideally there should be evidence from high quality randomised controlled trials that the screening programme is effective in reducing mortality or morbidity. Where screening is aimed solely at providing information to allow the person being screened to make an informed choice, there must be evidence from high quality trials that the test accurately measures risk. The information that is provided about the test and its outcome must be of value and readily understood by the individual being screened.
12		There should be evidence that the complete screening programme (test, diagnostic procedures, treatment/ intervention) is acceptable and can be implemented.

Criterion No.	NSAC Grouping	Criterion
13		The benefit gained by populations and individuals from the screening programme should outweigh the harms. The public should be informed of these harms and of their associated undesirable physical and psychological consequences.
14		The opportunity cost of the screening programme (including testing, diagnosis and treatment, administration, training and quality assurance) should be economically balanced in relation to expenditure on medical care as a whole (value for money). Assessment against these criteria should have regard to evidence from cost benefit and or cost effectiveness analyses and have regard to the effective use of available resource.
15	Implementation Criteria	Clinical management of the condition and patient outcomes should be in place before a screening programme is initiated.
16		Adequate staffing and facilities for testing, diagnosis, treatment and programme management should be available prior to the commencement of the screening programme.
17		All other options for managing the condition should have been considered (such as improving treatment or providing other services), to ensure that no more cost-effective intervention could be introduced, or current interventions increased within the resources available.
18		There should be a plan for managing and monitoring the screening programme against an agreed set of quality assurance standards. This should include monitoring performance against different sub-groupings in the population.
19		The potential benefits and harms of screening, investigation, preventative intervention or treatment, should be made available and explained to the eligible participants to assist them in making an informed choice. There should be a clear system of communication incorporated into each screening programme to ensure patients are kept aware of any developments in their case.
20		Decisions about commencing, expanding or ceasing a programme should be based on scientifically validated evidence.

Key: NSAC – National Screening Advisory Committee.

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