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ULTRASOUND

Reliability of 2D and 3D ultrasound for infant hip dysplasia in the hands of novice users

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Abstract

Purpose Developmental dysplasia of the hip (DDH) diagnosis by two-dimensional ultrasound (2DUS) can have poor inter-rater reliability. 3D ultrasound (3DUS) may be more reliably performed, particularly by novice users. We compared intra- and inter-rater reliability between expert and novice operators performing 2DUS and 3DUS for DDH.

Materials and methods Infants with suspected DDH were assessed with 2DUS and 3DUS. Novice operators had 1.5 h of training and Experts had 5–15 years' experience. Images included two 2DUS static and two 3DUS sweep images per operator. Image quality was assessed by 5-point system (yes/no: full femoral head; full acetabular roof; horizontal iliac wing; os ischium; absent motion/artifact). 2DUS indices (alpha angle, coverage) were measured centrally by a blinded reader with 2 years DDH US experience. 3DUS was post-processed by semi-automated custom software generating acetabular surface models, indices and estimated probability of DDH. Gold-standard diagnosis of each hip as normal, borderline or dysplastic was based on radiologist review of expert 2DUS.

Results Thirty infants, mean age 10.8 weeks were enrolled. Quality scores were 2.7±1.2 Novice versus 4.9±0.3 Expert for 2DUS ($p = 0.04$), and 4.2±1.0 Novice versus 4.9±0.3 Expert for 3DUS ($p = 0.99$). Inter-rater reliability was poor for 2DUS (ICC=0.10 for alpha angle, 0.04 for acetabular coverage) and moderate to high for 3DUS (ICC=0.73-0.83 for alpha angle, 0.55 for acetabular coverage). Intra-rater reliability and diagnostic accuracy was higher for 3DUS than 2DUS.

Conclusion Novice operators can perform 3DUS for DDH with reliability and accuracy approaching expert sonographers. Novices perform 2DUS with poor reliability and accuracy.

Key Points

- *Novice/expert inter-rater reliability improved from poor with 2DUS to moderate/high with 3DUS.*
- *Novice operators using 3DUS correctly classified 57/58 (98%) of infant hips.*
- *DDH can be reliably assessed by novice operators using 3DUS.*

Keywords Diagnostic imaging · Hip dislocation, congenital · Observer variation · Reproducibility of results · Ultrasonography

Abbreviations and Acronyms

2DUS	Two-dimensional ultrasound
2Dα	2D Alpha Angle
3DUS	Three-dimensional ultrasound
ACR	American College of Radiology
DDH	Developmental dysplasia of the hip
ICC	Intraclass correlation coefficients
POCUS	Point-of-care ultrasound

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Introduction

Developmental dysplasia of the hip (DDH) is a treatable condition with a prevalence varying from 1.6 to 28.5 cases per 1,000 live births depending on the study population [1]. If detected early, treatment with a Pavlik harness is curative in 85–95% of cases [2]. However, if missed at early screening, DDH requires costly and challenging surgical management, and risks the lifelong morbidity of hip osteoarthritis [3].

The current preferred imaging modality for the diagnosis of DDH is two-dimensional ultrasound (2DUS). In most nations, screening with 2DUS is not universal, but rather is performed selectively with the presence of risk factors or suspicious clinical examination. Unfortunately, given the limited sensitivity (60%) of the Barlow and Ortolani clinical manoeuvres for

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assessment of hip instability in the infant [4], this approach inevitably misses cases of DDH. In fact, some of the most at-risk patients who may lack easy access to tertiary care are easily missed by this type of referral-based screening. For example, infants in remote rural area, often present later for diagnosis and treatment of DDH in comparison to inner city infants [5]. In many cases this is too late for brace management alone to be effective [6].

Resistance to universal screening through imaging of infants is justifiable based on existing evidence [1], and is primarily due to limitations inherent in 2DUS. Conventional 2DUS is difficult to perform even for expert examiners and has many false-positive or indeterminate results, requiring follow-up examinations, which are costly for healthcare systems and stressful for families. 2DUS is highly operator dependent with poor inter-rater reliability: a study where four different sonographers (two radiologists and two orthopaedists) made alpha angle measurements from hip 2DUS images found an agreement rate of 3.6–44.5% [7]. Variations in the angle at which the ultrasound probe is held can change the diagnosis of the same hip from normal to dysplastic or vice versa [8]. Since the alpha angle is the main quantitative measure used to diagnose DDH, this variability can greatly influence over- and under-treatment rates. Three-dimensional ultrasound (3DUS) has advantages over 2DUS including multiplanar visualisation, reduced operator-dependence and standardisation of measurements [9]. 3DUS may provide a more accurate and reliable method of diagnosing DDH through universal screening efforts.

To implement 3DUS imaging toward the goal of eventual universal screening in diverse settings including rural clinics and community health settings, it would be highly desirable for 3DUS to be reliable even when performed by relatively inexperienced operators. Therefore, in this study we sought to compare the intra- and inter-rater reliability between novice and expert operators performing both 2D and 3D ultrasound scans of infant hips for the diagnosis of DDH.

Materials and methods

Enrolment of patients

This was a prospective study performed at University of Alberta Hospital, Edmonton, Canada. The study was approved by the institutional ethics committee (PRO00032107). Written informed consent for participation was obtained from a parent or guardian of each infant prior to enrolment. Consecutive infants presenting to the radiology department for an ultrasound scan requested for clinical suspicion of hip dysplasia, aged less than 6 months, were eligible. We included 30 infants, mean age 10.8 weeks (range 6 weeks–4 months), 14 males, 16 females.

Enrollment of operators

We had two groups of ultrasound operators: Novices and Experts. The Novices were seven medical students with no prior experience in ultrasound imaging (never held a probe before). Each novice was trained briefly just prior to data collection. Training involved a 30-min explanation by the study investigators on the Graf criteria and quality of ultrasound images, as well as 1 h of hands-on proctored training on both 2DUS and 3DUS scanning of infant hips using the ultrasound transducer during a hip dysplasia clinic. Following training the novices performed 2D and 3DUS scans of infant hips while the expert was either not present in the room or instructed not to assist in the procedure in any way. The Experts consisted of four ultrasound technicians with 5–15 years of professional experience conducting ultrasound scans of infant hips at tertiary paediatric hospitals, including at least 1 year of 3DUS scanning in our department.

Imaging

Each infant had both hips scanned while in a supine position, hips flexed 90°, by Novice and Expert operators using 2D and 3D ultrasound. Imaging was performed on a Philips IU22 machine using a 12-MHz L12-5 linear probe for 2DUS and 13-MHz 13VL5 probe for 3DUS. For 2DUS, imaging was completed in the Graf standard plane, conforming to the American College of Radiology (ACR) recommendations [10]. The 3DUS images were also obtained in this coronal orientation; we performed a 3.2-s automated sweep, through a range of ±15° to produce US slices of 0.22 mm thickness. To assess both inter- and intra-rater reliability, we had each participant attempt to perform two 2DUS scans and two 3DUS automated sweeps of each hip (Fig. 1). However, in some cases of particularly challenging infants and at the request of parents, our novices were only able to perform one scan of the hips.

Image processing

Image quality was assessed on both 2DUS and 3DUS images for Novices and Experts by having an observer with 2 years' experience dedicated to hip imaging, blinded to whether a scan was by a Novice or an Expert, grade each scan semi-quantitatively based on the sum of criteria related mainly to Graf standard plane landmarks (1 point each for: full width of femoral head visible; acetabular roof clearly visible; straight and level iliac wing; os ischium visible; absence of artefacts obscuring image; maximum score 5).

This same observer also measured alpha angle and acetabular coverage measurements for each saved 2DUS image, blinded to 3DUS images and the diagnostic outcome of each case. For 3DUS, index measurements were obtained

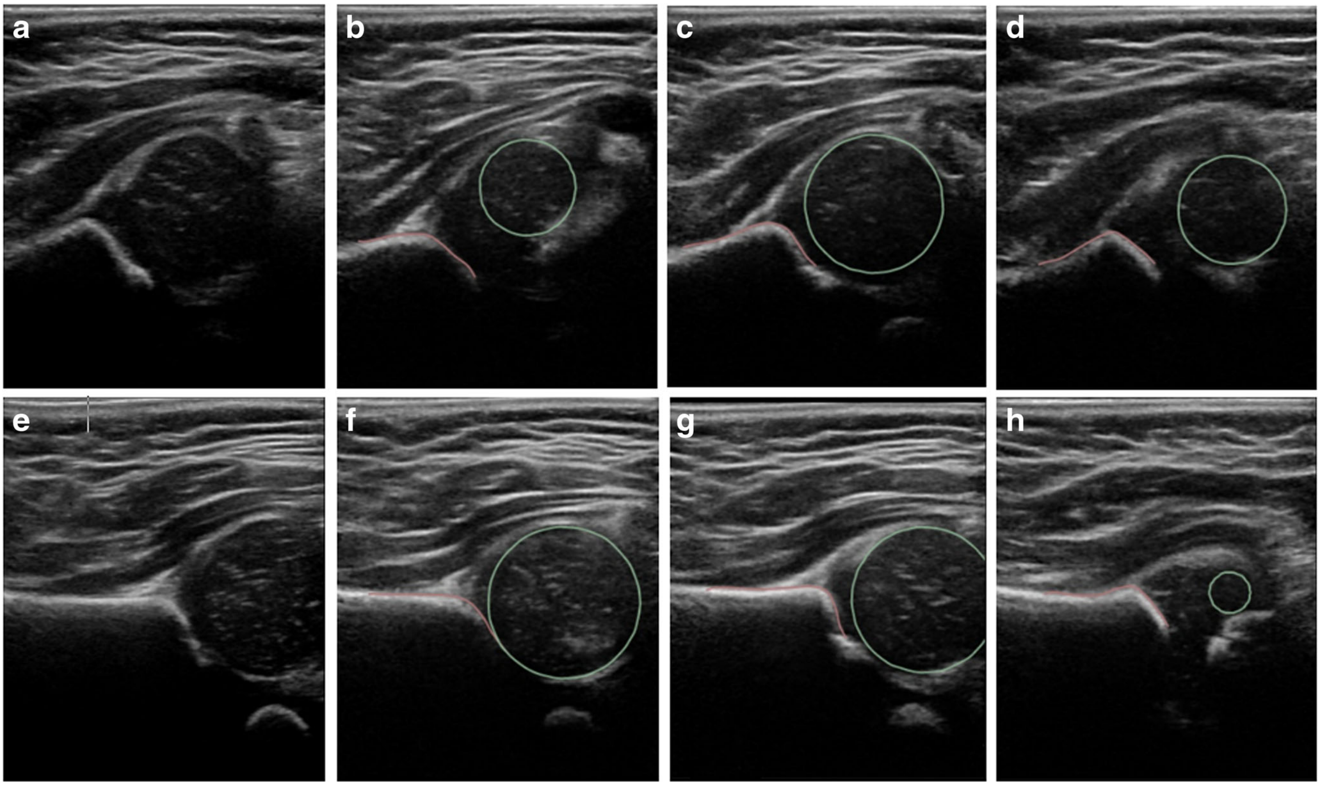

Fig. 1 Example of 2D and 3D scans performed on the same hip in a challenging infant. **a** 2DUS scan performed by novice operator demonstrating lack of a straight iliac wing and poorly visualised os ischium (quality 3/5). **b–d** Anterior, middle and posterior slices, respectively, of 3DUS scan performed by novice operator. The red line conveys automated tracing of the acetabulum by the custom program; femoral head tracing is shown in green. **e** 2DUS scan performed by expert operator. This received a quality score of 5/5. **f–h** Anterior, middle and posterior slices of 3DUS scan performed by expert operator

automatically by a custom post-processing software algorithm written in Python (version 2.7) using the Visualisation Toolkit (VTK, Kitware). This algorithm generates 3D acetabular surface models and indices (3D alpha, 3D anterior alpha angle, 3D posterior alpha angle and 3D coverage) for DDH diagnosis [11]. The gold-standard diagnosis was made as 'normal', 'borderline' (indeterminate requiring further follow-up, usually Graf IIa) or 'dysplastic' based on central re-review of Expert 2DUS images by a paediatric musculoskeletal radiologist, blinded to other clinical and imaging data, considering alpha angle, acetabular coverage and acetabular morphology as per ACR diagnostic criteria [10].

Statistics

Intraclass correlation coefficient (ICC) estimates and their corresponding 95% confidence intervals were computed using SPSS statistical package version 22 (SPSS Inc). Calculations were based on single ratings, consistency, two-way fixed-effects model ICC (3,1). Simple logistic regression was also performed to predict the clinical diagnosis of each infant hip according to basic demographics such as age and sex, and the 3D indices generated.

Results

Novices each performed between 12 and 20 scans. At study completion we had scans of 60 hips from 30 patients for analysis of hip dysplasia using 2DUS and 3DUS.

On 2DUS, the quality score (maximum possible 5) averaged 2.7 ± 1.2 (mean ± standard deviation SD) for Novice versus 4.9 ± 0.3 for Expert scans (p = 0.04), with 19/60 (32%) Novice and 60/60 (100%) Expert scans having quality of at least 4/5. On 3DUS, the quality score was 4.2 ± 1.0 for Novice versus 4.9 ± 0.3 for Expert scans (p = 0.99), with 48/60 (80%) Novice and 60/60 (100%) Expert scans having quality of at least 4/5. The most common error in scans performed by novice operators was difficulty producing a straight level iliac wing (Fig. 1).

Our novice operators managed to perform 2 3DUS scans of each hip on 23/30 infants in the study. For the remaining seven patients, three patients had one of their hips scanned only once and the other four cases had only a single scan of each hip; at the completion of the study novices had performed 109 total scans (Fig. 2). We excluded eight 3DUS scans (3.4%) from further processing due to motion, lack of a visible acetabulum or if there was no

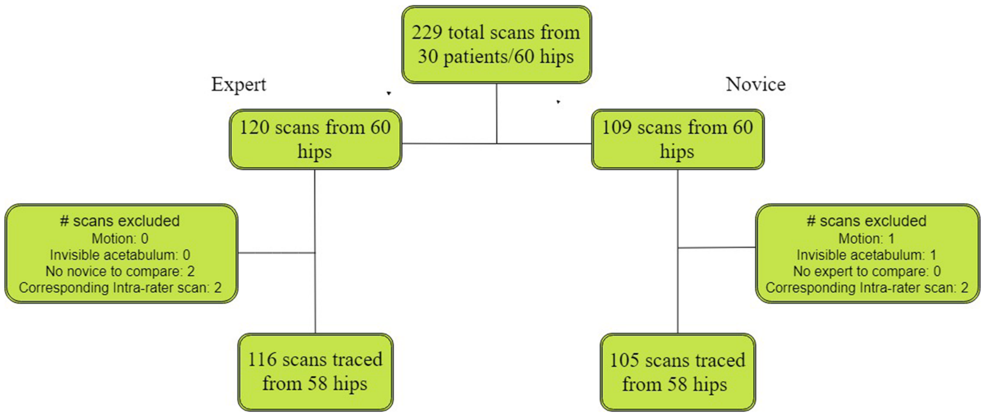

Fig. 2 3D scan exclusion flowchart

comparative scan available (Fig. 2). Two 3DUS hip scans by Novices were excluded from the study as uninterpretable due to very poor quality (lack of a visible/traceable acetabulum and excess motion); the corresponding expert and intra-rater scans were also excluded leaving 58 hips for further analysis (Fig. 2). Nearly all hips in this study were normal: 56/58 hips (96.6%) classified as 'Normal', 1/58 (1.7%) as 'Borderline' and 1/58 (1.7%) as 'Dysplastic' according to the Expert 2DUS assessment.

Inter-rater/intra-rater reliability

Inter-rater reliability between novices and experts differed substantially between 2DUS and 3DUS, being poor for both 2DUS indices (0.04–0.10), fair for 3DUS acetabular coverage (0.55) and good to excellent for 3DUS alpha angles (0.73–0.83) (Table 1). Intra-rater reliability was also somewhat higher for 3DUS indices than 2DUS indices (0.65–0.74 vs. 0.54–0.62; Table 1). Absolute differences between novice and expert values were also substantially higher for 2DUS than 3DUS indices (Table 1).

Diagnostic accuracy

At 3DUS, our computer model, generated automatically from surface models in Novice scans, predicted 57 hips in the study to be 'Normal' and one hip as 'Dysplastic', while classifying as 'Normal' the hip identified at gold-standard Expert 2DUS as being 'Borderline' (Fig. 3). Conversely, at 2DUS the 2D alpha angle (2Dα) computed from the Novice scans classified only 21 hips at Normal (2Dα >= 60°), 21 hips as Borderline (2Dα 51° – 59.9°) and 16 hips as Dysplastic (2Dα < 51°), widely discrepant from gold-standard diagnosis.

Discussion

Three-dimensional US is an emerging imaging technique for the diagnosis of DDH. In this study we assessed the reliability of 3DUS versus 2DUS when performed by novice operators with no prior experience in either modality, and minimal training. 3DUS was superior in three ways. First, image quality scored semi-quantitatively was significantly better for Experts than Novices at 2DUS but similar between Novices and Experts at 3DUS. Second, inter-rater reliability of indices

Table 1 Two-dimensional (2D), three-dimensional (3D) inter-rater (Expert/Novice) and 3D intra-rater (Novice and Expert) ICC values (± 95% CI). The mean values for each index and absolute difference are also reported

Imaging index	Inter-rater ICC (95% CI)	Novice intra-rater ICC (95% CI)	Expert intra-rater ICC (95% CI)	Index values: mean+ / -SD		Novice / Expert absolute difference (mean ± SD)
				Expert	Novice	
2DUS						
Alpha	0.10 (-0.26–0.43)	0.54 (0.16–0.78)	0.80 (0.69–0.88)	66.7 ± 5.0°	57.1 ± 10.8°	13.2 ± 6.6
Coverage	0.04 (-0.31–0.39)	0.62 (0.27–0.83)	0.67 (0.50–0.79)	56.2 ± 6.7%	45.0 ± 15.3%	17.2 ± 9.0
3DUS						
Alpha	0.83 (0.72–0.89)	0.74 (0.58–0.84)	0.77 00.57–0.88)	67.8 ± 9.2°	66.5 ± 8.2°	3.9 ± 3.5
Alpha ant	0.82 (0.72–0.89)	0.65 (0.45–0.79)	0.79 (0.61–0.89)	62.4 ± 10.4°	61.0 ± 10.7°	5.2 ± 3.8
Alpha post	0.73 (0.58–0.74)	0.74 (0.58–0.85)	0.67 (0.42–0.82)	72.8 ± 8.3°	70.9 ± 6.9°	4.6 ± 3.7
Coverage	0.55 (0.35–0.52)	0.70 (0.53–0.82)	0.65 (0.39–0.81)	0.57 ± 0.2%	0.54 ± 0.2%	0.1 ± 0.1

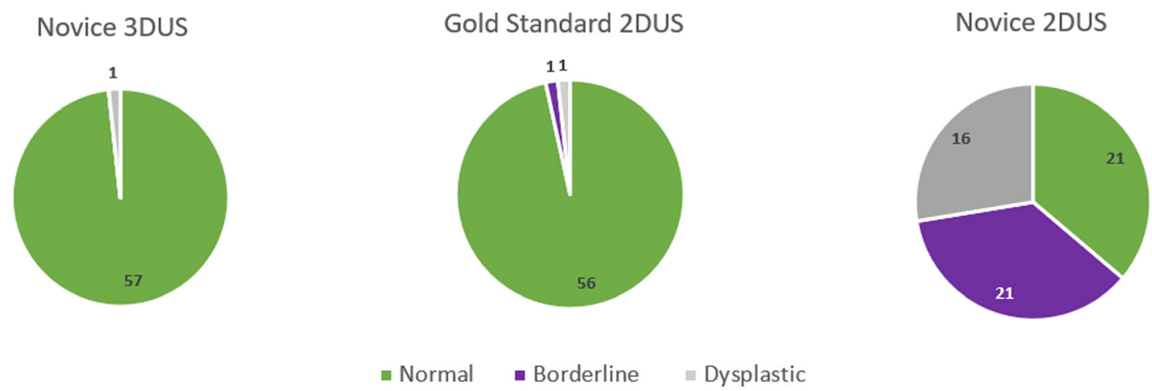

Fig. 3 Diagnosis from images obtained at novice 3DUS and novice 2DUS relative to the expert's gold standard 2DUS diagnosis. The diagnoses from novice 3DUS scans closely matched the expert scans; the novice 2DUS scans produced many false-positive dysplastic-appearing hips

derived from the scans performed by Novices versus Experts was much higher for 3DUS (ICC 0.55–0.83) than for 2DUS (ICC=0.04–0.10). Third, 3DUS scanning by Novices correctly classified all 56 'Normal' hips in this study, and identified the single 'Dysplastic' case, while classifying the single 'Borderline' hip as 'Normal', which may eventually be correct since most 'Borderline' (Graf IIa) hips normalise spontaneously at follow-up [12]. This diagnostic performance closely matched that of Expert 2DUS. In contrast, conventional 2DUS performed by Novices misclassified 35 of the cases; most importantly, 15 'Normal' hips were declared 'Dysplastic' by indices measured from the novice 2DUS scans, false-positive results that would potentially lead to unnecessary treatment at substantial costs to the healthcare system and stress for families.

The improved subjective image quality, inter-scan index reliability and diagnostic accuracy of 3DUS over 2DUS when performed by Novices after brief training highlight an advantage of 3DUS as an imaging modality – hip 3DUS appears to be easier to learn and perform reliably than 2DUS. This agrees with previous work demonstrating the improved accuracy of 3DUS over conventional 2DUS in classifying hips as normal, borderline or dysplastic [11]. The reduced dependence on user skill that is intrinsic to 3DUS compared to 2DUS could improve on screening accuracy and cost-effectiveness.

This study has limitations. First, we did not assess the effect of different levels of training on the reliability of participants; future studies can assess to what extent more rigorous, but still relatively brief, training (e.g. a weekend course on 2DUS and 3DUS) would further improve Novice scan quality, reliability and diagnostic accuracy. Second, in this population there was a low proportion of hips with dysplasia (1/60 = 1.7%, matching the expected population incidence of 1–2%) [1]. Further studies on populations with a higher incidence of DDH, such as in patients with known dysplasia who are attending for follow-up visits, would more fully evaluate the performance of Novices at detecting dysplastic hips of differing levels of severity. However, the population tested in this study does match that of the ultimate intended screening program, and scanning of dysplastic hips during management is likely to continue to be performed by expert sonographers in tertiary settings. Third, because there is no external reference standard conveniently available for the diagnosis of hip dysplasia within a study time frame, we used Expert 2DUS diagnosis as the reference standard for DDH diagnosis despite the well-known flaws in 2DUS assessment [8]. It is difficult to avoid this problem in any study of DDH without multi-year follow-up to determine long-term clinical and imaging outcomes. Fourth, while time to image acquisition was subjectively similar between the novice and expert groups, this parameter was not formally assessed in this study and warrants further examination in the future. Finally, the study was relatively small at n=30 patients; however, clear significant differences emerged even at this sample size.

This study has important clinical implications in the diagnosis of DDH. Infants that are diagnosed with DDH at an early age fare better with treatment than those that are missed; however, universal screening programs are rarely implemented worldwide due to limitations in 2DUS imaging [13–15]. Specifically, universal screening with 2DUS leads to increased rates of (costly) treatment without a corresponding reduction in late diagnosis of DDH or surgical management [1]. These studies used expert sonographers for their assessments; our study suggests that accuracy would be lower, and particularly that false-positive rates would likely be higher, if the 2DUS scans were performed by inexperienced operators. 3DUS can address these limitations by providing a semi-automated method of diagnosis that outperforms conventional 2DUS by minimising the number of borderline cases and reducing the need for follow up imaging in children assessed for DDH [11]. Our study also demonstrates that 3DUS significantly outperforms 2DUS in reliability and correct diagnostic classification of hips when performed by Novice operators. Ultimately, 3DUS may facilitate the introduction of universal screening policies for DDH, which would improve early detection of this condition.

The benefits of 3DUS have the potential to reach beyond diagnosis DDH in infants. Recent evidence has emerged demonstrating the utility of point-of-care ultrasound (POCUS) ultrasound in a variety of settings, from the emergency department to military bases [16]. POCUS in the emergency setting has been shown to accurately detect fractures of the clavicle, humerus, rib, femur, ankle and foot [17], as well as for triaging abdominal injuries in military personnel during war times [18]. The World Health Organization (WHO) has also recognised the value of POCUS for medical care in remote areas [19]. To address concerns that providers in developing countries have not received adequate training [18], 3DUS allows a semi-automated method of diagnosis that minimises reliance on the sonographer. In the future portable 3DUS equipment could be applied even in remote locations to provide POCUS of infant hips and other conditions. The images could be uploaded to radiologists at tertiary care centres, and/or to automated deep-learning analysis algorithms, for review and diagnosis. This would reduce or ultimately eliminate the unfortunate situation where a child with DDH is not screened and presents beyond the window for non-invasive treatment. The combination of 3DUS technology and artificial intelligence analysis could allow for cost-effective screening, followed by expert tertiary sonographer/radiologist assessment of the cases referred for screening.

In conclusion, we have demonstrated that unlike 2DUS, 3DUS of infant hips can be performed by operators with little to no ultrasound experience with relatively high image quality, fair to excellent inter-rater index reliability, and diagnostic accuracy sufficient to appropriately identify hip dysplasia in a small screening population. Performed by novice users, 3DUS was much more reliable than 2DUS. This motivates further study to optimise 3DUS user training and also to potentially expand 3DUS use to other clinical indications.

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Compliance with ethical standards

Guarantor The scientific guarantor of this publication is Dr. Jacob Jaremko, M.D., Ph.D, FRCPC.

Conflict of interest The authors of this manuscript declare no relationships with any companies whose products or services may be related to the subject matter of the article.

Statistics and biometry One of the authors has significant statistical expertise.

Informed consent Written informed consent was obtained from all subjects (patients) in this study.

Ethical approval Institutional Review Board approval was obtained.

Methodology
• Retrospective
• Cross-sectional study
• Performed at one institution

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