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Positive predictive values in clinical screening for developmental dysplasia of the hip

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Abstract

Aim

To establish the positive predictive value (PPV) of clinical hip examinations performed by referrers in the Danish screening programme for Developmental Dysplasia of the Hip (DDH) utilising three definitions of true positive DDH diagnosis.

Methods

We retrospectively identified 290 children (169 female) referred during a four-year period to the orthopaedic outpatient clinic at our institution with a positive clinical hip examination.

PPV was calculated for clinical hip examinations across three definitions of a true positive clinical hip examination for all referrers and subgroups consisting of general practitioners, midwives and paediatricians. The PPV for clinical hip examinations was calculated for paediatric orthopaedic surgeons using one of the three definitions.

Results

PPV of clinical hip examinations for all referrers were 5.4%, 3.6% and 1.8% with the definition of a true positive DDH diagnosis defined as clinical instability found by orthopaedic surgeon, ultrasound classification \geq Graf IIc or both definitions combined, respectively.

PPV of clinical hip examinations performed by orthopaedic surgeons was 33.3% with a true positive clinical examination defined as an ultrasound classification \geq Graf IIc.

Conclusion

We conclude that the positive predictive value of clinical hip examinations made by referrers in the Danish screening programme for DDH is low.

Keywords: Developmental dysplasia of the hip, paediatric orthopaedics, predictive values, screening

Key notes

The success of a universal clinical screening for developmental dysplasia of the hip (DDH) is dependent on the quality of the primary clinical examinations.

The positive predictive value of clinical hip examinations made by primary screeners in the Danish screening programme for DDH is low.

This study serves demonstrates that further education of the primary screeners in the Danish screening programme for DDH is needed

List of abbreviations

DDH, developmental dysplasia of the hip; PPV, positive predictive value; AAUH, Aalborg University Hospital; NPV, negative predictive value; TP, True positive; AP, all clinically positive referrals

Introduction

Background

Developmental dysplasia of the hip (DDH) is a term which incorporates a spectrum of disorders of the hip, ranging from mild dysplasia to complete irreducible hip dislocation [1].

In Denmark, a combination of universal clinical screening and selective ultrasound screening for DDH is implemented. Official national guidelines recommend a clinical hip examination by a midwife after birth which is repeated at a 5-week follow up by a general practitioner [2]. In supplement to the nationally recommended clinical examinations, screening for recognized risk factors for DDH have been implemented regionally.

Hence, if one of the clinical hip examinations are positive or the child has a recognized risk factor for DDH, it is referred to a specialized hip ultrasound examination and orthopaedic evaluation.

The Danish national guidelines specify that the Barlow and Ortolani manoeuvres should be used in the clinical examination for DDH [2]. These manoeuvres are difficult to perform correctly, especially by non-orthopaedic health professionals [3]. In Denmark there is no formalized education in these difficult provocative hip tests, but the training is part of the non-formalised apprenticeship for paediatricians, midwives and general practitioners.

The positive predictive value (PPV) of the primary clinical hip examination is 4% in a neonatal screening program where examinations were performed by general practitioners, midwives and nurses and when defining a true positive clinical examination as a child referred with a positive clinical hip examination and a subsequent diagnosis of clinically unstable hip made by a paediatric orthopaedic surgeon [4]. The PPV for the infant clinical hip examination by a group of referrers consisting of paediatricians, midwives and general practitioners across varying definitions of a true positive hip exam, as well as comparison to the PPV of the orthopaedic examination have not previously been investigated.

Objectives

The primary objective of this study was to assess and compare the PPV of the clinical hip examinations, performed by health professionals involved in the screening for DDH using three different definitions of a true positive clinical examination.

Patients and methods

This was a retrospective cohort study of children referred to the orthopaedic outpatient clinic at Aalborg University Hospital (AAUH), Aalborg, Denmark during a four-year period. Reporting follows the STROBE guidelines for reporting on observational studies.

We retrospectively identified children referred from our recruitment area (population: 400,000) for suspected DDH with a positive clinical examination between January 1. 2016 to December 31. 2019 during which there were 16.495 births in our region.

The inclusion criteria were: children referred to the orthopaedic outpatient clinic through the DDH screening programme, due to a positive clinical hip examination of one or both hips.

The term positive clinical hip examination is defined in its broadest possible form for referrals, ranging from referrals mentioning specific tests (Barlow/Ortolani/Galeazzi) to unspecific clinical findings (hip “clunk/click”, “positive clinical examination”).

Exclusion criteria: child referred with a recognized risk factor for DDH but no positive clinical hip examination. Age at referral > 3 months or examined side not specified by referrer.

Children referred on suspicion of DDH secondary to other primary pathology (e.g. neuromuscular syndromes or skeletal dysplasias) were seen in a different clinical setting, and data for these children was not included in this study (figure 1).

All included children were clinically examined for hip instability by one of three different groups of health professionals: within the first day of life by a midwife, or if the midwife was in doubt, a paediatrician and by the general practitioner at a 5-week routine follow-up, if the initial clinical examinations were negative.

Once referred, the children were initially sonographically examined by a musculoskeletal radiologist using the method described by Graf [5] and subsequently examined by one of three paediatric orthopaedic surgeons in the outpatient clinic. The orthopaedic examination consisted of hip stability assessment using the Barlow, Ortolani and Galeazzi tests as well as examining for limited hip abduction. It was standard procedure for the orthopaedic examiners to receive the results of the ultrasound examination after the clinical examination to prevent observation bias of the result.

Referrals, patient records and radiological reports were collected and reviewed retrospectively for study data by three independent reviewers. Study data was collected and managed using REDCap electronic data capture tools hosted at AAUH. REDCap is a General Data Protection Regulation /Health Insurance Portability and Accountability Act compliant web-based software platform designed to support data capture for research studies.

This study was approved by the Danish Patient Safety Authority with case number 31-1521-260.

Ethical approval was not required in accordance with the guidelines of the Danish National committee on health research ethics for non-interventional studies.

Primary outcomes:

PPV of clinical hip examinations performed by four groups of health professional were calculated using three different definitions of a true positive clinical hip exam. Definitions can be seen in table 1.

For the group of orthopaedic surgeons, we calculated the PPV of the clinical hip examination using a sonographic Graf classification \geq IIc as golden standard (True positive definition #2, Table 1), as the true positive definition 1 and 3 were dependent on the result of the orthopaedic clinical examination.

Statistical methods

PPV was calculated as (true positives/all clinically positive examinations performed by the group) using three definitions of a true positive clinical hip examination (one for orthopaedic surgeons) across four different groups of health professionals (paediatricians, midwives, general practitioners and orthopaedic surgeons).

Characteristics of included children were grouped by agreement between referrers and orthopaedic surgeons in clinical hip examination. Mean age at referral and examination in outpatient clinic was compared using independent two-sample *t*-tests. Distributions of gender and referrers between the agreement and disagreement group were compared using chi-square statistics.

Statistical analyses were performed using Stata version 16.1 (StataCorp, College Station, TX, USA).

Results

We retrospectively identified 290 children (169 female) referred during a four-year period with a positive clinical hip examination. Of these a positive clinical examination was reported for 336 hips (figure 1). One hip was missing sonographic information.

Mean age of children at referral and examination in the outpatient clinic was significantly higher for the group of children with disagreement between referrer and orthopaedic surgeon in the clinical hip examination. The mean interval between referral and examination in the outpatient clinic was not significantly different between the agreement and disagreement groups. Demographics of included children grouped by agreement can be seen in table 2.

The PPV of the clinical hip examination for referrers as a group was 5.4%, 3.6% and 1.8% when the true definition was defined as clinical instability found by orthopaedic surgeon, ultrasound classification \geq Graf IIc or both definitions combined, respectively. PPV of clinical hip examinations performed by the orthopaedic surgeons was 33.3% when the true definition was defined as an ultrasound classification \geq Graf IIc.

PPV for all subgroups across true positive definitions can be seen in table 3.

Discussion

In this study we found the PPV of clinical hip examinations performed by referrers in the Danish screening programme for DDH to range from 1.8% to 5.4% across three different definitions of a true positive clinical examination.

We defined a positive clinical examination in the broadest possible term as midwives, general practitioners and paediatricians in Denmark receive no formalized education in clinical hip exams including hip provocative tests and therefore may not be familiar with the correct wording or nomenclature in the referral. As an example, a positive Ortolani is often confused with clicky hips. Therefore, all referrals based on clinical suspicion were included.

Since no clear definition of what constitutes a DDH diagnosis exists, it is difficult to define one true positive clinical hip examination for this disorder. When assessing the PPV of clinical hip examinations performed by referrers, we aimed to demonstrate how this value varies when different definitions were applied. In the widest definition of a true positive clinical hip exam in this study, the referrer group produced a PPV of 5.4%. By using a separate ultrasound examination classification in our second definition of a true positive clinical hip examination, we were able to demonstrate a significant difference in the PPV of clinical hip examinations performed by referrers and paediatric orthopaedic surgeons.

We recognize that including potential immature hips lowers the threshold for a true positive clinical examination in definition #1, which could explain why the PPV was higher than previous studies have found [4]. However, this highlights how differing definitions of pathological DDH results in varying rates of DDH diagnosis.

The timing of the clinical hip examination is important as hip instability may resolve spontaneously within the first 2-6 weeks of life [6] [7]. As the paediatricians and midwives examine the children within the children's first days of life, they may detect clinical hip instability that will resolve spontaneously before the child is examined by the orthopaedic surgeon in the outpatient clinic. In Denmark the general practitioners generally examine the child after this period of spontaneous resolution, but interestingly had the lowest PPV across all definitions compared to the other groups of referrers in this study.

The interval between referral and orthopaedic examination could allow for spontaneous resolution of any detected hip instability resulting in a lower PPV of the clinical examination performed by referrers. However, we did not find a significant difference in intervals when comparing the groups of children with agreement and disagreement between referrers and orthopaedic surgeons (Table 2). Additionally, the group of children, with disagreement between referrer and orthopaedic surgeon in clinical hip examination, was significantly older at both referral and initial visit to the outpatient clinic when compared to the agreement group.

As referrers did not report negative clinical hip examinations, we were not able to calculate the negative predictive value (NPV) of the clinical hip examination.

When interpreting PPV and NPV it is important to recognize the impact of prevalence on both values. As the prevalence of a condition decreases the PPV of any test testing for that condition, regardless of sensitivity, will likewise decrease and NPV will increase. As DDH is a low-prevalence condition this partly explains the low PPV of the referrer's clinical examination.

In the United Kingdom, a screening programme for DDH is implemented, similar to the Danish programme [10]. The clinical examiners are general practitioners, midwives and nurse practitioners. Choudry et al. found a decrease in clinical PPV from 28% to 4% over a period of 25 years. While the examinations and referral criteria had stayed the same during this period, the group of referrers had been expanded, and the authors commented that the large number of inexperienced examiners may have been associated with the decrease in PPV of referred clinical hip examinations [4]. Similarly, Swedish investigators found that an increase in referrers resulted in higher referral and surgical treatment rates with no reduction in late diagnosis [11]. In this study, the clinical hip examinations had a PPV of 33%, when performed by paediatric orthopaedic surgeons and held up against a sonographic DDH diagnosis but only 3.6% when performed by midwives, paediatricians and general practitioners with the same criteria for a true positive examination.

In two of the largest randomised controlled trials on DDH screening to date, two teams of Norwegian investigators found no significant impact on the rate of late diagnosis of DDH when implementing a universal ultrasound screening programme to their population. [12] [13]. But as Holen et al. commented; this depended upon the clinical screening being of high quality, as is arguably the case in Norway where the clinical screening is centralized to expert paediatric orthopaedic examiners and paediatricians.

To put the quality of clinical screening in perspective, during our study period, 2.5% of all children born were referred for further examination based on clinical examination alone and 6.7% based on risk factors for DDH and clinical examination combined. In the UK, corresponding rates are 0.4% and 4.7% respectively [14][4]. The comparable high referral rates in our study further underlines the need for experienced examiners in the clinical screening of DDH. The positive predictive value of screening will always be low in a relatively rare disease as DDH. However, it is mandatory to seek the highest level of education in those performing the screening as a false positive test both results in unnecessary psychological stress to the family and in an economical burden to the health care system.

The present study demonstrates the relatively low predictive value of clinical hip examinations in the screening of DDH when compared to expert examiners. This is worrying as the clinical examination is the only national recommendation in the Danish national guidelines for screening of DDH [2].

We conclude that the positive predictive value of clinical hip examinations made by referrers in our screening programme for DDH is low. It might be that clinical screening for DDH would be improved by concentrating it to a small group of health professionals who have received formalized training in the clinical examinations used.

Other information

No external funding was obtained for this study

The authors declare that they have no conflict of interests

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Tables:

Table 1 – definitions of a true positive clinical hip examination	
	Definition – referred with a positive clinical hip examination and:
True positive #1	Clinically unstable at examination by orthopaedic surgeon
True positive #2	Hip ultrasound classification \geq Graf IIc
True positive #3	Definition #1 and #2 needs to be fulfilled

Table 2 – Characteristics of children grouped by agreement between referrer and orthopaedic surgeon in clinical hip examination.

	Disagreement (n=318)	Agreement (n=18)	P-value
Mean age at referral (SD), days	13.7 (17.8)	7.3 (10.9)	<0.05
Mean age at examination in outpatient clinic (SD), days	40.7 (19.7)	32.6 (15.2)	<0.05
Mean interval between referral and examination in the outpatient clinic (SD), days	27.1 (10.8)	25 (8.7)	0.32
Female:male ratio	1.3	2	0.40
Referred by:			
General practitioners, n (%)	90 (27.7)	2 (11.1)	<0.05*
Midwives, n (%)	140 (44)	5 (27.8)	
Paediatricians, n (%)	88 (28.3)	11 (61.1)	

Table 3 – PPV comparison across groups stratified by true positive definitions							
Group	subgroup	True positive definition					
Referrer		True positive 1		True positive 2		True positive 3	
		TP/AP	PPV	TP/AP	PPV	TP/AP	PPV
	General practitioners	2/92	2.2%	0/91	0%	0/91	0%
	Midwives	5/145	3.4%	6/145	4.1%	2/145	1.4%
	Paediatricians	11/99	11.1%	6/99	6.0%	4/99	4.0%
	All referrers	18/336	5.4%	12/335	3.6%	6/335	1.8%
Orthopaedic Surgeons		-	-	6/18	33.3%	-	-

Figures

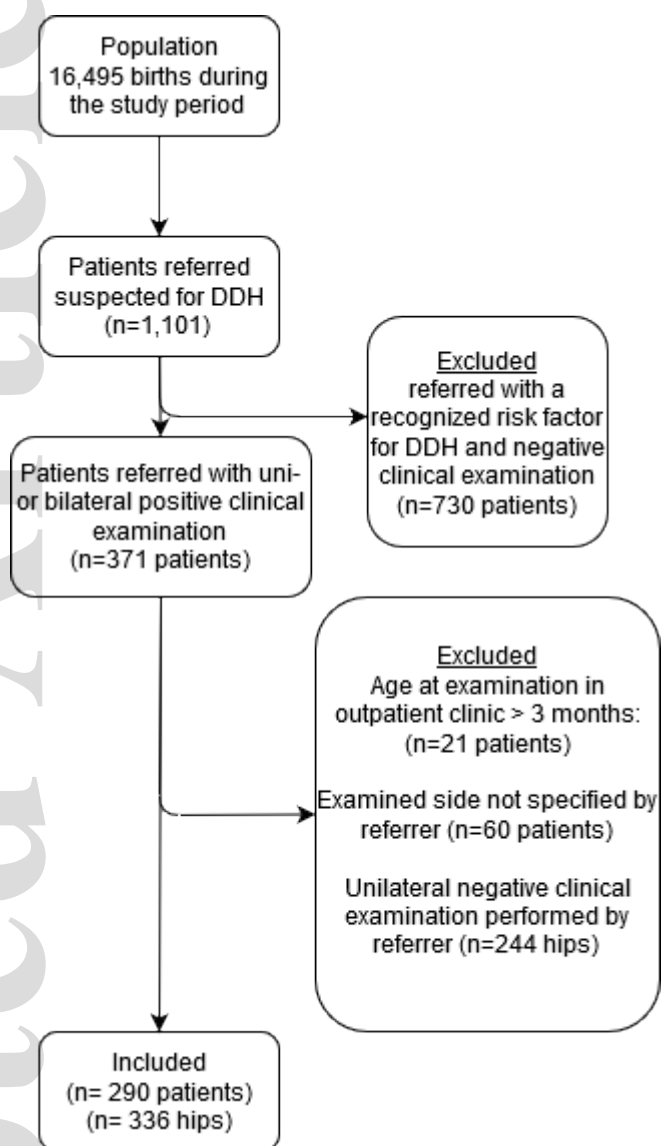


Figure and table legends

Table 1: definitions of a true positive clinical hip examination used in this study

*Table 2: Characteristics of children grouped by agreement between referrer and orthopaedic surgeon in clinical hip examination. Statistically significant results are marked as bold. *calculated using chi-square statistics.*

Table 3: TP = true positive, AP = all clinically positive referrals, PPV = positive predictive value. True positive 1,2 and 3 defined as - A positive clinical examination and: clinically unstable at examination by orthopaedic surgeon, hip ultrasound classification \geq Graf IIc or both combined, respectively.

Figure 1: Consort diagram of the inclusion and exclusion process for this study