







The reality of sonography of the infant hip in Brazil: results and recommendations

Giovanna Galvão Braga Motta^{1,2*} , Natasha Vogel Majewski Rodrigues^{1,3} , Susana Reis Braga^{1,4} , Márcio Luís Duarte^{1,5} , Patricia Moreno Grangeiro^{1,6} , Alexandre Francisco de Lourenço^{1,7} 

¹Instituto Brasileiro de Apoio a Saúde da Criança com DDQ - Reinhard Graf, São Paulo-SP 04080-917, Brazil

²Department of Evidence-Based Health, Escola Paulista de Medicina da Universidade Federal de São Paulo, São Paulo-SP 04024-002, Brazil

³Department of Pediatric Orthopedics, Hospital do Servidor Público Municipal, São Paulo-SP 01532-000, Brazil

⁴Department of Orthopedics, Santa Casa de Misericórdia de São Paulo, São Paulo-SP 01221-020, Brazil

⁵Department of Radiology, Universidade de Ribeirão Preto Campus Guarujá, São Paulo-SP 11440-003, Brazil

⁶Department of Pediatric Orthopedics, Hospital das Clínicas da Universidade de São Paulo, São Paulo-SP 05403-010, Brazil

⁷Department of Pediatric Orthopedics, Escola Paulista de Medicina da Universidade Federal de São Paulo, São Paulo-SP 04023-062, Brazil

***Correspondence:** Giovanna Galvão Braga Motta, Instituto Brasileiro de Apoio a Saúde da Criança com DDQ - Reinhard Graf, São Paulo-SP 04080-917, Brazil. giovannabragam@gmail.com

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Abstract

Since 1980, when Professor Reinhard Graf developed a novel technique, sonography of the infant hip has played a prominent role in the early diagnosis of developmental dysplasia of the hip (DDH). Brazil is a vast country, and disparities exist among regions and between the public and private health systems. In addition, healthcare professionals often have limited knowledge of DDH, and patients need to be referred to larger urban centers for definitive diagnosis and appropriate treatment. These reasons, and more, may hinder the early diagnosis of DDH. Following the successful implementation of similar neonatal screening programs for other conditions in Brazil, developing a screening program for DDH in Brazil is imperative. Such a program should consider training professionals to use Graf's technique as a diagnostic procedure. It is recommended that more studies be conducted in every region of the country to provide a broader understanding of the current role of sonography in the screening and diagnosis of DDH in infants in Brazil.

Keywords

Developmental dysplasia of the hip, ultrasonography, diagnosis, diagnostic screening programs, Brazil



Introduction

Developmental dysplasia of the hip (DDH) is an orthopedic condition that involves morphological changes in the hip at birth or early infancy, ranging from delayed maturation to hip dislocation [1]. Although the exact cause of DDH remains unknown, it is the most common congenital anomaly of the musculoskeletal system, with an incidence ranging from 0.06 per 1,000 live births among some groups of Africans to 76.1 per 1,000 live births among Native Americans, as demonstrated by Loder and Skopelja [2]. These authors identified significant variability in incidence within ethnic groups by geographic location, which also occurs in Brazil [3], mainly because it is a vast country in which there are many people of mixed ethnic and racial backgrounds.

In 1984, Graf [4] developed a technique for sonography of the infant hip, which has played a prominent role in the early diagnosis of DDH. In 1989, Milani et al. [5] introduced the technique to Brazil, and the first examinations were performed at Escola Paulista de Medicina, in the city of São Paulo; this was the first step towards the dissemination of Graf's method in Brazil.

In 1988, Guarniero et al. [6] estimated the prevalence of the disease, based on Ortolani maneuver positivity, to be approximately 0.5% among newborns at Associação Maternidade de São Paulo. More recently, Motta et al. [7] reported the prevalence of DDH to be considerably higher (5.4%) using Graf's method of performing ultrasound to screen for changes in all newborns' hips at a maternity hospital in São Paulo. Globally, hip sonography using Graf's method has been adopted as a key measure to complement early DDH diagnosis, as clinical screening alone has low sensitivity in the neonatal hip [8]. It has been reported that 54% of neonatal hips show no clinical signs of the disorder [9].

DDH is a prevalent subject in specialty conferences, meetings, and symposia in the field of healthcare in Brazil. This is because of the difficulty of its diagnosis, limited access to prompt treatment, and the significant impact which its potential sequelae may have on quality of life during all stages of development and into adulthood. It is challenging to implement a screening program for DDH and to organize patient flow in a country that is vast and socio-economically heterogeneous, as in Brazil.

It is noteworthy that studies reporting the diagnosis of DDH by infant hip ultrasound in Brazil mostly originate from particular regions of the country. Moreover, studies related to the use of this diagnostic method aimed at the development of public policies for DDH screening are scarce, making it difficult to develop strategies for the early diagnosis and treatment of this condition.

The objective of this article is to describe the current state of infant hip sonography in Brazil, develop a critical assessment of the accessibility of hip sonography performed with correct technique, and propose recommendations for implementing DDH screening in the country. To that end, we conducted a narrative review of the literature on the application of ultrasound in diagnosing DDH in Brazil.

Particularities of Brazil

Brazil is the fifth largest country in the world, with an area of 8,510,418 km², and is divided into five regions, comprising 26 states and one federal district, with 5,568 cities and a total population of approximately 203,000,000 inhabitants (approximately 104 million women and 98 million men) [10]. A substantial portion of the population resides along the coast, where many state capitals are located. The coastal state of São Paulo, in the southeastern region of the country, is equal in size to the United Kingdom. The southeastern and southern regions, which comprise only seven states, are collectively home to approximately 113 million individuals, corresponding to 55% of the country's population [10]. The non-uniform population distribution in Brazil may limit patient access to healthcare and trained healthcare professionals, as has been reported in several other countries [11] and demonstrated by the low quality of imaging examinations reported in the review conducted by Walter et al. [12]. There are also heterogeneous socioeconomic conditions and extensive ethnic diversity throughout the country. In the last Brazilian census conducted in 2022, 47.51% of the population self-identified as white, whereas only 7.52% self-identified as black [10]. There is a direct correlation between ethnicity and DDH, which has higher rates

among Caucasians [3]. In the southern region of Brazil, there is a predominance of individuals of European descent, whereas individuals of African descent are predominant in the northern and northeastern regions. A nationwide genetic study found the prevalence of European ancestry to be up to 85% in some municipalities in the southern region of Brazil [3]. Given that DDH is more common in individuals of European descent, the incidence of this disorder may differ significantly among regions of the country.

Universal healthcare in Brazil

SUS, which stands for Sistema Único de Saúde (Unified Health System), is the Brazilian public health system created by the Federal Constitution of 1988 and effectively implemented in 1990 [13]. The five basic principles that guide the legal system of SUS are universality, integrality, equity, decentralization, and social participation [13]. Among countries with more than 200 million inhabitants, Brazil is the only one that has a universal public health system fully financed by the state [14]. SUS guarantees full free access to the entire country's population [14].

Even though every citizen has the right to use SUS, approximately 71.5% of the population uses the public system, whereas the remainder uses the private sector mainly in the form of healthcare plans. In regions with better socioeconomic conditions, such as the southeastern and southern regions, the proportion of the population with private healthcare plans (37.5% and 32.8%, respectively) is higher than the national average (28.5%). The state of São Paulo has the highest proportion in Brazil (41.2%) [15].

Souza et al. [3] found that the rate of cases of DDH submitted to surgery in Brazil was 0.29 per 1,000 live births, a value lower than that previously reported in the literature [16], and ranged from 0.72 per 1,000 live births in the southern region to 0.05 submitted to surgery per 1,000 live births in the northern region. However, their study sample included only patients treated by SUS, excluding the 25.6% who underwent surgery at private facilities. In the southern region of Brazil, which has a considerable proportion of European immigrants, the rate of surgical treatment for DDH is more than twice the national average and is 15 times higher than that in the northern region.

Gonçalves et al. [17] showed that the frequency of primary total hip arthroplasty (THA) via SUS increased from 2008 to 2015. The mean annual number of THAs in Brazil is $11,842.1 \pm 868.3$, with a mean total annual cost of US\$9,250,466.99 \pm US\$1,682,324.36. Of the patients who underwent THA, 62% were under 50 years of age and were referred for the procedure because of hip arthrosis. More than 40% of adults undergoing THA before the age of 50 years have radiographic findings suggestive of DDH. These alarming numbers underscore the need to establish a screening program for DDH in the pediatric population to avoid an increase in the number of THA performed and to minimize the costs involved with late diagnosis and treatment—delay in diagnosis of three months or more increases the likelihood that surgery will be required to correct the problem [18].

Various screening programs for different childhood conditions have been successfully implemented in Brazil. One example is the National Neonatal Screening Program, which investigates a variety of diseases including phenylketonuria, congenital hypothyroidism, sickle cell disease, other hemoglobinopathies, cystic fibrosis, congenital adrenal hyperplasia, and biotinidase deficiency. Another important program is the Brazilian National Immunization Program, which has gained international respect and has been recognized as a global reference by the Pan American Health Organization, a branch of the World Health Organization.

For DDH, the Brazilian National Ministry of Health recommends the Ortolani and Barlow maneuver in the first two days of life of all newborns and subsequent early childhood evaluations, however, there are no clear guidelines on referral to ultrasound examination [19]. The centralized structure of SUS could facilitate the implementation of a national screening program for DDH in Brazil.

Sonography of the infant hip in Brazil

During the neonatal period, the hip joint is mainly composed of cartilage, which makes ultrasound the most reliable method for diagnosing DDH. Sonography of the hip detects 52% more pathological hips than the

Ortolani and Barlow maneuvers [20]. In addition, ultrasound allows dynamic studies to be performed simultaneously with Ortolani and Barlow maneuvers [21]. Furthermore, it presents significant advantages over a pelvis radiograph; it does not involve the use of radiation, the examinations can be performed rapidly, the equipment is often portable, and it is highly accurate [21, 22]. A topic of much discussion is the fact that pelvic organs are vulnerable to radiation, and radiation-induced genetic damage is cumulative in a growing body with long years of life, which makes ultrasound examinations more recommendable for infants [11].

As with many other diagnostic methods, knowledge of the proper technique for sonography of the hip is essential because the correct interpretation of the examination relies on the technique being strictly followed. A defining feature of ultrasound is that it is operator-dependent [23], which makes it necessary to train professionals to perform each specific ultrasound technique. In a study conducted in Brazil, Souza et al. [24] concluded that there is a lack of knowledge of DDH among health professionals who screen newborns, demonstrating that 81.1% of such professionals (students, residents, pediatricians, and orthopedists) had never made a diagnosis of DDH in practice.

In Brazil, the method devised by Graf has previously been considered difficult and complex partly because it is associated with a recommendation to use a positioning cradle which is not yet commercially available in Brazil. Misdiagnosis through this method, when it does occur, is due to a systematized failure to identify the anatomical landmarks and to acquire images in the standard plane. However, these arguments against Graf's method have progressively been deconstructed nationwide as courses, in which the method is presented and demonstrated to help interested doctors carry out the examination, have been developed and offered by the Graf Institute and medical societies. The Graf Institute (<https://institutograf.com.br>) was created in 2022 by a group of professionals to facilitate the diagnosis, disseminate information about DDH and promote courses to teach doctors from all over Brazil how to perform ultrasound using Graf's method. It also presents educational courses on how to treat children with DDH. Above all, it supports families and guides children's parents and caregivers.

It is estimated that 7,759 ultrasound devices are needed to meet the demands of the SUS. There are approximately 50,000 ultrasound devices in Brazil [25], 28,030 of which are used by SUS [26]. Despite the sufficient number of appliances with a mean of 17.83 ± 5.04 devices per 100,000 persons, there is an inequity in the distribution among the regions of the country (26.65/100,000 in the Federal District of Brasília compared with 8.71/100,000 in the state of Amazonas). There is also disproportionality in the distribution of ultrasound devices between the public and private sectors, for which the national averages are 20.25% and 79.75%, respectively [27].

The inequality between the public and private healthcare sectors, along with the regional disparities, impedes the early diagnosis of DDH; some patients inevitably have difficulty accessing ultrasound examinations and receiving appropriate treatment. Also, those who may not have a geographical barrier to receiving an ultrasound examination may still experience a delay in diagnosis due to long wait times in SUS. Although several centers in Brazil have adopted Graf's method, it is not always performed appropriately, as has also been reported in several European countries [12]. Currently, there is a pressing need for additional training of the professionals involved.

Challenges of physician training in DDH care in Brazil

Graf's method, although relatively simple and reproducible, requires extensive training for reliable appraisal as the image of the hip must be acquired in a specific spatial plane and the anatomical landmarks must be properly identified [28]. Those requirements can be challenging, especially for less experienced examiners—some studies have demonstrated low levels of intraobserver and interobserver agreement [29].

Brazil has different realities. In large centers, there are radiologists, pediatricians, obstetricians, and pediatric orthopedic surgeons who are well-trained and are aware of DDH, carrying out the diagnosis and treatment of the condition appropriately. However, in other regions, no professionals are qualified to carry

out those tasks. For example, de Oliveira et al. [30] showed that in the small city of Itanhém (in the northeastern state of Bahia, the fourth most populous state in Brazil), 99.3% of hospitalizations for DDH between 2011 and 2021 were of patients ≥ 15 years of age, the remainder being of patients 9–14 years of age. During that same period in Salvador, which is the largest city in Bahia, 71.9% of hospitalizations for DDH were of patients ≤ 14 years of age, indicating that diagnostic performance is better and patients receive treatment earlier at large centers. However, even when professionals are accessible, they often do not have the necessary knowledge about DDH. In 2016, Souza et al. [24] assessed knowledge of DDH by professionals involved in its diagnosis and found that, among pediatricians and orthopedists, as well as residents in those fields, 12.8% believed that, if left untreated in the early stages of life, severe DDH has a good prognosis, with minimal sequelae and no need for surgery. Likewise, 20.9% of the respondents were unaware of the maneuvers for diagnosing DDH in a physical examination.

The difficulty in training health professionals to diagnose DDH is seen in countries other than Brazil. At healthcare facilities in the city of Kahramanmaras, in Turkey, Uzel et al. [31] assessed physician knowledge regarding DDH before and after a lecture and found that, before the lecture, 18.6% of the participants did not recognize DDH as a preventable disease with controllable consequences. According to the German National Association of Statutory Health Insurance Physicians [32], 48% of incorrect ultrasound diagnosis of DDH are due to failure to identify the relevant anatomical landmarks. Walter et al. [12] carried out a review of 131 studies from 25 countries to assess whether the objective quality criteria established for the Graf's method were met and concluded that only 68 (51.9%) of those studies presented ultrasound images that were considered to have met Graf's criteria. It is noteworthy that in Germany, Austria, and Switzerland, hip ultrasound can be performed only with a special license obtained by completing a course with instructors certified and authorized by Professor Graf or one of his representatives [33].

Conclusions

In 2020, Duarte et al. [34], with a team of pediatric radiologists and pediatric orthopedic surgeons, conducted a systematic review and meta-analysis on the topic of DDH to determine the detection rates for and diagnostic accuracy of various ultrasound techniques, using the Graf's method as the reference. In their systematic review, they analyzed fifteen studies in which a collective total of 16,736 hips were evaluated and concluded that Graf's method is the most appropriate choice for diagnosing DDH.

In a systematic review of DDH conducted by Harper et al. [35], the four studies that compared universal ultrasound screening (for all newborns) and selective ultrasound screening concluded that, in the United Kingdom, the former would be more cost-effective than the latter because it would reduce the rates of late detection and surgery. Similar conclusions have been drawn from studies conducted in countries such as Germany, Austria, and Switzerland [33], suggesting that DDH screening in Brazil should be carried out by using the Graf's method in the entire neonatal population. The economic impact of screening programs is multifactorial and requires careful consideration on the part of governments. The costs of neonatal screening may be offset by the savings associated with early diagnosis, favorable outcomes, and the avoidance of surgery. As of yet, no such economic analysis has been conducted in Brazil. The physical and psychological benefits for newborns receiving early diagnosis and treatment are pronounced and should also be considered, although they are difficult to quantify objectively in a research setting [35].

It is imperative to develop a neonatal DDH screening program, similar to the existing Brazilian National Neonatal Screening Program, and train professionals to use Graf's technique as a diagnostic procedure. Plans should be made to scale up the current program from selective screening to universal screening in the long term [24]. The majority of studies conducted in Brazil have been concentrated in the city of São Paulo. Therefore, there is a need for additional studies conducted in other cities and regions of the country. In addition, the structure of the ultrasound reports should be standardized to improve the available evidence [7].

The goal for Brazil is to achieve a radical reduction in the need for surgical treatment of DDH, such treatment typically being performed in infants older than 1 year of age. This can be achieved by greater

integration of medical societies involved in the care of pediatric patients, the government, and non-governmental organizations. The Graf Institute is committed to training more professionals on the sonography of the hips of infants to promote successful conservative treatment at the appropriate age.

Abbreviations

DDH: developmental dysplasia of the hip

SUS: Sistema Único de Saúde

THA: total hip arthroplasty

Declarations

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Author contributions

GGBM and MLD: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Validation, Visualization, Writing—original draft, Writing—review & editing. NVMR and PMG: Investigation, Methodology, Project administration, Software, Supervision, Validation, Visualization, Writing—original draft, Writing—review & editing. SRB: Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Validation, Visualization, Writing—original draft, Writing—review & editing. AFdL: Formal analysis, Funding acquisition, Resources, Software, Writing—review & editing.

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The authors declare that they have no conflicts of interest.

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