International Interdisciplinary Consensus Meeting on the Evaluation of Developmental Dysplasia of the Hip

Internationales interdisziplinares Konsensustreffen zur Evaluation der Diagnostik und Therapie der angeborenen Hüftdysplasie

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ABSTRACT
In September 2018, an international meeting of doctors of various disciplines, with expertise in the detection and treatment of DDH, was held in Csolyospalos, Hungary. The aim was to achieve consensus on the detection and early treatment of the condition and to develop a standardized system of teaching and training for hip ultrasound. There was strong agreement that US screening is essential. Specifically the Graf technique was selected as the technique of choice. Universal US screening was strongly favored. Screening should be carried out as soon as possible, but not later than the sixth week of age. US screening is cost-effective, does not result in overtreatment, and contributes to a reduction of long-term consequences. The essential principle of treatment is timely application of a device to achieve reduction, retention and maturation, by holding the hips in flexion, and a safe degree of abduction. It was agreed that the effectiveness of any screening policy depends on the correct scanning technique. Therefore, standardization of teaching and training of the
Introduction

Developmental dysplasia of the hip (DDH), formerly known as congenital dislocation of the hip (CDH), is a condition that includes a wide spectrum of severity, which, if left untreated, may cause significant long-term complications (including intra-articular lesions in the pre-arthritic stage and early onset osteoarthritis [1, 2]) thus leading to severe disability during early adult life.

Most cases of DDH, if diagnosed and treated early, are potentially reversible. All aspects of DDH have long been a source of debate, and universal agreement has not been reached. There is disagreement regarding the etiology and pathogenesis (genetic vs. mechanical), the preferred diagnostic approach (clinical vs. ultrasound (US)), the most effective screening policy (selective vs. universal) and the appropriate treatment approach.

Screening by US is well established as a critical element in the evaluation of the infant hip. However, there are many different techniques in use [3–6]. The value of US screening is dependent on adherence to correct technique which underlines the critical importance of standardized training, audit and quality control [7].

The aim of this meeting was to achieve consensus on all of the above-mentioned issues, to propose an evidence-based approach to the problem of DDH, and to develop a system for early detection, diagnosis and treatment.

Meeting background

The meeting was held in Csolyospalos, Hungary in September 2018. 24 medical doctors (radiologists, orthopedic surgeons, pediatricians, general practitioners), with expertise in DDH diagnosis and management, were invited to participate. Several countries were represented, including Austria, Cyprus, Germany, Greece, Hungary, Iran, Ireland, Italy, Switzerland, Turkey and the UK.

Participants prepared a summary of the current evidence on specific topics, which then served as the basis for discussion. Questionnaires about the most important points were then distributed to the participants, who had to vote either for or against the proposals.

Depending on the proportion of votes received, the strength of the recommendations was recorded as follows:

- a) strong agreement supports the recommendation (≥ 90 %)
- b) general agreement supports the recommendation (≥ 70 % – < 90 %)
- c) general agreement weakly supports the recommendation (≥ 50 % – < 70 %)
- d) there is not enough evidence to support the recommendation (< 50 %)

Following the meeting, the moderators of each session provided a summary of their topics which is summarized in this paper, under the following headings:

- Terminology
- Etiology and pathogenesis
- Epidemiology and risk factors
- Detection of DDH
- US examination and techniques
- Timing of DDH
- Selective vs. universal US
- Treatment
- Training/auditing/accreditation
- Future plans

Terminology

The term “developmental dysplasia of the hip” (DDH) (dysplasia: Greek word meaning “abnormal growth or development of an
organ or tissue”) [8] has replaced congenital dislocation of the hip (CDH) (Klisic, 1984), [9]. There are two reasons for this: (a) it better describes the spectrum of pathology [10, 11], which ranges from simple immaturity to complete dislocation of the hip joint; (b) it introduces the term “developmental” (instead of “congenital”), implying that the disorder can develop even after birth.

Developmental dysplasia of the hip (DDH) is in fact a spectrum of disorders of the developing hip [12, 13], encompassing:
1. Hip abnormalities found on US and/or radiography without any clinical findings. Left untreated, these may present later as a frank dislocation.
2. Hip instability, such that the femoral head can be dislocated partially or fully from the acetabulum by an examiner, but relocates spontaneously.
3. Dislocated but reducible hip.
4. Dislocated hip that cannot be reduced.

**Etiology and Pathogenesis**

Many theories have been proposed to explain the etiology and mechanism of DDH.

Embryologically the femoral head is fully formed with a spherical configuration, a short neck and a primitive greater trochanter at 11 weeks [14]. With the subsequent development of the capsule, the femoral head is held centered in the acetabulum by the surface tension of the synovial fluid. Further interaction between the femoral head and the joint cartilage results in the normal development of the hip joint both antenatally and postnatally. The etiology of DDH is thought to be multifactorial; a combination of genetic/constitutional [15] and mechanical factors [16, 17] is believed to be involved in the pathogenesis of the disorder.

The higher frequency of dislocations in females, familial/ethnic predisposition and geographical variation [18] support the genetic theory [19]. Abnormal forces transmitted to the femoral head, either before birth (breech presentation, left-sided predominance, etc.), at delivery (vaginal birth with breech presentation) or after birth (swaddling effect) constitute the basis of the mechanical theory [14, 19].

Regarding breech presentation, a study in 2010 [20] compared the incidence of DDH between breech twins (all with flexed hips and knees) and breech singletons (more of whom had flexed hips and extended knees); the breech singletons had a significantly higher incidence of DDH, supporting the specific importance of breech position with extended knees (frank breech).

**Detection of DDH**

Clinical examination of the newborn can detect hip instability but not acetabular dysplasia. The development of US has advanced our understanding of the normal development of the hip and allows monitoring of the dislocated, unstable and dysplastic hip [31 – 35].

Clinical examination is not sensitive enough to identify every child with DDH. The reported specificity is 90%. The sensitivity, however, is reported as being as low as 50%. US has been proved to be more sensitive and more specific for the diagnosis with significantly higher reported sensitivity and specificity (> 90%). The latter arguably is an underestimation, if we exclude sonographers who have not been properly trained [36 – 40].

Other imaging modalities (X-ray, CT, MRI) are not routinely used for the detection of DDH. X-ray, which has been the main-
US examination techniques

The commonly known US techniques used worldwide are the Graf technique which is mainly used in Europe [3], the Terjesen technique [4] originating in Scandinavia (Terjesen et al, 1989), the Harcke technique developed in the United States [5] and the Suzuki technique developed in Japan [6]. A detailed description of the abovementioned techniques, is beyond the scope of this article.

A comparative literature review of these techniques was carried out via PubMed and seven papers were identified that compared two or more of these methods with each other. 

Diaz A et al. [46] examined 208 babies (416 hips) with the Graf, Harcke and Suzuki techniques. Overall the three methods correlated well with regard to the severity of the pathology, but they found the Graf technique to be the most reliable.

Czubak et al. [47] compared the Graf and Terjesen techniques in 657 newborns (1312 hips) with a mean age of 23 days. The authors found that both techniques had similar results, but favored the Terjesen technique, as more specific and simpler. It is worth mentioning that in this study, 2 hips that were classified as Type III hips according to Graf, were classified as normal according to Terjesen.

Langford et al. [48] compared the Graf and Terjesen techniques and concluded that sonographers need to be trained in the Graf technique in order to get accurate results.

Falliner et al. [49] compared the Graf and Terjesen techniques in 232 neonates under 4 days old. In order to determine the reproducibility of the methods, 50 hips were evaluated by two skilled examiners. Statistical comparison showed a good correlation between the two techniques and proved no obvious difference in regard to inter-observer reliability. However, the Graf technique offered better reproducibility and intra-observer reliability.

Peterlein et al. [50] examined 207 newborns with both the Terjesen and Graf techniques and investigated the inter- and intra-observer reliability for the measurements of alpha angle, beta angle and femoral head coverage (FHC). They found better reliability for the alpha angle, followed by the beta angle and finally by the FHC, regardless of the experience level of the investigator.

Pacheco et al. [51] investigated 225 infants at a mean age of 7.79 weeks with the Graf, Harcke and Terjesen techniques and calculated the specificity and sensitivity for each. They found the Graf technique to be the most sensitive and specific, and proved that the alpha angle was the most reliable criterion on which to base a treatment decision.

Kotlarsky et al. [52] published a review article in 2015, comparing the Graf, Suzuki and Harcke techniques. Their conclusion was that the Graf technique fulfilled the requirements for an effective US technique, offering simple, precise, quantitative and consistent definitions for examination and diagnosis.

Comparative presentation of the results of the abovementioned studies are presented in Table 1.

In summary, our literature review strongly suggests that the Graf technique seems to be superior to the other techniques. A recent review has also reinforced this position [53].

The Graf technique correlates the pathology and severity of acetabular dysplasia, with the patient’s age, thus offering a patient-specific diagnostic and treatment algorithm [7]. The scanning technique per se is standardized, and its application and principles have been thoroughly described [54]. The value of structured training by authorized teachers has been recognized [55]. Mistakes may be avoided by strict adherence to the technique, so only appropriately trained and certified sonographers should be allowed to scan. Continuous quality control and audit should also be offered [7].

The technical details of the technique were also thoroughly discussed during the meeting. Adoption of the scanning principles (including equipment, technique and documentation) as presented in detail in the teaching manual [54] is mandatory.

Timing of US screening

Several papers make the point that closed treatment of DDH is more successful if instituted earlier [56–59]. From this, the view
may be taken that the earlier the scan is carried out and any necessary treatment is begun, the better.

On the other hand, some authors have expressed concern regarding overdiagnosis arising from scanning occurring very early [23, 60]. In the German language literature, Grill and Müller [61] similarly expressed concern about overdiagnosis, when scans are performed in the first week.

There is a range of recommendations in the literature regarding the time to scan. The German language literature generally recommends scanning before six weeks of age [62–65]. There is quite a variation in the English language literature. Tyagi et al. [66] described scanning at an average of 5 weeks. Bacche et al. [67], Imrie et al. [68] and Clarke et al. [27], all described scanning at 6 weeks. Kolb et al. [69] and Schaeffer et al. [70], both recommended scanning at 6 to 8 weeks. Choudry et al. [71] recommended scanning babies with unilateral limitation of abduction at 8 weeks. Pillai et al. [72] reported that the specificity and accuracy of US with the Graf technique was at its highest at 3 months of age.

Some authors have made a distinction between babies who have clinical signs of instability and those who are being screened without any clinical abnormality [27, 73].

To summarize, the picture which emerges is that earlier diagnosis facilitates earlier treatment, which is more successful, but it has been suggested that scanning too early can risk overtreatment, as many hips which appear immature at first will mature later. There is support for early scanning in babies with clinical signs. Otherwise, in the German language literature, the recommendation is to scan before six weeks of age, while in the English language literature, the range of recommended ages for scanning babies without clinical signs goes from just under six weeks to three months.

At this meeting, it was pointed out that the standard practice in Austria is to scan the baby’s hips twice: first at birth and then again between the 4th and 7th week of life. This allows treatment to begin as soon as possible, and therefore more successfully. However, even if dysplasia is only detected on the second scan, between the 4th and 7th week, the hip is still within the maturation curve. There was strong support for the view that, given the maturation process of the hip, the earlier a problem is identified and treated the better. It should be possible for a trained examiner to distinguish between clearly abnormal hips and those which may simply be immature and can be followed up, thus avoiding the danger of overtreatment.

### OUTCOME

There was strong agreement at this meeting that US screening should be carried out as soon as possible, but no later than the sixth week of life. (A)

### Selective vs. universal US screening

The literature surrounding this question is very varied. To some extent, it reflects an evolution of understanding over time, but also variation in practice between different countries.

There have been some reviews that cast doubt upon the whole practice of US screening [11, 74–76]. A Cochrane review [75, 76] concluded that neither US strategy had been demonstrated to improve clinical outcomes, including late diagnosed DDH and surgery. A Canadian review in 2001 [74] concluded there was “fair evidence” to exclude both “general US screening” and “selective screening”. Shipman et al. [11], based in the USA, carried out a meta-analysis, in which they were critical of the literature on screening and interventions for DDH.

Leaving these criticisms of the evidence in the literature aside, the debate about universal vs. selective screening revolves to a large extent around effectiveness, cost, and the possibility of overdiagnosis and overtreatment.
Several papers have claimed good results with a regime of selective US screening [27, 77–80]. In a Norwegian study, Holen et al. [77] argued based on their findings that "when clinical screening is of high quality", universal US was not necessary. Instead they favored selective screening based on clinical findings and risk factors. Mahan et al. [78] in the USA described an analytic model and they argued that the best approach was to clinically screen all neonates and then to use US selectively for infants at high risk. Laborie et al. [79] from Norway referred to a regime of selective screening resulting in an "acceptable" rate of early treatment and low rates of subluxated/dislocated hips. In an Australian study Leba et al. [80] highlighted the cost of screening but showed good results from selective screening. Clarke et al. [27] in the UK also showed good results using a system of selective screening based on breech presentation, family history and foot deformity.

However, there have been a number of studies pointing to patients with DDH who would have been missed by a selective US screening regime [67, 81, 82]. A review in the USA [82] looked at skeletally mature patients who had had surgery for the late consequences of DDH. 85.3% of them would not have been screened in a selective US screening program.

Starting in the early 1990s, evidence of the potential of universal US screening began to be published in the English language [83, 84] and since then a number of papers reinforcing this have been published [56, 69, 85–88]. In 2004 Wirth et al. [85] from Germany reported that a program of "general" US screening had resulted in a dramatic decrease in surgical procedures, hospitalizations and late presentation. Tschauner et al. [56] from Austria reported that a cohort of babies who had come through a universal screening program had a 98.9% success rate with treatment. A year later, von Kries et al. [86] from Germany reported a reduction in the rate of operative procedures due to "general" US screening, but cautioned that further assessment was required to balance this benefit against potential overtreatment and adverse effects.

Reporting on the outcomes of the Austrian universal screening program, Thalinger et al. [87] showed a significant decrease in the rates of open reduction, pelvic surgery, and hospital admissions for DDH. Kolb et al. [69], also from Austria, observed that risk factors had a low impact within their group. Therefore, they were also in favor of universal screening. Guler et al. [88] from Turkey reported on their population of babies who underwent US screening with satisfactory results and thus recommended its use.

At this meeting, a speaker from Ireland described how a system of US screening for DDH had been initiated over the previous four years, starting from a highly variable situation in the country. A system of selective US screening was now almost in place. At this time, resources were not available for universal screening. However, an added dimension in Ireland was an extremely litigious climate, in which other health service screening programs had been subjected to legal action because of false negatives. This meant that caution was necessary in initiating any screening program, while making sure the objectives were realistic and clearly communicated.

The Swiss team presented the design and application of a nationwide universal screening program, which was introduced under the name "The Mongolian Project" [89–91]. The project, which included more than 300,000 scans and over 4000 successfully treated children, was commended as a very worthy and successful initiative. It was seen as a good example of how a universal screening, early-scan and early-treatment national policy can be successfully implemented nationwide, even when healthcare resources are scarce.

A presenter from Austria described how universal US screening had been established in that country since 1992, and had been shown to be successful and cost-effective. Universal US screening has been in place in Germany since 1996. The German language literature supports this position, in terms of reduced late diagnosis and reduced rates of late surgery related to dysplasia [61, 63, 92].

One of the presenters at this meeting, having practiced both in Austria and in the UK, reported on the striking contrast between the two systems. In the UK, where selective rather than universal US screening is employed, there is an incidence of late diagnosis of DDH which is almost unknown in Austria. Over a four-year period, one pediatric center in the UK had 114 patients requiring operations for DDH. 91 of these were late presentations (63 with no risk factors). 62 patients had open surgery, and 52 had closed reduction and spica application, with 8 subsequently requiring open surgery. It was stated that there appeared to be resistance in a system employing selective screening to moving to universal screening despite the evidence, probably largely due to issues regarding logistics and resources.

One issue for debate is whether the benefits of universal US screening justify the cost. Paton et al. [93] from the UK called its value into question, suggesting that it could not be justified on the grounds of cost. However, Clegg et al. [84], also from the UK, showed as far back as 1999 that the cost appeared justified in terms of the treatment savings. Further UK studies [84, 95] suggested that universal screening did not incur increased costs.

More recently, Thaler et al. [96] from Austria demonstrated that the cost of a universal US screening program was more than offset by the costs of surgical and non-surgical treatments that had been avoided, and that therefore the policy was indeed cost-effective. The German language literature supports this position. Ihme et al. [92] compared the costs of hip surgery due to DDH at two different time periods (patients who received US and a historical group) and demonstrated that the costs of hip US were lower than the costs of hip surgery. This position was further supported by the study by Farr [65].

At this meeting, the presenters expressed the view that a policy of universal US screening is indeed cost-effective, citing the studies by Clegg et al. [84] and Thaler et al. [96]. This is true, particularly when we include the prospective costs, including late surgery which is avoided by early diagnosis and treatment. The difficulty is in making the case for the initial investment, based on the expectation of overall cost savings in the longer term.

Another concern which has been expressed is the possibility of overdiagnosis and overtreatment, with the associated risk of avascular necrosis [29, 60, 66]. Most notably, in their guidelines published in 2016, the American Academy of Pediatrics [29] recommended against universal US screening in North America, citing "expense, inconvenience, inconsistency, subjectivity, and high false-positive rates."
However, the point has been made that a universal US screening system can actually reduce the non-operative treatment rate since treatment is based on objective rather than subjective clinical criteria [97, 98].

At this meeting, the prevailing view was the latter, i.e. that the objectivity provided by US screening that is carried out and interpreted correctly, should not result in babies being treated unnecessarily. However, some attendees observed that in a system in which babies are treated according to US findings at an early age, overtreatment might happen.

Another area of expressed uncertainty concerns later outcomes of babies with hip dysplasia [56, 99]. Thalinger et al. [87], reporting on the late outcomes of the Austrian screening program, did show a decrease in the frequency of late pelvic osteotomies, quoting in particular the findings of Grill and Muller [61] and Thalinger et al. [87].

In summary, while the German language literature is supportive of universal US screening, the English language literature presents a mixed picture. To a large extent this is reflective of geographical variation. It is clear that universal US screening results in a low rate of late diagnosis. What varies between studies is the extent to which this approach outperforms selective screening, and whether this extra benefit is offset by cost, or the possibility of overdiagnosis and overtreatment, with its attendant risk. Some believe that the degree to which this lowered rate of late diagnosis translates into a reduction of the sequelae of dysplasia at skeletal maturity also remains to be established.

At this meeting, there was strong agreement that a system of universal US screening results in a reduction in the need for later pelvic osteotomies, quoting in particular the findings of Grill and Muller [61] and Thalinger et al. [87].

In summary, while the German language literature is supportive of universal US screening, the English language literature presents a mixed picture. To a large extent this is reflective of geographical variation. It is clear that universal US screening results in a low rate of late diagnosis. What varies between studies is the extent to which this approach outperforms selective screening, and whether this extra benefit is offset by cost, or the possibility of overdiagnosis and overtreatment, with its attendant risk. Some believe that the degree to which this lowered rate of late diagnosis translates into a reduction of the sequelae of dysplasia at skeletal maturity also remains to be established.

At this meeting it was stated that this variation in findings reported in the literature was probably due to studies not being comparable. Techniques could vary, the scans could be carried out by individuals with varying levels of training, the definition of “risk factors” could vary, the quality of clinical screening could vary and different outcome measures may have been used.

In addition, the definition of “cost” could vary depending on what factors were included. The consensus group emphasized two points:

Firstly, there should be a differentiation between clinical effectiveness and cost-effectiveness. The former is what healthcare providers should aim to practice and must be the gold standard, whereas the latter (cost-effectiveness) is a compromise and should be honestly explained to parents and to the public.

Secondly, all studies that assessed cost-effectiveness compared the cost of a universal screening program to the cost of surgical treatment for DDH. None of the studies included long-term effects such as quality of life, the need for future hip replacement, and loss of the ability to work. The published German, Austrian, Swiss and Coventry studies demonstrated that although there is a marginal increase in the universal screening cost, it is a good value for the money.

Reference was made to a recent report by Biedermann and Eastwood [98] from Austria and the UK, respectively. This was a joint review calling for a “paradigm shift” towards universal US screening, stating that it results in the lowest late presentation rates, low treatment rates, a lower rate of surgery on the infant hip, and a reduction in the rate of surgery for DDH in later life. They also make the point that avascular necrosis can be avoided, and that modern orthoses should have a zero rate of AVN. The dominant outcome of the discussion was one of support for this “paradigm shift”.

### OUTCOME

There was strong agreement in favor of universal US screening. (A)

There was strong agreement that, when all short- and long-term costs are taken into account, a system of universal US screening is cost-effective. (A)

There was general agreement that a system of universal US screening using the Graf technique would not result in overtreatment. (B)

There was strong agreement that a system of universal US screening using the Graf technique will result in a reduction of later problems with dysplasia. (A)

### Treatment

According to the classification system developed by Graf [41, 54], Type I hips are mature hips, so they do not require any treatment. Type IIa hips are immature hips and as long as their degree of maturity is age-appropriate, treatment is not required. However, they should reach maturity by three months of age. Otherwise, they are re-classified as Type IIb and require treatment. Treatment is required for any hip that is classified as Type IIa minus or worse. Therefore, Type IIc, D, III & IV hips always require treatment.

**Table 2** summarizes the abovementioned classification/treatment scheme.

DDH treatment according to Graf consists of (a) femoral head reduction, (b) maintenance of head relocation (retention) and (c) correction of any residual acetabular dysplasia (maturity).

Early initiation of treatment is strongly recommended as a substantial part of maturation of the hip joint occurs within the first three months. By six weeks, the baby’s hip is already half-way along this steep part of the maturation curve. Therefore, it is very important for treatment to be initiated before six weeks of age. Correct use of an abduction device is the mainstay of treatment. A combination of flexion (110°) and abduction (45°) prevents avascular necrosis of the femoral head, as this position avoids excessive pressure on the femoral head against the hyaline cartilage of the acetabulum.

The essential elements of early treatment are:

a) Reduction: This is required in Types D, III and IV. Provided treatment is initiated early enough, this should be achieved by the position of flexion and a safe degree of abduction as has been described above. Treatment position may be reached by a spica cast, although in newborns some centers prefer the use of an external harness.

b) Retention: This is required in Type IIc unstable and in Type D, III and IV hips that have been effectively reduced. Retention is
Training, auditing, accreditation

It was recognized at this meeting that teaching of the Graf technique must be standardized. Who is trained will vary from country to country, but at a minimum, it must be a health care professional. In order to help with this standardization, a set of slides was prepared and presented. They summarize the theoretical content of the basic course. Future work on these will include further refinement of the slides and translation into languages other than English.

Regarding future development of the course, there was support for including a section on the basic physics of US and for making the theory of the course available online prior to attendance.

Practical sessions should include all theoretical and practical aspects of the technique, including scanning demonstration on a phantom and/or babies.

The ideal duration of the course should be two days.

Regarding further training, it was agreed that the update/accreditation course should only be taken by those who have done the basic course, normally at least six months previously. This course should also be standardized. Further issues, to be addressed at future meetings, include required practical training and experience following the basic course, accreditation of instructors, and re-accreditation.

Another area for future development will be the formation of national committees whose roles would include approval of training courses, examiners and instructors, re-accreditation of examiners and instructors, and quality management. Such committees would in turn connect with the international organization (ICODE – the International Interdisciplinary Consensus Committee on DDH Evaluation).

Future plans

The group has now been formalized as the International Interdisciplinary Consensus Committee on DDH Evaluation (ICODE). The objectives of the group are:

- To work towards consensus on the various issues regarding DDH, in particular policies for detection and early treatment.
- To promote, standardize and continually improve teaching and training in the Graf technique of infant hip US.
- To use its international network to support research, audit and quality improvement in the field of detection and early treatment of DDH.
An ICODE website has been set up and is being developed. It can be accessed via www.icode.expert

The group will continue to meet, with the next meeting scheduled for Heraklion, Crete, in May/June 2019. As it develops, the group will aim to widen its efforts internationally with the goal of achieving the best possible outcomes from DDH worldwide.

Conflict of Interest

The authors declare that they have no conflict of interest.

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