Is it worthwhile to screen the hip in infants born with clubfeet?

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Abstract

Purpose There is some disagreement about whether idiopathic congenital talipes equinovarus (CTEV) increases the risk of neonatal developmental dysplasia of the hip (DDH). This study aimed to investigate the incidence of DDH in our infants with idiopathic CTEV.

Methods We conducted an observational cohort study over a three-year period to assess the relationship between idiopathic CTEV and DDH. All neonates younger than six weeks with idiopathic CTEV who were treated in our medical centre were admitted to this study. Each subject underwent hip ultrasound examination using the Graf method at the age of six weeks. DDH was diagnosed when a hip was type IIa(–) or worse according to the Graf classification of sonographic hip type.

Results A total of 184 patients were diagnosed with idiopathic CTEV and underwent hip sonography. In total, seven hips of five individuals underwent treatment (four girls and one boy).

The results indicated that 2.7 % of babies (five of 184) with idiopathic CTEV had DDH. However, we did not find any statistically significant difference ($p=0.5776$) in the Pirani scores between the DDH group and group with normal hips. **Conclusions** This study revealed that the idiopathic CTEV group had a greater incidence of DDH in comparison with the general population. It is recommended that hip sonography be undertaken particularly in patients with idiopathic CTEV.

Keywords Idiopathic clubfoot · DDH · Infant · Hip sonography · Graf method · Ultrasound

Introduction

Congenital talipes equinovarus (CTEV), also known as clubfoot, and developmental dysplasia of the hip (DDH) are both

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common paediatric orthopaedic conditions, although the aetiology of both conditions remains unclear. An association between these two conditions has been proposed, but no consensus has been reached [1–8]. Wynne-Davies [7] reported that only one of 165 patients with CTEV had DDH. Paton and Choudry [4] identified seven patients with type II DDH among 60 children with CTEV (11.7 %) using ultrasound examination. The hips from these seven patients progressed to normal hip status without any treatment. In another similar study, only one DDH patient was found among 349 patients with CTEV [6]. In these studies, the incidence of clubfoot did not lead to an increased incidence of DDH. On the other hand, the results from some other studies have suggested an association between clubfoot and DDH [1, 2]. Carney and Vanek [2] identified eight children with radiographic signs of hip dysplasia among 51 children with clubfoot who underwent surgical release, while another study reported that 7.7 % of children with idiopathic clubfoot required treatment using the Pavlik harness [1]. Lochmiller et al. [3] reported that 2.4 % of their population with CTEV had DDH. Yang et al. [8] recognised 12 DDH patients among 801 patients with clubfoot. DDH has a wide spectrum of pathology from complete dislocation to mild dysplasia, which may develop to a more severe condition after the child starts to walk. In terms of clinical examination, therefore, it is more difficult to identify DDH during the neonatal period than clubfoot, which is a visual deformity and can be recognised at birth. In terms of radiological examination, it is also difficult to find the mild and even moderate forms of DDH at the early neonatal stage because of the poor ossification of the femoral head and acetabulum. However, the presumed correlation between CTEV and DDH may provide a target population for hip screening, especially for the cases in which mild DDH is suspected.

Currently, with the widely accepted Graf method [9] in Europe, hip sonography has become the most effective and popular means of early DDH detection in a number of countries, such as Austria, Germany, Switzerland, Norway, the UK, France and Hungary [10]. Universal screening has been adopted in Germany, Austria and Switzerland, while hip screening selectively in patients with one or more risk factors has been applied in the UK, Norway and France [10]. Selective hip ultrasound screening, which is directed towards the patients with DDH risk factors, is more suitable for countries with a large neonate population. To test the hypothesis of a higher incidence of DDH in the population of CTEV patients, hip sonography was performed in all of the CTEV patients using the Graf method in our medical centre over the past three years (2010–2012). The purpose of this study was to investigate the incidence of DDH in CTEV patients in our population and to assess the feasibility and efficacy of hip sonography and management of DDH in neonatal CTEV patients undergoing treatment with the Ponseti method.

Patients and methods

We reviewed a consecutive series of patients with CTEV who were treated at our medical centre between January 2010 and December 2012 and were younger than six weeks of age at their referral. The inclusion criteria included: (1) clinical diagnosis of idiopathic CTEV, (2) the absence of any other abnormalities and family history and (3) no previous treatment. The demographic data included sex, laterality, date of birth, gestational age at the time of delivery and the Pirani score for assessing the severity of deformity. Risk factors for DDH such as breech position, family history, oligohydramnios or birth order were also recorded. The diagnosis and Pirani score prior to treatment were determined by a single consultant paediatric orthopaedic surgeon (LZ). When performing the ultrasound examination, premature birth should be taken into account and the timing of examination and measurement should be based on the infant's corrected gestational age. The definition of premature in our study was that the baby was born at less than 37 weeks of gestation. If the child was born preterm, the corrected age, which was obtained by subtracting preterm days from actual age, was used in our study. All neonates with CTEV underwent routine clinical examination at the initial visit and hip ultrasound examination at the age of six weeks. The hip was graded using the Graf classification [11–16]. The maturation of the neonatal hip was assessed according to the maturation curve in the Graf method of hip sonography. The concept of linear maturation was taken to simplify this assessment by setting the threshold beyond the border of suspected condition so that misdiagnosis would be almost impossible. In a newborn, 50° is the minimum value of the α angle in the hip which can be expected to become a type I hip by the end of the third month of age [11–16]. Ossification progresses in a linear pattern from birth to 12 weeks of age and must reach a certain degree of ossification by each week. A hip joint that attains this minimum degree of maturation or more is regarded as type IIa(+). For example, 55° or more indicates a type IIa(+) hip in a six-week-old baby. If the value of the α angle is less than 55°, for example in a six-week-old baby, the hip is regarded as the lagging one, e.g. type IIa(–) [11–16]. According to the therapeutic implications of the Graf method [16], the hip, as type IIa(–) which does not attain a minimal degree of maturity or worse, requires treatment. In our study, DDH was defined as a hip of type IIa(–) or worse, which was taken as an abnormal case. All type II hips underwent repeat ultrasound examination at 12 weeks of age; if a child continued to have a type II hip or worse, he or she underwent ultrasound examination again at six months. The sonographic examinations were performed and the sonograms were collected by two sonographers (YC and LL) in this study. These two sonographers learned the method under the instruction of Dr. Graf in 2008 and were qualified in the practice of the Graf method of hip sonography. Statistical significance

was obtained by the chi-square test as to the number of CTEV patients distributed yearly. The Wilcoxon rank sum test was used to compare the data of Pirani scores between DDH patients and the other patients. A p value of <0.05 was considered significant.

Results

Between January 2010 and December 2012, 184 patients were diagnosed with idiopathic CTEV and were treated at our medical centre using the Ponseti method (Table 1). There was no statistical difference in the distribution of patient numbers at each year during this period of the study ($p=0.9785$). There were 137 boys (74 %) and 47 girls (26 %). Of the patients, 91 (49 %) had bilateral clubfeet, while 93 (51 %) had unilateral clubfoot, including 56 on the right side and 37 on the left side. Most children (95 %) were full term, without complications during gestation or delivery.

Of the 184 children who underwent clinical examination and hip sonography, none had clinical signs of hip dysplasia. There were 40 hips categorised as type II or worse, including 33 type IIa(+) hips, three type IIa(−) hips, three type IIc and one type III hip, based on the sonographic assessment at six weeks of age (Table 2). The 33 type IIa(+) hips were regarded as benign and the other seven hips of five babies (four girls and one boy) without any risk factors for DDH, which were classified as type IIa(−) or worse, were regarded as abnormal. Three type IIc and one type III hip were prescribed treatment using a Pavlik harness or Otto Bock splint for hip abduction, while three patients with type IIa(−) hips were just advised to keep the hip in flexion and abduction as far as possible. All 40 hips that were type II or worse underwent repeat ultrasound examination at 12 weeks of age, when all of the type IIa(+) hips develop into type I hips and there were six hips that remained type II or worse, including four type IIb hips, one type IIc hip and one type III hip (Table 3). All of these six hips came from those five babies with abnormal hips at six weeks of age (Table 4). Only one type IIb hip still required treatment at six months, although one child was lost to follow-up (Table 4). The mean Pirani score of DDH patients and the

Table 1 Observed distribution of CTEV patients younger than 6 weeks of age seen at our medical centre during the 3-year study period

Year	Cases
2010	62
2011	62
2012	60
Total	184

There was no statistical difference in the distribution of patient numbers at each year during the period of this study ($p=0.9785$)

Table 2 Hip grading with the Graf classification at 6 weeks of age

Hip type	No. of left hips	No. of right hips	No. of babies with corresponding hip type
I	162	166	159 ^a
II			
IIa(+)	18	15	20 ^b
IIa(−)	1	2	2 ^c
IIc	2	1	2 ^d
D	0	0	0
III	1	0	1 ^e
IV	0	0	0
Total	184	184	184

^a Type I hip was noted bilaterally in 159 babies

^b Type IIa(+) hip was noted in 20 babies

^c Type IIa(−) hip was noted in two babies

^d Type IIc hip was noted in two babies

^e Type III hip was noted in one baby

other patients was 4.125 ± 1.026 (range, 3.0–5.5) and 3.840 ± 1.198 (range 0.5–6.0), respectively, but we did not find any statistically significant difference ($p=0.5776$) in mean Pirani score between these two groups. (Table 5)

Discussion

The presumed correlation between CTEV and DDH may provide a target population for hip screening. However, selective screening of the hip in the CTEV population remains controversial. The debate also continues as to the choice of method for hip sonography [1–8, 10]. DDH was evaluated by Westberry et al. [6] using anteroposterior pelvic radiographs, the value of the acetabular index (AI), the presence of a disrupted Shenton's line and the Tönnis grade. Carney and

Table 3 Hip grading with the Graf classification at 12 weeks of age

Hip type	No. of left hips	No. of right hips	No. of babies with corresponding hip type
I	180	182	180 ^a
II			
IIb	2	2	2 ^b
IIc	1	0	1 ^c
D	0	0	0
III	1	0	1 ^d
IV	0	0	0
Total	184	184	184

^a Type I hip was noted bilaterally in 180 babies

^b Type IIb hip was noted in two babies

^c Type IIc hip was obtained in one baby

^d Type III hip was noted in one baby

Table 4 Hip classification of five patients at 6 weeks, 12 weeks and 6 months of age

Patient	Laterality	Hip type		
		6 weeks	12 weeks	6 months
1	Left	IIC	IIB	IA
	Right	IIC	IIB	IB
2	Left	IIC	IIC	IB
	Right	IIa(–)	IIB	IB
3	Right	IIa(–)	IB	Null
4	Left	III	III	Lost to follow-up
5	Left	IIa(–)	IIB	IIB

Vanek [2] also used the conventional method of radiograph in their study for evaluation of DDH. It is known that radiographs have low sensitivity for ruling out mild DDH because of the poorly ossified nucleus of the femoral head and acetabular roof in infants and possibly altered babies' postures when the radiograph is taken due to crying. It was presumed in these reports that all of the radiographs were centred without rotation. In the report of Westberry et al. [6], radiographs were taken of only 127 children, while the other cases of DDH were detected only by clinical examination. Lochmiller et al. [3] reported five cases of DDH among the group of 285 propositi with idiopathic clubfoot (1.75 %). However, the information for 73 propositi was unknown or not available at the time of the study. Therefore, it is presumed that some more cases of DDH might have been ignored in their study group.

As the foundation of hip examination, the hip morphology can be precisely and comprehensively described and evaluated by means of hip sonography, especially for infants younger than six months of age. In a study of 119 patients with idiopathic clubfoot, Perry et al. [5] reported a DDH incidence of 5.9 % using hip sonography. Despite the authors' deliberate efforts to categorise the degree of dysplasia according to the Graf classification, they failed to distinguish the subtypes of the type IIa hip and to recognise the relationship between type IIa and type IIB hips. According to the Graf method, a type IIa hip is defined as that when the value of the α angle is between 50 and 59° in a baby younger than 12 weeks of age. The type IIa hip of a baby who is between six and 12 weeks of age can be divided into type IIa(+), in which the α angle is between 55

and 59°, and type IIa(–), in which the α angle is between 50 and 55° [13, 14, 16]. The babies with a type IIa(+) hip which has reached the minimal level of hip maturity only require clinical follow-up. The type IIa(–) hip which only exists in infants between six and 12 weeks of age was defective during development. The value of the α angle for this type did not reach the minimal requirement in terms of linear pattern of hip maturity and therefore treatment was required according to the therapeutic implications of the Graf method [14, 16]. If the value of the α angle is less than 60° while the baby is older than 12 weeks of age, the hip becomes a type IIB hip. The only distinction between type IIa and type IIB hips is age [14]. In the study of Perry et al., type IIa hips were not subdivided into IIa(+) and IIa(–), while type IIB at the age of six weeks was not in accordance with the Graf classification [14, 16]. Moreover, they did not take premature delivery into consideration for hip sonography and classification. These similar studies mentioned above reached different conclusions supposedly due to different methods of hip examination, definitions of hip dysplasia, ages and races of patients.

During the period of our study, there was no statistically significant difference in the number of annual presentation of CTEV infants ($p=0.9785$). This non-fluctuating research population during the three-year study period is consistent with that as reported by Perry et al. [5]. In our study, all of the babies with CTEV had their hips clinically examined for instability at the initial visit using the Barlow and Ortolani tests; these tests could not rule out mild hip dysplasia due to low sensitivity. Therefore, ultrasound examination is absolutely necessary, particularly for infants younger than six weeks. Previous studies have indicated that sonographic hip classification was correlated with the pathological changes in the hip joint, and the requirement of the Graf method for a “sonogram in the standard plane” made the assessment independent of the posture and projection because the measurement is performed based upon the standard plane [9, 11–14]. Using the Graf method [11–16], we found three type IIa(–) hips, three type IIC hips and one type III hip in five babies at the six-week ultrasound examination (Table 2). Since we defined DDH as a hip which is type IIa(–) or worse, five patients, with DDH detected at the age of six weeks, constituted 2.7 % (5/184) among the study group of 184 patients with idiopathic CTEV, which was the incidence of DDH in the clubfoot population in our study. Of these seven hips, three type IIC and one type III hips were treated using the Pavlik harness or Otto Bock splint for hip abduction; the other three type IIa(–) hips were just maintained in flexion and abduction according to our recommendations. Nevertheless, one patient (patient 2 in Table 4) with bilateral clubfeet was type IIC in the left hip and type IIa(–) in the right hip. An Otto Bock splint was also prescribed for this baby. When these babies underwent repeat ultrasound examination at 12 weeks, four type IIB hips, one type IIC hip and one type III hip were found in four babies (Table 3). At this point in

Table 5 Comparison of mean Pirani scores per foot between the groups with the hips diagnosed as DDH and normal hips

Groups according to the hip condition	No. of feet involved	Mean Pirani score (mean±SD)
Group with hips diagnosed as DDH	8	4.125±1.026
Group with normal hips	266	3.840±1.198

There was no statistical difference in the Pirani score between the DDH group and the control group ($p=0.5776$)

time, one type IIa(–) hip had become a type I hip, and two type IIa(–) and two type IIc hips had become type IIb hips, while one type IIc hip and one type III hip remained. Only one type IIb hip remained at six months of age, while one baby previously with type III hip was lost to follow-up (Table 4). However, all type IIa(+) developed into type I hips, while two of three type IIa(–) hips turned into IIb. In addition, only one of these babies (type IIb) required continuous treatment at six months of age. These observations indicate that it might be necessary to classify subtypes of type IIa hips between six and 12 weeks, and a type IIa(–) hip might need medical intervention, because two of three type IIa(–) hips did not develop into type I hips at 12 weeks although one (the right hip of patient 2 in Table 4) accepted treatment with an Otto Bock splint. The treatment using the Pavlik harness or Otto Bock splint was convenient and economical, but did not greatly increase the burden of patient care, especially in cases of CTEV undergoing treatment with the Ponseti method. As soon as the hip developed into type I, treatment was terminated. In addition, no statistically significant difference ($p=0.5776$) was found in the mean Pirani scores of CTEV between the DDH group and the normal group (Table 5). Consistent with previous studies [5], this result may reveal that the need for hip treatment is not directly associated with the severity of clubfoot and may suggest that factors that affected the severity of clubfoot did not impact the incidence of DDH in patients with CTEV.

The results from our study supported an association between the incidence of CTEV and DDH. Our study identified five DDH cases in 184 babies (2.7 %) with CTEV. However, our results differed from those of previous studies [1–8], but we insist on recommending hip ultrasound screening in patients with idiopathic CTEV in our area. Firstly, two studies from our area reported using the Graf method for hip screening in the general population. The prevalence of DDH in Hong Kong was 0.87 % [17] and the incidence of DDH in Taiwan was lower than 8 % at six weeks after birth [18]. According to the figures from the reported studies performed in the same region with a population having supposedly the same genetic predisposition as mentioned above, our CTEV group had a much greater incidence of DDH requiring treatment. Even though we exclude two patients with unilateral type IIa(–) hips, which remains a controversial issue, we still have a significantly higher DDH incidence of 1.6 % (3/184) in CTEV babies. Secondly, unlike other risk factors for DDH, CTEV can be easily diagnosed at birth. Currently, an increasing number of medical centres have been using the Ponseti method to treat clubfoot during the neonatal period. When these babies are six weeks old, they may still remain at the stage of manipulation and casting; therefore, it is beneficial for these babies to undergo a clinical and ultrasound examination. This may be suitable for some developing countries with a large population as it is difficult for the public health care system to afford hip ultrasound screening in all neonates. If DDH is

identified in infancy, less invasive treatment can be provided at an early stage. Canavese et al. [1] found that two cases of newborns with unilateral clubfoot developed unilateral hip dysplasia on the same side as the involved foot during clubfoot treatment. The parents reported that their babies slept on the involved side keeping the leg with the cast raised. The authors supposed that raising the long leg cast might have caused hip adduction which contributed to hip dysplasia. A recent study has revealed that earlier swaddling and prolonged swaddling with the lower extremities extended and adducted were more detrimental to hip development in their experimental model [19]. No similar cases occurred in our series because we taught the parents how to use a Pavlik harness or Otto Bock splint during the Ponseti casting or bracing period, let them use

Fig. 1 Application of Otto Bock splint during Ponseti treatment (patient 2 in Table 4). **a, b** The patient was wearing an Otto Bock splint after the fourth Ponseti cast at the age of 6 weeks. **c** The patient was wearing an Otto Bock splint when the phase of foot abduction orthosis was started

it under our supervision and advised that the babies maintain a supine position for as long as possible. As a result, the weight of the long leg cast or the brace with connecting bar were able to stabilise the hip in flexion and abduction (Fig. 1). As a result, no DDH case was found during the period of clubfoot treatment.

This study faced a dilemma due to the theory that hip sonography for DDH diagnosis might have a high sensitivity, which might lead to the over-diagnosis of DDH, and that even the definition and implication of type IIa(–) have not yet been widely accepted. The over-diagnosis of DDH can be avoided by using a standard procedure [20], dependent on the examiner's comprehension of and adhesion to the principles and detailed techniques of the Graf method. The diagnosis using the Graf method is based on the standard plane, which requires three points in space to be defined. If the lower limb of the os ilium, for an example, is not seen on the sonogram, the sectional plane is not taken as the standard plane [9, 11–14]. As a result, the inter-observer and intra-observer agreement of ultrasonographic measurement of trained examiners showed good results due to operating with the standard plane [21], and the inter-rater reliability and intra-rater agreement of the Graf classification were also reliable [22]. Moreover, our previous study showed that inter-observer agreement of two sonographers (YC and LL) in this study was reliable. We found that the reliability of hip ultrasonography using the Graf method in DDH diagnosis mainly depended on the basic technique of the performers and the standardisation of the performance, rather than operating experience [23].

In summary, a higher incidence of DDH exists in the population of our patients with idiopathic CTEV compared to the general population of the same race. Moreover, even with treatment using the Otto Bock splint or recommendation of hip flexion and abduction, only one of the three type IIa(–) hips developed into normal type I. This indicates that CTEV patients should be the candidates particularly for selective hip screening in our population. It is of significant value to recognise at an early stage cases of even mild dysplasia, which run the risk of leading to the development of more severe DDH as the child grows, and to prevent the worsening condition by close follow-up and intervention. Consequently, early intervention should be strongly advised.

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