Original Article

Should We Continue to Screen for Developmental Dysplasia of the Hip in Clubfoot? Our Experience and Review of the Literature

Teixeira R MD [1], Ovídio J MD [2], Arcangelo J MD [2], Campagnolo J MD [2], Tavares D MD [2]

Dr. Teixeira R

Dr. Ovídio J

Dr. Arcangelo J

Dr. Campagnolo J

Dr. Tavares D

Address of Correspondence
Dr. Raquel Teixeira,
Rua dos Quartéis 80, r/c direito, 1300-483 Ajuda, Lisbon, Portugal.
E-mail: rt.corda@gmail.com

[1]Hospital São Francisco Xavier, Lisbon, Portugal.
[2]Hospital Dona Estefânia, Lisbon, Portugal.

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Abstract

Objective: The association between clubfoot and developmental dysplasia of the hip (DDH) remains uncertain, with only a few studies linking both. However, clubfoot is considered as a risk factor for DDH. The aim of this study was to determine the incidence of DDH and evaluate the need for routine hip imaging in our population of children with clubfoot.

Methods: Retrospective analysis of all patients treated for clubfoot in our center between 2010 and 2019. We included patients with hip imaging for DDH in the first 12 months of life.

Results: There were 108 children with clubfoot who underwent DDH screening. 92 had idiopathic clubfoot and 16 had syndromic clubfoot. Of the patients with idiopathic clubfoot, 2 (2.2%) had DDH; one had a clinically unstable hip and the other patient underwent hip screening on account of the clubfoot alone. Among patients with syndromic clubfoot, 3 (18.8%) had developmental dysplasia of the hip. Two of them had an abnormal hip examination while the other had normal hip clinical examination but other established risk factors for DDH.

Conclusion: A targeted ultrasound or radiological screening programme for DDH in idiopathic clubfoot diagnosed hip dysplasia in only 1 child that would have otherwise been missed by clinical examination alone. We conclude that hip imaging is not warranted in children with idiopathic clubfoot and regular clinical screening may suffice. In syndromic clubfoot, due to the higher incidence of DDH, we recommend specific ultrasound screening even in the presence of a normal hip examination.

Keywords: Clubfoot, Screening, Developmental dysplasia of the hip

Introduction

Clubfoot and developmental dysplasia of the hip (DDH) are two of the most common pediatric congenital deformities [1]. Clubfoot, also known as Congenital talipes equinovarus, is characterized by cavus, forefoot adductus, varus and equinus [2] and occurs in 1 in every 1000 live births [3]. The incidence of DDH is estimated in 1.5 to 20 cases per 1000 live births, depending on the population and the screening method used [4].

The etiology of both clubfoot and DDH remain unknown [5]. DDH is considered to be multifactorial and its etiology may include genetic, ethnic, and environmental components [1]; a positive familiar history being the most significant risk factor [6]. Risk factors for DDH such as breech presentation,

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primiparity, oligohydramnios and the association with congenital torticollis and metatarsus adductus support the theory of a "crowding phenomenon" in its pathogenesis [1]. However, only one in 75 children with risk factors have DDH [4].

The majority of cases of clubfoot are isolated birth defects, described as "idiopathic clubfoot" [7]. The incidence of other malformations, chromosomal abnormalities and known genetic syndromes in patients with clubfoot vary substantially among studies (11% [7] to 50% [2]) and these are termed "syndromic clubfoot" [8]. Clubfoot was suspected to arise from mechanical or constraining forces, such as breech delivery, oligohydramnios and multiple gestations [9]; thereby sharing a common pathological mechanism with DDH. Ponseti showed that clubfoot developed in the foetus early in the gestation period, long before intrauterine compression became significant [10]. As the understanding of the etiology of clubfoot improved [9], other theories arose, including vascular deficiency [2], environmental factors, muscle or bone lesions [1] and genetic factors [2, 7, 8]. Consequently, the link between DDH and clubfoot became less clear.

Even so, concerns remain regarding an increased rate of hip dysplasia in patients with clubfoot [10]. Indications for hip imaging screening are controversial, as most textbooks and screening protocols still cite a potential association and state lower limb deformity as a risk factor for DDH, recommending hip screening for these patients [1]. The purpose of this study is to determine the incidence of DDH in the population of children with clubfoot treated in our institution.

Methods:

We identified patients treated with the Ponseti method for clubfoot from 2010 to 2019 at our institution, a pediatric hospital and a pediatric orthopedics reference center.

We included all clubfoot patients with imaging hip screening for DDH (hip ultrasound or X-ray) in the first 12 months of life. The decision to screen for DDH was not based on a specific protocol, but on clinical observation, attending physician preference or the presence of other risk factors for DDH.

We excluded patients without hip screening or if the imaging was performed after the age of 12 months.

The association of clubfoot with other deformities or genetic abnormalities (syndromic clubfoot) was recorded. Charts were reviewed for family history, risk factors for DDH, physical examination findings, imaging results and treatment for DDH (where applicable). We included the following as risk factors for DDH: breech presentation, positive family history, oligohydramnios, congenital torticollis and multiple congenital anomalies.

Results:

During the study period, 251 children were treated for clubfoot at our institution. Of these, 108 were screened for DDH in the first year of life (45 with pelvis X-ray and 63 with hip ultrasound) and were included in our study. Of the 108 children included, 92 (85.2%) had idiopathic clubfoot and 16 (14.8%) had syndromic clubfoot (Table 1).

Table 1: List of etiologies of syndromic clubfoot

Etiologies of Syndromic clubfoot	
Central nervous system	
Cerebral malformation	1
muscle	
Congenital muscular dystrophy	4
Congenital myotonic dystrophy	1
Distal arthrogryposis	1
Arthrogryposis multiplex	1
Chromosomal abnormality	
Ring chromosome 18	1
Trisomy 21	1
Known genetic syndromes	
Weaver syndrome	1
Prader-Willi syndrome	1
Collagen type II disease	1
Probable genetic syndrome	
Multiple congenital anomaly	3

Of the 92 patients with idiopathic clubfoot, 6 had risk factors for DDH (4 with breech presentation and 2 with congenital torticollis) and 2 patients (2.2%) had DDH. One patient had a clinically unstable hip and an ultrasound at 1 week of age confirmed a dislocated hip. After failed conservative treatment, an adductor tenotomy was performed at four months. At 1 year of age, the child required corrective osteotomy. The other child had no risk factors for DDH and a normal physical examination. A hip ultrasound at 5 weeks of age revealed DDH. Conservative treatment was successful resulting in a sonographically normal hip at 3 months.

Of the 16 children with syndromic clubfoot, 3 had DDH (18.8%):

- 1 patient with multiple congenital anomalies (inherited metabolic disorder with bilateral clubfoot, left hip and left knee congenital dislocation) had a positive Barlow test. A hip ultrasound was positive for hip dysplasia at 3 weeks of age. Conservative treatment with a harness was successful, with a normal ultrasound at 9 weeks.
- 1 patient with congenital myotic dystrophy and congenital torticollis had a normal hip examination but a hip ultrasound at

Table 2: Description of patients with positive screening for DDH: idiopathic or syndromic, risk factors for DDH, clinical observation, screening results, treatment, final result of treatment.

Idiopathic/syndromic clubfoot	Risk factors for DDH	Clinical findings	1st ultrasound				2nd ultrasound				X-ray		Treatment	Result
			Age (Weeks)	Alpha angle	Beta angle	Graf	Age (Weeks)	Alpha angle	Beta angle	Graf	Age	Tonnis		
Idiopathic	No	Abnormal (clinical sign not described in reports)	1W	R 49º L 56.5º	R 62.5º L 58º	R IIc	-	-	-	-	4 months	III	Osteotomy	Resolution
Idiopathic	No	Normal	5W	R 56.3º L 39,5º	R 51º L 46.3º	R IIb L III	10W	R 60º L 60º	R 48.3º L 53.7º	R Ib L Ib	-	-	Correctio brace	Resolution
Syndromic: polymalformative syndrome	Polymalformation	Barlow positive	3W	R 68º L 48.5º	R 48.8º L 51.2º	R Ib L IIc	9W	R 69º L 65.9º	R 40º L 30º	R Ib L Ib	-	-	Correctio brace	Resolution
Syndromic: Congenital myotonic dystrophy	Torticollis, muscular dystrophia	Normal	6W	R 56.5º L 63º	R 29º E 44.5º	R IIb L Ib	12W	R 72.9º L 71.9º	R 58º L 54º	R Ia L Ib	-	-	Spica cast 4 weeks	Resolution
Syndromic: polymalformative syndrome	Polymalformation	Limited abduction, hip creases asymmetry	-	-	-		-	-	-	-	11 months	III	Osteotomy	Residual dysplasia with subluxation

6 weeks of age was positive for DDH. Conservative management in a hip spine resulted in a normal ultrasound at 3 months of age.

• 1 patient with multiple congenital anomalies (clubfoot, hip dislocation and cerebral malformation) had a positive clinical evaluation (limited abduction and asymmetry of hip creases) underwent femoral osteotomy at 3 years of age, and currently has residual subluxation of the hip.

Discussion:

While the diagnosis of clubfoot is obvious, clinical examination can be normal in neonates with DDH. As early diagnosis for DDH is associated with a more favorable outcome, screening is important. It is recognized that infants with at least one risk factor have twice the risk of having DDH compared to infants without risk factors [11]. However, only 25% to 30% of infants with DDH have identifiable risk factors, making the development of an effective screening program challenging [12].

Clinical screening tests for DDH, such as Barlow and Ortolani, while specific have low sensitivity [13, 14]. Consequentially, other methods of screening population at risk for DDH started to gain popularity, such as ultrasound or pelvis X-ray. The American Academy of Pediatrics recommends screening infants for DDH by physical exam of all infants and by ultrasonography considered at high risk for hip dysplasia (breech presentation, family history of DDH, or positive physical exam) [10]. The Canadian Task Force recommends serial clinical examinations of the hips of all infants until the age of 12 months and a supervised period of observation for newborns with clinically detected DDH, and does not recommend general ultrasound or radiographic screening for high-risk infants [15]. The European Society of Pediatrics Radiology considers that the only risk factors that indicate the need for hip screening with a normal physical examination are breech presentation and positive family history [16]. The recommendation for screening for DDH of the Portuguese Society of Orthopedic and Traumatology includes every child with positive examination or risk factors for DDH, including foot deformities, without specifying which deformities should be included.

Though universal screening for DDH leads to early identification in many infants, studies suggest that 90% of newborn hips with mild dysplasia identified by ultrasound resolve spontaneously between 6 weeks and 6 months of age [15]. Therefore, with universal screening there is a risk of overdiagnosis and overtreatment.

Among children with clubfoot, the prevalence of DDH was thought to be higher due to the presumption that both result from intrauterine compression [10]. Many reports and textbooks continue to quote "foot deformities" as a risk factor for DDH [6, 11, 13–15, 17, 18]. However, with the increasing knowledge of the pathophysiology of both conditions, they seem less related, and only a few studies have cited cases having both clubfoot and DDH, with low incidences of DDH in clubfoot patients [1, 5, 10, 19, 20].

Wynne-Davis [19] reported only 1 patient with both clubfoot and DDH in 165 patients with primary skeletal deformities. In 127 patients with idiopathic clubfoot who underwent hip X-rays, Westberry et al [1] only found 1 patient with DDH and concluded that hip screening in idiopathic clubfoot was probable not warranted. Lochmiller et al [20] studied 285 patients with idiopathic clubfoot and found 5 patients with DDH. Chou and Ramachandran [5] only reported 1 patient with DDH of 101 patients with clubfoot, and this patient had already been diagnosed due to routine ultrasound for breech presentation. Mahan et al [10] compared 677 patients with idiopathic clubfoot with a control group and found that 5 patients had clubfoot and hip dysplasia (0.74%) versus 5 patients with DDH in the control group (0.25%), without statistically significant difference. Perry et al [13] reported that 1 in 17 babies (5.7%) with congenital clubfoot had hip dysplasia, and recommended hip screening. Even though it only included idiopathic clubfoot, the authors recognized that the patients with both DDH and clubfoot may have an underlying and undiagnosed syndrome.

Our study included 108 patients with clubfoot and hip screening exams, including idiopathic and syndromic clubfoot.

Only 2 patients (2.2%) with idiopathic clubfoot had DDH, and one had a positive clinical exam. Therefore, the specific screening resulted in early diagnosis of DDH in one infant with idiopathic clubfoot, that had a normal ultrasound at 3 months, probably reflecting a mild dysplasia that would have resolved spontaneously.

We identified a higher incidence of DDH in patients with syndromic clubfoot (18.8% vs 2.2%). However, of the 3 patients with both syndromic clubfoot and DDH, 2 had abnormal hip examination, and the other had other risk factors for DDH (Congenital torticollis and muscular dystrophy). The imaging for DDH was therefore indicated for reasons other than the clubfoot.

Conclusion:

Children with idiopathic clubfoot without risk factors for DDH benefit from a careful physical exam of the hips by their treating orthopedic surgeon. If the hip examination is normal, we do not recommend further imaging (x-rays or ultrasound). We acknowledge the increased prevalence of DDH in syndromic clubfoot and therefore recommend hip imaging in these children.

Limitations:

There are several limitations to this study. It was a retrospective study. Risk factors for DDH and other clinical details may not have been recorded consistently. After exclusions, only a small group of patients were available for further analysis. There was no control group to compare the incidence of DDH in patients without clubfoot.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the Journal. The patient understands that his name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil; **Source of support:** None

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