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EARLY DIAGNOSIS OF DEVELOPMENTAL HIP DYSPLASIA IN THE DISTRICT OF PIROT, SERBIA

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Summary. *Developmental dysplasia of the hip (DDH) is a major health problem leading in untreated babies to permanent disability. Costs for treatment, surgery and rehabilitation of these cases are much higher than for relatively simple prevention. A relatively simple ultrasound screening method, according to Graf, has permitted an early diagnosis and treatment of DDH. The aim of this work was to analyze the early diagnosis and prevention of DDH in the district of Pirot. The following periods were analyzed:1) From 1985 to 1988, when diagnosis of DDH was made on the basis of clinical examination and radiology of the hips in the sixth month of life; 2) From 1989 to 1999, when diagnosis of DDH was made by clinical examination and ultrasound of the hips; from 1989 to 1992 in the fourth/fifth month, and from 1993 to 1999 in the third month of life. Hip ultrasound screening from 1989 to 1992 revealed DDH in 0.88% of boys and 2.49% of girls. Screening in the third month of life, from 1993 to 1998 has revealed DDH in 0.21% of boys and 1.35% of girls, significantly less than in the previous period. The treatment of DDH is recommended in the first six weeks of life, at least in the third month of life. Organized teamwork for an early detection and treatment of DDH has given impressive results. Coordinated work of obstetricians, pediatricians-neonatologists, radiologists and children's surgeons-orthopedists has led to the early diagnosis and treatment of DDH, leading to the exclusion of surgery as a method of treatment.*

Key words: *Developmental hip dysplasia, early diagnosis, epidemiology, ultrasound, treatment*

Introduction

Developmental dysplasia of the hip (DDH) is the preferred term to describe the condition in which the femoral head has an abnormal relationship to the acetabulum. DDH includes frank dislocation (luxation), partial dislocation (subluxation), instability wherein the femoral head comes in and out of the socket, and an array of radiographic abnormalities that reflect inadequate formation of the acetabulum.

Prevalence of the clinical condition has been reported to vary from 0.8 to 1.6 per 1000 births in populations not screened neonatally, but with high rates of 10 to 100 per 1000 births among ethnic communities, where infants are traditionally cradled or clothed with their hips extended and adducted; in screened populations, rates of 2.5 to 20 per 1000 births have been reported, but reach 40-90 per 1000 births in some communities (1-4). Differences in reported prevalence may be due to genetic differences and differences in clinical skills and methods used in detection as well as definition of the condition (5).

Several regions of Serbia have a high prevalence of DDH, especially in Vojvodina and South Serbia. In Vojvodina a rate of 28.2 per 1000 of live births was reported (5). Approximately 400 newborn babies in Vojvodina each year were found affected by moderate to severe DDH. Nagorni has described a very high rate of DDH in villages of the county Crna Trava (6). Overall rate of DDH was 15.4, however, in the most affected villages it was 28.7 (village Jabukovik with 591 inhabitants, 17 with lunation) and 32.5 (village Kozilo with 215 inhabitants, 7 with lunation). In the eight counties of the district of Leskovac the rate of DDH varied from 0.73 (Brestovac) to 2.4 (Vlasotince). In his doctoral thesis Lazic has described 2,814 (2167 female and 647 male) children with luxative DDH, out of 56,167 babies born from 1960 to 1979 in the district of Nis (7).

Early identification of affected infants is important for optimal outcome, as results of treatment become worse with delayed diagnosis after the neonatal period (5). Neonatal clinical screening programmes for the condition have been operative since the 1950s, but have varying levels of sensitivity. Attempts to improve sensitivity have been based on the identification of infants at increased risk and ancillary procedures such as ultrasound scanning, which may identify dysplastic hips that are clinically normal (5).

The aim of this work was to analyze early diagnosis and prevention of DDH in the district of Pirot.

Material and methods

District of Pirot has a population of over 116,000 people and about 1,000 live births every year. Details of

mother and baby for births occurring in the Medical Center of Pirot have been collected from the Registry of the Department of Neonatology. The data comprise more than 99.5% of all births.

The number of the inhabitants in four counties (Pirot, Dimitrovgrad, Babusnica, Bela Palanka) of the district of Pirot, according to the Census from 1981 and 1991, is given in Table 1. Due to the migration, the number of inhabitants has declined for more than 13,000 in the year 1991 compared to the year 1981.

Table 1. Number of inhabitants of the district of Pirot

County	1981	1991
Pirot	70.797	67.658
Dimitrovgrad	15.518	13.488
Babusnica	24.453	19.333
Bela Palanka	19.266	16.447
District Pirot	130.034	116.926

The following periods were analyzed:

1. From 1985 to 1988, when the diagnosis of DDH was made on the basis of clinical examination and radiology of the hips in the sixth month of life.

2. From 1989 to 1999, when the diagnosis of DDH was made by clinical examination and ultrasound of the hips; from 1989 to 1992 in the fourth/fifth month, and from 1993 to 1999 in the third month of life.

Ultrasound of the hips was made by the Aloca-260, with a linear transducer of 5 MHz. Examinations were performed according to Graf's procedure and reported according to its classification (8,9).

Results

From 1985 to 1988 hip radiographies were performed in 66.7 to 43.4% of babies born in the district of Pirot. Thereafter a rapid decline was evident, being only 1.0% in 1993, and 0% in 1999 (Table 2).

In the period from 1985 to 1988, 4234 babies were born, but in 2398 hip radiographies were done (Table 3). Positive findings related to the number of radiographies were as follows: hip dysplasia in 18.5%, subluxation 0.71%, luxation 0.87% of children. Positive findings related to the number of live births from 1985 to 1988 were: hip dysplasia 10.5%, subluxatio 0.40%, lunation 0.50%, normal finding in 88.6% of babies.

Table 2. Number of hip radigraphies in the Pirot district of babies in the neonatal period, from 1985 to 1999

Year	No of radiographies	No of newborn babies	Radiographies, %
1985	699	1048	66.7
1986	661	1073	61.6
1987	584	1070	54.6
1988	453	1043	43.4
1989	146	1051	13.9
1990	60	968	6.2
1991	34	941	3.6
1992	13	987	1.3
1993	9	912	1.0
1994	12	1008	1.2
1995	9	963	0.93
1996	6	938	0.64
1997	8	845	0.95
1998	2	764	0.26
1999	0	823	0.0

The ultrasound examination of hips was introduced in the Medical Center of Pirot in 1988. However, at the beginning it was performed as radiography of hips in the fifth and sixth month of life. Later on, the understanding that early diagnosis and treatment are important has changed the policy to performing ultrasound in the third month of life. In the period from 1989 to 1992 ultrasound screening was performed in the 14.4 weeks, but in 11.1 weeks in the period from 1993 to 1999, the difference been statistically significant, $p<0.05$ (Table 4).

Table 4. Age of children at the time of screening of congenital hip dysplasia

Year of investigation	N	Median	min	max
1989-1992	3947	15.4	2	63
1993-1999	6253	11.1*	2	80

Difference between groups was estimated by Mann-Whitney rank sum test $p < 0,001$

The diagnosis of DDH is performed by clinical examination immediately after birth; positive clinical finding or the presence of risk factors requires ultrasound examination of the hips, according to Graf. Ultra-

Table 3. Congenital hip dysplasia diagnosed on radiographies in the period from 1985 to 1988

Year	N	Dysplasia		Subluxatia		Luxatia		Normal	
		Related to number of live born babies							
1985	1048	95	(9.1%)	11	(1.05%)	10	(0.95%)	932	(88.9%)
1986	1073	161	(15.0%)	2	(0.19%)	6	(0.60%)	904	(84.3%)
1987	1070	128	(12.0%)	2	(0.19%)	0	(0.00%)	940	(87.8%)
1988	1043	59	(5.7%)	2	(0.19%)	5	(0.48%)	977	(93.6%)
1985-1988	4234	443	(10.5%)	17	(0.40%)	21	(0.50%)	3753	(88.6%)
		Related to number of radiographies							
1985	699	95	(13.6%)	11	(1.57%)	10	(1.43%)	583	(83.4%)
1986	661	161	(24.4%)	2	(0.30%)	6	(0.91%)	492	(74.4%)
1987	584	128	(21.9%)	2	(0.34%)	0	(0.00%)	454	(77.7%)
1988	453	59	(13.0%)	2	(0.44%)	5	(1.10%)	387	(85.4%)
1985-1988	2397	443	(18.5%)	17	(0.71%)	21	(0.87%)	1916	(79.9%)

sonography in 149 newborn babies has revealed: 44 (29.53%) with type IIa, 6 (4.03%) with type IIb, and 10 (6.71%) with type IIc (Table 5). Based on these findings treatment of DDH has started at the Maternity Unit.

Table 5. Ultrasound findings in newborns, in the period from 1989 to 1999

Finding	Number of children	%
Ia	2	1.34
Ib	87	58.39
IIa	44	29.53
IIb	6	4.03
IIc	10	6.71
IID	0	0.0
IIIa	0	0.0
IIIb	0	0.0
IV	0	0.0
Total	149	100.0

The results of ultrasonography screening performed from 1989 to 1992 in 3497 children are presented in Table 6. Primary normal ultrasonography of hips (types Ia and Ib) was found in 87.98% of neonates. Type IIa was found in 0.63% of boys and 0.96% of girls, type IIb in 9.65% of neonates. Unstable-critical hip (type IIc) was found in 0.63% of boys and 1.80% of girls, type IID in 0.05% of boys but 0.15% of girls. Type IIIa,b was detected in 0.14% of boys and 0.49% of girls; type IV in 0.05% of children. Positive ultrasound findings of DDH (types IIc, IID, IIIa, IIIb and IV) were documented in 0.88% of boys and 2.49% of girls.

Table 6. Ultrasound screening in the neonatal period, from 1989 to 1992

Finding	Boys %	Girls %	Total %
Ia	33.11	27.85	29.50
Ib	59.27	57.69	58.48
IIa	0.48	0.93	0.70
IIb	6.27	13.05	9.65
IIc	0.63	1.80	1.21
IID	0.05	0.15	0.10
IIIa	0.14	0.34	0.24
IIIb	0.00	0.15	0.07
IV	0.05	0.05	0.05

The results of ultrasonography screening performed from 1993 to 1998 in 6253 children are presented in Table 7. Normal ultrasonography of hips (types Ia and Ib) was found in 96.38% of boys and 91.64% of girls. Type IIa was found in 0.36% of boys and 0.68% of girls, type IIb in 3.04% of boys and 6.33% of girls. Unstable-critical hip (type IIc) was found in 0.18% of boys and 1.17% of girls, type IID in 0.03% of boys but 0.06% of girls. Type IIIa,b was detected in 0.09% of girls; type IV in 0.03% of girls. Positive ultrasound findings of DDH (types IIc, IID, IIIa, IIIb and IV) were documented in 0.21% of boys and 1.35% of girls.

Table 7. Ultrasound screening in the neonatal period, from 1993 to 1999

Finding	Boys %	Girls %	Total %
Ia	5.31	4.39	4.86
Ib	91.07	87.25	89.19
IIa	0.36	0.68	0.51
IIb	3.04	6.33	4.66
IIc	0.18	1.17	0.67
IID	0.03	0.06	0.05
IIIa	0.00	0.09	0.05
IIIb	0.00	0.00	0.00
IV	0.00	0.03	0.02

Discussion

A clinically unstable hip in a newborn may be an early sign of congenital dysplasia. Unless followed and treated at a young age, it can progress to a degenerative hip joint disorder with considerable functional disability in adult life. For this reason, the early diagnosis of neonatal hip instability is crucial. In a study with 9199 neonates, examined independently by clinical and ultrasonographic techniques, instability was diagnosed in 0.8% of the hips (10). Only 47% of the unstable hips were diagnosed by the initial clinical examination, in the remainder the dysplasia was recognised only by sonography. Sonographic changes were also detected on re-examination in 6% of the unstable hips following the recognition of clinical instability. It is evident that combined clinical and ultrasonographic examination significantly improves the detection rate of dysplastic hips in newborns (10).

The reliability of Graf's technique in diagnosing DDH was investigated in several reports. In a prospective study, 6,548 neonates were examined clinically and sonographically; 470 children were reexamined at least once (11). Sonographic alpha angles and radiographic acetabular index (AI) angles were followed up and compared. Results were as follows: 84.6% of the hips were mature; 14.3% were physiologically immature; 1.1% were dysplastic. Of the sonographically dysplastic hips, 63% were clinically normal. The neonatal sonographic hip status was affected by family history, breech delivery, birth weight, and gestational age. At follow-up, none of the primarily mature hips had worsened. Of the type IIa hips, 89% matured spontaneously, and 11% needed abduction. The 68 dysplastic hips had matured after a maximum of 80 days' abduction, with normal alpha and AI angles by the end of treatment. At 1 year, the pitch had deteriorated again in six children. Graf's sonographic technique reliably diagnoses infantile DDH. Regular orthopedic checkups are needed to detect secondary deterioration of dysplastic hips.

A meta analysis from the USA was based on the literature primarily from Europe, British Isles, Scandinavia, and their descendants (12). There were 5 controlled trials, each with a sample size less than 40. The remainder were case series. The evidence was available for 17 of the desired 30 probabilities. The evidence quality

ranged primarily between one third and two thirds of the maximum attainable score (median: 10-21; interquartile range: 8-14). Based on the raw evidence and Bayesian hierarchical meta-analyses, the estimate for the incidence of DDH revealed by physical examination performed by pediatricians is 8.6 per 1000; for orthopaedic screening, 11.5; for ultrasonography, 25. The odds ratio for DDH, given breech delivery, is 5.5; for female sex, 4.1; for positive family history, 1.7. The postneonatal cases of DDH were divided into mid-term (younger than 6 months of age) and late-term (older than 6 months of age). The estimates for the mid-term rate for screening by pediatricians is 0.34/1000 children screened; for orthopedists, 0.1; and for ultrasonography, 0.28. The estimates for late-term DDH rates are 0.21/1000 newborns screened by pediatricians; 0.08, by orthopedists; and 0.2 for ultrasonography. The rates of AVN for children referred before 6 months of age is estimated at 2.5/1000 infants referred. For those referred after 6 months of age, the estimate is 109/1000 referred infants. The decision model (reduced, based on available evidence) suggests that orthopedic screening is optimal, but because orthopedists in the published studies and in practice would differ, the supply of orthopedists is relatively limited, and the difference between orthopedists and pediatricians is statistically insignificant, pediatric screening is to be recommended. The place of ultrasonography in the screening process remains to be defined because there are too few data about postneonatal diagnosis by ultrasonographic screening to permit definitive recommendations.

Whether ultrasonography screening should be performed on all newborns is debated, as is the optimal age for such screening (13-15). However, ultrasonography is clearly indicated in the newborn that has unexplained abnormal findings on clinical exam, such as limited abduction. Ultrasonography may also be justified in a newborn who has a normal clinical exam yet has major risk factors for DDH. Hip ultrasonography gives progressively poorer image resolution after the femoral head begins to ossify at approximately six months of age. Plain radiographic imaging can show DDH with adequate reliability after age 3-4 months (16).

The cause of DDH is unknown, but the proposed etiologies include intrauterine positioning, primary acetabular dysplasia and ligament laxity (17). Important risk factors include breech presentation (20% have DDH), congenital muscular torticollis (8%), congenital foot deformities (6%), and positive family history (10%) (17-19). The risk factors of less significance include first-born female, oligohydroamnios, multiple births, prematurity, and postnatal swaddling. Hip involvement is 60% left, 20% right, and 20% bilateral (17).

In the district of Pirot systematic hip ultrasonography was introduced in 1988. The introduction of hip sonography has resulted in a marked decrease of hip radiographies from 54.6% of neonates to 0% in 1999. Hip ultrasound screening from 1989 to 1992 revealed DDH in 0.88% of boys and 2.49% of girls. Screening in the third month of life, from 1993 to 1998 has revealed DDH in 0.21% of boys and 1.35% of girls, significantly less than in the previous period.

DDH could be discovered early after the birth and successfully treated by conservative methods. Late diagnosis of DDH is associated with surgery and a degree of disability. Overall, less surgery and less disability in a population is in favor of an effective screening of DDH and an early treatment. In a previous study surgery the treatment of the inhabitants of the district of Pirot in the last 20 years was analyzed in relation to the screening procedure: ultrasound of the hip-early screening (since 1988), or radiology-late screening (ten years before) (20). Twenty-seven children from the county of Pirot, 11 from Dimitrovgrad, and 7 from Babusnica, born from 1979 onwards, underwent hip surgery at the Children's Clinics in Nis and Belgrade. Compared to the number of inhabitants, the number of DDH was markedly higher in the county of Dimitrovgrad than in Pirot or Babusnica. In 12 children with DDH a major surgery was performed, representing 0.58 surgeries per 1000 newborns. In children born from 1993 to 1999, no hospitalization for DDH was performed, testifying a successful screening of DDH and successful conservative treatment of DDH (20).

DDH is a major health problem leading in untreated babies to permanent disability. Costs for treatment, surgery and rehabilitation of these cases are much higher than for relatively simple prevention. The relatively simple ultrasound screening method, according to Graf, has permitted an early diagnosis and treatment of DDH. Recently, the treatment of DDH starts in the maternity unit or at the first control examination. The treatment of DDH is recommended in the first six weeks of life, or at least in the third month of life. Organized teamwork for an early detection and treatment of DDH has given impressive results. Coordinated work of obstetricians, pediatricians-neonatologists, radiologists and children's surgeons-orthopedists has led to the early diagnosis and treatment of DDH, leading to the exclusion of surgery as a method of treatment (21).

In the last ten years the problem of DDH has been successfully dealt with, although not eradicated. It is still not possible to detect every case of DDH at the birth, nor it is possible to predict new cases after the neonatal period. It is also impossible to predict spontaneous healing of DDH. Therefore, a clinical and ultrasonography follow-up of hips is necessary until the period when normal walking of a child is established.

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RANA DIJAGNOZA RAZVOJNOG POREMEĆAJA KUKA U OKRUGU PIROT

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Kratak sadržaj: *Razvojni poremećaj kuka (RPK) predstavlja značajan medicinski problem, dovodeći kod nelečene dece do trajne invalidnosti. Troškovi lečenja, hirurškog tretmana i rehabilitacije ove dece su mnogo veći od relativno jednostavne prevencije. Ultrazvučni metod skrininga, prema Grafu, omogućuje ranu dijagnozu i lečenje RPK. Cilj ovog rada bio je da se analizira rana dijagnoza i prevencija RPK u okrugu Pirot. Analizirani su sledeći vremenski periodi: 1) od 1985. do 1988. godine, kada je dijagnoza RPK postavljana na osnovu kliničkog pregleda i radiološkog pregleda kukova u šestom mesecu života deteta; 2) od 1989. do 1999. godine, kada je dijagnoza RPK postavljana na osnovu kliničkog pregleda i ultrazvučnog nalaza na kukovima, i to od 1989. do 1992. u četvrtom/petom mesecu, a od 1993. do 1999. u trećem mesecu života. Ultrazvučni skrining kukova od 1989. do 1992. otkrio je RPK kod 0,88% dečaka i 2,49% devojčica. Skrining u trećem mesecu života, od 1993. do 1998, otkrio je RPK kod 0,21% dečaka i 1,35% devojčica, značajno manje nego u prethodnom periodu. Lečenje RPK se preporučuje u prvih šest nedelja ili bar u trećem mesecu života. Organizovani timski rad za rano otkrivanje i lečenje RPK dao je impresivne rezultate. Koordinacija rada akušera, pedijatra-neonatologa, radiologa i dečjeg hirurga-ortopeda omogućila je da se hirurgija ne primenjuje kao metod lečenja RPK.*

Ključne reči: *Razvojni poremećaj kuka, rana dijagnoza, epidemiologija, ultrazvuk, terapija*