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Hip joint development in multiple pregnancy infants – Graf method morphometric assessment. Is a multiple pregnancy a risk factor of developmental hip dysplasia?**

Ocena morfometryczna rozwoju stawów biodrowych u dzieci z ciąż wielopłodowych według metody Grafa. Czy ciąża wielopłodowa jest czynnikiem ryzyka rozwojowej dysplazji stawów biodrowych?

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S u m m a r y

The aetiology of the developmental dysplasia of the hip (DDH) is multifactorial. There are no unambiguous views regarding the effect of multiple pregnancy (MP) on DDH development.

The aim of the study is to determine whether MP is a DDH risk factor and to determine the influence of MP on hip joint (HJ) development.

The first examination included 200 MP infants (400 HJ) and a control group of 63 single pregnancy (SP) infants (126 HJ). Graf method hip joint ultrasonography was performed three times at similar intervals. The first examination was performed in the first postnatal days, the second at 12 weeks and the third at 6 months.

During the first examination of MP infants 28 HJ (7%) were allocated to group IIa. In the control group there were 19 Type IIa HJ (15.1%). During the first and second examination no dysplastic HJ were reported in both groups with Type IIa occurring more commonly in the controls. In the subsequent examinations there was an increasing percentage of Type Ia HJ in both groups. In the MP infants the mean value of the α angle increased in subsequent examinations and in the control group the values were lower in examination II and III. The mean value of β angle decreased in subsequent examinations in both groups.

MP does not predispose to increased incidence of Type IIa HJ. Starting from approx. the 12th postnatal week the osseous part of HJ acetabular roofs was developed better in MP than in SP infants.

Key words: developmental dysplasia of the hip, twin, ultrasound examination, Graf method

S t r e s z c z e n i e

Wstęp. Etiologia rozwojowej dysplazji stawów biodrowych (R.D.S.B) jest złożona. Nie ma jednoznacznych poglądów dotyczących wpływu ciąży wielopłodowej (c.w.) na powstawanie R.D.S.B. Celem pracy jest ustalenie czy c.w. jest czynnikiem ryzyka R.D.S.B., oraz określenie wpływu c.w. i czasu jej trwania na rozwój stawów biodrowych.

Materiał i metody. Badaniem pierwszorazowym objęto 200 dzieci z c.w. (400 stawów biodrowych), oraz grupę kontrolną 63 dzieci (126 stawów biodrowych), urodzonych z ciąż pojedyńczych. Badanie ultrasonograficzne wykonywane było metodą Grafa. Dzieci badane były trzykrotnie w analogicznych odstępach czasu: pierwsze badanie kontrolne w pierwszych dobach życia, drugie badanie kontrolne w 12 tygodniu życia, trzecie badanie w 6 miesiącu.

Wyniki. W pierwszym badaniu dzieci z c.w. 28 bioder (7%) zakwalifikowano do grupy IIa. W grupie kontrolnej typ IIa odnotowano w 19 biodrach (15,1%). W terminie pierwszego i drugiego badania w obu grupach nie stwierdzono stawów biodro-

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wych dysplastycznych, a typ IIa występował częściej u dzieci z grupy kontrolnej. W kolejnych badaniach dzieci z obu grup stwierdzono zwiększający się odsetek bioder typu Ia. W grupie dzieci z c.w. średnia wartości kąta α wzrastała w kolejnych badaniach, a w badaniu II i III oceniany parametr przyjmował niższe wartości w grupie kontrolnej.

Średnia wartości kąta β w obu badanych grupach zmniejszała się w kolejnych badaniach.

Wnioski. C.w. nie jest czynnikiem zwiększającym częstość występowania stawów biodrowych typu IIa.

Począwszy od około 12 tygodnia życia niemowlęcia, cześć kostna dachów panewek stawów biodrowych dzieci z c.w. była lepiej rozwinięta w porównaniu ze stawami dzieci urodzonych z ciąż pojedynczych.

Słowa kluczowe: rozwojowa dysplazja stawów biodrowych, bliźnięta, badanie ultrasonograficzne, metoda Grafa

INTRODUCTION

Gemellology is an interdisciplinary domain dealing with the development of twins. Multiple pregnancy is simultaneous development of two or more foetuses in the uterus. The incidence of spontaneous multiple pregnancy is approx. 1.05-1.35% (1). An increasing number of multiple pregnancies have been observed within the last twenty years in developed countries. At the same time, there has been a decrease in the general number of births. The factors contributing to this phenomenon, including assisted reproductive technology (ART), hormonal stimulation and hormonal contraceptives, have not been fully elucidated.

The developmental dysplasia of the hip (DDH) refers to the abnormal formation of this joint, possibly leading to the dislocation of the hip, occurring during intrauterine development, in the perinatal period or within the first postnatal weeks.

The aetiology of the disorder has a complex nature, with the factors contributing to the deformity of the hip joint development subdivided into hormonal, genetic and mechanical (2).

No unambiguous views regarding the effect of multiple pregnancy on DDH development have been presented so far. Some authors indicated twin pregnancy as a factor predisposing to DDH development (3-7). Apparently, the number of DDH risk factors, with intrauterine crowding being the leading one, should increase the risk of DDH development in multiple pregnancy.

Recent decades have faced an increase in the number of multiple pregnancies, but their effect on DDH development has remained unknown and the number of studies (predominantly retrospective ones) has been negligible. Therefore, the present authors have decided to conduct a prospective study concerning this issue. The study aims to refute or confirm the hypothesis that multiple pregnancy is a DDH risk factor and to determine the effect of multiple pregnancy on hip joint development.

MATERIAL AND METHODS

The study group consisted of infants born in the 2nd Department of Obstetrics and Gynaecology of the Medical University of Warsaw. The first examination was performed between 1 June 2003 and 2 December 2004 in the Department of Neonatology and Intensive Newborn Care of the Medical University of Warsaw and involved 200 children (400 hip joints) from 95 multiple pregnancies, including 97 female infants (48.5%) and 103 male infants (51.5%). The study group comprised 172 infants from twin pregnancies, 24 from triplet pregnancies and 4 from a quadruple pregnancy. The mean duration of a multiple pregnancy was 36 weeks (27 to 41 weeks).

The control group consisted of 63 single pregnancy infants (126 hip joints) including 29 female (46.0%) and 34 male infants (54.0%). The study did not involve infants diagnosed with neuroorthopaedic disorders (meningomyelocoele), congenital syndromes or those in poor overall condition placed at the ward of intensive newborn care. Only neonates with a birth weight exceeding 1000 g were enrolled (8).

A unified examination protocol was used for all participants. It involved the following elements:

- history taking to identify factors increasing the risk of DDH,
- a physical examination to assess the presence of clinical manifestations of DDH,
- an ultrasonographic study to evaluate hip joint development using morphometric indices.

The study and control group subjects underwent three examinations at similar intervals. The first examination was performed in the first postnatal days, the second at 12 weeks and the third at 6 months (mean of 25 weeks).

The second and third examination were conducted at the Clinic of Luxation Prevention, Department of Orthopaedics, Centre for Medical Postgraduate Education (CMKP), Prof. A. Gruca Memorial Independent Public Teaching Hospital in Otwock. The clinical and sonographic examinations were carried out by a specialist in orthopaedics and traumatology (the first author of the present paper) with 14 years of professional experience. During the first examination the parents received written information concerning the aim of the study.

Clinical assessment of the multiple and single pregnancy infants was always conducted according to the same examination procedure, which included the evaluation of hip joint compactness, range of motion, asymmetry of thigh skin folds and coexisting skeletal malformations.

Hip joint ultrasonography was performed according to the Graf method (9) with children placed in a Graf cradle and their hips flexed at approx. 35° and rotated internally at approx 10°. On the basis of alpha and beta angle values, hip joints were classified into one of 9 groups using a Graf sonometer. The examination of neonates was performed with a 12.5 MHz linear probe and the examination of infants was performed with a 7.5 MHz probe. The first ultrasonographic examination of multiple

pregnancy infants was conducted with ATL Ultrasound HDI 3500 device manufactured by Advanced Technologies Laboratories, Bothell, WA, USA, and subsequent examinations and examinations of single pregnancy infants were conducted with Siemens SL-1 device manufactured by Siemens AG, Erlagen, Germany.

Statistical analysis was conducted with the U-test, Wilcoxon test and the t-student test. The threshold of statistical significance was assumed at $p = 0.05$. The calculations were performed with StatSoft's STATISTICA software package.

RESULTS

Hip joint development – Graf method morphometric assessment

Multiple pregnancy infants

Basing on the results of the first sonographic examination of 400 joints in 200 multiple pregnancy infants, 28 joints (7.0%) were assigned to Type IIa group. No pathologically dysplastic hip joints were recorded.

The analysis of hip joint development in multiple pregnancy infants in subsequent studies revealed an increasing percentage of Type Ia joints and a decreasing percentage of Type Ib and IIa joints. Only one male infant had his both hip joints classified as Type IIa in the second examination and Type IIb in the third one. Therefore, it was necessary to apply hip abduction orthosis in this case. Specific numbers of joint types in multiple pregnancy infants are presented in table 1 and figure 1.

Control group

The first examination of 126 joints in 63 controls revealed 19 Type IIa joints (15%).

Subsequent follow-up examinations of single pregnancy infants revealed an increasing percentage of normal Type Ia hip joints. The third follow-up examination revealed 2 Type IIb dysplastic hip joints (3.7%) – one in a girl and the other in a boy. The particulars are presented in table 2 and figure 2.

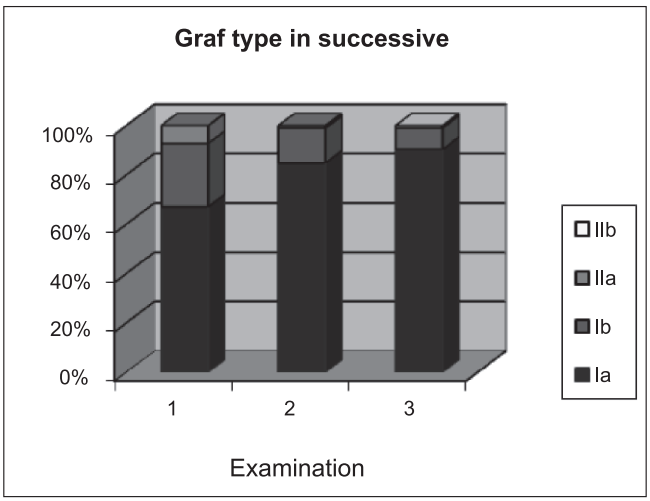

Fig 1. The percentage of Graf hip joint types in successive sonographic examinations of multiple pregnancy infants.

Table 1. Graf hip joint types in successive sonographic examinations of multiple pregnancy infants.

	Graf hip joint type				Total
	Ia	Ib	IIa	IIb	
Examination I	269 67.25%	103 25.75%	28 7.0%		400 100.0%
Examination II	252 84.9%	42 14.1%	3 1.0%	0 0%	297 100.0%
Examination III	211 90.5%	20 8.6%		2* 0.9%	233 100.0%

Table 2. Graf hip joint types in successive sonographic examinations of single pregnancy infants.

	Graf hip joint type				Total
	Ia	Ib	IIa	IIb	
Examination I	79 62.7%	28 22.2%	19 15.1%		126 100.0%
Examination II	87 82.1%	15 14.2%	4 3.8%	0 0%	106 100.0%
Examination III	49 90.7%	3 5.6%		2* 3.7%	54 100.0%

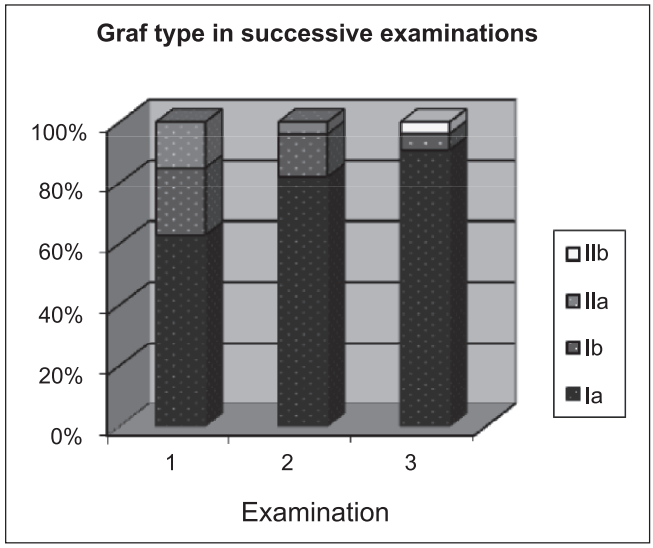

Fig. 2. The percentage of Graf hip joint types in successive sonographic examinations of single pregnancy infants.

Comparative analysis of hip joint development in both groups revealed a more marked percentage of Type IIa hips in the controls in the first examination. The difference was statistically significant ($p = 0.006$, U-test). Similar results were obtained on analysis of the second follow-up examination outcomes ($p = 0.046$). The third examination revealed no statistical significance ($p=0.111$) of differences in the percentage of Type IIa and IIb hip joints between multiple and single pregnancy infants.

Osseous roof – the angle of inclination

Multiple pregnancy infants

In the multiple pregnancy infants the mean value of the α angle was 65° in the first examination and it was increasing in subsequent examinations. The

increases between the first and second examination and between the second and third examination were statistically significant ($p < 0.001$; t-Student test). The particulars are presented in table 3 and the trend is presented in figure 3.

Table 3. The mean value of the α angle in multiple pregnancy infants in successive examinations.

Examination	The alpha angle (°)				
	mean	median	minimum	maximum	standard deviation
I.	65.0	65.0	51.0	78.0	4.6
II.	68.3	69.0	52.0	78.0	4.5
III.	70.2	70.0	55.0	82.0	4.2

Control group

The mean value of the α angle was 65.2° in the first examination in the controls and increased in the subsequent examinations. The increase between the first and second examination was statistically significant ($p < 0.001$), but between the second and third examination there was no statistically significant difference ($p = 0.053$; Wilcoxon test). The particulars are presented in table 4 and the trend is presented in figure 3.

Table 4. The mean value of the α angle in single pregnancy infants in successive examinations.

Examination	The alpha angle (°)				
	mean	median	minimum	maximum	standard deviation
I	65.2	65.0	45.0	80.0	5.9
II.	66.4	67.0	49.0	82.0	5.7
III.	66.6	67.0	56.0	78.0	5.7

The comparison of the α angle between groups in successive examinations revealed that the angle was lower in the control group in examinations II and III compared with multiple pregnancy group (fig. 3). In the first examination the difference was not statistically significant ($p = 0.585$). On the contrary, in the second examination it was $p = 0.001$, and in the third $p < 0.001$ (t-Student test).

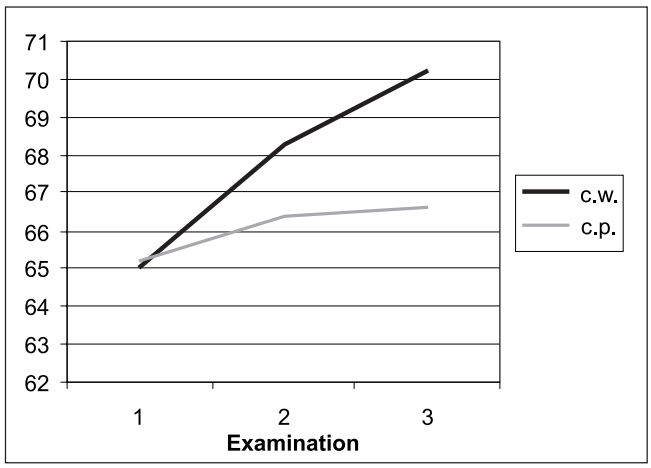

Fig. 3. Comparative assessment of the α angle changes in multiple preganacy (c.w.) and single pregnancy (c.p.) infants.

Cartilaginous roof – the angle of inclination

Multiple pregnancy infants

The mean value of the β angle of the hip joints of multiple pregnancy infants was 52.6° in the first examination and it was decreasing in subsequent examinations. Detailed values of this angle in individual examinations are presented in table 5 and figure 4.

The differences both between the first and second examination and between the second and third examination were statistically significant. The respective values were $p < 0.001$ and $p < 0.001$ (t-Student test).

Table 5. The mean value of the β angle (°) in multiple pregnancy infants in successive examinations.

Examination	Beta angle (°)				
	mean	median	minimum	maximum	standard deviation
I.	52.6	53.0	35.0	72.0	6.1
II.	51.3	52.0	35.0	64.0	5.3
III.	49.7	50.0	36.0	63.0	4.8

Control group

In the control group the mean value of the β angle was 49.9° in the first examination and gradually decreased in subsequent examinations. The differences both between the first and second examination and between the second and third examination were not statistically significant. The respective values were $p < 0.177$ and $p < 0.084$. Detailed beta angle values in successive examinations are presented in table 6 and figure 4.

Table 6. The mean value of the β angle in single pregnancy infants in successive examinations.

Examination	Beta angle (°)				
	mean	median	minimum	maximum	standard deviation
I.	49.9	50.0	30.0	70.0	7.9
II.	49.4	49.0	26.0	79.0	7.5
III.	49.0	50.0	36.0	70.0	6.2

The comparison of mean β angle values between individual groups revealed its lower values throughout the observation period in single pregnancy infants (fig. 4). The differences were statistically significant for the first and second examination. The respective values were $p < 0.001$ and $p < 0.003$. The third examination revealed no statistically significant difference with $p = 0.740$ (t-Student test).

DISCUSSION

Since Graf et al. (2, 9-13) introduced a sonographic method of examination and classification of hip joints, numerous studies describing DDH incidence have been published. A vast majority of studies concerning

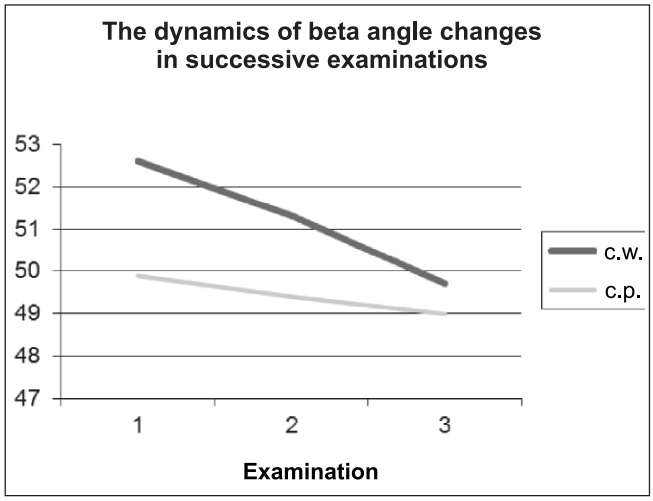

Fig. 4. Comparative assessment of the dynamics of beta angle changes in multiple preganacy (c.w.) and single pregnancy (c.p.) infants.

the outcomes of screening several thousands of neonates and infants have not distinguished the hip joints of multiple pregnancy infants. Available professional literature does not provide information concerning DDH incidence in such a large group of multiple pregnancy neonates and infants as in the present study.

Moreover, no unambiguous views regarding the effect of multiple pregnancy on DDH development have been presented so far. To date, the results of scarce DDH research in multiple pregnancy infants have been obtained predominantly from retrospective studies (4, 14-17). Some authors indicated multiple pregnancy as a factor predisposing to DDH development (3-5, 7, 18). These authors subscribed to the opinion that multiple pregnancy is accompanied by intrauterine crowding, which is one of the main DDH risk factors in single pregnancy.

In the study by Hatzmann et al. (4) the authors compared the development of hip joints in a group of 58 infants born from breech presentation and the control group of 66 infants in which no risk factors were identified. The first group consisted of multiple and single pregnancy infants. In the first group the authors noted a higher incidence of Type IIa hip joints and pathologically dysplastic Type IIc and IIIa hip joints. On the basis of obtained results they suggested that multiple pregnancy is a risk factor of developmental dysplasia due to commonly occurring breech presentation (4). Similar opinion was shared by Ömeroğlu et al. (5). They examined only 6 twin pregnancy neonates and diagnosed one (16.6%) with DDH. Therefore, they included multiple pregnancy in DDH risk factors. In Polish professional literature Zwierzchowski et al. (7) reported multiple pregnancy to be a DDH risk factor.

Other authors (14-19) presented an opposite hypothesis. Hensinger (14) presented the results of retrospective screening study including a group of 150918 Australian infants. DDH incidence was 7.5 per 1000 in this group with breech presentation of the foetus being the most significant DDH risk factor. Multiple pregnancy was viewed as DDH preventive factor. Basing on the sonographic examination of hip joints in 1000 neonates De Pellegrin (19) also reported no pathological hip joints in multiple pregnancy infants.

This issue was also tackled by Rühman et al. (15, 16). They sonographically examined and classified (Graf method) hip joints of 4476 infants. There were 97 twin pregnancy infants (2.2%).

DDH incidence was not increased in the twin population compared with single pregnancy controls. Types Ia, Ib, IIa (+) were reported in the hip joints of 95 multiple pregnancy infants (97.9%) and 4112 (93.9%) single pregnancy infants. Only 1 pair of female twins (2.1%) needed the treatment of DDH. In the single pregnancy group 267 infants (6.1%) required such a treatment.

The present authors observed the tendency towards the decreased percentage of physiologically immature hip joints in successive examinations of multiple pregnancy infants. These trends remained compliant with those reported in the present control group and with the studies of other authors (20, 22, 23,). Marks et al. (23) performed a sonographic examination in 14050 neonates. They reported 6% of immature hip joint types, with 90% of them diagnosed normal during the subsequent follow-up examination performed at 9 weeks. Similar trends were confirmed by Czubak et al. (21) in the study involving 657 children. During the first sonographic examination the developmental dysplasia of the hip was reported in 3.9% of infants and 29% of hip joints were categorized as Type IIa. During two subsequent examinations performed at the intervals of 6 weeks all of the assessed parameters of physiologically immature joints underwent spontaneous normalization, on average at approx. 10 weeks.

The above authors did not conduct their research in a group older than 4 months. Therefore, it is impossible to compare the present results obtained during the third examination performed at around 6 months.

During that period two Type IIa hip joints in one multiple pregnancy infant and two Type IIa joints in one control group infant deteriorated and were classified as Type IIb. These joints were classified as late dysplasia. According to Clarke et al. (24,25) its incidence was 0.2 per 1000.

CONCLUSIONS

1. The results of the present study are not grounds for the confirmation of the hypothesis that multiple pregnancy is not a DDH risk factor.
2. Multiple pregnancy does not predispose to increased incidence of Type IIa physiologically immature hip joints.
3. Starting from approx. the 12th postnatal week the osseous part of hip joint acetabular roofs was developed better in multiple pregnancy than in single pregnancy infants.
4. During the first 3 postnatal months the cartilaginous part of hip joint acetabular roofs was less developed in multiple pregnancy than in single pregnancy infants.

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