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Original Article / Оригинални рад

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Congenital pes metatarsus varus: role of arterial abnormalities in feet and treatment duration and outcome in children

Конгенитални метатарзус варус: значај васкуларних абнормалности и дужина лечења код деце

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SUMMARY

Introduction/Objective We aimed to examine proportion of patients with arterial abnormalities of feet due to age and severity degree of pes metatarsus varus (PMV), and to evaluate treatment duration and outcome.

Methods The prospective longitudinal study included 240 patients with congenital PMV classified into 3 age groups: group <3 months of life (Group-1), group 3-9 months (Group-2), and group 9-12 months (Group-3). Three categories of PMV were analyzed: mild/moderate/severe. Groups with arterial anomalies (Group-A) and without (Group-B) were analyzed. Clinical outcome was graded as: good/satisfactory/poor.

Results There is statistically significant difference in distribution of children regarding age and severity degree on first visit and presence of feet arterial abnormalities ($p<0.01$). For Group-A younger children had longer physical therapy, while for Group-B, older children had longer duration of physical therapy. Same trend applies as severity degree of foot deformity increase. In Group-A, the most frequent treatment outcome was poor (for Group-1–46.7%; Group-2-60%; Group-3-62%), while in Group-B for Groups 1-2 it was frequently good (Group-1-90%; Group-2-40%), and for Group-3 frequently satisfactory (Group-3-53.3%).

Conclusion In children with PMV it might be advisable to perform ultrasound evaluation of arterial structure of feet, and particularly in cases were such deformity is more severe.

Keywords: Metatarsus varus; Arterial abnormalities; Age; Severity degree; Physical therapy; Treatment outcome

САЖЕТАК

Увод/Циљ Циљ рада је био да се испита учесталост болесника са артеријским абнормалностима стопала у односу на узраст и степен тежине метатарзус варус деформитета (МВ), као и да се евалуира дужина и исход лечења.

Методе Проспективна лонгитудинална студија је обухватала 240 испитаника са конгениталним МВ који су уписани у три узрастне групе: Група-1: <3 месеца, група-2: 3–9 месеци, група-3: 9–12 месеци живота. Три категорије МВ деформитетза су анализиране: блага/средње тешка/тешка. Анализиране су групе са (Група-А) и без (Група-Б) артеријских абнормалности. Исход лечења је клинички класификован: добар/задовољавајући/лош.

Резултати Постоје статистички значајне разлике у дистрибуцији деце у односу на узраст и степен тежине деформитета на првом прегледу као и у присуству артеријске абнормалности стопала ($p<0.01$). У Групи-Б, старија деца су имала дужу физикалну терапију и лошије резултате лечења. Исти тренд је за степен тежине деформитета стопала. У Групи-А најчешћи исход терапије је лош (Група-1–46.7%; Група-2-60%; Група-3-62%), док је у Групи-Б најчешћи исход лечења добар за Групе 1-2 (Група-1-90%; Група-2-40%), и за Групу-3 задовољавајући (Група-3-53.3%).

Закључак Код деце са МВ деформитетом се саветује ултразвучна евалуација артеријске структуре стопала, у случајевима са деформитетом тешког степена и старије деце.

Кључне речи: метатарзус варус; артерисјке абнормалности; узраст; степен тежине деформитета; физикална терапија; исход лечења

INTRODUCTION

Congenital pes metatarsus varus (PMV) is a term denoting deformity of children's foot in which the forepart of the foot is in adduction and supination and could be as well presented as contracture at the tarsometatarsal joints. The etiology of PMV and its associated skeletal, neuromuscular and vascular changes have not been fully elucidated. Despite the fact that it can be presented as isolated abnormality, possible neuromuscular deformities and syndromic conditions should be considered [1]. The incidence is estimated to be around 1-2 in 1000 births [2]. PMV is usually diagnosed at birth, however such deformity in certain proportion of children could not be noticed until first year of life [3]. Although deformities of the foot have been study extensively [4-6], there are a few studies of the

effect of vascularization of these structures. Doppler sonography has also been used to establish the possible presence of vascular anomalies in association with foot disorders [7, 8].

The purpose of the study was to examine proportion of patients with arterial abnormalities of feet with regards to age and severity degree of PMV deformity. Additional aim was to evaluate treatment duration and outcome in defined age groups and severity degree of PMV deformity that is related to presence of arterial blood vessels abnormalities in feet.

METHODS

The prospective longitudinal study included 240 patients with congenital PMV (50 had unilateral deformities, and 190 had bilateral deformities), that were treated at University Children's Hospital in Belgrade, Serbia. The study period was 4 years from December 2013 to December 2017 year. The diagnosis was established by board certified physiatrist with experience of more than 4 years in pediatric rehabilitation. Other conditions and diseases (including other congenital anomalies, syndromes and neuromuscular diseases) were excluded during complete clinical examination that included examinations done by: board certified pediatrician, board certified pediatric surgeon, board certified physiatrist and board certified radiologist. All patients were treated by combined electrotherapy, kinesiotherapy and applications of corrective plaster cast. The physical therapists that were included into treatment performance were specialized for pediatric population. The criteria for termination of physical therapy were same results in foot correction on two consecutive examinations by board certified physiatrist.

According to the age of participants on first visit, patients were classified into 3 groups due to the walking modality: group <3 months of life included children (Group 1), group from 3 to 9 months of life (Group 2), and group from 9 to 12 months of life included (Group 3).

Based on clinical findings on first visit we classified the degree of PMV deformity into three categories according to Bleck: mild (axis passes thru 3rd toe), moderate (axis passes between 3rd and 4th toe) and severe (axis passes between 4th and 5th toe) [9].

The patients were examined by use of Doppler sonography at the beginning and at the end of applied therapy. The following arteries were examined: dorsalis pedis artery, anterior tibial artery and posterior tibial artery. Pressures were recorded over the customary anatomic positions: for the posterior tibial artery scans were taken posteriorly to the medial malleolus; for the anterior tibial artery, above the lateral malleolus on the anterior aspect, dorsal portion of the mid-front [10]. Doppler display was provided by placing the probe over the examined artery with acoustic gel for appropriate probe/skin contact. The spectrum corresponding to the arterial circulation was best represented in heterodynamic form showing bi-directional flow. Regarding the presence of arterial anomalies we classified patients into 2 groups: group with changes (hypoplastic and aplastic changes of dorsal pedis artery and tibial anterior artery) (Group A; N=90) and group without changes (Group B; N=150).

Physical treatment was individually prescribed according to the severity degree of foot deformity. In severe degree of such deformity kinesiotherapy with kinesiotaping was administered along with corrective plaster cast. After increase in motion range of anterior part of foot abduction electrostimulation of muscles that were inervated by peroneal nerve was included. Exponential currents were applied with stimulation duration of 10 ms and pause of 20 ms for patients without neurogenic lesions. In some cases galvanization was applied. In moderate cases kinesiotherapy with kinesiotaping was administered along with corrective plaster cast, while in mild cases of foot deformity we performed only kinesiotherapy and kinesiotaping. Kinesiotherapy was performed as an exercise mode for increase in abduction, elevation and dorsiflexion of anterior parts of feet. Kinesiotaping was performed for increase in strength of peroneal muscle group.

After applied physical therapy, a clinical outcome was graded as: good, satisfactory and poor. Passive stretching range in relation to normal foot position was used to assess above mentioned parameters, where for good treatment outcome passive stretching was above projected axis of normal foot, satisfactory outcome was defined as range motion of passive stretching to but not above projected axis of normal foot, while poor outcome was defined as passive stretching not reaching projected axis of normal foot.

To present frequencies of participants age in defined groups, severity degree of feet deformities in defined groups and treatment outcome we used whole numbers and percents. Treatment duration was presented as median values (MV) with standard deviation (SD). Analytic statistical method of χ^2 test was used to assess statistical difference between age groups and severity degree of foot deformity in patients with and without changes on Doppler ultrasonography. For statistical analysis of treatment duration for age groups and severity degree of feet deformities we used one-way ANOVA test, while for separate comparisons regarding treatment duration between different age groups and severity degrees of feet deformities we used Students T test. Statistical significance was set at $p<0.05$.

RESULTS

In Table 1 we presented frequencies of children of different age regarding the presence of arterial anomalies. There is statistically significant difference in distribution of children regarding age on first visit and presence of feet arterial abnormalities ($\chi^2=32.8$; $p<0.01$) (Table 1). Statistically significant distribution of patients is noticed in each age group regarding presence of feet arterial abnormalities (Group 1- $\chi^2=9.1$; $p<0.01$, Group 2- $\chi^2=8.3$; $p<0.01$, and Group 3- $\chi^2=32.0$; $p<0.01$) (Table 1).

Table 1. Frequencies of children regarding age groups and presence of feet arterial abnormalities.

AGE	Group 1	Group 2	Group 3	χ^2 value
Group A	30 (33.3%)	10 (11.1%)	50 (55.6%)	32.8*
Group B	80 (53.3%)	40 (26.7%)	30 (20.0%)	
χ^2 value	9.1*	8.3*	32.0*	

In table 2 we presented frequencies of children of different severity degree of feet regarding the presence of

Table 2. Frequencies of children regarding foot deformity severity degree and presence of arterial abnormalities.

Severity degree of deformity	Mild	Moderate	Severe	χ^2 value
Group A	0 (0.0%)	20 (22.2%)	70 (77.8%)	74.5*
Group B	70 (46.6%)	40 (26.7%)	40 (26.7%)	
χ^2 value	59.3*	0.6	59.2*	

*p<0.01

arterial anomalies. We found statistically significant difference in distribution of children regarding deformity severity degree of feet on first visit and presence of feet arterial abnormalities (χ^2=74.5; p<0.01) (Table 2). Statistically significant distribution of patients is noticed in groups 1 and 3 of severity degree of feet regarding presence of feet arterial abnormalities (Group 1- χ^2=59.3; p<0.01, Group 2- χ^2=0.6; p>0.05, and Group 3- χ^2=59.2; p<0.01) (Table 2).

Table 3. Statistical analysis between age groups and deformity degree groups.

Parameters		χ^2 value	
		Group A	Group B
Age	Group 1/ Group 2	20.0*	26.7*
	Group 1/ Group 3	10.0*	45.5*
	Group 2/ Group 3	53.3*	2.9
Deformity degree	Mild/ Moderate	40.0*	16.4*
	Mild/ Severe	140.0*	16.4*
	Moderate/ Severe	55.6*	0.0

*p<0.01

Statistical analysis between age groups and deformity degree groups was presented in table 3. Our findings showed presence of significant difference in frequencies between age groups and between groups of different severity degrees of deformity for the group with present feet arterial anomalies (p<0.01). For the group without arterial abnormalities of feet there is significant difference in frequencies between observed age groups (p<0.01) except for comparisons between Group 2 and Group 3, and between groups of different severity degrees of deformity (p<0.01), except for comparisons between moderate and severe degree deformity.

Table 4. Mean duration of physical therapy and treatment outcome.

		Treatment duration	Treatment outcome		
		Months (MV±SD)	Good	Satisfactory	Poor
Group A	Group 1	5.6±2.7	4	12	14
	Group 2	3.7±0.9	1	3	6
	Group 3	3.2±1.4	2	17	31
	Mild	0.0±0.0	0	0	0
	Moderate	4.8±2.3	5	9	6
	Severe	2.7±2.2	2	23	45
Group B	Group 1	2.4±1.7	72	8	0
	Group 2	6.2±4.7	16	11	13
	Group 3	18.4±11.9	0	16	14
	Mild	5.4±2.1	64	6	0
	Moderate	8.8±4.5	19	9	2
	Severe	16.9±10.3	5	20	15

In table 4, we presented mean time of physical therapy in patients with PMV regarding age and severity degree of feet deformity separately for those with and those without feet arterial abnormalities. In group with feet arterial abnormalities, younger children had longer physical therapy, while those with severe deformity had shorter physical therapy. In group without feet arterial abnormalities, older children had longer duration of physical therapy, same trend applies as severity degree of feet deformity increases. Regardless the age, in Group A, the most frequent treatment outcome was poor (for Group 1 – 46.7%; Group 2- 60%; Group 3- 62%), while in

Group B in younger patients Groups 1-2 treatment outcome was frequently good (Group 1- 90%; Group 2- 40%), while for Group 3 treatment outcome was frequently satisfactory (Group 3- 53.3%) (Table 4).

Statistical comparisons of treatment duration for age groups of patients and for different severity degrees separately for group with and group without feet arterial abnormalities was presented in table 5. For the group of patients with arterial abnormalities of feet, our results showed statistical significance in deference between age groups except between Group 2 and Group 3. There is no significant difference in frequencies of treatment outcome regarding age (χ^2=3.3; p>0.05) while we found significant differences in frequencies of treatment outcome regarding severity degree of feet deformities (χ^2=13.7; p<0.01) (Table 5). For the group of patients without arterial abnormalities of feet, we found significant differences between groups of different degree of feet deformities (p<0.01), and between age groups (p<0.01) except comparisons between Group 1 and Group 3. There is significant difference in frequencies of treatment outcome regarding age (χ^2=84.1; p<0.01) and severity degree of feet deformities (χ^2=73.5; p<0.01) (Table 5).

Table 5. Statistical analysis of treatment duration between age groups and severity degrees of feet deformities.

		Treatment duration			Treatment outcome
		t test (T value)		ANOVA (F value)	χ^2 value
Group A	Group 1/ Group 2	2.169*	Groups 1–3	15.123†	3.3
	Group 1/ Group 3	5.235†			
	Group 2/ Group 3	1.081			
	Moderate/ Severe	3.728†	Moderate/ Severe	-	13.7†
Group B	Group 1/ Group 2	6.457†	Groups 1–3	79.133†	84.1†
	Group 1/ Group 3	11.796			
	Group 2/ Group 3	5.910†			
	Mild/ Moderate	5.390†	Mild/ Moderate/ Severe	47.547†	73.5†
	Mild/ Severe	9.047†			
	Moderate/ Severe	4.558†			

*-p<0.05; †-p<0.01

DISCUSSION

According to the post-mortem examination [11], vascular abnormalities were found in 2.4-7.1% of patients with normal feet [12]. It is pointed out as well that 3.7-12% of population with normal feet has certain degree of anterior tibial artery abnormality [13]. Abnormal angiograms showing a vascular model similar to the vascular structures seen in fetal angiograms after up to 3 months of gestation were found in another study [14]. These findings are compatible with the theory of discontinuited embrional development as a cause of congenital anomalies of the lower limbs. Thus, foot deformity may result from the persistent model of the fetal circulation and occur as consequence of injury or infection during a specific level of development. They represent a discontinuation of normal development and are usually present at birth.

In our study from the group with present arterial abnormalities of feet we have noticed that as children are older, the more frequently PMV is diagnosed and more severe degree of deformity.

Contrary to these observations, further, for the group without arterial abnormalities, PMV was more frequently diagnosed in younger children, and more severe deformities were seen in younger patients as well.

The more frequent vascular changes we detected in older children in our study were probably the result of adaptation rather than the cause of developmental deformity. Thus, bearing in mind the anatomy of arterial vessels in the foot, it might be postulated that lesions of the dorsalis pedis artery could occur only when a child with foot deformity begins to walk and therefore are more likely to be the result of injuries. This hypothesis could be justified by the results of our study where we found significant increase of frequencies in PMV deformity as children are older between all age groups in case where arterial abnormalities of feet were diagnosed, while significant changes in frequencies of PMV between age groups (Group 2 and Group 3) were not noticed for those without feet arterial abnormalities. Same applies with the degree of foot deformity.

So far, there are a few studies and no clear consensus regarding physical treatment duration and outcome for children with diagnosed congenital metatarsus varus deformity. In the review article of Williams et at, recommendations for the treatment of metatarsus adducts were analyzed, and authors stressed out that there is limited high-level evidence regarding flexible metatarsus adductus non-surgical treatment and contradictions in treatment of semi-flexible metatarsus adductus [15]. Therefore, we wanted to present the findings of our study from professional experience in physical treatment of PMV deformity.

We have pointed out that children with PMV and arterial abnormalities as they are older have significantly shorter duration of physical therapy with more frequently poorer treatment outcome regardless the age. Shorter duration of physical therapy in older children with arterial abnormalities of feet are due to the fact that regardless the age, physical treatment outcome is frequently poor, thus other treatment modalities (eg. corrective surgery) might be of benefit in overall treatment outcome. Despite the fact that these children are referred to pediatric surgeon, physical therapy is shown to have to the certain degree beneficial effects on treatment outcome. However, it should be stressed out that physical therapy improves passive stretching of feet with PMV in children with arterial foot abnormalities and reduces the extensivity of surgical procedures enabling reduction of possible postoperative complications.

Regarding degree of deformity severity, our study have stressed out that severe degree of deformity had shorter duration of physical treatment. This was due to the fact that such deformity in cases with present arterial abnormalities of feet is more resistible to physical treatment modalities.

Previously it was stated that mild deformities of metatarsus adductus have good response to physical treatment [16], and that younger children have better correction outcome, particularly within first months of life [17]. Our results for the group without arterial abnormalities of feet are in line with previous reports. We have demonstrated that for this group of patients, younger children have shorter duration of physical treatment and significantly better treatment outcome.

CONCLUSIONS

Our findings have demonstrated how complex is diagnostic and treatment approach to the metatarsus varus deformities in children, particularly in treatment duration and treatment outcome. However, in older children (with walking independence) with metatarsus varus deformities it might be advisable to perform ultrasound evaluation of arterial structure of feet, and particularly in cases were such deformity is more severe. This is justified by the fact that severe deformities in young children within first months of life, with arterial abnormalities could be due to the presence of such abnormalities, while in older one such abnormalities might be the secondary complications of non-physiological structures of feet.

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