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The Prevalence of Minor Physical Anomalies in Mentally Retarded Children

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A B S T R A C T

The prevalence of minor physical anomalies was examined in a sample of 109 children with idiopathic mental retardation (65 boys and 44 girls). Control group consisted of 246 healthy schoolchildren (123 boys and 123 girls) aged 8 to 12 years. A comparison was made between number of found minor anomalies per child (W1) and their Waldrop weight scores (W2) in healthy and mentally retarded (MR) children. The MR children were found to have a higher number of minor anomalies per child. In their group predominated those with four or more anomalies (56.9%), whereas among healthy children only 7.7% had four anomalies or more. In contrast to the high weighted score value (W2) of five or greater in 36.7% of MR children, it was absent in all control group subjects. There were highly significant differences between the MR and healthy children in the average value of the number of minor anomalies per child (W1) and in the average weighted score (W2). The average number of minor anomalies per child (W1) in MR and well children was 3.65 and 1.7, respectively. In MR children the average weighted score (W2) was 3.82, being 1.46 in healthy children. Our results suggest that common etiological factors, which had led to a physical and mental disorder, were active early in the development of MR children. The finding of high incidence of multiple minor anomalies in MR children indicates that genetic factors may play an important role in the etiology of the underlying disorder in the child group studied.

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Introduction

It has been established that the occurrence of small structural deviations in physical development is linked with the action of prenatal and perinatal factors. They were noted to arise more commonly in children with minimal dysfunction of the central nervous system, behavioral disorders and mental retardation[1]. These deviations or minor anomalies are considered to be a result of morphogenetic disorders[2]. One usually feels that minor anomalies are abnormal morphological characteristics of no serious consequence for the patient[3]. In 1968, Waldrop et al. made a list of 18 anomalies, developing a system quantifying their frequency and weight for each subject[4]. Such approach has facilitated their research and assessment of severity. Later, Waldrop and Halverson revised this minor anomaly list[5]. Of recent, preference has been given to the term »informative morphogenetic variants«, intended to denote mild morphogenetic errors that are prenatal in origin[6]. Although they are clinically and cosmetically insignificant, it is possible to use them effectively for diagnostic, prognostic and epidemiological purposes. Nevertheless, there are indications that their presence in children makes the existence of major malformations more likely[7]. Another finding, in several epidemiological investigations, is the significantly larger presence of minor anomalies in schizophrenic patients, in persons affected by mental retardation, and those having cognitive disorders and behavioral disorders[1,8–10]. For changes arising before and during organogenesis Opitz[11] proposed the term »mild malformations«. However, Trixler et al.[6] showed the frequency of both changes to be significantly higher in the schizophrenic than healthy people group.

Mental retardation is definable as an intellectual function considerably below the average that is attended by incompleteness of adaptive behavior. An intelligence that is more than two standard deviations below the population's mean value is regarded as being considerably below average[12]. Mental retardation (insufficient mental development) can be classified as mild, moderate, severe and profound[12,13]. Many reports, however, distinguish only two degrees of mental retardation: mild (IQ 50–70) and severe (IQ < 50)[14]. About a third of the cases are accounted for by the severe degree of mental retardation[15]. The prevalence of mental retardation in children ranges between 5.2 and 16.6 per 1,000[14–18]. Although, etiologically, these factors can be different, for a large group of people with mental retardation the cause of mental retardation is considered unknown. The cause of mental retardation in as many as 55% of cases being unknown; it is termed so-called idiopathic or sporadic retardation[19].

The basic hypothesis of the present study was that due to the action of factors, which also cause disorders in morphogenesis and early development, the frequency of minor physical minor anomalies in the mentally retarded children's group is significantly higher than in well schoolchildren.

The objective of this study was to look at the prevalence, frequency and severity of minor anomalies in general child population. Equally, it did so in the group of mentally retarded children in order to explain their importance for prediction and timely recognition of mentally insufficiently developed children.

Subjects and Methods

This study was done on 109 mentally retarded children (65 boys and 44 girls) aged 7–17 years. Our control group involved 246 normal schoolchildren (123 boys and 123 girls) aged 8–12 years. Of a

total of 105 wards of the Center for Education and Training of Children and Youths (with milder forms of mental retardation), Zagreb, a group of 94 mentally retarded children was available to our study. Because among mentally retarded about a third are affected by more severe forms of mental retardation – about 50% of which is accounted for by known causes that were excluded from thus study – we randomly selected 15 children from among the 256 children with more severe forms of mental retardation at the Center for Autism, Zagreb. Our control group consisted of a group of normal schoolchildren, i.e. three, four and five formers attending the Primary School »Jordanovac« in Zagreb. We excluded from the study mentally retarded with recognizable genetic defects. Mental retardation is a condition of arrested or incomplete development of the mind[12,13,20]). Used in this study were the International Statistical Classification of Diseases and Related Health Problems (ICD-10), which classify children with intelligence quotients (IQ) below 70 as mentally retarded[13]. Assessment of minor anomalies and their severity was according to the list of 18 minor anomalies on the Waldrop and Halverson scale[5]. The study analyzed the prevalence of individual minor anomalies, minor anomaly total and the sum of their weighted scores[5].

In order to establish differences between the mentally retarded and normal children, two variables suggested by Waldrop and Halverson, i.e. the number of anomalies and the sum of there weighting scores per child were taken. Structural differences in the finding of observed minor anomalies in these two groups were examined by means of binary variables that register only their absence or presence[5]. To test differences in the frequency of anomalies use was made of the Pearson χ^2-test with Yates' correction; for small frequencies we used Fisher's exact test. To test differences in the frequencies of anomalies we used Pearson's χ^2-test with Yates' correction; in the case of small frequency, we used Fisher's exact test. With the t-test, differences in metric variables were tested. The degree of correlation between metric variables was determined by calculating Pearson's correlation coefficient and linear regression.

Results

Comparison of frequencies between individual minor anomalies in mentally retarded children and normal schoolchildren revealed the presence of significant group differences (Table 1). The minor anomalies analyzed occurred significantly more frequently in mentally retarded than in healthy child group. In relation to sex, there were no significant differences in the prevalence of minor anomalies.

Waldrop's scoring system was applied to both groups of children analyzing number of anomalies per child. Comparison also comprised weighted scores (W2) between the groups (Table 2). We added scoring weights to minor anomalies in both subject groups, expressing their weight as Waldrop's weighted scores. Number of minor anomalies per child was regarded as Waldrop score one – W1, and weighted scores of anomalies as Waldrop score two – W2. There was a higher frequency of mentally retarded children with a greater number of minor anomalies. In contrast, dominating in the well-child group were children with a smaller minor anomaly total (Figure 1). Nevertheless, the greatest number of healthy children had one single minor anomaly (about 33% of children), whereas in the group of mentally retarded children those with four minor anomalies (about 30%) were most numerous. While among the mentally retarded children a significant portion had five or six anomalies, in healthy children so large a number of minor

TABLE 1
THE PREVALENCE OF MINOR ANOMALIES IN A MENTALLY RETARDED CHILDREN AND IN THE CONTROL GROUP

Variables	Mentally retarded N = 109	Control group N = 246	
	%	%	p
Fine electric hair	12.8	0.8	< 0.001
Two or more hair whorls	40.4	27.6	0.013
Head circumference outside normal range	46.8	42.3	0.487
Epicanthus	15.6	4.5	0.001
Hypertelorism	13.8	1.6	< 0.001
Low-seated ears	3.7	0.4	0.033
Adherent ear lobes	31.2	10.2	< 0.001
Malformed ears	3.7	0.0	0.009
Asymmetrical ears	9.2	0.4	< 0.001
Soft and pliable ears	12.8	1.6	< 0.001
High-steepled palate	64.2	43.1	< 0.001
Furrowed tongue	15.6	3.3	< 0.001
Tongue with smooth-rough spots	4.6	27.6	< 0.001
Curved fifth finger	7.3	0.8	0.002
Single transverse palmar crease	5.5	2.0	0.100
Third toe longcr than second	2.8	0.0	0.028
Partial syndactylia of two middle toes	5.5	0.8	0.012
Big gap between first and second toes	69.7	2.8	< 0.001

p value tests the independence between the groups with Fisher´s exact test

TABLE 2
DIFFERENCE OF W1 AND W2 BETWEEN MENTALLY RETARDED AND CONTROL GROUP

	Group	N	Mean	SD	St. error of mean	Mean difference	t	df	p
W1	MR	109	3.65	1.55	0.150	1.95	13.12	353	< 0.001
	K	246	1.70	1.16	0.074				
W2	MR	109	3.82	1.86	0.180	2.36	14.48	353	< 0.001
	K	246	1.46	1.17	0.075				

anomalies per child were almost inexistant. While those with four or more anomalies predominated (56.9%) in the MR child group, only 7.7% of the healthy child group had four anomalies or more. Comparison of minor anomaly weighted scores between the two children's groups display even clearer differences (Figure 2). There was a clear shift toward higher weighting scores (from 3 to 5) among MR

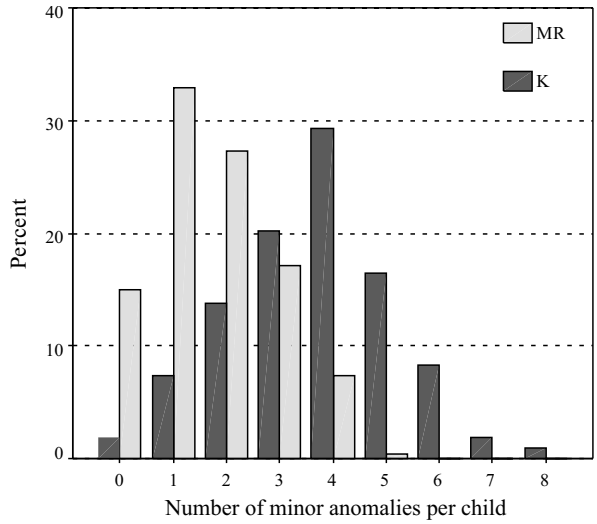

Fig. 1. Mentally retarded and control children groups according to the number of minor anomalies per child after Waldrop. MR = mentally retarded children (N = 109), K = control group (N = 246).

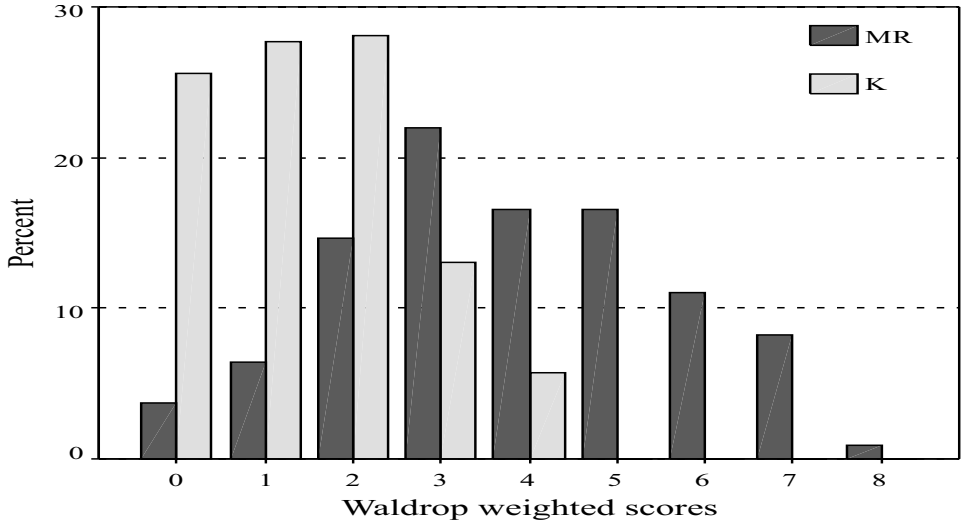

Fig. 2. Waldrop weighted scores for mentally retarded children and their controls. MR = mentally retarded children (N = 109), K = control group (N = 246).

children. In well children, however, lower weighted scores (from 0 to 2) predominated. High weighting scores (5 or higher) were found in 36.7% of MR children. Not a single child in the well-child group had a weighting score 5 or higher. Among

TABLE 3
PREDICTION OF MENTAL RETARDATION BY LOGISTIC REGRESSION MODEL OF VARIABLES FOR DESCIBING THE STATUS OF MENTAL RETARDATION

Free variables	B	SEB	p	EXP(B)
FEH – Fine electic hair (Yes)	4.256	1.013	<0.001	70.554
HYP – Hypertelorism (Yes)	2.427	0.757	0.001	11.328
AEL – (Yes)	1.196	0.525	0.023	3.305
FT – Furoved tongue (Yes)	–2.058	0.691	0.003	0.128
CFF – Curved fifth finger (Yes)	3.520	1.017	<0.001	33.793
PSTMT – Partial synodactylia of two middle toes (Yes)	3.110	1.125	0.006	22.421
BGFST – big gap between firs and second toes (Yes)	4.770	0.507	<0.001	117.948
Const.	5.955	1.076	<0.001	
–2LL	173.703			
χ^2	264.164			
df	7			
p	<0.001			

B regression coefficients; SEB standard error of regression coefficients

MR children, the average frequency of minor anomalies per child (W1) was 3.65, this being 1.7 in healthy children. The MR children's average weighting score of 3.82 compares with 1.46 in the control group. By testing differences in the average values of Waldrop's score one (W1) and Waldrop's score two (W2) it was established that the tested groups differed significantly on both (Table 3).

All variables for recording the presence or absence of 18 analyzed minor anomalies were included in a logistical regression model with a dependent variable, which marks a child as belonging to the group of mentally retarded or to the group of normal schoolchildren. For specific minor anomalies the odds ratio was determined. In the final model (Table 4), those 7 of the 18 minor anomalies were retained whose finding contributes significantly to the increase, and in one case actually to a decrease, in the odds ratio to classify a subject into group of mentally retarded.

Big gap between the first and second toes (117.948), fine electric hair (70.554), clinodactyly of fifth finger (33.793), par-

TABLE 4
CLASSIFICATION TABLE FOR MENTALLY RETARDED AND CONTROL GROUPS (N = 355)

Observed	Predicted		Percent correct
	Control group	Mentally retarded	
Control group	235	11	95.53%
Mentally retarded	19	90	82.57%
Overall			91.55%

tial syndactylia of two-middle toes (22.421) and hypertelorism (11.328) were the most influential variables. Adherent ear lobe (3.305) was a less influential variable. Conversely, the occurrence of a tongue with smooth-rough spots reduces sevenfold the chances to classify a child into mentally retarded children's' group. The model has a greater predictive power (Table 5) in the case of normal schoolchildren (95.53%) than mentally retarded children (82.57%).

On the average, mentally retarded children had 3.65 of the 18 minor anomalies per child, normal schoolchildren having only 1.70. The situation of weighting scores for anomalies per child is similar. Mentally retarded children were on average loaded with a weighted score 3.82, while normal schoolchildren were loaded with a weighted score 1.46 (Table 3).

Differences between mentally retarded children and normal schoolchildren are manifested by significant differences in the number of minor anomalies and their weighted score values per child. It was also noted that correlation between the number of minor anomalies per child and their weighted scores were very high (Figure 3). Correlations in both groups were significant; amounting to 0.816 between mentally retarded children ($p < 0.001$) and 0.733 for normal schoolchildren ($p < 0.001$). Correlation coefficient was significantly higher in mentally retarded children than in normal children. Accordingly, also significantly higher was the regression coefficient in the mentally retarded children's group in the regression model in which the value of weighting scores of minor anomalies is assessed in function of the number of anomalies per child. Thus, whereas the occurrence of one minor anomaly in mentally retarded children raises the value of their weighting score by 0.978, in normal schoolchildren the increase amounts to 0.739.

Discussion

Findings of this study display a wide distribution (from 0 to 8) of the number of minor anomalies in MR children per individual. Most children exhibited three or four minor anomalies. Compared with MR children, the number of detected minor anomalies per child in healthy children was half the MR's. It ranged from 0 to 5. Most healthy children exhibited only one to two minor anomalies. More severe forms of individual minor anomalies (i.e. anomalies with higher weighting scores) were not found in the healthy schoolchild group. They are: fine electric hair (very fine hair that will not comb down), epicanthus (where upper and lower lids join the nose, point of union is deeply covered – seen in only one subject), clinodactyly of fifth finger (markedly curved inward toward other fingers), third toe longer than second. Of other minor anomalies, the only unrecorded finding in the well schoolchild group was malformed ears. A more common phenomenon, tongue with smooth-rough spots, however, is associated with healthy individuals. Other workers have also stated that certain minor anomalies are more common in a healthy child group than in children with mental retardation[4,5]. As to healthy schoolchildren, the literature notes wide differences in the average frequency of minor physical anomalies per child. Thus for boys Steg et al.[21] found an average value of 2.88, while Firestone et al.[22] found 1.4 minor anomalies per child. In girls, Waldrop et al.[9] found this value to be 3.54, against 2.61 minor anomalies per child found by Rosenberg et al.[23].

Another cause of this might lie in the application of different criteria when assessing minor anomalies. Nonetheless, differences are mainly explainable by different characteristics of the populations' studied[24]. They're being no universal models for investigating minor physical ano-

malies, it is on the researcher to design it. Even if there were a uniform concept of minor anomalies, one could not achieve absolute objectivity since researcher's subjectivity in making their assessment cannot be excluded. Various deviations from normal morphogenesis are most striking in the head, hands, and feet. Numerous differences that one notes in these body parts in individuals (including members of the same family, siblings and even twins) are easy to observe. Those, however, who have no clear concept of »normal« and »abnormal« for these structures, will be unable to provide a clear description of the existing changes they encounter in examining an individual with dysmorphism of any etiology.

The fact that 3% of children with one minor anomaly also have a major malformation underscores the importance of minor anomalies. As regards children who have three of more minor anomalies, in as many as 90% of cases they have a major malformation[7]. Reports do not normally mention the exact criteria used in establishing individual anomalies and the total score for an individual subject, rendering inter-study comparisons difficult.

In the MR subject group, this study found an average of 3.7 anomalies per child, the figure in control group being only 1.7 minor anomalies. In other words, mentally retarded children exhibit twice as many minor anomalies than well children do. The obtained results are in agreement with the set hypothesis that the frequency of minor physical anomalies in the mentally retarded child group is higher than in the group of healthy schoolchildren. In the mentally retarded children, the value of weighting score per child was 3.7, whereas in the healthy schoolchildren's group it was 1.5. Observing minor physical anomalies through their weighted scores, one may note that control group has a greater number of subjects with smaller weighted score values per child. Typical of this group, weighted scores of persons with few minor anomalies lack the weight, namely they have a zero value. Unlike control group, MR subjects (who have more minor anomalies) are in relation to detected physical anomalies mainly affected by their severer forms (anomalies with higher weighting score). Namely, there is a greater number there of subjects with higher weighted scores per child. The literature also refers to the data on significantly greater average frequency of total physical anomalies in the MR than healthy children[22]. In this study, we verified it using a linear regression model as well. There was a high positive correlation per person between Waldrop's weighted scores of minor anomalies per child (W2) and frequency of minor anomalies per child (W1). This was borne out by other studies[4,9,24]. The corresponding regression line is significantly steeper in the MR than healthy child group as evident in Figure 3. While in MR children one minor anomaly was weighted with an average weighted score of 0.98, in healthy schoolchildren this was a mere 0.74.

Although minor anomalies also occur in healthy population, their presence is significantly larger in individuals having undergone developmental disorders during morphogenesis, regardless of the factors that had led to them[2,10]. Of great practical significance is the fact that children with some major anomaly also have a greater number of minor anomalies[7]. The reverse is also true, namely in children having a greater number of minor anomalies one may expect to find also a major anomaly. Smith[3] has reported that in children with three of more minor anomalies some major anomaly as well was present in 90% of cases. Consequently, any new knowledge of the number and severity of minor anomalies, especially in children, may contribute to an early recognition of mentally retarded children.

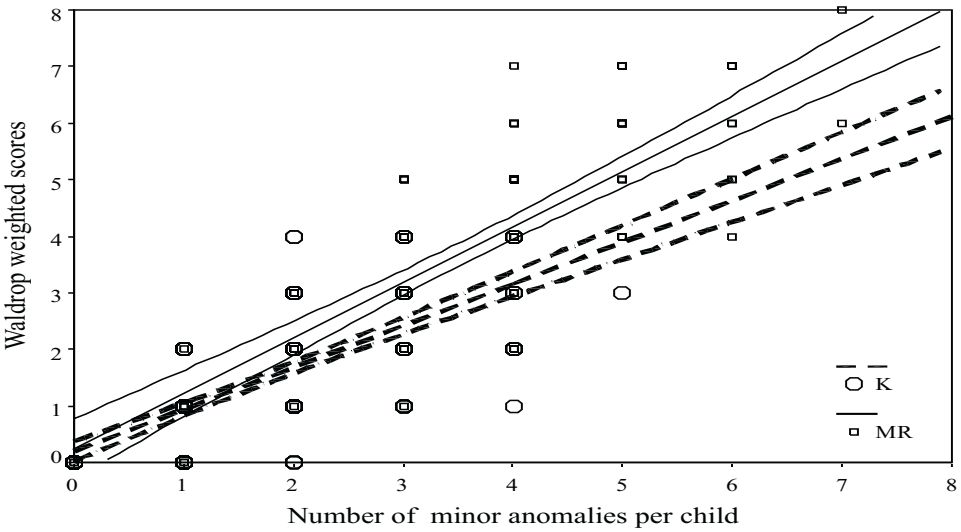

Fig. 3. Scatter diagram and correlations between the number of minor anomalies per child and Waldrop weighted scores for the mentally retarded and control groups of children. MR = mentally retarded children (N = 109), K = control group (N = 246).

Thus the finding of a greater number of minor anomalies in the same child could use as an indication to carry out complex examinations and tests in order to detect the underlying developmental disorder. Because multiple minor anomalies are strictly determined genetically, their finding in a disorder could suggest a condition's genetic determination[21,25]. Therefore, the finding of high percentage of subjects with MR with three or more minor anomalies (77.1%) is ascribable to the dominant genetic etiology in examined children. This suggests a need for detailed medical examination and analysis of each apparently healthy child having a greater number of minor anomalies, as well as a higher sum of their weighting scores. This would increase the possibility of timely recognition of individuals with mental retardation. Whereas manifestation of mental retardation comes later, minor anomalies can be recorded (if looked for) much earlier. In this way, earlier recognition of mental retardation and an earlier start of casework with these children, and hence an earlier rehabilitation would be enabled. Such rehabilitation, in turn, gives children more chance of completing a higher degree of training. This study shows that healthy schoolchildren and mentally retarded children are clearly distinguishable by frequency of anomalies and the strength of their expression, namely according to the weighting scores of these anomalies. A significantly greater number of minor anomalies per child and their higher Waldrop weight score in a group of mentally retarded children than in healthy children' findings suggests the action of a common factor or factors during early development. These simultaneously lead to the occurrence of underlying developmental disorders (mental retardation) and of minor anomalies. The diversity of minor anomalies in a part of the children mental retardation is indicative of the complexity of their etiology, as well as of the need for further research.

REFERENCES

1. PASAMANICK, B., A. LILIENFELD, JAMA., 159 (1955) 155. — 2. SMITH, D. V., K. E. BOSTIAN, J. Pediatr., 65 (1964) 189. — 3. SMITH, D. W.: Recognizable patterns of human malformation. (Saunders, Philadelphia, 1982). — 4. WALDROP, M. F., F. A. PEDERSEN, R. Q. BELL, Child Development, 39 (1968) 391. — 5. WALDROP, M. F., C. F., HALVERSON, Jr., Minor physical anomalies and hyperactive behavior in young children. In: HELLMUTH, J. (Ed.): Exceptional infant: Studies in abnormalities: Vol. 2. (Brunner/Mazel, New York, 1971). — 6. TRIXLER, M., T. TÉNYI, G. CSÁBI, G. SZABÓ, K. MÉHES, Am. J. Psychiatry, 154 (1997) 691. — 7. MARDEN, P. M., D. W. SMITH, M. J. McDONALD, J. Pediatr., 64 (1964) 357. — 8. LOHR, J. B., K. FLYNN, Schizophr. Bull., 19 (1993) 551. — 9. WALDROP, M. F., J. D. GOERING, Am. J. Orthopsychiatry, 41 (1971) 602. — 10. MÉHES, K.: Informative morphogenetic variants in the newborn. (Akadémiai Kiadó, Budapest, 1988). — 11. OPITZ, J. M., Eur. J. Pediatr., 144 (1985) 252. — 12. AMERICAN PSHYCHIATRIC ASSOCIATION. Diagnostic and statistical manual of mental disorders: DSM IV. (APA, Washington DC, 1994). — 13. WHO: ICD 10: International Statistical Classification of Diseases and Related Health Problems. (WHO, Geneva, 1992). — 14. FERNELL, E., Acta Paediatr., 85 (1996) 584. — 15. BAIRD, P. A., A. D. SADOVNICK, Am. J. Ment. Defic., 89 (1985) 323. — 16. BOYLE, C. A., M. YEARGIN-ALLSOPP, N. S. DOERNBERG, P. HOLMGREEN, C. C. MURPHY, D. E. SCHENDEL, Mor. Mortal Wkly Rep. CDC Surveill. Summ., 45 (1996) 1. — 17. STRØMME, P., K. VALVATNE, Acta Paediatr., 87 (1998) 291. — 18. RICHARDSON, S. A., H. KOLLER, M. KATZ, Ups. J. Med. Sci., 44 Suppl. (1987) 83. — 19. HAGBERG, B., G. HAGBERG, A. LEWERTH, U. LINDBERG, Acta Paediatr. Scand., 70 (1981) 445. — 20. ANONYMOUS, Narodne Novine, 39 (1998) 830. — 21. STEG, J. P., J. L. RAPOPORT, J. Autism Child Schizophr., 5 (1975) 299. — 22. FIRESTONE, P., S. PETERS, M. RIVIER, R. M. KNIGHTS, J. Child. Psychol. Psychiatry, 19 (1978) 155. — 23. ROSENBERG, J. B., G. M. WELLER, Dev. Med. Child. Neurol., 15 (1973) 131. — 24. FIRESTONE, P., S. PETERS, J. Autism Dev. Disord., 13 (1983) 411. — 25. MEGGYESSY, V., M. RÉVEHELYI, K. MÉHES, Acta Paediatr. Acad. Sci. Hung., 21 (1980) 175.

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ZASTUPLJENOST TJELESNIH MINOR ANOMALIJA U MENTALNO RETARDIRANE DJECE

SAŽETAK

Prevalencija tjelesnih minor anomalija istraživana je u uzorku od 109 djece s idiopatskom mentalnom retardacijom (65 dječaka i 44 djevojčice). Kontrolnu skupinu činilo je 246 zdrave školske djece (123 dječaka i 123 djevojčice) u dobi od 8 do 12 godina. Komparirani su brojevi nađenih minor anomalija po djetetu (W1) i njihovi težinski skorovi po Waldropu (W2) u zdrave i mentalno retardirane (MR) djece. Utvrđen je veći broj minor anomalija po djetetu u MR djece. U skupini MR djece prevladavali su oni s 4 i više anomalija (56.9%), dok je u zdrave djece svega 7.7% imalo 4 ili više anomalija. Visoka vrijednost težinskog skora (W2) od 5 ili više utvrđena je u 36.7% MR djece, a niti u jednog ispitanika kontrolne skupine. Razlike između MR i zdrave djece u prosječnoj vrijednosti broja minor anomalija po osobi (W1) i prosjeku težinskog skora (W2) bile su visoko značajne. Prosječna vrijednost broja minor anomalija po osobi (W1) za MR djecu bila je 3.65 i 1.7 za zdravu djecu. Prosječna vrijednost težinskog skora (W2) za MR djecu bila je 3.82, a za zdravu djecu 1.46. Dobiveni rezultati ukazuju da su tijekom ranog razvoja u MR djece djelovali zajednički etiološki faktori koji dovode do tjelesnog i mentalnog poremećaja. Nalaz visoke zastupljenosti multiplih minor anomalija u MR djece ukazuje na značajnu ulogu genetskih faktora u etiologiji temeljnog poremećaja u skupini analizirane djece.