Value of Oro-Dental and Physical Minor Anomalies in the Discrimination of Children with Developmental Disorders/Impaired Development

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Summary

The incidence of oral and physical minor anomalies is higher in children with developmental disorders compared with healthy children. The aim of this study was to determine the possibility of discriminating children with developmental disorders from healthy children on the basis of oro-dental and physical minor anomalies. The study was performed on a sample of 303 children with developmental disorders (DD) and a control group of 303 healthy children (HC). The sample of children with developmental disorders comprised 176 mentally retarded children (MR), 70 children with impaired hearing (IH) (partially deaf and deaf), and 57 children with impaired vision (IV) (partially blind and blind). The control group consisted of 303 healthy children of the same age and sex. Multivariate discriminate analysis was performed in the manifest space of 18 oro-dental and physical minor anomalies. The initial space of 18 original variables was reduced to three discriminative functions. Clear discrimination and great distance was determined between the centroids of the control group and the groups of children with developmental disorders. The first two discriminate variables are significant for discrimination between the groups, and they explain 95.7% of the total variance. The first variable contains 87.2% of the information and is defined by oro-dental and toe anomalies. The second variable explains 8.5% of the total variability, and is defined by dental and auricle anomalies. The third function, which is not significant, contains just 4.3% of the total variability. The discriminate functions obtained enable clear discrimination between the three groups of children with developmental disorders, but not clear mutual discrimination of individual entities within the groups of children with developmental disorders.

Key words: *children with developmental disorders, minor anomalies, dental anomalies, discriminate analysis*

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Introduction

Slight deviations from normal morphogenesis are known as minor physical anomalies. They do not usually have medical repercussion per person but can have important clinical and genetic significance (1-4). They occur in the general population of completely healthy people, although their incidence is significantly increased in the case of different genetic disorders and syndromes (5-9). It has been observed that children with developmental disorders demonstrate a significantly increased frequency of minor anomalies compared to healthy children (5, 11-15).

The occurrence of an increased number of minor anomalies in one person may indicate seriously disturbed morphogenesis and consequently their recognition and registration has diagnostic and genetic importance (1-3, 14). Waldrop et al developed a system for evaluating their weighted scores with the aim of assessing the significance of minor anomalies in some people (9, 10). Significant correlation has been determined between the weighted score of minor anomalies according to Waldrop and some oral and dental anomalies in children (13, 14). In some people an increased number of minor anomalies and their weighted score can be an indicator of disturbed embriogenesis (1, 3, 6). It is considered that a high frequency of minor anomalies and high values of their weighted scores in a certain population or persons indicate possible genetic determination of disorders during early development (5, 6, 12). Significant increase in the frequency of minor anomalies has also been observed in children with mental retardation and disturbed behaviour (1, 5, 7, 14-16). Investigations to date have shown that it is possible to discriminate groups of healthy children from children with developmental disorders on the basis of certain minor anomalies (1, 5, 7, 14, 15).

The aim of this study was to perform a discriminate analysis of healthy children and children with developmental disorders in the space of 18 manifest variables of minor physical anomalies and to determine whether these variables contribute to discrimination of the groups and to what extent. The intention was also to determine whether the variables from the category of minor abnormalities provide stronger contribution to discrimination of the groups than those from the groups of minor anomalies.

Subjects and methods

An analysis of physical and oral minor anomalies was performed on a sample of 303 children with developmental disorders and 303 healthy children. The sample of children with developmental disorders consisted of three sub-groups: a) 176 mentally retarded children (MR), 70 children with impaired hearing (IH) and 57 children with impaired vision (IV). The structure of the sample is presented in Table 1.

A multivariate discriminate analysis of children with developmental disorders and healthy children was carried out on the basis of 18 physical minor anomalies, including oral and dental anomalies.

Oral and dental anomalies and other physical minor anomalies in children were determined by an intraoral and systematic physical examination. Eighteen of the most frequent physical minor anomalies were included in the multivariate discriminative analysis, including oral and dental: large gap between the first and second toe, epicanthus, thin electrified hair, high palate, impacted teeth, frenulum of tongue, diastema mediana, partial syndactyly of the second and third toes, hypodontia, asymmetry of the auricles, two or more hair vortices, fissured tongue, soft and pliant auricles, coalesced earlobes, low positioned auricles, microdontia, monkey grooves on the palm, malformed auricles, abnormal head diameter, dens invaginatus, third toe longer than the second, clynodactyly, hypertelorism, spotted tongue and frenulum of the upper lip.

Results

The initial space of 18 manifest variables was reduced by multivariate discriminate analysis to three discriminate functions (Table 2). Of the three discriminative functions the first two statistically significantly differentiate the groups of subjects, while the third does not contribute significantly to explanation of the structure of the difference between the groups included in the analysis. The first function explains 87.2% of the total variance, the second 8.5% and the third, which is not significant, 4.3% of the total variability.

The initial dimensionality of the analysed groups can be shown in reduced three-dimensional discriminative space. The content of each of these dimensions (discriminative variables) can be determined on the basis of their correlation with the initial manifest variables. These correlations, in order of size, are shown in Table 3.

The first discriminate function is initially determined by the variables of the large gap between the first and second toe, epicanthus, thin and electrified hair, high palate, impacted teeth, frenulum of tongue, diastema and partial syndactyly of the second and third toe. Thus it can be said that the first function is equally explained by physical and oral minor anomalies. The second function is defined by hypodontia, asymmetry of the auricles, two or more hair vortices, fissured tongue, soft and pliant auricles, coalescent earlobes, low positioned auricles and microdontia.

The position of individual groups of subjects in the discriminate space is shown in Figure 1. The position of each subject is marked by small dots, while large circles mark the position of the centroids of individual groups. The first two discriminate variables are significant for discrimination of the control group from the groups of children with developmental disorders.

The discriminate variables enable determination of the relationship of certain entities with determined groups. The probable appurtenance/relationship of a particular group of subjects is expressed in percentages and shown in Table 4. The percentages in the table diagonals, shown in bold print, mark the subjects, who according to the prediction of the discriminative analysis, remained in their initial groups. Outside the diagonals are the percentages of subjects, who, according to their characteristics (values of variables describing oro-dental and physical minor anomalies) should be in another group.

Table 4 shows the high stability of the control group, in which 93.4% of all subjects remained in their group. In contrast to the control group the groups of children with developmental disorders showed great redistribution of subjects. The group of MR children showed stability and 63.1% remained in the initial group. There is clear discrimination and great distance between the centroids of the control group and the groups of children with developmental disorders. Such clear discrimination of the control group of healthy children is determined by the first discriminate function, which in the initial discriminate solution explains 87.2% of the total variability. Of the children with impaired vision 21.1% remained in the initial group, and of the group of children with impaired hearing 15.7%. All other variables which correlate with the third function do not participate significantly in differentiating the individual groups of children with developmental disorders.

Discussion

In the present study the results of the discriminate analysis indicate that it is possible to recognise groups of children with developmental disorders and a control group on the basis of dental and physical minor anomalies. Previous investigations of subjects with developmental disorders showed that the total number of minor anomalies per person and their weighted score contributed most to differences between groups (14).

In this study the best discrimination was achieved between healthy and mentally retarded children on the basis of the variables used in the discriminate analysis (Figure 1). Greatest stability was shown by the control group, in which 93.4% of subjects remained in the initial group after the analysis, and mentally retarded subjects of which 63.1% remained in the same group (Table 4). The variables which highly correlate with the first function have the greatest share in discrimination of the groups. These are oro-dental anomalies and anomalies of the toes. The variables which correlate with the third discriminative function do not significantly contribute to discrimination of the groups.

Investigations so far have shown that children with developmental disorders significantly differ from control groups of healthy children with regard to the number and severity of certain minor anomalies (5, 7, 13, 14, 16-18). Minor anomalies occur in completely healthy people, although their incidence is much higher in people with different developmental disorders (2, 4, 5). Minor anomalies with higher weighted scores were not found at all in the group of healthy schoolchildren (for example, low positioned earlobes, marked epicanthus, clynodactyly, third toe longer than the second). In children with developmental disorders the number of minor anomalies per child most frequently ranged from 3 to 5, and in the group of healthy children from 1 to 2 anomalies (13, 14).

Because of diagnostic values of minor anomalies Gillberg and Soderstrom (19) proposed that an analysis of minor physical anomalies should always be included in clinical examinations of children with developmental disorders.

A finding of several minor anomalies in a child with developmental disorders is a powerful indicator of the need to carry out karyotypisation and other genetic tests. Children with such a finding have a stronger genetic burden for a fundamental disorder, leading to handicap. Consequently, identification of a minor anomaly in such children is an important step in explaining the aetiology of the disorder and share of genetic factors in the occurrence of the disorder (19).

Previous investigations of the average number of minor anomalies per child showed that their frequency in the control group amounted to 1.993, and in the group of children with developmental disorders 3.81. At the same time the average weighted score according to Waldrop in the group of subjects with developmental disorders was much higher and amounted to 3.80, and for healthy children only 1.818 (14). These data show that minor physical anomalies are significantly more frequent in the group of subjects with developmental disorders. Particularly high frequency of minor physical anomalies and share of more severe anomalies has been observed in mentally retarded and autistic children (7, 18, 20).

Miles and Hillman (18) carried out an analysis of the severity of minor anomalies in autistic persons. They determined that those persons with more minor anomalies, categorised as "phenotypically abnormal" had a completely different reason for the basic disorder than other persons in the group. Five syndromes were found in approximately 20% of the group with more severe minor anomalies, and in the remaining 80% with mild minor anomalies only one person with a syndrome was found.

The frequency of minor anomalies and their weighted score per person significantly varied between the individual groups of children with developmental disorders. The highest average number of minor anomalies per person (3.955) was found in the group of mentally retarded children, followed by the group of children with impaired hearing with an average of 3.900 anomalies, and then the group of children with impaired vision with 3.263 anomalies. The mean value of the minor anomalies per person in the control group was 1.993 (14). A high prevalence of oral and dental anomalies was determined and their significant correlation with Waldrop's weighted score in the same group of subjects with developmental disorders (13).

In their investigation Ulovec et al (15) found a high incidence of minor anomalies and high value of their weighted scores in mentally retarded children. In a number of disorders the prevalence of congenital anomalies was significantly increased. Their investigation in a group of children with different disorders is important in order to determine the biological value of each of those anomalies in a particular disorder and for such anomalies to obtain their weighted value. Particularly valuable is the study of minor anomalies in the case of disorders which are suspected of being the consequence of developmental disorders (21).

The results of the obtained discriminative analysis should be interpreted in the light of this knowledge. The results of this study show that it is possible to discriminate children with developmental disorders from healthy children on the basis of oro-dental and physical minor anomalies, and that these anomalies provide a very varied contribution to discrimination of groups. Variables which correlate with the first and second function provide a significant contribution to discrimination of the groups of children with developmental disorders from the control group, although they are not sufficient for mutual recognition of individual groups of children with different types of developmental disorders (e.g. deaf and partially blind children).

Merks et al (22) stress the clinical value of evaluating phenotypic anomalies in a population. Such anomalies can be the basis for discrimination of different groups of patients in a heterogenic population, which have specific pathogenesis, treatment and prognosis.

The latest approach to evaluation of small developmental disorders in different groups with developmental disorders take into account the heterogeneity of the minor anomaly. Starting from the principle of the need to separate analysis of minor malformations, such as disturbed morphogenesis, from minor variants or disturbed phenogenesis (21-24). Minor malformations are indicators of more serious developmental disorders and can be more informative than minor variants in genetic investigations of different developmental disorders (19, 21, 24). Such as for example an excessive number of teeth, hypodontia, abnormally formed teeth, enamel and dentine abnormalities (24).

Regardless of the fact that they do not offer the possibility of mutual discrimination of groups of children with developmental disorders, minor anomalies clearly discriminate each of these groups from the healthy children. This indicates that analysis of developmental abnormalities in children with developmental disorders can be useful in assessing the aetiology of the basic disorder in the child.

Conclusions

In this study the results of the discriminate analysis show that the applied oro-dental and physical minor anomalies enabled good discrimination between healthy children and a group of children with developmental disorders. The great distance of the centroids of the control group from the groups of children with developmental disorders indicates the existence of clear discrimination among the subjects. However, the applied variables do not enable clear mutual discrimination of the three groups of children with developmental disorders. Further investigation is needed of sensitive variables which will enable discrimination between certain entities within the sample of children with developmental disorders. Thus further study should be based on a separate analysis of minor abnormalities and minor anomalies in groups of subjects with developmental disorders.