The Prevalence of Oral and Dental Anomalies in Children with Developmental Disturbances

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Summary

The prevalence of oral and dental anomalies was assessed in children with developmental disturbances (DD). The study sample consisted of 606 children and young adults aged 5-20 years (303 DD and 303 control subjects). The group of DD children included 176 children with mental retardation (MR), 70 children with hearing impairment (HI), and 57 children with visual impairment (VI). The association of oral and dental anomalies with minor physical anomalies was studied. Significant differences were found between the two groups in the prevalence of oral and dental anomalies. Hypodontia was more common in DD children (9.2%) than in control subjects (2.0%) ($p < 0.001$). Median diastema was also more common in DD children than in control subjects (20.8% and 11.6%, respectively) ($p < 0.002$). The prevalence of maxillary labial frenum was higher in DD children (6.6%) than in control subjects (1.3%) ($p < 0.001$). Both groups showed a high association of labial frenum and median diastema. A significant association between hypodontia and microdontia was observed in control subjects. A high value of weighted Waldrop score (W) was obtained in the group of VI children. So, W was 6.00 in the group with microdontia, 5.43 in MR children with hypertrophic lingual frenum (ankyloglossia), 4.80 in HI children with dens invaginatus, and 4.45 in MR children with ankyloglossia. The high prevalence of oral and dental anomalies, and high weighted Waldrop score of minor anomalies in DD children suggests that developmental disturbances during early development in this group could be attributed to a common underlying factor.

Key words: *oral and dental anomalies, minor physical anomalies, prevalence*

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Introduction

Minor physical anomalies such as deviations in the development of the body normally occur in the population, and entail no medical or cosmetic consequences for the affected individual. These anomalies may also occur as part of a syndrome or disease, where they do have diagnostic and medical significance. Minor anomalies are mild malformations with primarily morphological involvement. They represent a morphological defect, an irreversible sequel of disturbed morphogenesis. External or visible minor anomalies may involve any region of the body. However, they are usually localized on the head, hands and feet.

The majority (> 71%) of minor anomalies are found in the head and neck region, and on the hands (1). Mulitple anomalies in neonates are potential indicators of a severe morphogenesis disorder. The presence of three or more minor anomalies in a neonate points to the existence of one or more major malformations in 90% of cases (2).

Anomalies of the oral and dental structures can be isolated, although they may also be a symptom of a major defect or manifestation of a general disorder or syndrome. About 50% of all chromosomal aberrations and 25% of genetically determined disorders manifest themselves in the craniofacial region (1). Epidemiologic data show that some 10% of children are born with lesions that entail permanent sequels affecting the physical, mental or emotional life of the child worldwide. In industrialized countries, there are about 10% of children with developmental disturbances, whereas in developing countries their percentage is higher, ranging between 15% and 20% (3). The recognition and identification of oral/dental and minor anomalies are of great importance for timely and accurate diagnosis of numerous genetic abnormalities of the craniofacial region.

Although minor physical anomalies can develop in healthy individuals, they are more common in those with congenital disorders (2). The prevalence of a single minor anomaly in the general neonatal population is 14% and is not considered a serious disturbance. However, the occurrence of two minor anomalies in this age group points to a fivefold prevalence of major malformations. The presence of multiple minor anomalies in an individual may indicate a disorder that occurred at the time of morphogenesis (4). Multiple minor anomalies usually occur as part of a syndrome. Marden et al. (5) defined minor anomalies as structural anomalies without medical and genetic sequels, and with a prevalence below 4% in the general population. Méhes et al. (2) consider that minor anomalies develop consequentially to morphogenesis disturbances, and are frequently associated with congenital disturbances in wider terms. Two, three or more minor anomalies have been observed in children with malignant lymphoproliferative diseases.

Analyzing minor malformation score in healthy elementary school children, Firestone et al. (7), and Steg and Rapoport (8) report on a mean score of 1.4 and 2.88 for boys, respectively. In girls, Waldrop and Goering (9), and Rosenberg and Weller (10) found a mean score of 3.54 and 2.61, respectively. In the general population, the mean score ranges from 2.7 (11) to 3.53 (12). In a Swedish study in a group of children aged 3-17 years, a higher prevalence of minor anomalies was recorded among mentally retarded children (13).

The hypothesis of the present study was that groups of children with developmental disturbances (mental retardation, hearing and vision impairments) have a significantly higher prevalence of oral and dental structure anomalies and minor physical anomalies than healthy children from the general population. To test the hypothesis, anomalies of oral and dental structures, and minor physical anomalies were studied in the groups of children with specific developmental disturbances, i.e. in children with mental retardation, children with vision impairments (partial and complete loss of vision), and children with hearing impairments (partial and complete hearing loss).

The aim of the study was to assess the prevalence of oral and dental structure anomalies in the three groups of children with developmental disturbances, and to compare these findings to those obtained in the control group of healthy children. Also, our intention was to compare the findings obtained with the values of minor malformation score according to Waldrop et al. (4, 9) in children with various developmental disturbances, and to assess the degree of association for oral and dental anomalies.

Subjects and methods

The prevalence of minor physical anomalies, and of oral and dental anomalies was studied in a sample of 606 subjects aged 5-20 years, including 303 subjects with developmental disturbances (176 with mental retardation, 70 with hearing impairment, and 57 with vision impairment), and in a control group of 303 school children free from any of the above mentioned developmental disturbances on systematic examination. The distribution of study subjects according to sex and diagnosis is shown in Table 1. The group of subjects with developmental disturbances included 165 boys and 138 girls, whereas the control group consisted of 152 boys and 151 girls. Total male to female ratio of 52.3% : 47.7% did not differ significantly from the expected ratio in the population of the Republic of Croatia. A higher proportion of boys in the group of children with developmental disturbances of 54.5%, yielded by X^2-test, also did not differ significantly from the expected ratio.

After analysis of the data obtained, the children with developmental disturbances were divided according to diagnosis, whereas those with identifiable syndromes were excluded from the study. The nine oral and dental anomalies analyzed in the study are listed in Table 2. In case of six oral and dental anomalies found in children with developmental disturbances, weighted score of minor physical anomalies according to Waldrop et al. (4, 9) was calculated.

Mental retardation is a mental disorder due to arrested or inadequate development of the child's intellectual functioning. In the present study, Tenth International Classification of Diseases and Injuries was used, according to which children are classified according to intelligence quotient (IQ) into three grades of mental retardation: (a) mild (IQ 50-69); (b) moderate (IQ 35-49); (c) severe (IQ 20-34); and (d) profound (IQ below 20) (14). In the present study, the children with these grades of mental retardation were included in the group of mentally retarded subjects and were observed together.

Partial hearing loss was defined as hearing impairment at frequencies of 25 to 80 dB, and deafness as hearing loss at speech frequencies of > 81 dB (500 to 4000 Hz) (14). The children with these impairments were included in the group of subjects with hearing impairments.

Partial loss of vision is divided into (a) visual acuity on the better eye with the best possible correction of 0.1 to 0.3 or less; and (b) visual acuity on the better eye of 0.3 to 0.4. Visual acuity of (0.05 on the better eye, with the best possible correction of 0.25 with vision field narrowing to 20 degrees or less, and inability to read letters or marks of Jaeger 8 size on near vision is considered as blindness. These individuals need to learn Braille.The children with these impairments were defined as a group of subjects with vision impairments.

In both groups of subjects, minor physical anomalies and their weighted quantification were assessed according to the method of Waldrop et al. (4, 9, 15). Standard tests for nonparametric analysis (X^2-test and rank of correlation) were used on data analysis.

Results

Analysis of the prevalence of the nine oral and dental anomalies showed the group of children with developmental disturbances (DD) to significantly differ from the control group of healthy children (C) in the findings for four anomalies (Table 2). Consequently, the anomalies of hypodontia, median diastema, superior lip frenulum and pronounced tongue frenulum, forming the anomaly of lingua acreta, were significantly more common in DD children (Table 2). Other anomalies showed no significant differences.

The calculation of Pearson's coefficients of correlation pointed to significant correlation between the anomalies of upper lip frenulum and median diastema in DD children ($p < 0.01$) (Table 3). In the control group of healthy children, significant correlation was found between upper lip frenulum and diastema, microdontia and hypodontia, and microdontia and frenulum of the tongue (Table 4).

Weighted Waldrop score (W) was calculated for those oral and dental anomalies that occurred at a certain rate in all groups of children (Table 5). The highest W values were recorded in the groups of children with partial loss of vision and microdontia (W = 6.00), children with mental retardation and microdontia (W = 5.43), children with hearing loss and dens invaginatus (W = 4.80), and children with mental retardation and frenulum of the tongue (W = 4.45). These findings indicated that the men-

tioned oral and dental anomalies most frequently occur in children with most severely pronounced minor physical anomalies, i.e. in those with highest W values.

Discussion

The analysis of oral and dental anomalies revealed a significant difference between the group of children with developmental disturbance and the control group of healthy children (Table 2). Particular anomalies such as cleft lip and palate, microglossia, premature tooth eruption, impacted teeth, and hyperdontia were not found in the control group. The prevalence of these anomalies in the general population is very low (1, 16-21). Analysis of the prevalence of oral and dental anomalies included nine anomalies with a higher rate of occurrence (Tables 2 and 5). Analysis of the prevalence of anomalies according to sex showed no significant sex difference, thus the male and female subjects were considered together.

The oral and dental anomalies belong to a group of nonspecific abnormalities and may present in a number of syndromes. When associated with congenital malformations, they may point to disorders of various tissues and systems, and may occasionally present as the leading symptom of these states, thus being the key element in their diagnosis and genetic counseling.

Most of the anomalies of the oral and dental structures as well as minor physical anomalies can be detected by inspection on dental examination, some can be detected by anthropometric measurements, whereas others require confirmation by diagnostic methods such as x-ray analysis or by some specific methods, e.g., Graber's test for true superior lip frenulum (22, 23).

Hypodontia, as one of the most common dental anomalies, showed a very high prevalence in the group of children with developmental disturbances (Table 2). The prevalence of hypodontia in the general population is between 2% and 10% (24-27). A high prevalence of hypodontia has been reported in children with mental retardation and those with hearing impairment (26, 28). In the present study, the highest prevalence of hypodontia was recorded in children with partial and total loss of vision (15.8%), followed by mentally retarded children (10.2%), whereas the rate of hypodontia in children with partial and total hearing loss was similar to that found in the control group of healthy children (Table 2). A high prevalence of hypodontia of 11% in mentally retarded children has also been reported by Dixon and Stewart (29), whereas Ericson found a significant rate of hypodontia in children with hearing loss (28).

Median diastema was present at a significant rate in the group of children with developmental disturbances, primarily in association with mental retardation. In the general population, the prevalence of median diastema varies among different populations, ranging from 1.6% to 25.4%, with a slight male predominance (22, 23, 30). In the present study, there was no sex difference, but the prevalence of diastema in the subjects with developmental disturbances showed statistically significant differences according to diagnoses (Table 2). This anomaly was significantly more common in all the three groups of children with developmental disturbances than in the control group of healthy subjects. In case of median diastema, the weighted Waldrop score for minor physical anomalies differed between the group of children with developmental disturbances and control group of healthy children. Analysis of correlations for oral and dental anomalies yielded a significant positive correlation between median diastema and superior lip frenulum (Tables 3 and 4), supporting the hypothesis on the role of persistent and deeply inserted upper lip frenulum in the etiology of median diastema (23).

In all the three groups of children with developmental disturbances, the prevalence of upper lip frenulum differed from that recorded in the control group. All the three diagnoses were equally associated with its occurrence, yielding higher rates compared with its general prevalence. In the control group, the prevalence of this anomaly was significantly lower than its overall mean rate in the children with developmental disturbances (Table 2). In all the three groups of children with developmental disturbances, analysis of the weighted Waldrop score produced considerably higher values in the subjects with this anomaly (Table 5).

Frenulum of the tongue or lingua acreta is an oral anomaly which frequently occurs in some syndromes, but may rarely also occur in the general pop-

ulation of children. In a study conducted in Memphis, this anomaly was found in 4.4% of 500 neonates (31). In the present study, frenulum of the tongue was recorded in 6.6% of the children with developmental disturbances, and in only 1.3% of control group children (Table 2). All the three groups of children with developmental disturbances showed a comparable prevalence of the anomaly.

Other anomalies showed a very low prevalence in the children with developmental disturbances, so their contribution to the weighted Waldrop score of minor anomalies was very small.

Analysis of correlation for most common oral and dental anomalies, presented in Tables 3 and 4, showed significant correlation between particular anomalies. This primarily refers to median diastema and superior lip frenulum in all groups of subjects. In the control group, even more significant correlation was found between microdontia and frenulum of the tongue, and between microdontia and hypodontia, supporting the hypothesis according to which microdontia of particular teeth could be considered a variability in the hypodontia gene expression. In this study, microdontia as a rare anomaly was recorded in 2.8% of the subjects only, however, at a rate exceeding the mean prevalence in all the three groups of children with developmental disturbances. Analysis of the minor anomaly weighted Waldrop score yielded a mean W of (5 for the prevalence of microdontia in the subjects with developmental disturbances, as compared with only 3.75 for healthy subjects. Therefore, the presence of microdontia in the subjects with developmental disturbances could be related to the occurrence of more pronounced minor physical anomalies.

In the children with developmental disturbance, the findings of all oral and dental anomalies differed from those obtained in the control group of healthy school children. Statistically significant differences were recorded for hypodontia, median diastema, upper lip frenulum and frenulum of the tongue. High values of weighted Waldrop score for minor anomalies were obtained in the groups of children with vision impairment and microdontia (W = 6.00), mentally retarded children with microdontia (W = 5.43), children with hearing loss and dens invaginatus (W = 4.80), and mentally retarded children with frenulum of the tongue (W = 4.45). The significantly greater number of oral and dental anomalies, and higher weighted Waldrop score for minor anomalies in the children with developmental disturbances point to the action of common factors during the early development in these groups of children. It can well be postulated that these factors, besides their contribution to the underlying disturbance, also lead to the increased prevalence of oral and dental anomalies, thus increasing the severity of minor physical anomalies in children with developmental disturbances.