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Taurodontism Of Premolars In Turner's Syndrome Patients

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Abstract:

Introduction: *Taurodontism is a dental anomaly defined by enlargement of the pulp chamber of multirooted teeth with apical displacement of the pulp floor and bifurcation of the roots. It can be an isolated trait or part of a syndrome. The complete or partial absence of an X chromosome in the karyotype of phenotypic females has an impact on tooth morphology. The association of taurodontism and X-chromosome aneuploidy indicates that the X chromosome is involved in the regulation of root morphogenesis.*

The purpose of this study was to investigated the root morphology of premolars in girls with Turner syndrome, and to determine the influence of various karyotypes on the study variable.

Material and methods: *The study population consisted of 40 Turner syndrome patients, aged from 9.2 to 18 years, and 40 healthy girls, aged from 9.3 to 18 years, as the control group. The TS patients were subdivided according to karyotype (monosomy X, mosaic, and isochromosome). The occurrence of taurodontism in premolars were analyzed from orthopantomograms and classified as normal, hypotaurodont, mesotaurodont, or hypertaurodont. All statistical calculations were performed by computer programs Statistica 7.1 for Windows and SPSS 23,0. Fisher's Exact 2 test were performed to tested the differences in the prevalence rates of taurodontism between Turner and control group.*

Results: *The results showed significant differences in the frequency of taurodontism between the groups. Taurodontism was registered in eight TS females. Five of them showed unilateral and the other bilateral taurodontism. In the control group only one patient showed unilaterally taurodontism. All affected teeth were mandibular premolars and classified as hypotaurodont. These findings also demonstrate that the karyotype has no effect on taurodontism and indicate that the genes affecting morphogenesis of roots may be the same genes that affect the development of enamel.*

Conclusion: *The development of the teeth involves very complex biological processes (interaction of mesenchymal and epithelial tissuec, morphodifferentiation, apposition and mineralization). Anomalies of teeth present in TS indicate that some of these processes are affected by the lack of X-chromosome.*

Keywords: *Taurodontism, premolars, Turner syndrome, root morphology, tooth morphology.*

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I. Introduction

Turner syndrome (TS) is one of the most frequent genetic anomalies caused by a complete or partial absence of one of the X chromosomes in the karyotype of phenotypic females with gonadal dysgenesis. It affects approximately 1 in 2,000 to 5,000 live female births worldwide [1-3]. The incidence of TS in Macedonia is approximately one in 2,500 girls [4]. The most common karyotype is monosomy X, found in 50-60% of the females, and the less common are the mosaic and isochromosome karyotypes for the long arm of the X chromosome [5]. The most common features of this disease are short stature and gonadal dysgenesis [6]. In addition to short stature, reduced size of the craniofacial complex and retrognathic profile have also been registered [7-11]. The most frequent oral findings are altered occlusal morphology, small teeth, thin enamel, and short roots [12-21].

Taurodontism is a dental anomaly defined by enlargement of the pulp chamber of multirooted teeth with apical displacement of the pulp floor and bifurcation of the roots [22]. The etiology of this condition has not been clearly explained. Many authors assume that the anomaly forms in early fetal life due to the failure of Hertwig's epithelial sheath diaphragm to invaginate at the proper horizontal level [23]. Taurodontism is associated with several craniofacial developmental conditions [24]. Only a few studies have investigated taurodontism in TS females. The association of taurodontism and X-chromosome aneuploidy indicates that the X chromosome is involved in the regulation of root morphogenesis and it more prevalent in individuals with extra X chromosomes [25-27]. The results of these studies show that a higher number of X chromosomes tends to enhance the expression of the trait [26].

The aim of the present study was to investigated the root morphology of premolars in girls with Turner syndrome, and to evaluate the influence of various karyotypes on the study variable.

II. Material and Methods

This investigation was part of a systematic study whose purpose was to study development specific to children with Turner syndrome and to determine the influence of various karyotypes on the study variables. Study was approved by Teaching and Science Research Council of Ss. Cyril and Methodius University -Skopje. The karyotyping was done by chromosome analysis of peripheral lymphocytes. The study population consisted of 40 individuals with TS, aged from 9.2 to 18 y, who were patients at the Pediatric Clinic, Medical Faculty, University of Skopje. Forty healthy girls, aged from 9.3 to 18 y, patients at the Department of Orthodontics, Faculty of Dentistry, University of Skopje, were selected as the control group. Written permission has been obtained from the parents of the children included in the study. None of the patients had undergone previous orthodontic treatment. Those with TS were subdivided according to karyotype (monosomy X, mosaic, and isochromosome) so that karyotypic phenotypic correlations could be studied. The karyotypes, age ranges, and mean ages of the study groups are presented in Table 1. The occurrence of taurodontism in premolars were analyzed from orthopantomograms and classified as normal, hypotaurodont, mesotaurodont, or hypertaurodont [28]. All radiographs were examined in a blind fashion by the one investigator (CBM).

All statistical calculations were performed by computer programs Statistica 7.1 for Windows and SPSS 23,0. Fisher's Exact 2 test were performed to tested the differences in the prevalence rates of taurodontism between Turner and control group.

Table 1. TS Patients and Controls According to Age and Karyotype.

Karyotype	n	Age (y) range	mean
Monosomy X 45,X	26	9.2-18	14.7
Mosaic	11	9.3-18	15.1
Isochromosome 46,X,i (Xq)	3	9.8-18	14.1
Turner syndrome (total)	40	9.2-18	14.8
Control group	40	9.3-18	14.9

III. Results

Significant differences were found in the frequency of taurodontism between the TS and control groups (Tab.2). Taurodontism was registered in eight TS females. Five of them showed unilateral and the other bilateral taurodontism. In the control group only one patient showed unilaterally taurodontism. All affected teeth were mandibular first and second premolars and classified as hypotaurodont. The frequency of taurodontism in TS patients was 20% and in the control group 2,5%. The investigation revealed no significant differences between the karyotypes.

Table 2. Comparisons of the prevalence of taurodontism between TS Groups and Control Group.

	TS 45,X		TS other types		Controls		
	n	%	n	%	n	%	P
Taurodontism	4	15.4	4	28.6	1	2.5	P < 0.001
No taurodontism	22	84.6	10	71,4	39	97.5	

Fisher's exact test; ***P < 0.001.

IV. Discussion

The results showed significant differences in the frequency of taurodontism between the two groups. Comparison of these obtained values with the results from other authors could not be done due to the absence of such data in the literature. Varrela et al. [27] examined the frequencies of taurodontism in mandibular molars in 45,X females, first-degree female relatives of these females, and a population sample of normal females and normal males. They suggested that the X chromosome influencing development of enamel may also be involved in the development of taurodontism. Examining the root morphology of molars in girls with Turner syndrome, Bajraktarova Mishevska et al. [29], concluded that taurodontism in molars occurs in TS women with a frequency similar to that in healthy women.

The frequencies in 45,X females and in normal females are near what is expected on the basis of a model with a single dominant gene. However, as also indicated by the earlier family data, the inheritance of taurodontism is more complex, probably involving a polygenic system. It has been suggested that a delay in the growth of the processes would cause the formation of a taurodont molar [30].

Growth and its regulatory mechanisms are under the influence of genes on the X chromosome, and because of this, these genes have an impact on the size of the maxilla and teeth, as a result of the interaction between mesenchyme and epithelium [31]. Numerical aberrations of the X chromosome influence the quantitative and qualitative excretion of amelogenin, which causes a reduction in the dimensions of the dental crown and enamel hypoplasia [32]. Disturbances of odontogenesis in these individuals happens at an early stage of morphogenesis [21].

The genes on the human X chromosome also influence the root morphogenesis of the teeth. Since both the frequency and expressivity of taurodontism seem to be positively affected by extra X chromosomes [25,26], a reversed trend was expected in 45,X females [27]. Several authors have also been observed increased numbers of first and second premolar roots [33-36]. The most frequently observed deformities are two rooted mandibular first and second premolars. Kusiak et al. [36], examined the morphology of the mandibular premolar roots in 40 TS females; they reported that two-rooted mandibular first premolars were observed in 34% of cases in the 45,X group, and in 31% with mosaicism and structural aberrations of X chromosome. On the other hand, approximately 98% of the mandibular first premolars were single rooted; the incidence of two roots was 1.8% and three roots were found in 0.2% of all teeth studied [37]. Furthermore, considering that the general prevalence of taurodonts was reported to range 2.5 - 11.3% of the human population, it is assumed to be a very rare case that all the mandibular premolars were diagnosed to be taurodonts [37]. Taurodontism also has been reported that it is sometimes associated with several syndromes and anomalies, including Down syndrome, Klinefelter's syndrome, amelogenesis imperfecta, ectodermal disturbance and others [38].

V. Conclusion

The development of the teeth involves very complex biological processes (interaction of mesenchymal and epithelial tissuec, morphodifferentiation, apposition and mineralization). Anomalies of teeth present in TS indicate that some of these processes are affected by the lack of X-chromosome. Our findings show significant differences in the frequency of taurodontism between the groups. These results also demonstrate that the karyotype has no effect on taurodontism and indicate that the genes affecting morphogenesis of roots may be the same genes that affect the development of enamel.

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