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Taurodontism in Turner Syndrome Karyotypes.

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ABSTRACT

The complete or partial absence of an X chromosome in the karyotype of phenotypic females has an impact on tooth morphology. The purpose of this study was to investigated the root morphology of molars in girls with Turner syndrome, and to determine the influence of various karyotypes on the study variable. 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 molars were analyzed from orthopantomograms and classified as normal, hypotaurodont, mesotaurodont, or hypertaurodont. The differences in the prevalence of taurodontism between Turner and control patients were statistically tested by Fisher's Exact test. The results showed non-significant differences in the frequency of taurodontism between the groups. 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.

Keywords: Taurodontism, permanent molars, tooth morphology, Turner syndrome.

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INTRODUCTION

Turner syndrome (TS), also referred to as Ullrich-Turner syndrome, 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 [1]. It affects approximately 1 in 2,000 to 5,000 live female births worldwide [2-4]. The incidence of TS in Macedonia is approximately one in 2,500 girls [5]. 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 [6,7]. Short stature and gonadal dysgenesis are the main characteristics of this disease [8]. In addition to short stature, cranial growth reduction has also been registered. Comparison of craniofacial proportions with those of unaffected children showed reduced size of the craniofacial complex, retrognathic profile, and increased cranial base angle [1,9-14]. The most frequent oral findings are small teeth, thin enamel, and short roots [15-21].

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 [22-24]. The results of these studies show that taurodontism is more prevalent in individuals with extra X chromosomes. They found that a higher number of X chromosomes tends to enhance the expression of the trait [23].

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

MATERIAL AND METHODS

This research 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. The karyotyping was done by chromosome analysis of peripheral lymphocytes. The study population was comprised 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. The TS patients were subdivided according to karyotype (monosomy X, mosaic, and isochromosome) so that karyotypic phenotypic correlations could be studied.

The occurrence of taurodontism in molars were analyzed from orthopantomograms and classified as normal, hypotaurodont, mesotaurodont, or hypertaurodont [25]. All radiographs were examined in a blind fashion by the one investigator (CBM). The differences in the prevalence of taurodontism between Turner and control patients were statistically tested by Fisher's Exact test.

RESULTS

No significant differences were found in the frequency of taurodontism between the syndrome and control groups. Taurodontism was registered in four TS females. Two of them showed unilateral and the other bilateral taurodontism. In the control group three patients showed taurodontism, one unilaterally and two bilaterally. All affected teeth were first mandibular molars and classified as hypotaurodont. The frequency of taurodontism in TS patients was 10% and in the control group 7,5%. The investigation revealed no significant differences between the karyotypes.

DISCUSSION

The results showed that taurodontism occurs in TS females with a frequency similar to that in normal females. Comparison of these obtained values can be made only with the findings of Varrela et al. (1990) [24] due to the absence of such data in the literature. Varrela et al. (1996) [24] examining 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. The findings are similar but the frequencies of taurodontism differ.

Quantitative and qualitative changes in the X chromosomes in TS, due to different mechanisms, influence the processes of development and contribute to dysmorphology and changes in craniofacial morphology. Studying the complex mechanisms of craniofacial development, and the possible reasons for dysmorphology, Hall (1988) [26] stated that the development of the craniofacial morphology represents the culmination of a series of different situations that are superposed. All these events are associated with the three basic developmental processes – cell-term differentiation, morphogenesis, and growth – disorders in the development of any of which can cause irreversible effects on craniofacial morphology, as the result of their impact on pathogenesis in TS.

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 [27]. 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 [28]. Disturbances of odontogenesis in these individuals happens at an early stage of morphogenesis [29].

The genes on the human X chromosome also influence the root morphogenesis of the molar teeth. Since both the frequency and expressivity of taurodontism seem to be positively affected by extra X chromosomes [22, 23], a reversed trend was expected in 45,X females [24]. Curiously, 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 [24]. It has been suggested that a delay in the growth of the processes would cause the formation of a taurodont molar [30].

Our results show non-significant differences in the frequency of taurodontism between the groups. 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.

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