

Dental arch dimensions and palatal height in children with Turner syndrome

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ABSTRACT

Introduction: A complete or partial absence of an X chromosome in girls with Turner syndrome (TS) has an impact on dental arch morphology. The aim of this study was to analyze the sizes of dental arches as well as the height of the palatal vault in patients with TS, and to determine the influence of various karyotypes on the study variables.

Materials and methods: The study population sample consisted of 40 TS female patients, aged 9.2 to 18 years, and 40 healthy females aged 9.3 to 18 years, as the control group. The TS patients were subdivided according to karyotype (monosomy X, mosaic, and isochromosome). Dental arch width and length, and palatal height were measured directly on plaster dental casts.

Results: The results showed differences in the dental arch dimensions between TS and control groups. The maxillary dental arch was narrower and shorter, but the mandibular arch was shorter and of normal width in the TS group. The palate in TS patients were normal in height and did not differ from those of the control subjects. Although the dental arch dimensions and palatal height were slightly reduced in mosaic and isochromosome karyotypes compared with those in 45,X females, the investigation revealed no significant differences between the karyotypes.

Conclusions: Our findings indicate that the reduction of X chromosomal genetic material in Turner syndrome females results in the reduction of dental arch dimensions, which is further reflected in the increased frequency of malocclusion in these individuals. Importantly, this study demonstrates that palatal height is normal, contrary to some reports in the literature. It is further significant that the TS karyotype does not affect dentoalveolar morphology and palatal height.

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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. 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.^{1,2} Short stature and gonadal dysgenesis are the main characteristics of this disease.³ while reduced cranial growth has also been reported in some cases. Cranial growth reduction has also been registered.⁴⁻⁹ The most frequent oral findings are small teeth, thin enamel, and short roots.¹⁰⁻¹⁶ The commonly observed malocclusions include distal molar occlusion, tooth crowding, lateral crossbite, and open bite associated with a narrow maxilla.^{11,17-19} In addition, mandibular arches are reported to be wide and short in TS females.^{11,15,20,21}

In several studies²²⁻²⁴, a high arched palate has been described as a specific feature of TS; however, the majority of these studies are based on subjective assessment of the investigated features.

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The aim of this study was to analyze the sizes of dental arches as well as height of the palatal vault in patients with Turner syndrome compared with healthy controls, and to determine the influence of various karyotypes on the study variables.

MATERIALS 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. Study was approved by Teaching and Science Research Council of Ss. Cyril and Methodius University of Skopje. The karyotyping was done by chromosome analysis of peripheral lymphocytes.

The study population consisted of 40 Turner syndrome patients aged 9.2 to 18 years, at the Pediatric Clinic, Medical Faculty, University of Skopje, and 40 healthy females aged from 9.3 to 18 years, patients of the Department of Orthodontics, Faculty of Dentistry, University of Skopje, selected as the control group. Written permission has been obtained from the parents of the children included in the study. No patients had received previous orthodontic treatment. TS patients and control were age - matched. The TS patients 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.

Dental arch width and length, according to Nance,²⁵ were measured directly on plaster dental casts by means of a Korkhaus orthodontic caliper (029-360-01; Dentaureum GmbH, Ispringen, Germany). The intercanine width was measured as the distance between the cusp tips of the right and left canines, the intermolar width as the distance between the mesiobuccal cusp tips of the right and left first molars, and the dental arch length as the sum of the right and left distances from mesial anatomic contact points of the first permanent molars to the contact point of the central incisors or to the midpoint between the central incisors if spaced (Figure 1). A Korkhaus three - dimensional orthodontic caliper (028-353-00; Dentaureum GmbH, Ispringen, Germany) was used to measure palatal height (Figure 2). All measurements were made twice by one investigator who was blinded to the karyotypes of the females during the measurements. The error of measurement was estimated by τ , calculated by the formula $\tau = \Sigma d^2/2n$, where d is the difference between the double measurements and n is the sample size (Dahlberg, 1940).²⁶

All statistical calculations were performed by computer programs (Minitab, 1991).²⁷ Student's t - test was used to analyze the dental arch and palatal height differences between the TS and control groups, and between the 45,X karyotype and the other karyotypes. One - way analysis of variance (ANOVA) was used to evaluate the influence of various karyotypes on these variables. $P < 0.05$ was considered statistically significant.

Table 1. TS patients and controls according to age and karyotype

| Karyotype | Sample Size (n) | Age (years) | |
|-------------------------------|-----------------|-------------|------|
| | | 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 |

Figure 1. Dental cast measurements.

A + B = arch length
C = intercanine width
D = intermolar width

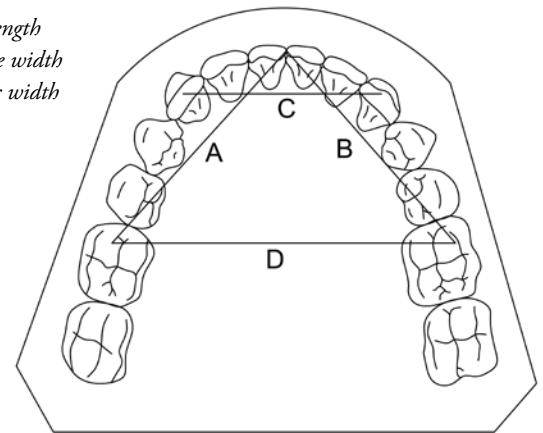
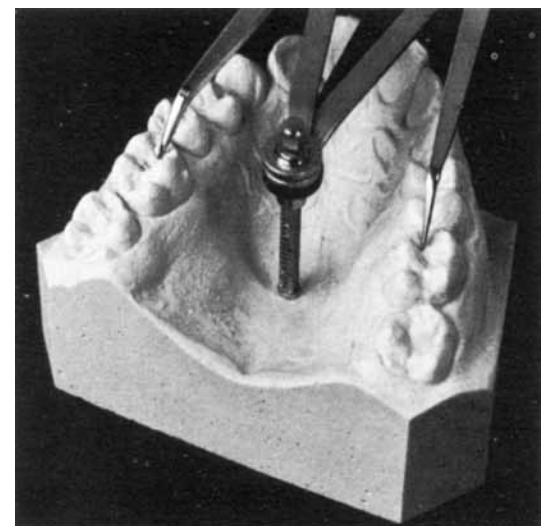


Figure 2. Palatal height measurement.



RESULTS

A comparison of the means of the dentoalveolar arch and palatal height variables between 45,X, mosaic, and isochromosome karyotypes and healthy controls is presented in Tables 2 and 3. The results showed significant differences in the dental arch dimensions between the two groups. TS females had a smaller maxillary intercanine and intermolar width ($P < 0.001$ and $P < 0.05$, respectively) and shorter maxillary and mandibular dental arch length ($P < 0.01$ and $P < 0.05$, respectively) (Table 2).

No significant differences were found in the mandibular intercanine and intermolar widths between the syndrome and control groups. The palate in TS females was normal in height and did not differ from that in the control group (Table 3).

Although the dental arch dimensions and palatal height were slightly reduced in mosaic and isochromosome karyotypes compared with those in 45,X girls, the investigation revealed no significant differences between the karyotypes.

Table 2. The width and length of the maxillary and mandibular dental arches in TS groups compared with the control group

| Variable | TS 45,X (n=26) | | TS Other Types (n=14) | | Controls (n=40) | |
|-------------------------------|-------------------|------|--------------------------|------|--------------------|------|
| | Mean | SD | Mean | SD | Mean | SD |
| Maxillary intercanine width | 25.46*** | 2.45 | 25.08*** | 4.13 | 29.67 | 3.42 |
| Maxillary intermolar width | 48.95* | 2.39 | 48.29* | 3.95 | 50.13 | 2.31 |
| Mandibular intercanine width | 21.32 | 2.29 | 21.88 | 2.36 | 22.31 | 2.09 |
| Mandibular intermolar width | 45.92 | 2.85 | 44.88 | 2.48 | 44.58 | 2.56 |
| Maxillary dental arch length | 66.92** | 4.69 | 63.75*** | 6.71 | 69.83 | 2.68 |
| Mandibular dental arch length | 60.08* | 3.61 | 59.16* | 3.09 | 61.95 | 3.78 |

*P < 0.05; **P < 0.01; ***P < 0.001.

Standard deviation (SD)

Table 3. The height of the palatal vault in TS groups compared with the control group

| Variable | TS 45,X (n=26) | | TS Other Types (n=14) | | Controls (n=40) | |
|-----------------------------|-------------------|------|--------------------------|------|--------------------|------|
| | Mean | SD | Mean | SD | Mean | SD |
| Height of the palatal vault | 34.60 | 5.42 | 32.79 | 4.10 | 35.01 | 4.97 |

DISCUSSION

The width and length of the dental arches in Turner syndrome patients were significantly reduced compared with controls. The maxillary dental arch was narrower and shorter, but the mandibular arch was shorter and of normal width. The similar dentoalveolar features in females with TS have been reported in earlier studies.^{15,20,21,28} Ogiuchi et al. (1985)⁵, identified a greater width in the coronal and basal arch, while Szilagyi et al. (2000)¹⁵ noted a narrower and normal (in length) maxillary dental arch and a wider but shorter mandibular arch in TS females. Analysis of these data indicates the retarded development of the maxillary jaw in the forward direction, forming wide, flat facial characteristics of patients with TS. According to Laine

and Alvesalo (1986),²⁸ the broader but shorter mandibular dental arch, compared with the narrower but normal (in length) maxillary arch, has a negative impact on facial growth in individuals with a deficiency of one X chromosome. Our findings of a reduction in the lengths of the dental arches are in agreement with those of Ogiuchi et al. (1985),²⁰ who also found shorter dental arches. The low position of the tongue among females with TS increases the pressure of the cheek on the maxillary dental arch, which results in a reduction in its size.²⁹ The narrow maxillary dental arch and normal width of the mandibular arch result in the increasing prevalence of malocclusions such as distal molar occlusion, tooth crowding, lateral crossbite and deep bite. These malocclusions are reported to be common among females with TS.^{11,15,18,19}

In the scientific literature, there are conflicting data on the width and height of the palate in individuals with Turner syndrome. According to Gorlin (1963),²² the appearance of a high palate is present in approximately 35% of this population. Laine et al. (1985),³⁰ Pospieszynska and Korman (1998),³¹ Perkiömäki and Alvesalo (2008),²⁹ and Rizell et al. (2013),²¹ researching the changes in the palatal dimensions of individuals with Turner syndrome, identified narrow but normal (in height) palates. Our findings of normal palatal height in Turner syndrome patients support these results. In contrast, López et al. (2002),²⁴ studying oral characteristics of 23 Argentinean girls with Turner syndrome, noted the appearance of a high gothic palate in all those with this syndrome. Opinions on the appearance of a high palate as a common characteristic of several syndromes are usually based upon the subjective judgment of the researcher and not on the basis of concrete investigation. When palatal dimensions in these individuals are measured, they are often found to be narrow but normal in height. This reflects the effects of sex chromosomes on width but not other dimensions of the palate, resulting in deficient transverse growth of the palate, possibly through decreased growth of the palatal shelves or through disturbances in the growth of the nasal septum.³⁰ It is assumed that the short stature homeobox (SHOX) gene, which plays a key role in the appearance of reduced growth in girls with Turner syndrome, is involved in the development of the characteristic palatal shape in these individuals.³² The width of the palate is under the strong influence of the sex chromosomes. Research by Laine and Alvesalo (1993)³³ indicates an increase in palatal width with increased numbers of chromosomes, but men with the 47,XXY karyotype have palates narrower than those of normal 46,XY males, pointing to the fact that the role of the sex chromosomes is not decisive for the achievement of a certain palatal width.³⁴ No statistically significant differences in the dimensions of dental arches and palatal height were determined between different karyotypes among the individuals with Turner syndrome. Comparison of these obtained values can be made only with the findings of Rizell et al. (2013),²¹ due to the absence of such data in the literature. Rizell et al. (2013),²¹ examining the impact of karyotype and age on the

dimensions of maxillary and mandibular dental arch and palatal height, noted that karyotype did not affect the examined parameters, while age influenced only the size of the dental arch. In this study we examined only the impact of karyotype on the dental arch dimensions and palatal height in children with Turner syndrome, and not include the impact of age on the examined parameters which is a limitation of the study. Our findings indicate that the reduction of X chromosomal genetic material in Turner syndrome females results in the reduction of dental arch dimensions. This study is also important because it indicate that palatal height is normal in these individuals. It is

primarily significant for the finding that the TS karyotype does not affect dentoalveolar morphology and palatal height.

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