

Morphological characteristics of the occlusal surfaces of the first permanent molars in individuals with Turner Syndrome

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Abstract:

Introduction: The complete or partial absence of an X chromosome in the karyotype of phenotypic females has an impact on tooth morphology. The aim of this study was to determine the morphological characteristics of occlusal surfaces of first molars in girls with Turner syndrome (TS).

Material and methods: The study population was comprised of 40 individuals with TS, aged from 9.2 to 18 years, and 40 healthy girls, aged from 9.3 to 18 years, selected as the control group. The number and size of the cusps, and the shape of the occlusal surface of the first molars of both jaws were analyzed on intraoral images and plaster dental casts. A chi-square test was used for analysis of the differences in first molar occlusal morphology between TS and control group.

Results: The results showed significant differences in the occlusal morphology of the first molars between the study groups manifested as, missing or reduction in the size of the distopalatal cusp of the maxillary first molar and distobuccal cusp of the mandibular first molar. The changes were registered equally from both sides of the jaws, twice as often in the lower than the upper jaw. Carabelli's trait was less present in subjects with Turner-syndrome, with a frequency approximately twice lower compared with the control group. Reduced size and number of cusps of first molars resulted in the transformation of occlusal surfaces, with the consequent loss of fissure system.

Conclusion: The development of the teeth involves very complex biological processes (interaction of mesenchymal and epithelial tissue, morphodifferentiation, apposition and mineralization). Anomalies of teeth present in TS indicate that some of these processes are affected by the lack of X-chromosome. Deviations in occlusal morphology of the first permanent molars in individuals with Turner-syndrome defined in the survey, confirm this.

Keywords: Carabelli's trait, first permanent molar, occlusal morphology, tooth morphology, Turner syndrome.

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]. Short stature and gonadal dysgenesis are the main characteristics of this disease [5]. Cephalometric studies have reported reduced size and an altered morphology of the craniofacial complex, retrognathic profile, increased cranial base angle, and reduced posterior cranial base in females with TS [6-11]. The most frequent oral findings are reduced tooth crown size, thin enamel, short roots and early eruption of permanent teeth [12-18]. Only a few studies have investigated the first molar occlusal morphology in TS females. Macesic et al. (2003), found reduced distolingual cusp on the upper molars and distal cusp on the lower molar [19]. Identical findings were also made by Midtbo and Halse (1994), who found altered tooth morphology in Turner patients [14]. Mayhall and Alvesalo (1992) [20] have noted a reduction of cusp areas and volumes of the maxillary first molars in the 45,X0 females, while Nakayama et al. (2007) [21] showed association between Carabelli's trait and mesiodistal tooth crown diameter in 45,X females.

The purpose of this study was to determine the morphological characteristics of occlusal surfaces of first molars in girls with Turner syndrome, by analyzing the number and size of the cusps, and the shape of the first molars occlusal surfaces.

II. Material and Methods

This research was part of a systematic study whose purpose was to study development specific to children with Turner syndrome. The study population was comprised of 40 individuals with TS, aged from 9.2 to 18 y (mean age 14.8 y), who were patients at the Pediatric Clinic, Medical Faculty, University of Skopje. Forty healthy girls, aged from 9.3 to 18 y (mean age 14.9 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. The number and size of the cusps, and the shape of the occlusal surfaces of the first molars of both jaws were analyzed on intraoral images and plaster dental casts. Teeth with large restorations or damaged by trauma or caries, were excluded from this analysis. Because in only 6 patients in the control group were observed changes in the number and size of cusps of the first molars, which is the individual variation in the development of these teeth, there was no logic to making intraoral images of patients studied in this group.

All statistical calculations were performed by computer programs (Minitab, 1991) [22]. The differences in morphological characteristics of occlusal surfaces of first molars between Turner and control patients were compared by means of the chi-square test. $P < 0.05$ was considered statistically significant.

III. Results

The analysis of the number and size of cusps of first permanent molars of both jaws showed significant differences ($p < 0.001$) between patients with TS and the control group (Table 2). The changes were observed with equal frequency on both sides of the jaws, so in Table 1 shows the relative frequency of the characteristic changes in the occlusal surfaces of first molars only on the right side of the jaws.

In TS females, reduced distobuccal cusp or hypoconid was found in 26,5% of the mandibular first molar. Of the first maxillary molars, 13.5% showed reduced hypocone (Fig.1). Missing distobuccal cusp occurred in 35.3% of the mandibular first molar (Fig.2). In maxillary ones 10.8% missing the distopalatal cusp. Analysis of cusps of the first molars in the control group showed changes in the number and size only in molars of the lower jaw (missing distobuccal cusp in 7.5% and reduction in its size in 5%). Carabelli's trait in patients with Turner syndrome was observed in 8.1% of the maxillary first molars, while in the control group was present in 15% of the examined teeth.

The characteristic changes of the occlusal surfaces of the first permanent molars in Turner syndrome patients, was twice more common in the mandible than in the maxilla.

Table 1. Relative frequencies of each trait distributed in accordance with type of tooth in TS group and the control group

Trait	Tooth affected	Turner syndrome		Control group	
		No. of teeth	Relative frequency (%)	No. of teeth	Relative frequency (%)
Reduced distopalatal cusp (hypocone)	M ¹ max	37	13.5	40	0
Reduced distobuccal cusp (hypoconid)	M ₁ mand	34	26.5	40	5.0
Missing distopalatal cusp (hypocone)	M ¹ max	37	10.8	40	0
Missing distobuccal cusp (hypoconid)	M ₁ mand	34	35.3	40	7.5
Carabelli's trait	M ¹ max	37	8.1	40	15.0

Table 2. Relative frequencies of teeth with traits in TS group and the control group

	No. of patients	No. of teeth		Teeth with traits		Relative frequencies (%)	
		Max.	Mand.	Max.	Mand.	Max.	Mand.
Turner syndrome	40	74	68	24	42	27.0	61.8
Control group	40	80	80	12	10	17.5	22.5

Maxilla $\chi^2 = 415.0, p < 0.001$;

Mandible $\chi^2 = 506.0, p < 0.001$;

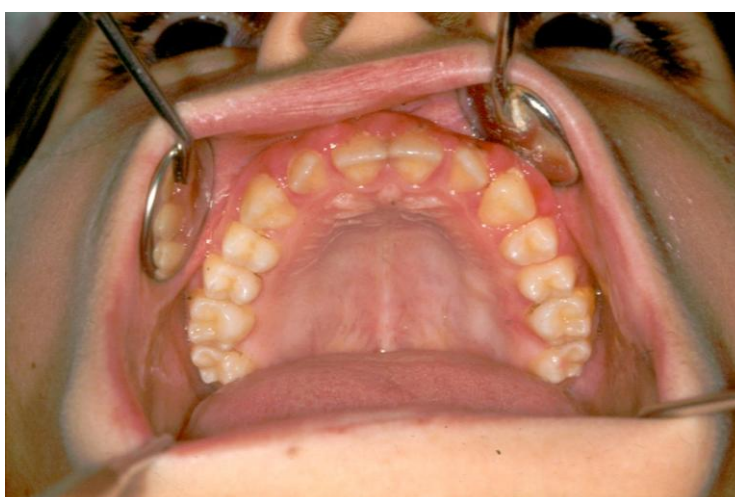


Figure 1. Reduced distopalatal cusp of the maxillary first permanent molars in girls with Turner-syndrome



Figure 2. Missing distobuccal cusp of the mandibular first permanent molars in girls with Turner-syndrome

IV. Discussion

The analysis of the occlusal surface of the first permanent molars showed significant morphological differences between patients with TS and the control group manifested as, missing or reduction in the size of the distopalatal cusp of the maxillary first molar and distobuccal cusp of the mandibular first molar. The changes were registered equally from both sides of the jaws, twice as often in the lower than the upper jaw. The similar findings in females with TS were reported in earlier studies [14,19,20]. Macesic et al. (2003) [19], noted

reduced size of occlusal surface and number cusps on upper and lower molars, while Mayhall and Alvesalo (1992) [20] found decreased basal area, cusp volume, intercuspatal distances and sharper cusps of the permanent upper first molars in 45,X females. According to these authors, the reduction in the number and size of cusps was most pronounced in the distal tuber of the molars.

Carabelli's trait was less present in children with Turner-syndrome, with a frequency approximately twice lower compared with the control group, which is in accordance with the findings of Midtbo and Halse (1994) [14], and Nakayama et al. (2007) [21]. The Carabelli's trait originates from the lingual cingulum region of the maxillary molars, being evident particularly in the first molar [23]. Alvesalo et al. (1975) reported that the existence of the Carabelli's trait was controlled by the hereditary factor, however, the phenotype of Carabelli's trait had large variation [24].

The presence of Carabelli's trait affect the dimensions of the teeth. The molars with Carabelli's trait has larger mesiodistal and buccolingual tooth crown dimensions [25]. According to Nakayama et al. (2007), mesiodistal tooth crown diameter is a key factor to develop the Carabelli's trait in 45,X females [21]. Deviations in the tooth crown morphology in TS patients is a result of insufficient time to develop Carabelli's trait [21]. But, given the way the formation of the occlusal surface (formation of cusps), thinking in this direction may be different; it is more logical to assume that the absence of Carabelli's trait in TS females due to the absence of a genetic signal for its formation than the time factor. The expression of the Carabelli's trait showed low frequency in the maxillary second deciduous and first permanent molars [26].

Dental development and eruption of teeth in individuals with Turner syndrome are more advanced in terms of healthy females [27]. The sex chromosomes (X, Y) have an impact on the development of dental crowns. Y-chromosome triggers the formation of enamel and dentin and the impact of X-chromosome is limited to the apposition of enamel [17,28,29]. The reduced thickness of enamel is the main reason for the smaller dimensions of dental crowns in individuals with TS [17]. Reduced tooth crown dimensions reflect the occlusal morphology of molar teeth. Disturbances of odontogenesis in these individuals happens at an early stage of morphogenesis.

The observed reduction in number and size of the cusps in first permanent molars resulted in occlusal surface shape change of these teeth in both jaws: in the maxillary first permanent molars, the normal rhomboid shape transforms into triangular occlusal shape, while the mandibular teeth lose their regular contours (in the form of latin Y) on the occlusal surface of this molar. These changes are accompanied by a loss of the fissure system regular form.

V. Conclusion

The genes on the human X chromosome are of great importance for the harmonious growth and development of the craniofacial complex and also for normal occlusal relationships and occlusal morphology. The development of the teeth involves very complex biological processes (interaction of mesenchymal and epithelial tissue, morphodifferentiation, apposition and mineralization). Anomalies of teeth present in Turner-syndrome indicate that some of these processes are affected by the deficit of X-chromosome. Deviations in occlusal morphology of the first permanent molars in individuals with Turner-syndrome defined in the survey, confirm this.

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References

- [1] Warburton D, Kline J, Stein Z, Susser M. Monosomy X: A chromosomal anomaly associated with young maternal age. *The Lancet*. 1980;1:167-9.
- [2] Lippe B. Turner syndrome. *Endocrinol Metab Clin North Am*. 1991; 20(1):121-52.
- [3] Frias JL, Davenport ML. Health supervision for children with Turner syndrome. *Pediatrics*. 2003 Mar;111(3):692-702.
- [4] Kočova M. Turner syndrome-challenge for early diagnosis and successful therapy. *Reviews in Pediatrics*. 2010;12:61-7.
- [5] Szilagyí A, Madlena M, Nagy G. The role of the pediatric dentist to initially diagnose and provide early treatment of Turner's syndrome: a case report. *Eur J Paediatr Dent*. 2000;1(4):185-90.
- [6] Jensen BL. Craniofacial morphology in Turner syndrome. *J Craniofac Genet Dev Biol*. 1985;5(4):327-40.
- [7] Peltomaki T, Alvesalo L, Isotupa K. Shape of the craniofacial complex in 45,X females: Cephalometric study. *J Craniofac Genet Dev Biol*. 1989;9(4):331-8.
- [8] Midtbo M, Wisth JP, Halse A. Craniofacial morphology in young patients with Turner syndrome. *Eur J Orthod*. 1996;18:215-25.
- [9] Perkiömäki M, Kyrkanides S, Niinimaa A, Alvesalo L. The relationship of distinct craniofacial features between Turner syndrome females and their parents. *Eur J Orthod*. 2005;27(1):48-52.

- [10] Dumancic J, Kaic Z, Lapter Varga M, Lauc T, Dumic M, Anic Milosevic S, et al. Characteristics of the craniofacial complex in Turner syndrome. *Arch Oral Biol.* 2010;55(1):81-8.
- [11] Bajraktarova Misevska C, Kocova M, Kanurkova L, Curcieva Cuckova G, Bajraktarova B, Maneva M, et al. Craniofacial morphology in Turner syndrome karyotypes. *South Eur J Orthod Dentofac Res.* 2015;2(1):14-20.
- [12] Laine T, Alvesalo L, Savolainen A, Lammi S. Occlusal morphology in 45,X females. *J Craniofac Genet Dev Biol.* 1986;6(4):351-5.
- [13] Mayhall JT, Alvesalo L, Townsend GC. Tooth crown size in 46, X,i (Xq) human females. *Arch Oral Biol.* 1991;36(6):411-4.
- [14] Midtbo M, Halse A. Tooth crown size and morphology in Turner syndrome. *Acta Odontol Scand.* 1994;52(1):7-19.
- [15] Szilágyi A, Keszthelyi G, Madlena M, Nagy G. Morphologic alterations of tooth crown in patients with Turner syndrome and its association with orthodontic anomalies. *Fogorv Sz.* 2000;93(9):268-76.
- [16] Rizell S, Barrenäs ML, Andlin Sobocki A, Stecksén Bliks C, Kjellberg H. Turner syndrome isochromosome karyotype correlates with decreased dental crown width. *Eur J Orthod.* 2012;34(2):213-8.
- [17] Alvesalo L, Tammisalo E. Enamel thickness in 45,X females' permanent teeth. *Am J Hum Genet.* 1981;33:464-9.
- [18] Varrela J, Townsend G, Alvesalo L. Tooth crown size in human females with 45,X/46,XX chromosomes. *Arch Oral Biol.* 1988;33:291-4.
- [19] Mačesič M, Kaič Z, Dumančič J, Poje Z, Dumič M. Occlusal molar surfaces in females with Turner's syndrome. *Coll Antropol* 2003;27(2):761-8.
- [20] Mayhall J T, Alvesalo L. Dental morphology of 45,XO human females: Molar cusp area, volume, shape and linear measurements. *Archives of Oral Biology* 1992;37(12):1039-1043.
- [21] Nakayama M, Lähdesmäki R, Kanazawa E, Alvesalo L. The relationship between Carabelli's trait and Tooth crown size in 45,X females. *Int J Oral-Med Sci* 2007;6(2):61-66.
- [22] Minitab Inc. MINITAB Reference Manual PC Version Release 8. Rosemont (PA):Minitab; 1991.
- [23] Sakai T, Hanamura H. A morphological analysis of Carabelli's cusp. *Aichi-Gakuin Dent Sci* 1971;5:60-72.
- [24] Alvesalo L, Nuutila M, Portin P. The cusp of Carabelli. Occurrence in first upper molars and evaluation of its heritability. *Acta Odont Scand* 1975;33:191-197.
- [25] Kondo S, Townsend GC. Associations between Carabelli trait and cusp area in human permanent maxillary first molars. *Am J Phys Anthropol* 2006;129:196-200.
- [26] Nakayama M, Lähdesmäki R, Kanazawa E, Alvesalo L. Analysis of Carabelli's trait in maxillary second deciduous and permanent molars in 45,X and 45,X/46,XX females. In: Zadzińska E, editors. *Current Trends in Dental Morphology Research.* University of Lodz Press, Poland; 2005. p.325-331.
- [27] Filipsson R, Lindsten J, Almqvist S. Time of eruption of the permanent teeth, cephalometric and tooth measurement and sulphation factor activity in 45 patients with Turner's syndrome with different types of X chromosome aberrations. *Acta Endocrinol* 1965;48:91-113.
- [28] Alvesalo L. Human sex chromosomes in oral and craniofacial growth. *Arch Oral Biol.* 2009;54(1):18-24.
- [29] Zilberman U, Smith P, Alvesalo L. Crown components of mandibular molar teeth in 45,X females (Turner syndrome). *Arch Oral Biol.* 2000;45(3):217-225.