Coll. Antropol. **27** (2003) 2: 761–768
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Original scientific paper

Occlusal Molar Surfaces in Females with Turner's Syndrome

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A B S T R A C T

The aim of this study was to identify the molar occlusal features in 73 subjects with the Turner's syndrome (TS) and compared to a control group (CG) of 322 healthy females. The occlusal features were scored on dental plaster casts using the Scoring Procedures for Key Morphological Traits of the Permanent Dentition: The Arizona State University Dental Anthropology System (ASU). The results were analyzed through frequency, percentage and χ^2-test. TS subjects have more frequent reduction of cusp number, distolingual cusp on the upper molars and distal cusp on the lower molar, with the consequent reduction of the occlusal surface. Reduced size of occlusal surface and number cusps on upper molars resulted in the transformation of rhomboid occlusal shape into triangular, with the consequent loss of H-shaped groove system (in the upper right first molars H-shaped groove system was significantly less frequently found in TS ($p< 0.05$); in the upper left second molars H-shaped groove system was significantly less frequently found in TS ($p<0.01$). The X-chromosome aneuploidy can cause a decrease in developmental homeostasis, which results in the alteration of apposition of the enamel and in consequently substantial changes of the molar occlusal morphological features.

Key words: *molars, Turner's syndrome*

Introduction

The outlines of molar occlusal surfaces are defined polygenetically and are determined by the allelic combination on two or more points, and are completed in one of the terminal developmental stages of molars, as a result of terminal deposition

Received for publication December 3, 2002

of the enamel. In cases of gonadal dysgenesis, when one sex chromosome is absent, or aneuploid, or structurally damaged, many somatic anomalies are present[1–4].

Among other changes in dentition, one of the features is the reduction in size of the crown, which is particularly prominent in mesiodistal direction. The reduced enamel thickness caused by the aberrant sex chromosomes will affect the morphology of occlusal molar surfaces[5–9].

Turner's syndrome includes various clinical manifestations like stunted ovaries (in the form of projections), stunted growth, pterygium colli and cubitus valgus. Studies of the sex chromatin and sex chromosome have demonstrated that approximately one in 2,500–3,000 female babies is born with chromosomal aberrations leading to the gonadal dysgenesis or Turner's syndrome. Karyotype 45, X (monosomy) is responsible for 48–58%, X-mosaicism for 9–38% and X-chromosome defects for 5–43% cases of Turner's syndrome. X-monosomy is characterized by the complete absence of one sex chromosome. This karyotype is usually found in patients with the most serious clinical manifestations such as face deformities, pterygium colli, short stature, »shield« chest with broad-spaced nipples, multiple pigmented naevi, short 4th metacarpals, hypoplastic nails, coarctation of the aorta, amenorrhoea, failure of breast development and juvenile external genitalia. The ovaries are replaced by bilateral streaks of fibrous stroma which are usually devoid of developing ova[10].

Recent odontometric studies in subjects with aneuploid sex chromosomes indicated that both sex chromosomes may influence the growth and development of teeth[5,11,12].

In this study the features of molar occlusal surfaces were observed in a group of patients with Turner's syndrome and compared to the healthy population in relation to:

1. occlusal contour;
2. standard and additional cusps;
3. grooves pattern;
4. additional ridges, pits and grooves.

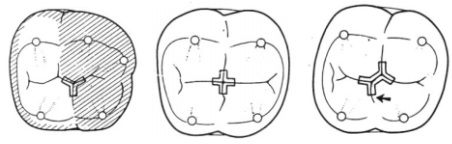
Material and Methods

The material comprised of plaster models of dental arches from the collection of School of Dental Medicine, University of Zagreb.

Seventy-three plaster models from patients with Turner's syndrome (TS) (X-monosomy n=46, X-chromosome defects n= 16, X-mosaicism n=11), as well as 322 plaster models from healthy females (control group–CG) were evaluated.

Turner's syndrome – a total number of 111 upper molars were examined. Of this number 23 were upper third molars (M^3), 47 upper second molars (M^2) and 41 upper first molars (M^1). A total number of 81 lower molars were examined. Of this number 20 were lower third molars (M3) m 42 lower second molars (M2) and 19 lower first molars (M1). Control group–a total number of 1,425 upper molars were examined. Of this number 395 were upper third molars (M^3), 644 upper second molars (M^2) and 386 upper first molars (M^1). A total number of 1,282 lower molars were examined. Of this number 441 were lower third molars (M3), 569 lower second molars (M2) and 272 lower first molars (M1).

Two examiners in mutually independent examinations performed analysis of the material for this study. The scoring of occlusal molar surfaces was performed according to the following standards for characterization of morphological variants of permanent dentition: ASU (The Arizona State University Dental Anthropology System)[13].



A *B* *C*

Fig. 1. A. Original Dryopithecus pattern of grooves. Y-shaped configuration. There distinct contact between the middle buccal cusp (hypoconid) and the mesiolingual cusp (metaconid). B. Plus (+) pattern of grooves in the quadritubercular lower molar, caused by reduction or complete loss of (commonly) the hypoconulid. C. X-pattern of grooves as a result of reduction of the metaconid end expansion of the entoconid. Distinct contact between the mesiobuccal cusp (protoconid) and the distolingual cusp (entoconid).

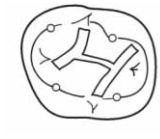


Fig. 2. Grooves between cusps of upper molars show H-shaped pattern.

Lower molars

Lower molar grooves were characterized by 3-stage scoring system introduced by Jorgensen and assessed by the use of 10x hand lens[14]:

Y – cusp 2 and cusp 3 are in contact,
+ – cusps 1–4 are in contact,
X – cusp 1 and cusp 4 are in contact (Figure 1).

Scoring of cusp nuber of lower molars proposed by Gregory was, with minor adaptations, incorporated into ASU system[15,16]:

1. Cusps 1–4 (1. protoconid (mesiobuccal – MB); 2. metaconid (mesiolingual – ML); 3. hypoconid (distobuccal – DB); 4. entoconid (distolingual – DL)) are present;
2. Cusp 5 (hypoconulid (distal – D)) is also present;
3. Cusp 6 (entoconulid) is also present.

Upper molars

Grooves between cusps of upper molars show H-shaped pattern (Figure 2).

Cusps 1–4: 1. protocon (mesiobuccal – MB); 2. metacon (mesiolingual – ML); 3. hypocon (distobuccal – DB); 4. entocon (distolingual – DL).

The following associations among parameters observed were statistically analysed:

1. The distribution of traits (absolute and relative), assessed according to scoring scales for each tooth individually.

2. Contingensy tables were set up to assess the association between observed traits for each tooth individually. Tables were not done for the traits showing minimal or no variability. The associations within tables were tested χ^2-test and the results of the analysis were presented as a probability of the null hypothesis for the non-existence of association.

Results

Upper molars

The upper right third molar: no differences in distributions neither of occlusal surface features nor in the presence of cusps on this tooth were found between patients (TS) and healthy subjects (CG). Additional ridges were more frequently seen in TS than in CG (Table 1).

The upper left third molar: triangular outline of occlusal surface was more frequently seen in TS, while rhomboid was found only in CG. Mesiolingual cusp was more frequently seen in CG. H-shaped groove system was found only in CG. Additional pits and grooves were more frequently in TS.

TABLE 1
DISTRIBUTION OF UPPER MOLARS' TRAITS (%) IN TURNER'S SYNDROME (TS) AND IN CONTROL GROUP (CG)

	M3				M2				M1			
	Right		Left		Right		Left		Right		Left	
	T S	CG	TS	C G	T S	C G	T S	C G	T S	C G	T S	C G
Shape												
Trian.	76.92	73.89	80.00	72.92	59.09	39.75	52.00	34.16				
Rhomb.	7.69	7.39	0.00	8.33	36.36	54.04	40.00	58.08	100.00	98.95	95.00	97.95
Ellip.	15.39	18.72	20.00	18.75	4.55	6.21	8.00	7.76				
Tetra.									0.00	1.05	5.00	2.05
Cusp												
MB	100.00	100.00	100.00	100.00	100.00	100.00	96.00	100.00	100.00	100.00	100.00	100.00
ML	100.00	99.51	90.00	99.48	100.00	100.00	100.00	99.69	100.00	100.00	100.00	100.00
DB	100.00	99.51	100.00	100.00	100.00	100.00	100.00	99.69	100.00	100.00	100.00	100.00
DL	23.08	22.17	20.00	22.92	45.45	60.25	52.00	66.77	100.00	100.00	100.00	99.49
H-sh. sys.	0.00	1.97	0.00	3.65	30.00	46.89	22.73	50.62	73.33	90.05	85.71	91.28
Add. ridg.	15.38	9.85	10.00	11.46	5.00	9.01	18.18	8.70	20.00	8.38	7.14	7.69
Add. pits	7.69	7.88	20.00	5.21	0.00	5.90	0.00	2.48	6.67	1.57	7.14	2.05
Add. groov.	15.38	14.78	20.00	15.63	10.00	8.70	9.09	9.94	6.67	4.19	0.00	4.62

The upper right second molar: triangular shape of the occlusal surface was more frequently seen in TS, while the rhomboid shape of occlusal surface was more frequently in CG. Distolingual cusp was more frequently seen in CG. H-shaped system of grooves appeared more frequently in CG. Additional ridges were more frequently seen in CG. Additional pits were found only in CG.

The upper left second molar: the outline of the occlusal surface of this tooth was in TS more frequently triangular than in CG, while the rhomboid outline was more frequently observed in CG. Distolingual cusp was more frequently seen in CG. H-shaped groove system was significantly more frequently found in CG ($p<0.01$). Additional ridges appeared more frequently in TS.

The upper right first molar: two groups did not show any differences in distributions of occlusal surface features nor in the presence of cusps. H-shaped groove system was signifiantly less frequently found in TS ($p<0.05$). Additional ridges and pits were more frequently seen in TS.

The upper left first molar: H-shaped groove system was more frequently found in CG. Additional pits were more frequently seen in TS. Additional grooves were found only in CG.

Lower molar

The lower right third molar: elliptic -shaped occlusal surface were more frequently seen in TS than in CG, while the pentagonal shape of the occlusal surface was present in CG only (Table 2). The distal cusp was present in CG only. Groove pattern Y was found only in CG. Groove pattern + was more frequently in TS, but groove pattern X was more frequently seen in CG. Additional ridges, pits and grooves were found in CG only.

The lower left third molar: tetragonal -shaped occlusal surface was more frequently seen in TS. Distal cusp was more

TABLE 2
DISTRIBUTION OF LOWER MOLARS' TRAITS (%) IN TURNER'S SYNDROME (TS) AND IN CONTROL GROUP (CG)

	M3				M2				M1			
	Right		Left		Right		Left		Right		Left	
	T S	CG	TS	C G	T S	C G	T S	C G	T S	C G	T S	C G
Shape												
Ellip.	33.33	17.35	18.18	21.62								
Tetr.	66.67	68.04	72.73	66.67	95.00	94.81	90.91	95.00	50.00	58.78	57.14	58.87
Pent.	0.00	14.61	9.09	11.71	5.00	5.19	9.09	5.00	50.00	41.22	42.86	41.13
Cusp												
MB	100.00	100.00	100.00	100.00	100.00	100.00	100.00	100.00	100.00	100.00	100.00	98.58
ML	100.00	100.00	100.00	99.55	100.00	100.00	100.00	100.00	100.00	100.00	100.00	100.00
DB	100.00	100.00	100.00	99.55	100.00	100.00	100.00	100.00	100.00	100.00	100.00	100.00
DL	88.89	92.24	90.91	91.89	100.00	99.65	100.00	100.00	100.00	100.00	100.00	100.00
D	0.00	36.07	9.09	36.04	5.00	5.54	9.09	5.36	58.33	70.99	57.14	71.63
Groov.pat.												
Y	0.00	4.12	0.00	2.96	0.00	4.55	7.69	4.71	83.33	70.99	66.67	70.07
+	83.33	71.76	75.00	73.96	93.33	90.21	84.62	89.49	16.67	29.01	33.33	29.93
X	16.67	24.12	25.00	23.08	6.67	5.24	7.69	5.80	0.00	0.00	0.00	0.00
Add. ridg.	0.00	12.39	0.00	12.61	0.00	0.35	0.00	0.36	0.00	1.53	0.00	2.84
Add. pits	0.00	1.83	0.00	1.35	6.67	0.35	0.00	0.36	8.33	1.53	0.00	1.42
Add. groov.	0.00	10.09	16.67	11.71	6.67	1.38	6.67	1.79	8.33	2.29	25.00	3.55

frequently seen in CG. Additional ridges and pits were found in CG while in TS were absent.

The lower right second molar: there were no significant differences between two study groups.

The lower left second molar: no differences were seen in any traits between two study groups.

The lower right first molar: tetragonal outline occlusal surface and distal cusp was more frequently seen in CG. Pentagonal outline occlusal surface was more frequently seen in TS. Groove pattern Y was more frequently seen in TS, but groove pattern+was more frequently seen in CG, while additional pits and grooves were more frequently present in TS.

The lower left first molar: the distal cusp was more frequently seen in CG. Additional grooves were more frequently seen in TS.

Discussion

Turner's syndrome is the eponym used to describe the clinical features of females with only one X chromosome (X-monosomy), X-mosaisism and X-chromosome defects. The typical affected newborn presents with marked dorsal lymphoedema of the hands and feet, and with lymphoedema or loose folds of skin over the posterior aspect of the neck. Characteristic features of girls and women are short stature, webbing of the neck, »shield« chest with broad-spaced nipples, multiple pigmented naevi, short 4th metacarpals, hypoplastic nails, coarctation of the aorta, amenorrhoea, failure of breast development and juvenile external genitalia. All permanent teeth in 45, X females are reduced in size, with mesiodistal diameters affected more than buccolingual[5,6,8,17,18]. The results of analysis of upper molar occlusal surfaces in subjects with Tur-

ner's syndrome presented in this study revealed increased frequency of triangular shape of occlusal surface on the upper left third molars in TS. Rhomboid shape was found only in CG. Mesiolingual cusp was more frequently seen in CG. The features of upper right third molars did not show any differences between two study groups except additional ridges who were more frequently seen in TS. Triangular shape of occlusal surface on the second upper molars was seen more frequently in TS, while rhomboid shape that was more frequently in CG. Distolingual cusp was more frequently seen in CG, as was the H-system of grooves, whose incidence on the upper left second molars was significantly higher ($p<0.01$) in CG than in TS. The upper first molars did not show any differences between two study groups, in distributions of occlusal surface features nor in the presence of cusps, but H-shaped of grooves were less frequently found in TS (in the upper right first molars was significantly less frequently in TS ($p<0.05$) than in CG).

Distal cusp was more frequently seen on the lower left third molars in CG than in TS; on the lower right third molars the cusp was present only in CG. No differences were seen between two groups in any feature of the lower second molars. The distal cusp of the lower first molars was more frequently found in CG. The reduction of distolingual cusp on upper molars and of distal cusp on lower ones was probably caused by the reduction of enamel thickness, with the consequent reduction of the occlusal surface. Reduced size and number cusps on upper molars resulted in the transformation of rhomboid occlusal shape into triangular, with the consequent loss of H-shaped groove system. Mayhall and Alvesalo have noted in 45,X females in permanent upper first molars, decreased basal area, cusp volume, intercuspal distances and sharper cusps[19]. Alvesalo concludet from studies on individuals with sex-chromosome anomalies have further showed that the X-chromosome genes regulate enamel apposition, while cellular division demarcated by the dentine-enamel junctions and also enamel growth are influenced by the Y-chromosome genes[17]. Studies of tooth size in individuals with chromosomal anomalities have indicated that the severity of growth defects observed may be related to their time of development and location in the jaw. The insult to enamel growth and cell division also provides a developmental basis for the observed change in cuspal shape in 45,X females reported by Mayhall and Alvesalo[4]. Subsequent studies on individuals with sex -chromosome anomalies have further showed that X-chromosome genes regulate enamel apposition. Recent discoveries at the molecular level provide additional confirmation for these results on enamel growth. Nakahori and associates have sequenced the gene for amelogenin from both sex chromosomes. Its location has been mapped to the short arm of the X chromosome and is now considered to be the source of the defect in X-linked amelogenesis imperfecta[20–23]. The study of Zilberman at al. confirmed that the X-chromosome promotes enamel apposition and that both chromosomes in normal females are active in amelogenesis. The relative reduction in »dentine« or the estimated mesiodistal width of the tooth germ in the 45,X females indicates that their tooth development is affected at an early stage of morphogenesis[5,6,8]. This study revealed the following morphological changes in molar occlusal surfaces in women with Turner's syndrome: reduced size and number of cusps, reduced size and changed shape of the surface, and the loss of H-shaped groove system in upper molars. The results of this study, together with already published data, indicate that X-chromosome, responsible for the apposition of enamel, probably induces

the changes or morphological features of occlusal molar surfaces in patients with Turner's syndrome. It may be possible that when only one X-chromosome affects enamel apposition, the magnitude of its effect is progressive and time related[5].

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ZNAČAJKE OKLUZALNIH PLOHA MOLARA U ŽENA S TURNEROVIM SINDROMOM

SAŽETAK

Svrha ovoga rada bila je da se utvrde pojedina svojstva okluzalnih ploha molara u 73 osobe s Turnerovim sindromom (TS), te da ih se usporedi s kontrolnom grupom (CG) koju su činile 322 zdrave žene. Klasifikacija značajki okluzalnih ploha molara određena je prema ASU standardima za karakterizaciju morfoloških varijanti trajnih zuba (Arizona State University – Dentoantropološki sustav Državnog sveučilišta u Arizoni). Rezultati su analizirani kroz učestalost, postotak, te χ^2-test. Osobe s Turnerovim sindromom češće imaju smanjen broj kvržica: distolingvalne kvržice na gornjim molarima, te distalne na donjim molarima, što ima za posljedicu smanjenje površine okluzalne plohe. Smanjenje površine okluzalne plohe i broja kvržica na gornjim molarima dovelo je do promjene romboidnog obrisa okluzalne plohe u trokutast, što je imalo za poslje-

dicu gubitak H-sustava brazdi (gornji desni prvi molar: H-sustav brazdi bio je statistički značajno rjeđi u TS ($p<0,05$); gornji lijevi drugi molar: H-sustav brazdi bio je statistički značajno rjeđi u TS ($p<0,01$)). Aneuploidija X kromosoma može uzrokovati usporavanje razvoja homeostaze koja rezultira promjenom u odlaganju cakline te za posljedicu ima bitne promjene okluzalnih morfoloških obilježja molara.