IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 15, Issue 5 Ver. V (May. 2016), PP 09-12
www.iosrjournals.org

Prevalence Of Malocclusion In Turner Syndrome Karyotypes

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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 occlusal morphology. The aim of this study was to determine the prevalence of malocclusion, by analyzing the sagittal, transversal and vertical occlusion in patients with Turner syndrome (TS) compared with healthy controls.*
Material and Methods : *The study population was comprised of 40 individuals with TS, aged from 9.2 to 18 years, and 50 healthy girls, aged from 9.1 to 18 years, selected as the control group. The occlusion was assessed on the basis of the clinical examination and the analysis of the plaster dental casts. A chi-square test was used for analysis of the differences in sagittal, transversal and vertical occlusion between TS and control group.*
Results: *The results showed significantly increased prevalence of malocclusion in TS females compared with the unaffected girls. The deviations comprised distal molar occlusion, crowding of teeth, lateral crossbite and deep bite. Mesial molar occlusion was registered only in two patients.*
Conclusion: *Individuals with structural and/or numerical aberrations on the X chromosome develop a specific model of craniofacial morphology with deviations in sagittal, transversal and vertical directions. Our findings show a high prevalence of occlusal anomalies in Turner syndrome as a result of imbalanced growth of the craniofacial skeleton in three dimensions. Early diagnosis of occlusal anomalies is of utmost importance in Turner syndrome patients and should be accompanied by an early orthodontic treatment of malocclusion in these individuals.*
Keywords: *malocclusion, occlusal anomalies, occlusal morphology, Turner syndrome.*

I. Introduction

Turner syndrome (TS), also referred to as Ullrich – Turner syndrome, is a combination of characteristic clinical signs and complete or partial absence of an X chromosome in the karyotype of phenotypic females with gonadal dysgenesis [1]. 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 [2,3]. Short stature, gonadal dysgenesis, pterygium colli and cubitus valgus are the most common features of this disease [4]. 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 [1,5-10]. An individuals with Turner syndrome had reduced dental arches [11-14], and narrow but normal (in height) palates [11,13,15-17]. In several studies [11,13,15-17] an altered first molar occlusal morphology has been described as a specific feature of TS [18-21]. Only a few studies have investigated the prevalence of malocclusion in TS females [22-27]. The commonly observed malocclusion include distal molar occlusion, tooth crowding, lateral crossbite and open bite.

The purpose of this study was to determine the prevalence of malocclusion, by analyzing the sagittal, transversal and vertical occlusion in patients with Turner syndrome compared with healthy controls.

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 was

comprised of 40 Turner syndrome patients, aged 9.2 to 18 years, who were patients at the Pediatric Clinic, Medical Faculty, University of Skopje. Fifty healthy girls, aged from 9.1 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. For each of these patients a panoramic roentgenogram and plaster dental casts were taken. None of the patients had undergone previous orthodontic treatment. 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. The occlusion was assessed twice by one investigator (CBM) on the basis of the clinical examination and the analysis of the plaster dental casts.

All statistical calculations were performed by computer programs (Minitab, 1991) [28]. Differences in sagittal, transversal and vertical occlusion between Turner and control patients were compared by means of the chi-square test. $P < 0.05$ was considered statistically significant.

Table 1. Patients distributed on the basis of age and karyotype

	Sample Size (n)	Age (years) Range	Mean
Monosomy X 45,X	26	9.2-18	14.7
Mosaics	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	50	9.1-18	14.7

III. Results

The frequency of malocclusion was 95 per cent in the group of Turner patients. The most common are distal molar occlusion, crowding of teeth, lateral crossbite and deep bite.

45,X karyotype

Distal molar occlusion occurred in 76.9 per cent of the 45,X patients ($P < 0.001$), while mesial molar occlusion was registered in one patient (Table 2). Unilateral and bilateral crossbite was observed in 15.4 per cent and crowding of teeth in 80.8 per cent of the cases (Table 3). None of the 45,X patients had spacing of teeth. Deep bite was registered in 76.9 per cent while lateral open bite in 7.7 per cent of the patients (Table 4).

Mosaic and isochromosome karyotypes

The prevalence of distal molar occlusion was 63.6 per cent in mosaic patients and 66.7 per cent in isochromosome patients (Table 2). Unilateral and bilateral crossbite and crowding of teeth occurred most frequently in the mosaic karyotype (Table 3). In mosaic patients 45.5 per cent showed deep bite (Table 4). None of the patients with isochromosome karyotype had mesial molar occlusion, open bite and deep bite.

Table 2. Sagittal occlusal anomalies in Turner patients subdivided on the basis of karyotype.

Karyotype	Distal		Mesial	
	n	(%)	n	(%)
45,H (n=26)	20	76.9	1	3.8
Mosaics (n=11)	7	63.6	1	9,1
46,X,i (Xq) (n=3)	2	66.7	0	0
Turner syndrome (n=40)	29	72.5	2	5
Control group (n=50)	10	20	3	6

Turner versus controls: $x^2 = 23.587$, $p<0.001$;

Table 3. Transversal occlusal anomalies in Turner patients subdivided on the basis of karyotype.

Karyotype	Unilateral cross bite		Bilateral cross bite		Crowding of teeth		Spacing of teeth	
	n	(%)	n	(%)	n	(%)	n	(%)
45,H (n=26)	4	15.4	4	15.4	21	80.8	0	0
Mosaics (n=11)	3	27.3	1	9.1	9	81.8	1	9.1
46,X,i (Xq) (n=3)	1	33.3	0	0	1	33.3	1	33.3
Turner syndrome (n=40)	8	20	5	12.5	31	77.5	2	5
Control group (n=50)	4	8	2	4	15	30	1	2

Turner versus controls: $x^2 = 60.654$, $p<0.001$;

Table 4. Vertical occlusal anomalies in Turner patients subdivided on the basis of karyotype.

Karyotype	Anterior open bite		Lateral open bite		Deep bite	
	n	(%)	n	(%)	n	(%)
45,H (n=26)	0	0	2	7.7	20	76.9
Mosaics (n=11)	1	9.1	1	9.1	5	45.5
46,X,i (Xq) (n=3)	0	0	0	0	0	0
Turner syndrome (n=40)	1	2.5	3	7.5	25	62.5
Control group (n=50)	2	4	3	6	6	12

Turner versus controls: $x^2 = 22.952$, $p<0.001$;

IV. Discussion

In general, the patients with TS had increased prevalence of malocclusion compared with the unaffected girls. The results showed that 95 per cent of the subjects had some type of anomaly. Comparison of these obtained values can be made only with the findings of Poje et al. (1996) [27] due to the absence of such data in the literature. Poje et al. (1996) [27] examining the frequencies of malocclusion in patients with different types of Turner syndrome, noted 93.9 per cent prevalence of malocclusion in individuals with TS.

The investigation revealed increased prevalence of distal molar occlusion, crowding of teeth, lateral crossbite and deep bite in Turner syndrome patients. The similar findings in TS females were reported in earlier studies by Laine et al. (1986) [22], Harju et al. (1989) [23], Midtbo and Halse (1996) [24], and Scilyagi et al. (1997, 2000) [25-26]. The findings are similar but the frequencies of malocclusion differ. Laine et al. (1986) [22], found an increased prevalence of occlusal anomalies in 45,X females. Identical findings were also made by Harju et al. (1989) [23], who found that the most common type of malocclusion in 45,X/46,XX- and 46,Xi(Xq)-women was lateral cross bite. Also distal molar occlusion, increased maxillary overjet and in 45,X/46,XX-women open bite was found. Compared to 45,X-women, the study groups showed milder expression of malocclusion.

In general, the TS females were characterized by smaller dimensions and an altered morphology of the craniofacial complex. 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, which is further reflected in the increased frequency of malocclusion in these individuals. High frequency of distal molar occlusion in this syndrome is due to abnormal anteroposterior growth of the alveolar processes of maxilla. On the other hand, a disturbance in the transverse craniofacial growth, leads to disharmony in the size of the jaws in this direction, resulting with crossbite in these individuals.

Also, a short posterior cranial base, a posteriorly rotated mandible and reduced posterior face height indicate a skeletal basis for the vertical anomaly [7]. Our findings of increased frequency of deep bite caused by disturbances in craniofacial growth in vertical direction are in agreement with those of Poje et al. (1996) [27], but is contrary to the findings of Laine et al. (1986) [22], Harju et al. (1989) [23], Midtbo and Halse (1996) [24], and Scilyagi et al. (1997, 2000) [25-26], who founds high frequenciy of open bite in TS patients.

Growth and its regulatory mechanisms are under the influence of genes on the X chromosome. The number of X chromosomes also has an effect on mandibular growth relative to maxillary development. The studies of occlusal anomalies in individuals with different chromosomal aberrations have shown the influence and different effects of the sex chromosomes on the occlusal morphology. Research by Laine et al. (1986) [22], indicates an increased frequency of distal molar occlusion in Turner syndrome patients, while Alvesalo and Laine (1992) [29], have determined increased prevalence of mesial molar occlusion in 47,XXY, Klinefelter syndrome. The extra X chromosome causes deviations in sagittal jaw relationships, while its absence affects mandibular form [30].

V. Conclusion

Individuals with structural and/or numerical aberrations on the X chromosome develop a specific model of craniofacial morphology with deviations in sagittal, transversal and vertical directions. Our findings show a high prevalence of occlusal anomalies in Turner syndrome as a result of imbalanced growth of the craniofacial skeleton in three dimensions. Early diagnosis of occlusal anomalies is of utmost importance in Turner syndrome patients and should be accompanied by an early orthodontic treatment of malocclusion in these individuals.

Acknowledgements

The authors would like to thank the staff of the Department of Endocrinology and Genetics at the University Clinic for Children's Diseases –Skopje and the staff of the Orthodontic Dental Laboratory at the University Dental Clinical Centre - Skopje, for the excellent cooperation and technical assistance in achieving the targets. We also thank all the girls with Turner's syndrome and their families, for their cooperation and understanding.

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