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ORIGINAL ARTICLE

Caries intensity and *Streptococcus mutans* in the saliva of patients with Turner syndrome

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Abstract *Aim:* The aim of this study was to evaluate the caries intensity and *Streptococcus mutans* (*SM*) counts in patients with Turner syndrome.

Materials and methods: Nineteen patients aged 20–40 years were clinically and cytogenetically diagnosed with Turner syndrome (45, X). The karyotype was determined by chromosome analysis of peripheral blood lymphocytes. The control group comprised 47 healthy women aged 21–40 years. Both groups included non-smokers with no specific diet, such as a vegetarian or vegan diet, who were generally healthy with good oral hygiene and periodontal condition. Patients treated with antibiotics or steroid preparations in the past 6 months or with diseases or conditions that might affect the oral mucosal environment, such as disorders of salivary secretion and diabetes, were excluded from the study. Decayed, missing, and filled teeth (DMFT) scores and *SM* counts in saliva were determined.

Results: No colony growth of *SM* was noticed in 53% of patients with Turner syndrome and 4.2% of controls ($p < 0.001$). Colony counts of $SM \geq 10^5$ in saliva were observed in none of the patients with Turner syndrome but in 66% of controls ($p < 0.001$). The mean DMFT score was 1.63 ± 2.52 in patients with Turner syndrome and 14.49 ± 6.88 in controls. Statistically significant differences between the two groups were observed ($p < 0.05$).

Conclusion: Patients with genetic disorders may have different severities of caries and *SM* counts in saliva compared to those without genetic disorders. Further studies on saliva properties and

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genes located on the X chromosome could contribute to determining the effect of the X chromosome on the pathological processes in the oral cavity.

1. Introduction

Turner syndrome (TS) is a complicated genetic disorder characterised by X-chromosome aberrations. Its clinical features include somatic abnormalities, such as infantile external genitalia, short stature, cubitus valgus webbed neck (Temtamy et al., 1992; Robinson and de la Chapelle, 1996; Lopez et al., 2002), and oral abnormalities, such as microdontia (Kusiak et al., 2000; Szilagyi et al., 2000a, 2000b; Zillberman et al., 2000; Maier et al., 2019), malocclusion (Szilagyi et al., 2000a, 2000b; Cazzolla et al., 2018), enamel hypoplasia (Lopez et al., 2002; Kusiak et al., 2008) and irregularities in the root morphology of the mandibular teeth (Varrela et al., 1990; Lopez et al., 2002; Kusiak et al., 2005). However, the caries prevalence has been reported to be reduced in patients with TS (Takala et al., 1985; Szilagyi et al., 2000a, 2000b). Alteration in results of saliva tests reported for patients with genetic disorders such as Down's syndrome (Stabholz et al., 1991; Yarat et al., 1999; Chaushu et al., 2002; Siqueira et al., 2005), Papilon–Lefevre syndrome (Lundgren et al., 1996), and epidermolysis bullosa (Harris et al., 2001), warrants research on saliva properties of patients with TS.

The aim of the present study was to evaluate the caries intensity and *Streptococcus mutans* (SM) counts of patients with TS.

2. Materials and methods

2.1. Patients' population

Nineteen patients aged 20–40 (26.4 ± 4.4) years with TS were studied. Both the diagnosis of TS (45,X) and karyotype evaluation were performed at the Department of Biology and Genetics of the Medical University of Gdańsk. The karyotype was determined by chromosome analysis of peripheral blood lymphocytes. The control group comprised 47 healthy women aged 21–40 (29 ± 3.2) years, who first reported to the Department of Conservative Dentistry, Medical University of Gdańsk for follow-up examinations and agreed to the examination and met the inclusion criteria for the control group. Both groups included non-smokers with nonspecific diet, such as a vegetarian or vegan diet, who were in generally healthy with good oral hygiene and healthy periodontal tissues. Patients treated with antibiotics or steroid preparations in the past 6 months or with diseases that might affect the oral mucosal environment, such as disorders of salivary secretion and diabetes, were excluded from the study.

2.2. Saliva collection

Mixed stimulated saliva was collected from each study participant. The caries intensity was studied for its prevalence evaluation. Decayed, missing, filled, teeth (DMFT) scores were calculated for all patients.

Saliva was collected into sterile silicone (Corning test tubes) in the morning 2 h after the last meal. To mechanically stimulate saliva secretion, subjects were directed to chew a paraffin cube for 1 min. Saliva secreted within 1 min of paraffin chewing was expectorated and that secreted during the next 5 min was collected into calibrated Corning test tubes, as previously described (Kusiak et al., 2011). Collected saliva was used to calculate *SM* counts in 1 mL of stimulated saliva.

Due to the closest reflection of teeth surfaces and tongue's microbiological status, stimulated saliva was used for microbiological evaluation. The CRT® bacterial test was used

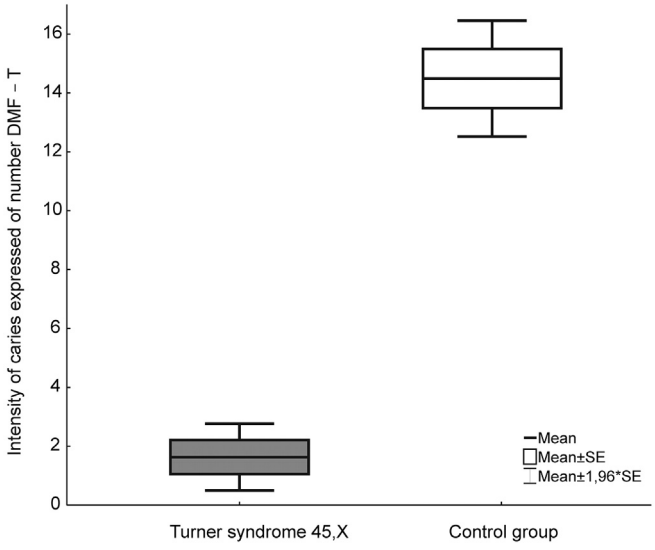

Fig. 1 Intensity of caries expressed of number DMF – T in Turner syndrome and the control group.

Table 1 Quantity of *Streptococcus mutans* colonies in stimulated saliva of patients in the Turner syndrome and control groups.

Karyotype	Total number of patients	Number of patients with a particular quantity of *SM* colonies in 1 mL of stimulated saliva					
	N	No growth		$SM < 10^5$ CFU/mL		$SM \geq 10^5$ CFU/mL	
		N	%	n	%	N	%
Turner syndrome 45, X	19	10	53[a]	9	47	0	0[c]
Control group	47	2	4,2[b]	14	29,8	31	66[d]

$p < 0.001$ for a vs. b and c vs. d; SM: *Streptococcus mutans*; CFU: Colony Forming Units.

Table 2 Caries intensity expressed as DMFT value in Turner syndrome and the control group.

Karyotype	DMFT - value		
	$\bar{x} \pm$ SD	Range	Median
Turner syndrome 45,X (N = 19)	1.63 ± 2.52[a]	0–8	1
Control group (N = 47)	14.49 ± 6.88[b]	5–41	13

$p < 0.05$ for a vs. b;$\bar{x}$: mean; SD: standard deviation; N:total number of patients; DMFT: decayed, missing, filled teeth.

(Ivoclar–Vicadent, Liechtenstein). *SM* counts were read in marked surface Colony Forming Units/mL of saliva and compared to the model pattern according to the manufacturer's guidelines. *SM* counts $< 10^5$ and $\geq 10^5$ in 1 mL of stimulated saliva were classified as low and high, respectively.

2.3. Statistical analysis

Statistical analyses were performed using the statistical suite STATISTICA (data analysis software system), version 12.0 (StatSoft. Inc., Tulsa, OK, USA). The Yates-corrected chi-square nonparametric test for unrelated values was used in the analyses of microbiological results in the TS and control groups. The caries intensity was analysed using the Kruskal–Wallis test. In all calculations, the statistical significance level was set at $p < 0.05$.

3. Results

Table 1 illustrates the *SM* counts in the stimulated saliva of patients in the TS and control groups.

No colony growth was noticed in ten women with TS(53%) and two controls(4.2%). A colony count of SM $< 10^5$in 1 mL of stimulated saliva was observed in nine women with TS (47%) and in 14 women in the control group (29.8%). Colony counts of SM $\geq 10^5$ in saliva were not observed in any patient with TS. However, they were present in 31 women in the control group (66%). Statistically significant differences could be identified between the TS and control groups ($p < 0.05$).

Table 2 and Fig. 1 illustrates the caries intensity expressed by the DMFT score. The mean DMFT score was 1.63 ± 2.52 in the TS group and 14.49 ± 6.88 in the control group. Statistically significant differences between the TS and control groups were observed ($p < 0.05$).

4. Discussion

Dental caries has a complex aetiology that has not been fully elucidated to date. According to the current state of knowledge, caries is conditioned by the presence of dental plaque and cariogenic bacteria occurring in it, mainly *SM,* which breaks down sugars into acids and creates large amounts of extracellular and insoluble glucans, components of the plaque matrix. Other elements are the supply of carbohydrates, which are associated with oral hygiene and regular dental plaque removal, and the host's resistance (susceptibility of the enamel surface and properties of saliva). Other researchers have indicated a lower incidence of caries, which may be related to the dammonths or with diseases or conditions that mightage or absence of the X chromosome in TS (Takala et al., 1985; Szilagyi et al., 2000a, 2000b). Takala et al. examined caries in 50 women with TS and found its low prevalence compared to the control group, particularly in relation to the premolars and molars.

There are no studies in the existing literature on the influence of saliva on oral cavity pathologies in patients with TS. Nevertheless, some authors have indicated a genetic aspect to some saliva irregularities noticed in other syndromes, such as Down syndrome (Barr-Agholme et al., 1998; Chaushu et al., 2002; Cogulu et al., 2006), Papillon-Lefévre syndrome (Lundgren et al., 1996) and epidermolysis bullosa (Harris et al., 2001).

The importance of bacterial factors in the course of caries has been pointed out by many authors (Grähn et al., 1988; Klock et al., 1990; Russell et al., 1990; Stabholz et al., 1991; Kirstilä et al., 1998; Gãbris et al., 1999; Nishikawara et al., 2006). In our study, a significant decrease in *SM* counts was observed in 1 mL of saliva of patients with TS compared to controls. The results obtained in the study indicated that patients with TS, whose saliva did not show increased *SM* counts, had a significantly lower caries intensity compared to the controls. When high colony counts of $SM \geq 10^5$ was present, caries intensification was also significantly lower in patients with TS than in controls.

As for previous studies, Stabholz et al. (1991) reported a similar correlation in the studied population of children with Down syndrome. Decreased counts of *SM* in the saliva and lower caries intensity were observed in children with Down syndrome. The study group was compared with two age-matched control groups living in the same institution: a group of healthy children and another mentally retarded children without Down syndrome. The caries experience showed significantly lower mean scores for the Down syndrome group in

comparison with both control groups, for whom maintaining good oral hygiene manually was similarly difficult. Among children with Down syndrome, 84% were free of caries.

Szilagyi et al. (2000a) and Szilagyi et al. (2000b) also observed reduced salivary *SM* counts and lower caries intensification in 29 patients with TS. A statistically significant correlation was found between mean DMFT scores and salivary microbiological counts. In our studies, similar results were obtained. Other properties of saliva, such as the buffering capacity, which may have an impact on caries intensity, were also altered in a study by Kusiak et al. (Kusiak et al., 2010), which revealed a significantly higher salivary buffering capacity in patients with TS compared to the control group. In the present study, the severity of caries was compared in the group of women with TS and in the control group in such a way that there were no additional disturbing factors, using strict exclusion criteria. Both groups of women were characterised by good oral hygiene and had no periodontitis, saliva secretion disorders, smoking habits or current antibiotic medication. The saliva of patients with TS had significantly lower *SM* counts than that of control subjects. In addition, DMFT scores were lower in women with TS than in control subjects.

The limitation of our study was the small sample size, which should be increased in the future to better assess this phenomenon.

5. Conclusion

Patients with genetic disorders may present with a different severity of caries and concentration of *SM* in the saliva compared to those without genetic disorders. Further studies on saliva properties and genes located on the X chromosome could contribute to determining the effect of the X chromosome on the pathological processes occurring in the oral cavity.

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Statement of Ethics

The study protocol was approved by the Independent Bioethics Committee for Scientific Research at the Medical University of Gdańsk. Ethical aspects of the study followed the World Medical Association Declaration of Helsinki.

Disclosure Statement

The authors declare no conflict of interest.

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