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ORIGINAL ARTICLE

Oral health in adult patients with congenital coagulation disorders – a case control study

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Summary. Inflammatory disorders of the periodontium, gingivitis and periodontitis are among the most prevalent diseases worldwide. A few studies have found poorer oral health in patients with congenital coagulation disorders (CCD) like haemophilia and von Willebrand's disease compared with non-affected controls. The aim of this study was to investigate the effect of congenital coagulation disorders on oral health and periodontal (alveolar) bone loss. This is a case control study comparing oral health and periodontal bone loss of patient with congenital coagulation disorders with matched healthy subjects. The examination included dental status (DMF-T), assessment of oral hygiene (modified Quigley-Hein-Index: QHI) and a dental panoramic X-ray for assessment of alveolar bone loss caused by periodontal disease. A total of 15 patients with CCD (Haemophilia A: n = 8, von Willebrand's disease: n = 7) were matched with 31 non-affected controls. We observed no clinical relevant difference of oral health (DMF-T, QHI) between patients with CCD and controls despite better oral hygiene (QHI) of patients with CCD. Moreover, there was a statistically significant difference in periodontal bone loss, but the observed difference is not clinically meaningful. Unlike previous studies carried out mainly in children we found no evidence that oral health or periodontal status in adult patients with CCD is worse than that in healthy subjects. However, larger studies and longitudinal studies in adults are needed to confirm our results.

Keywords: congenital coagulation disorders, haemophilia, oral health, periodontal (alveolar) bone loss, von Willebrand's disease

Introduction

The management of patients with congenital coagulation disorders (CCD) causes a considerable number of problems in dental medicine [1,2]. Affected patients require special attention; however, given the relative rarity of such disorders, most dentists have no experience in dealing with dental problems in patients with CCD. 'von Willebrand's disease affects about 1% of the population and is the most common CCD. Most patients have a mild to moderate form of the condition and are only diagnosed after a significant bleeding episode. [3,4]. Haemophilia affecting only about 1 in 5000 to 10 000 men is rarely missed [5]. Current recommendations for dental management of CCD like, e.g. haemophilia and von Willebrand's disease focus mainly on associated coagulation problems [2,6,7]. Only few small studies have examined dental and periodontal status mostly of children or young adults with haemophilia [8–10]. The studies found poorer oral health compared with normal controls. One study including three subjects with von Willebrand's disease was inconclusive [9]. Only one case report about aggressive periodontal disease described a family affected with von Willebrand's disease [11].

Inflammatory disorders of the periodontium, gingivitis and periodontitis are among the most prevalent diseases worldwide. About 80–90% of the population in the western world above age of 35 are affected [12]. Periodontitis leads to irreversible loss of connective tissue and bone around the teeth and is therefore the most frequent cause for tooth loss in adults [13]. The main cause is bacterial colonization of the oral cavity building up a biofilm (dental plaque) on the teeth [14]. However, the cause of disease is influenced by many other factors like smoking, alcohol and genetic factors

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[13]. Diabetes, HIV and other systemic disease are the associated risk factors [15–17].

It is conceivable that congenital coagulation disorders are risk factors for periodontitis and subsequent alveolar bone loss. The aim of this case control study was to investigate the effect of congenital coagulation disorders on oral health and periodontal (alveolar) bone loss.

Materials and methods

This is a case control study comparing oral health and periodontal bone loss of patient with congenital coagulation disorders with matched healthy subjects.

The study was reviewed and approved by the ethics committee of the Hannover Medical School, Germany.

Subjects

Congenital coagulation disorder group. Patients between 18 and 60 years suffering from congenital coagulation disease (haemophilia A/B or von Willebrand's disease) who attended the dental clinic of Hannover Medical School in 2007 were asked to give informed consent to participate in the study. To avoid confounding with other conditions affecting oral health, the following medical conditions were considered exclusion criteria: smoking in the last 5 years, heart disease (coronary artery disease, hypertension), diabetes mellitus, infectious diseases (hepatitis, HIV), seizures, drug addiction, chronic renal failure, organ transplantation, immunosuppressive medication, continuous medication and pregnancy. A dental panoramic X-ray (less than 1 year old) had to be available.

Control group. Healthy patients receiving routine dental check-up were selected randomly as controls. Patients were matched by age and gender: each CCD patient was matched with two healthy controls. Inclusion and exclusion criteria were identical.

Clinical examination

All subjects were examined once under standardized conditions by an experienced dentist (dentist 1 DZ). The dental examination was performed during a routine dental check-up in the dental office of the Hannover Medical School (CCD patients) or in the dental practice (control patients).

Oral health. The examination included dental status (DMF-T) and assessment of oral hygiene [modified Quigley-Hein-Index (QHI)] [18–20]. The DMF-T is a caries index used internationally to quantify the number of decayed teeth, missing teeth and filled teeth with values ranging from 0 to 28. The modified QHI evaluates oral hygiene by colouring dental plaque with a plaque detector (erythrosine solution). The plaque extension is graded on a scale from 0 to 5, values below 1 are considered as good oral hygiene.

Radiographic examination

Periodontal bone loss. Periodontal (alveolar) bone loss and the periodontal structures were assessed radiographically with a dental panoramic X-rays/tomography (DPT). All DPTs were performed under the same conditions (70 kV, 0.9 mA) with conventional films. All radiographs were taken with the same X-ray machine using the same program.

The evaluation of bone loss was carried out under standardized conditions. Each X-ray was examined by one dentist (dentist 2 CS) using identical assessment criteria. The examiner was blinded to which group patients belonged to. All X-rays were examined with 14 days between the examinations. The level of alveolar bone loss was evaluated for each tooth: the distance between the limbus alveolaris and the cemento-enamel junction was examined mesially as well as distally in millimetre (mm) with an mm-scaled periodontal probe (CP-15UNC, Hu-Friedy, Chicago, IL, USA) [21].

The level of periodontal status was graded according to recommendations from Page and Eke for definition of periodontitis [22]:

- severe periodontitis: alveolar bone loss >5mm (>30% of the sites)
- moderate periodontitis: alveolar bone loss 3–5mm (>30% of the sites)
- no or mild periodontitis: alveolar bone loss 1–2mm (>70% of the sites)

Statistical evaluation

Data were assessed for Normal distribution. Accordingly we used Student's t-test if Normal distribution could be assumed and Mann–Whitney *U*-test otherwise. Chi-square test was used for dichotomous data.

For description of alveolar bone loss in the two groups in a first step the figures obtained of mesial and distal alveolar bone were used to calculate the mean loss for each tooth. Afterwards the mean (mesial and distal summarized) of all teeth per patients was used for the Student's *t*-test. A *P*-value less than 0.05 was considered as significant. Statistical analysis was performed using SAS 9.2 (SAS Institute Inc., Cary, NC, USA).

Results

Description of the population

Congenital coagulation disorders group. A total of 37 patients (female = 21, male = 16) with CCD were asked to give consent. Five patients declined and another 17

were excluded because they did not meet the inclusion criteria (mostly smokers). Finally 15 patients (female = 5; male = 10) could be included (40%); 8 patients with haemophilia A (severe: $n = 6$, moderate: $n = 2$) and 7 patients with von Willebrand's disease Typ IIA. The average age was 39.2 (SD ± 8.3) years.

Control group. A total of 81 patients were approached to act as controls, of which 7 declined and 43 were ineligible mostly because of smoking. Finally 31 subjects (male = 21, female: $n = 10$) with an average age of 36.4 (SD ± 9.6) years were included in the study. From one male patient all data were not collected and therefore one more patient was recruited for a full data set.

The distribution of age and gender between the two groups was not significantly different (Table 1.).

Oral health

The comparison of patients and healthy controls is shown in Table 1. The median DMF-T of patients with CCD and healthy controls was not significantly different ($P = 0.41$). Moreover, no difference in sub-indices number of missing teeth (MT) ($P = 0.12$), number of decayed teeth (DT) ($P = 0.5$) as well as number of filled teeth (FT) ($P = 0.28$) was observed. Patients with CCD had a significantly better oral hygiene (modified QHI) ($P = 0.01$) (Fig. 1).

Periodontal bone loss

Patients with CCD had significantly more alveolar bone loss than controls (DPT) ($P = 0.003$) (Fig. 1). We also observed that patients with CCD had significantly more frequently moderate to severe periodontitis (80%) compared with the controls (48%) based on radiological assessment of alveolar bone loss ($P = 0.04$, Table 1).

A subgroup analysis comparing alveolar bone loss in patients with haemophilia to von Willebrand's disease did not show a significant difference ($P = 0.92$).

Discussion

Summary of the main results

We observed no clinical relevant difference of oral health between patients with CCD and healthy controls despite better oral hygiene of patients with CCD. Moreover, there was a statistically significant difference in periodontal (alveolar) bone loss, but the observed difference (less than 1 mm) is clinically not meaningful according to our clinical experience.

Comparison with existing literature

An Egyptian study found significantly higher DMF-T in haemophilic children (average age 7–8 years, $n = 30/30$) compared with non-haemophilic controls [23]. Another study in adolescents (average age 16 years) in Pakistan also observed significantly higher DMF-T in haemophiliacs ($n = 52/192$) [8]. Our study which was performed in adults cannot be directly compared with other studies on oral health of patients with CCD [8–10,23]. However, the results of the DMF-T in this study (CCD: median = 18; controls: median = 15) are similar to the findings of the age group 35–44 years (mean DMF-T = 14.5) in German Study on Oral Health (DMS-IV) [24]. A large Polish study ($n = 180$) in children with haemophilia and von Willebrand's disease from age 4–18 also found no significant difference in DMF-T [9]. Despite these non-significant findings, the author concluded that oral health in severe haemophiliacs is worse. A study in haemophilic children in Northern Ireland also found no difference [25]. The observed differences might be related to economic wealth and differences in provision of dental care.

Table 1. Comparison of patients with congenital coagulation disorders and healthy controls.

	CCD group ($n = 15$)	Control group ($n = 31$)	Statistical analysis (test)	Level of significance (P-value)
Gender (female)	5 (33%)	10 (32%)	Chi-squared test	0.94
Age (years ± SD)	39.2 ± 11	36.4 ± 11	t-test	0.41
DMF-T (median, range)	18 (9–28)	15 (4–24)	Mann–Whitney U-test	0.41
DT (median, range)	1 (0–8)	0 (0–7)	Mann–Whitney U-test	0.50
MT (median, range)	1 (0–11)	1 (0–5)	Mann–Whitney U-test	0.12
FT (median, range)	10 (3–17)	13 (2–21)	Mann–Whitney U-test	0.28
QHI (mean ± SD, range)	1.97 ± 0.36 (1.25–2.25)	2.00 ± 0.77 (1.25–2.95)	t-test	0.01*
Alveolar bone loss in mm (mean ± SD)	2.73 ± 0.73	2.00 ± 0.77	t-test	0.003*
Periodontal status				
No/mild	$n = 3$ (20%)	$n = 16$ (52%)	Chi-squared test	0.04*
Moderate/severe	$n = 12$ (80%)	$n = 15$ (48%)		

*Statistical significant.
DMF-T, number of decayed teeth, missing teeth and filled teeth (caries index); DT, decayed teeth; MT, missing teeth; FT, filled teeth; QHI, Quigley-Hein-Index (oral hygiene index); SD, standard deviation.

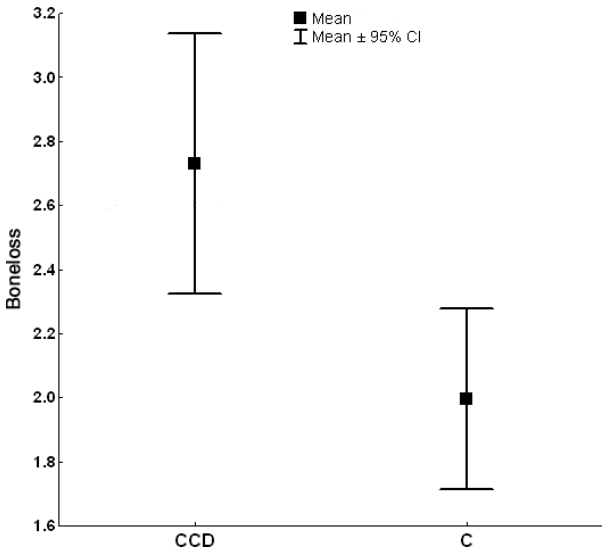

Fig. 1. Comparison of alveolar bone loss in mm for the CCD-group and control (C).

This is to our knowledge the first study examining periodontal (alveolar) bone loss in CCD patients. Only one case report, based on a family with four members, suggested a possible relationship between von Willebrand's disease and periodontitis [11]. The affected mother (45 years) as well as two affected sons (21 and 19 years) had all a severe periodontitis. Only one affected daughter (15 years) was periodontically healthy. Our results do not support this hypothesis, although we have observed a moderate statistically significant but clinically not relevant increased bone loss in the CCD group.

Strengths and limitations

This is to our knowledge the first study comparing oral health and periodontal status in adult CCD patients with healthy controls. Our study has several limitations. As a result of strict exclusion criteria to avoid confounding with other conditions affecting oral health, we could only include a small number of patients. Therefore, we aggregated patients with two different coagulation disorders in one group for comparison. We carried out a subgroup analysis which did not show differences. The examination of the periodontal (alveolar) bone loss was solely based on panoramic X-rays; however, for a better evaluation of the clinical situation, additional parameters (pocket depths, clinical attachment level) would have been helpful. As these invasive procedures might have caused prolonged bleeding and/or prolonged observation in CCD patients, we did not examine these parameters.

Conclusion

Periodontal disease is an underestimated public health problem. Although patients with CCD require special attention for invasive dental treatment, we found no evidence that oral health or periodontal status is worse than that in healthy subjects. However, larger studies and longitudinal studies in adults are needed to confirm our results.

Disclosures

The authors stated that they had no interests which might be perceived as posing a conflict or bias.

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