Radiographic assessment of the alveolar bone height in children and adolescents with familial dysautonomia

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Abstract

Purpose: Familial dysautonomia (FD) is a progressive neuropathy, characterized by somatic and skeletal abnormalities, and by a variety of oral and diet disturbances. The purpose of the study was to assess the alveolar bone height at the molar areas of children and adolescents with FD.

Methods: The distance from the cemento-enamel junction (CEJ) to the alveolar bone crest (ABC) was measured on routine diagnostic bitewing radiographs of nine males and seven females with FD (mean age=122 months) and in those of two matching groups (C1=119 months; C2=122 months).

Results: The mean values for the maxilla were significantly larger than those in the mandible. A positive significant correlation was found between the CEJ-ABC measurements of the primary and the permanent teeth, and between the CEJ-ABC measurements and age. The mean values per patient for the CEJ-ABC distances of the FD group were smaller than the control groups, but the difference was not statistically significant. The CEJ-ABC measurements in the primary and the permanent molars were smaller in the FD group, and in the premolars and permanent cuspids they were larger than those in the two control groups. These differences were not statistically significant. No differences were found between the FD and the control groups in the primary cuspids.

Conclusions: The alveolar bone height of children and adolescent with familial dysautonomia does not differ from that of healthy controls. (Pediatr Dent 23:61-65, 2001)

Familial dysautonomia (FD), also known as Hereditary Sensory and Autonomic Neuropathy type III, is an autosomal recessive disease, affecting exclusively Ashkenazi Jews (stemming from Eastern Europe), its prevalence being 1 per 3700 birth.[1] The disease is characterized by being a progressive degenerative disorder of the sensory and autonomic peripheral nervous system, and is associated with unusual somatic and skeletal abnormalities that include:[2-6]

1. Failure of somatic growth, short stature and kyphoscoliosis called for the assumption that nerve growth factor (NGF) may be missing or defective. However, NGF was found to be normal and the possibility that other "growth factors" may be defective has not yet been clarified.
2. Striking insensitivity to pain and dysesthesia that may result in unnoticed fractures, burns, dental trauma, and orodental self-mutilation.

Distinctive orofacial characteristics include:[6-12]

1. The craniofacial morphology of FD patients differs significantly from the classical norms. They exhibit an increased mandibular retrognathism and an enhanced horizontal mandibular growth. The occlusion is characterized by small dental arches, crowding, and a tendency toward class II malocclusion.
2. Measurements of tooth components show thick enamel on the occlusal surface and a small pulp size in the primary and permanent molars.
3. Caries prevalence is low despite high-risk etiologic factors, such as high sugar consumption and inadequate oral hygiene. The reasons for this are as of yet unknown.
4. Drooling has been attributed to hyper salivation and swallowing difficulties.
5. Frequent vomiting associated with gastrointestinal reflux is a serious source of acidity in the oral cavity.
6. Feeding habits in FD individuals are characterized by poor appetite since infancy, leading to a reduced consumption of food. Actually, a high percentage of FD patients have gastroesophageal fundoplication or gastrostomy from early childhood and therefore, most of their nutrition does not pass through their stomathognathic system.
7. Impaired taste and smell often modify their food choices and dietary habits. Based on the unique progressive degeneration of tongue fungiform papillae and their taste buds, former acceptance of a global loss or absence of taste in FD individuals was questioned by Gadoth et al., who found a selective hypoageusia and normal smelling in FD patients. They are similar in their ability to taste sour stimuli and to estimate intensity and appeal as normal healthy subjects.
8. Chronic gingivitis is common in FD patients in the permanent dentition, and is partly related to disuse atrophy, plaque, and calculus accumulation from early age, and disregard for oral hygiene. It should be noted that, as in children with no systemic disease,[13-16] FD children demonstrate a low gingival inflammatory response to relatively large amounts of dental plaque.

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Table 1. Number of Sites Examined by Type and by Group*

Tooth	Site	C1	C2	FD	Total
Maxillary permanent teeth					
First molar	M	26	22	19	67
Second premolar	D	10	7	7	24
	M	10	7	8	25
First premolar	D	10	7	7	24
	M	10	5	4	19
Canine	D	8	4	4	16
Maxillary primary teeth					
Second molar	D	18	18	13	49
	M	22	21	16	59
First molar	D	22	20	14	56
	M	19	18	16	53
Canine	D	19	15	16	50
Subtotal Maxilla		174	144	124	442
Mandibular permanent teeth					
First molar	M	29	26	24	79
Second premolar	D	10	8	8	26
	M	10	8	7	25
First premolar	D	10	8	7	25
	M	6	3	4	13
Canine	D	5	2	4	11
Mandibular primary teeth					
Second molar	D	22	21	18	61
	M	22	22	18	62
First molar	D	22	22	18	62
	M	18	11	14	43
Canine	D	16	11	12	39
Subtotal Mandible		170	142	134	446
Total		344	286	258	888

*C1= control 1, C2=control 2 and FD= familial dysautonomia.
M=Mesial; D=Distal

Occlusion, caries, feeding habits, and diet have a direct influence on the periodontal tissues.[17-19] It also has been demonstrated that facial growth and attrition have a direct influence on the distance from the CEJ to the alveolar bone crest.[20] Since FD children have characteristic skeletal occlusal and facial features, and their feeding habits are different from those of the general population,[7-12] it may also be possible that the periodontium of FD individuals differs from that of the general population. Therefore, the purpose of the present study was to assess, by radiographic means, the alveolar bone height at the cuspid to molar areas of children and adolescents with FD and to compare it to that of controls with no systemic disease.

Methods

Routine diagnostic bitewing radiographs of 25 children and adolescents with FD were examined by one of the authors (EM). Those with minimal or no distortion, minimal or no overlapping between the proximal surfaces and sites, and with a clear image of the interdental alveolar bone area, were selected for the study. The radiographs from 9 males and 7 females met the requested criteria (mean age = 122 months, range: 67 to 211 months). The same author examined bitewing radiographs of children with no systemic diseases, and selected those of 32 children of whom each two matched in age and gender each individual of the FD group and fit the criteria for the study. The radiographs were divided into 2 control groups (C1, mean age=119 months, range = 63 to 204 months; C2, mean age=122 months, range=61 to 212 months). All radiographs were coded, and the second author (EB), who was unaware as to which group the radiographs belonged, assessed the situation of the alveolar bone by examining the presence of the lamina dura over the alveolar bone crest and by measuring the distance from the cemento-enamel junction (CEJ) to the alveolar bone crest (ABC).[20] The measurements were taken from the mesial surface of the first permanent molar to the distal surface of the permanent or primary cuspid (12 sites in each radiograph). When the first permanent molar was still absent or partially erupted, the measurements were taken from the distal surface of the second primary molar to the distal surface of the primary or permanent cuspid (10 sites in each radiograph). The measurements were done on a light box using a magnifying glass with an attached micro-ruler (Viewcraft X10) in 0.1 mm increments.[20] Previous studies indicated a >0.80 level of reliability by the same author in these measurements.[21-24] The measurements for both sides of the mouth were pooled together, since previous studies have indicated no difference between left and right homologous sites.[20, 25, 26]

Statistical analysis

Mean CEJ-ABC distances were obtained by site (mesial of the maxillary first molar, distal of the second primary molar etc.), tooth type (primary/permanent, molar/premolar/cuspid), arch (mandible/maxilla), individual, and gender. ANOVA was utilized to examine the significance of the differences between age, by group and gender, and CEJ-ABC distances by gender, arch (mandible and maxilla), or group. Pearson correlation analysis was utilized to examine the significance of the correlation of the CEJ-ABC distances between arches, primary and permanent teeth, age, and CEJ-ABC distances. Linear regression analysis was utilized to examine the significance of the differences in CEJ-ABC measurements by age and group. A standard computer program for statistical analysis (JMP Version 3.2.6, SAS Institute Inc, Cary NC, 1889-1999) was utilized for the statistical examination. Differences at the 5% level of probability were considered statistically significant.

Results

No statistically significant differences in age were found between the FD, C1, and C2 groups (mean=122 months, SE=12; mean=119 months, SE =12; mean=122 months, SE=12 respectively). From a possible total of 972 sites, 888 were available for examination (Table 1). Missing sites were related to

Table 2. Mean Distances from the Cementoenamel Junction to the Alveolar Bone Crest by Tooth Class (primary/permanent), Child and Group.

	Primary teeth		Permanent teeth		By child	
Group*	Mean	SE	Mean	SE	Mean	SE
C1	0.8	0.8	0.3	0.6	0.6	0.1
C2	0.8	0.8	0.2	0.7	0.6	0.1
FD	0.8	0.9	0.2	0.7	0.6	0.1

*C1 and C2 = control groups; FD = familial dysautonomia
No statistically significant differences between the groups, $P>0.05$.

unerupted permanent teeth, exfoliated primary teeth, crown restorations, or an unclear radiographic image. The mean CEJ–ABC measurements per tooth type were found to be ≤2 mm in all but 3 children from control groups, who had measurements between 2 to 3 mm at the distal surfaces of maxillary first primary molars (one site in a 7-year old child and another site in a 10-year old) and distal surfaces of mandibular primary cuspids (two sites in a 10-year old child). The lamina dura over the alveolar bone crest appeared normal in all sites, in every child. No statistically significant differences were found between males and females in age (mean=112 months, SE=9 and mean=132 months, SE=10, respectively), in CEJ-ABC measurements of the primary teeth (mean=0.8 mm, SE=0.1 and mean=0.8 mm, SE=0.1, respectively) and in CEJ-ABC measurements of the permanent teeth (mean=0.2, SE=0.1 and mean=0.03, SE=0.1 respectively). The mean measurements for the maxilla (0.7 mm, SE=0.04) were found to be statistically significant larger ($P=0.04$) than those in the mandible (0.57 mm, SE=0.04), and a statistically significant correlation was found between the measurements of the mandible and the maxilla ($r=0.66$, $P<0.001$).

A positive significant correlation was found between the CEJ-ABC measurements of the primary and permanent teeth ($r=0.52$, $P<0.001$), and age and CEJ-ABC measurements of the primary teeth ($r=0.48$, $P<0.001$), age and CEJ-ABC measurements of the permanent teeth ($r=0.40$, $P<0.01$). The mean values for the CEJ-ABC distances of the FD group for the primary and the permanent teeth and per child were smaller, but not any more statistically significant than the control groups (Table 2). Comparison of the measurements by group and type of tooth (Table 3) indicated that in the FD group the CEJ-ABC measurements of the primary and permanent molars were smaller, but not statistically significant than in the 2 control groups. In the premolar and permanent cuspid areas, the FD group did not have statistically significant larger CEJ-ABC distances than the 2 control groups, and in the primary cuspid area, no definitive difference was found between the FD and the control groups. Analysis of the CEJ-ABC distances by age and group did not indicate statistically significant differences.

Discussion

One parameter that has been widely utilized for the diagnosis of periodontitis in children and adolescents is the distance between the CEJ and the ABC.[16, 18, 20-24] The normal CEJ-ABC distances in primary human teeth are into most sites ≤2 mm. Larger measurements have been considered normal when related to eruption (due to attrition or facial growth), a most anterior position of the tooth in the arch, or when the adjacent tooth is an exfoliating primary tooth or a newly erupted permanent tooth.[26-30] Therefore, the condition of the alveolar bone crest should also be taken in consideration when diagnosing alveolar bone loss in the primary dentition.[20] In the present study, three children of the control groups had >2 mm CEJ-ABC distances in primary teeth; two of them at ages and sites in which it is normal to have >2 mm measurements. The lamina dura above the alveolar bone crest was normal at all sites in all children. It may be therefore concluded that, in the present study, no definitive radiographic evidence of alveolar bone loss was found in any individual.

The present findings, in the two control groups, confirm previous ones that the CEJ-ABC distances in the primary teeth are larger in the maxilla than in the mandible.[25, 26, 30] Males have larger, but not statistically significant CEJ-ABC distances than females,[26, 30] and there is a positive significant correlation between the age and the CEJ-ABC measurements.[25, 26] Other studies that failed to demonstrate a correlation between age and the CEJ-ABC measurements[28, 30] do not necessarily contradict the present and previous findings, since they include a shorter age range (7-9-year olds), during which a pause in the increase of the CEJ-ABC distances may take place.[25-27, 29]

The purpose of the present study was to assess, by radiographic means, the alveolar bone height of children and adolescents with FD and to compare it to two matching control groups. We decided to utilize two control groups, to assure that any possible difference between FD and control patients would not be accidental. Indeed, the present study indicated no significant differences between either the FD and each of the control groups or between the two control groups.

Many FD patients get most of their nutrition through fundoplication and the others are known to eat mostly soft foods. In addition, FD individuals have impaired neural and muscular coordination with consequences reflected in their stomathognathic system and in their masticatory performances.[2-12] Since occlusion, caries, feeding habits and diet have a significant influence on the periodontal tissues,[17-19] we assumed that the particular characteristics of FD children and adolescents would have a distinctive influence on their periodontium. Differences could be related to: 1) the progressive neuropathy that definitely affects the skeleton; 2) the persistent known disesthesia in FD individuals; 3) the feeble utilization of their oral muscles in eating soft foods, or not using them for mastication on a regular basis; and 4) increased plaque and calculus accumulation.

One possibility that our findings did not demonstrate differences in their alveolar bone height is that the limited mastication of FD individuals results in minimal attrition and consequential reduced eruption, leading to shorter CEJ-ABC distances. This assumption was enhanced by a previous finding that FD individuals have a thicker than normal enamel in the occlusal table,[10] that could also be related to reduced attrition. Indeed, in the present study the mean per individual CEJ-ABC distances in the FD patients were smaller than in the control groups. However, these differences were not statistically significant (Table 2). It may be possible that a larger sample could have resulted in statistically significant differences.

Interesting is the finding that when the CEJ-ABC measurements were sorted by type of tooth (Table 3), the measurements were shorter (but not statistically significant) in the FD individuals only at the primary and the permanent molars. In the primary and the permanent cuspids and in the premolars, the measurements were larger in the FD individuals (but not statistically significantly larger). It should be noted that previous studies have indicated a pattern of larger CEJ-ABC measurements with a more anterior position in the arch of human primary teeth, apparently related to facial growth.[25, 26, 30] The reason for the changes in pattern between the present and previous studies still remains unclear.

A previous study indicated that compared to children with no systemic disease, FD individuals have a larger degree of plaque accumulation but without more severe gingival inflammation.[7] Despite large amounts of dental plaque, the limited gingival inflammation in children with no systemic disease is the result of an age-dependent inflammatory reaction of the gingiva to plaque accumulation.[13-16] Most likely, the same process takes place in the FD individuals. In the present study, no radiographic evidence of childhood periodontitis was found in any of the children. These findings do not necessarily eliminate the possibility of the presence of incipient periodontal disease that may be detected in these children with more subtle diagnostic means.

Table 3. Mean Distances from the Cementoenamel Junction to the Alveolar Bone Crest by Tooth Type and Group

Primary teeth

	Molars		Cuspids	
Group	mean	SE	mean	SE
C1	0.7	0.1	1.2	0.1
C2	0.8	0.1	1.1	0.1
FD	0.7	0.1	1.1	0.2

Permanent teeth

	Molars		Premolars		Cuspids	
Group	mean	SE	mean	SE	mean	SE
C1	0.3	0.1	0.4	0.1	0.4	0.1
C2	0.2	0.1	0.4	0.1	0.5	0.2
FD	0.2	0.1	0.6	0.1	0.8	0.2

No statistically significant differences between the groups, $P>0.05$.

Conclusions

The present study indicates that there is no statistically significant difference in the alveolar bone height between children and adolescents with familial dysautonomia and those with no systemic disease, when measured on bitewing radiographs. None of the examined children in the study had definitive radiographic evidence of childhood periodontitis.

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Abstract of the Scientific Literature

In Vitro Evaluation of Amalgam Bonding Agents

Previous studies have suggested that dental amalgam restorations may experience marginal leakage for periods as long as 18 months. Traditionally copal varnish was used to seal the interface between the tooth surface and amalgam restoration but, in recent years it has been suggested that bonding agents should be used in place of copal varnish. This study investigated the shear bond strength, microleakage, and degree of dentin penetration of 4 different dentin bonding agents (dba) recommended for amalgam restorations. The following 4 dba were used with amalgam alloy (Lojic, SDI): OptiBond Solo (OS, Kerr), AmalgamBond Plus (AB, Parkell), AmalgamBond Plus + high-performance methyl acrylate fibers (AB + HPA), and Prime& Bond 2.1 (PB, Dentsply). The values for mean shear bond strength (in MPa) of amalgam to dentin were: OS:2.0; AB:2.6; AB + HPA:7.2; PB 5.4. These shear bond strength values are low compared to those reported with composites (approximately 25 MPa). All 4 systems exhibited microleakage at the amalgam/bonding agent interface. In conclusion, the highest shear bond strength values of amalgam to dentin were obtained with Prime & Bond 2.1/base-catalyst and AmalgamBond Plus with high-performance methyl acrylate fibers. AmalgamBond Plus with high-performance methyl acrylate fibers had the highest variation of all 4 systems in both the bond strength and microleakage values.

Comments: The mean bond strength values of amalgam to dentin ranged from 2.0 to 7.2 MPa for these bonding agents. With fifth generation dentin bonding systems, shear bond strength values of ~ 25 MPa are reported for composite materials. Under the conditions of this study, microleakage was recorded at the amalgam/bonding agent interface with all 4 systems. PS

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Sias R. Grobler, Theunis G. Oberholzer, Roelof J. Rossouw, Anne Grobler-Rabie, Theunis J. Van Wyk Kotze. Shear bond strength, microleakage, and confocal studies of 4 amalgam alloy bonding agents. Quintessence Int 2000; 31:501-508.