ORIGINAL ARTICLE

Oro-Dental Manifestations of Pediatric Celiac Disease

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ABSTRACT

Objective: To document **Oro**-dental manifestations of pediatric celiac disease in our population.

Study Design: Prospective descriptive cross sectional study.

Place and Duration of Study: Department of Pediatrics, Post-Graduate Medical Institute/Ameer ud Din Medical College, Lahore, from 01-01-2023 to 15-02-2024.

Material and Methods: 45 pediatric patients, 2-18 years old, diagnosed as having celiac disease on the basis of small bowel biopsy were examined by a single consultant dental surgeon. Oro-dental findings were recorded through a predesigned proforma.

Results: Out of 45 patients, 19 (42.2%) were male, 26 (57.8%) were female with mean age of 8.16 ± 2.87 years. Dental lesions were most commonly located on incisors and molars combined. Defect in enamel color (35.6%) was the most common finding in our study. Among oral lesions, RAS (24.4%) was most common followed by atrophic glossitis (20%).

Conclusion: Oro-dental lesions are common in pediatric CD and must be screened for by early referral to dentist.

Key Words: *Pediatric celiac disease, Enamel defects, Recurrent atrophic stomatitis*.

INTRODUCTION

It is an established fact that systemic diseases can affect oral health. This is because systemic diseases may alter the function of immune system and usually cause chronic inflammation.[1] Celiac disease (CD) occurs in genetically susceptible individuals carrying the HLA DQ2 and HLA DQ8 haplotypes, due to immune related small intestine mucosal damage caused by immune related mechanisms triggered by dietary gluten and related triggers.[2] It can present as classical intestinal disease with diarrhea, poor weight gain, abdominal distention, abdominal pain, and malnutrition as common manifestations. The non-classical manifestations include short stature, arthritis, anemia, osteopenia, osteoporosis, dermatitis herpetiformis and other autoimmune diseases.[3]

Apart from systemic manifestations described above, oro-dental manifestations are also common which can include salivary disorders, delayed dental maturity, dental enamel defects (DED) and recurrent aphthous stomatitis (RAS).[4] These manifestations are sometimes helpful in identifying the patients with CD, particularly the asymptomatic type.[5] Oro-dental problems have been identified and reported to be higher in pediatric population.[6] Yet other researchers have findings that are contradictory to the above.[7] Delayed diagnosis of pediatric CD is also common as our health care professionals and dentists have

lesser understanding of disease symptoms and clinical presentation.[8] To the best of our knowledge, there is no comprehensive study that describes oral features of CD in our children. Therefore, we conducted this study to document the oro-dental findings of pediatric CD in our population.

MATERIAL AND METHODS

We conducted this prospective, cross-sectional study at the Department of Pediatrics, Post-Graduate Medical Institute/Ameer ud Din Medical College, Lahore, from 01-01-2023 to 15-02-2024. Nonprobability purposive sampling technique was used to recruit 45 male and female pediatric patients whose age was between 2-18 years. Prevalence of CD was taken as 3% in Pakistani population, confidence interval as 95% and margin of error 5%.[9]

$$n = \frac{3.8416 * (0.03 * (1 - 0.03))}{0.05 * 0.05} = 44.716224$$

Patients were diagnosed and labelled as having CD based on small bowel biopsy following ESPAGHAN criteria, by a single Pediatric Gastroenterologist.[10] The children with CD who were on corticosteroids, with physical or mental disabilities, chromosomal or congenital disorders, patients on tetracycline, undergoing orthodontic treatment or with dental fluorosis, were excluded from the study. All the subjects were examined by a single Consultant Dental Surgeon at hospital. Presence of oro-dental findings such as RAS, enamel defects (using Aine criteria) and lesions in oral soft tissue were investigated and recorded through a predesigned proforma after parental consent and approval from Institutional Review Board Committee.[11]

RESULTS

Out of 45 patients, 19 (42.2%) were male and 26 (57.8%) were female. The mean age of our patients was 8.16 ± 2.87 years. Dental lesions were most commonly located on incisors and molars combined. Defect in enamel color (35.6%) was the most common finding in our study. Among oral lesions, RAS (24.4%) was most common followed by atrophic glossitis (20%). The details of our findings are given in table 1.

TABLE:-1 Dental enamel defects, Aine Classification and location, distribution of oral soft tissue lesions

Dental enamel defects (Aine classification)	
Grade 0: no defect	14 (31.1%)
Grade I: deficits in enamel color	16 (35.6%)
Grade II: slight structural enamel defects	9 (20%)
Geade III: evident structural defects	6 (13.3%)
Localization	
Incisors	8
Incisors and canines	1
Premolars	1
Molars	3
Incisors and molars	14
Oral soft tissue lesions	
Recurrent atrophic stomatitis	11 (24.4%)
Atrophic glossitis	9 (20%)
Angular cheilitis	5 (11.1%)
Migratory exfoliative glossitis	7 (15.5%)
Glossodynia	6 (13.3%)

DISCUSSION

Recent epidemiological data demonstrates a continuous rise in the incidence of CD. Many factors attribute for these findings. There is increased awareness of the disease nowadays among physicians and patients. Improved recognition coupled by wide availability of the serological tests help in earlier screening of the disease. The real increase of this immune based disorder, independent of disease detection, is also responsible for rise in incidence of CD. However, the true reason related to rise in recent decades is still unknown. There has been no success in reducing the development of CD in any randomized control trials. The preventive efforts majorly focus on the quantity of gluten exposure during early life.[12] The major site involved by CD is the proximal part of intestinal mucosa. Nevertheless, presence of gluten derived T cells are found in whole of the gastrointestinal tract, including mouth. The occurrence of oral lesions in soft tissue and mineralized tissue present in mouth of patients with CD has been documented by various studies.[13,14]

Our study shows female preponderance (57.8%) with CD, a finding similar to AlNababteh and colleagues who narrated the higher prevalence of CD in female pediatric patients as 61% in their study.[15] However, El-Hodhod et al documented

increased frequency of CD in male patients being 51.43%, a finding in contrast to ours.[16] The mean age of our patients with CD was 8.16 ± 2.87 years. The findings are similar to Cantekin et al and El-Hodhod et al where mean age of children with CD was 8.94 ± 2.08 years and 8.33 ± 1.92 years respectively.[16,17] However, AlNababteh and colleagues have documented a lower mean age of 6.74 ± 3.3 years.[15] While a somewhat higher mean age of 13.2 ± 2.85 years has been mentioned by Maan et al.[18] Both of these are in contrast to our findings.

Mineralization disorders causing DEDs are classified as quantitative (e.g., hypoplasia) and quantitative (e.g., discoloration). Various disturbances can occur during matrix formation and mineralization of teeth. These factors may be locally acting agents, systemic or sometimes hereditary. These factors injure ameloblasts resulting in decrease production of matrix. Calcification of enamel matrix later leads to enamel hypoplasia, which is the quantitative defect. The qualitative defect results due to injury occurring during maturation phase, resulting in hypo-calcified areas. The exact cause of DED in patients with CD are unknown still. Various conditions can lead to hypoplasia of dental enamel in CD among which genetic susceptibility, hypocalcemia, or autoimmune reaction in enamel during odontogenesis are more common.[19] Our study has concluded that grade I DED were most common in our study group followed by grade II and grade III. De Carvalho and colleagues have also concluded that grade I DED are most prevalent in patients with CD, in agreement to our findings.[20] However, in contrast to us some authors have documented grade II DED as more common rather than type I.[21]

RAS is another condition commonly associated with CD; however, its etiology remains unknown still. The most plausible explanation in this regard can be the deficiency of micronutrients like Vitamin B12, folic acid and iron. Usually there are single or multiple oral mucosal ulcers with an erythematous base, round or oval, recurrent and are painful.[22] RAS was found to be 24.4% in our study group. A much higher prevalence of 50% and 44% has been documented by Villemur et al.[8] and Cantekin et al, respectively.[8,17] These findings are in contrast to ours. However, a range of 9.66% to 40.98% has been documented in literature for RAS.[17] It was followed by atrophic glossitis being 20% in our study group. In this condition the tongue becomes erythematous and smooth. Patients complain of difficulty in swallowing, chewing and speaking. Bramanti al conducted a similar study and found atrophic glossitis being 14% in their patients, somewhat similar to ours [23]

This was a pilot project to document of oro-dental manifestation of pediatric celiac disease in our population. Using stringent inclusion criteria and involvement of consultants for examining patients rather than mere interviewing were among strength of our study. Being a single center study and smaller sample size are some of the limitations of our study. Case control studies, with larger sample size, involving multiple pediatric centers from different parts of country are needed to formulate guidelines for screening and diagnosing oro-dental findings in our pediatric patients with CD as these are common and may go to adulthood, undetected.

It is of utmost importance that pediatricians, dentists and especially pediatric dentists should have a thorough knowledge of oro-dental manifestations of CD. Increased awareness can help earlier diagnosis and prevent complications of CD in a growing child in long term. Patient's dentist should be informed about the diagnosis as CD affects future dental development and dental treatment. Dentists can also pick features of CD who are asymptomatic, at initial stage of disease or in case of non-adherence to gluten free diet, guiding pediatricians for optimum management of CD.

CONCLUSION

Oro-dental lesions are common in pediatric CD and must be screened for by early referral to dentist.

Conflict of interest: None

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