CASE REPORT

Oral Manifestation of Autoimmune Systemic Disorder: A Case Report

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Abstract

Celiac disease is an autoimmune disorder that occurs in genetically predisposed people caused by gluten which attacks the lining of the small intestines and damages the villi, leading to malabsorption. Sometimes recurrent aphthous ulcers and benign migratory glossitis could be the only oral manifestation of this disorder. When pediatric dentists encounter these features, they should enquire about other symptoms, associated with celiac disease. In suspected cases, appropriate referral and a timely diagnosis can help prevent complications of this disorder.

Keywords: Benign migratory glossitis, Celiac disease (CD), Dental enamel defect, Gluten allergies, Recurrent apthous stomatitis.

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Introduction

Gluten, a protein present in various grains induces an autoimmune response in highly susceptible individuals which results in celiac disease (CD) or celiac sprue. It has four types- classical, atypical, subclinical, and underlying. The classical form includes developmental delays and gastrointestinal disorders. Such as chronic diarrhea, weight loss, vomiting, stomach ache, etc. The atypical, subclinical, and underlying forms display various symptoms from other sites. The atypical pattern predominates in extraintestinal features such as lesions of the oral mucosa, tooth enamel defects[1] iron deficiency anemia, dermatitis herpetiformis, and osteoporosis.

Multiple presentations of CD create a clinical challenge in diagnosis. Increased awareness and careful questioning helps in a timely diagnosis and obviate complications of untreated CD.

Case Description

A 4-year-old boy, accompanied by his parents, reported to the Department of Pediatric and Preventive Dentistry with frequent rotating ulcers on his tongue and buccal mucosa that have been self-limiting since 2 years of age. An extensive family, prenatal, postpartum, medical, and dental history was not reported. The patient displayed a moderate body construct, had no skeletal abnormalities, and occasionally complained of stomach aches.

On further extraoral examination, we observed pruritic papular eruptions over the skin of elbows and knees (Fig. 1). Intraoral examination revealed that there was the presence of benign migratory glossitis (Fig. 2) and recurrent aphthous stomatitis (RAS) (Fig. 3).

Recurrent aphthous stomatitis was solitary round punched-out ulcer (2 mm in diameter) with circumscribed margins; erythematous haloes, located on the lower lip, and was associated with pain and difficulty in eating. There were no aggravating/relieving factors present. On asking about the history of ulcer parents informed that they noticed the ulcer around 1 year back which was self-limiting and reoccurring in nature.

On the basis of history and clinical presentation, we suspected that there was a possibility of a systemic condition associated with these signs, so we initiated topical anesthesia that is lidocaine as 1% cream 3–4 times a day for 7 days for symptomatic relief and then we referred the patient to a pediatrician for systemic evaluation.

A week later, the patient came back with all pertinent reports and was diagnosed with celiac disease. The blood tests revealed that the patient was eosinophilic and the serological, tissue transglutaminase antibodies (tTG) (IgA) test revealed an increase in anti-tissue transglutaminase antibodies (tTG ANTIBODY-72.26 IU/mL) which substantiated the diagnosis so, a gluten-free diet was exhorted.

Discussion

In 1888, celiac disease was first described by Samuel Gee; however, the function that gluten played in the development of this pathology became pellucid in 1953.[2] Gliadin, hordein, secalin, and avenin were the parts of the gluten complex that play a role in the development of this sickness.

CD includes both genetic and environmental factors. Genetic factors are human leukocyte antigen (HLA) DQ2 and HLA DQ8 while the environmental factor is gluten. The CD is commonly reported after 6 months of life which coincides with the introduction of gluten in the diet. Gluten is not fully digested, its only breakdown into small peptides, which are transferred into the intestine where it interacts with tissue transglutaminase (TTG) and forms negatively charged peptides. This negatively charged peptide

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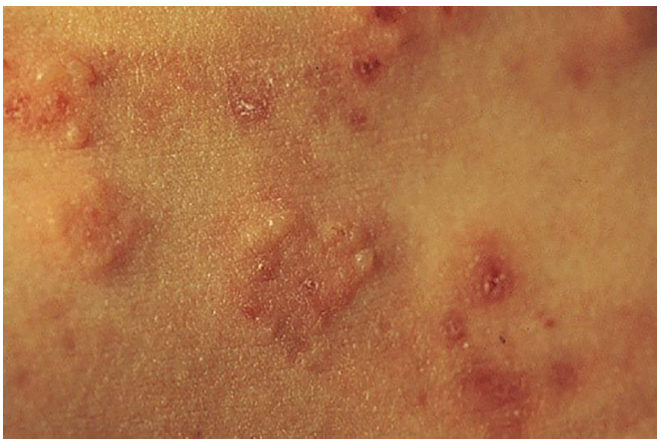

Fig. 1: Pruritic papular eruption over the skin of the elbow

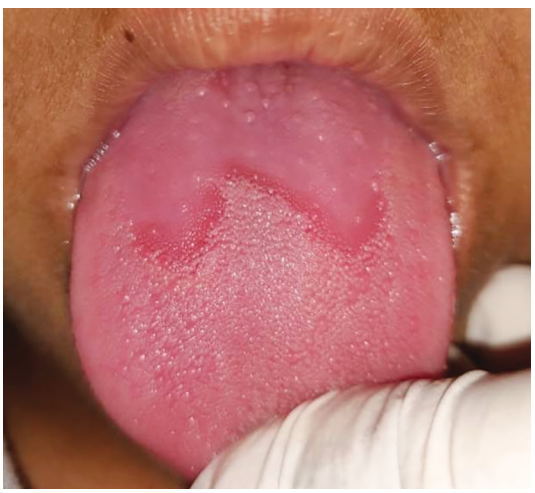

Fig. 2: Benign migratory glossitis

complements the positively charged HLA DQ2 and HLA DQ8 leads to the production of autoantibody,[3] which might be the substructure of disorder diagnosis. In this way, it can cause inflammation of intestinal villi.

The worldwide prevalence of celiac disease was 1.4%. In Asia, the occurrence of celiac sickness used to be 0.6% with girl predilection.[4] In India, the occurrence of celiac sickness was 0.8–1.04%.[5]

Signs and symptoms of celiac disease include persistent diarrhea, steatorrhea, stunted growth, muscle wasting, loss of appetite, failure to grow tall, and weight loss, all of which lead to lethargy and emotional misery.[6]

Extraintestinal manifestations of CD include iron deficiency anemia, dermatitis herpetiformis, and osteoporosis. Oral manifestation includes[7] glossitis, recurrent aphthous ulcer, stomatitis, burning mouth syndrome, enamel hypoplasia, and delayed tooth eruption. In celiac individuals, the cause of dental enamel abnormalities is still not clear. Some researchers believe that enamel defects are linked to hypocalcemia caused by intestinal malabsorption, which is necessary for enamel defects to occur. Celiac sickness sufferers with the HLA-DR3 genotype validated a higher hazard of enamel lesions, suggesting a genetic etiology.

Oral manifestation of soft tissue is due to insufficiency of vitamin $B_{12,}$ folic acid, and iron because the process of absorption is deficient in these patients. Although till now it is still not clear whether the oral lesions occur as a result of malabsorption or as a direct manifestation of CD.[8]

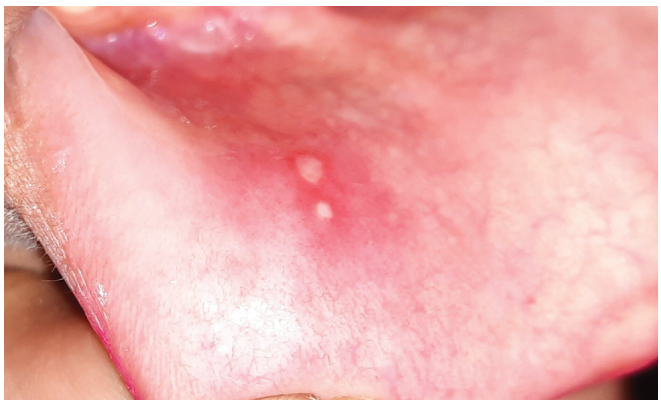

Fig. 3: Recurrent aphthous stomatitis

In this present case, we found frequent glossitis, recurrent aphthous ulcer, and dermatitis herpetiformis which was a clear indicator of gastrointestinal disease so with the help of a serological test a final diagnosis of CD was established.

A strict, lifelong gluten-free diet is the only treatment option available for celiac disease[9] if in case the treatment is not provided it will lead to various complications, such as sterility, osteoporosis, neurological and psychiatric disturbances. The intraoral visual examination of these sufferers gives an economical, speedy, and noninvasive procedure. When CD is suspected, dental practitioners need to refer to the pediatrician for analysis and substantiation.

After diagnosis, all celiac patients ought to be integrated into a preventive dental program aiming to provide professional oral hygiene, pits and fissures sealing, fluoride topical application, treatment of dental caries, and fractures of hypoplastic enamel.[10] It is necessary to make him aware that the soft tissue lesions will disappear after taking a gluten-free diet and if it reoccurs, it is advised to consult a pediatrician/pedodontist.

Conclusion

The oral cavity is the doorway to the gut therefore a meticulous inspection of the oral cavity, thorough history, and serological tests may contribute to an early diagnosis of celiac disease. Therefore, as a dentist, we should be aware of the signs and symptoms of celiac disease and help these patients to prevent complications of the disease.

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