Periodontitis as Manifestation of Crohn's Disease in Primary Dentition: A Case Report

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ABSTRACT

The purpose of this case report is to detail an initial periodontal manifestation of Crohn's disease in a 6-year-old boy. The first clinical diagnosis, localized aggressive periodontitis, was based on the microbiological isolation of Actinobacillus actinomycetemcomitans from subgingival sites. On examination, gingival lesions, together with bleeding on probing, edema, and erythema, were observed. Although an increased probing depth was detected, no radiographically visible alveolar bone loss was observed. According to these findings, periodontitis as a manifestation of a systemic disease was assumed. Furthermore, fissural ulcerations of the lips were noted. The patient also reported a swelling of the upper lip in the morning. Oral hygiene procedures, scaling, root planning, and the application of metronidazole and amoxicillin were not successful. Metabolic and several immunological tests, however, showed normal values. Two months after the first periodontal signs, the child suffered from severe malnutrition, accompanied by diarrhaea and abdominal pain. Active colitis with multiple granulomas was detected histopathologically from biopsies. Crohn's disease was then diagnosed by the internist. If in doubt, medical examinations in every case of childhood periodontitis are recommended to determine whether the findings speak for initial symptoms of a systemic disorder (eg, Crohn's disease). (*J Dent Child*. 2004;71:193-196)

KEYWORDS: CROHN'S DISEASE, PERIODONTITIS, ACTINOBACILLUS ACTINOMYCETEMCOMITANS

Inflammatory bowel diseases, ulcerative colitis, and Crohn's disease are chronic intestinal disorders of unknown etiology. Recently, genetic and environmental components have been assumed.[1] Moreover, frequent use of antibiotics is discussed as a possible risk factor.[2]

Enteritis regionalis (Crohn's disease), defined in 1932 by Crohn et al[3] as a separate syndrome, is a nonspecific, segmental, or plurisegmental inflammation of the intestines. Crohn's disease (CD) may affect any part of the gastrointestinal tract. Typically, it affects the terminal segment of the small and first segment of the large intestine.

In more than 50% of all cases, CD becomes manifest before the age of 30, the age of its peak frequency. Boraz[4] stated that CD often develops during childhood. In central Europe, the disease seems to occur in both sexes with equal frequency.[5] Authors who examined children and adolescents with CD found,[6] for example, 220 positive family case histories in 724 patients (30%). Bertrams et al suggest that CD is possibly associated with the HLA antigens B_{12} and B_{15}.[7]

It has also become increasingly evident that the syndrome does not affect the intestines exclusively. Extraintestinal manifestations involving other organs and organ systems have been reported. Besides the nervous system, joints, and eyes, manifestations of interest to dentists are the skin, oral mucosa, gingiva, and tonsils.[7-11] In 1991, Plauth et al[12] analyzed 79 cases of CD, mostly from reports in the literature. In 60% of all cases, oral symptoms were described; as a rule, however, these occurred simultaneously with the disease's intestinal course. The patients' average age was 22 years, and the ratio between male and female was approximately 2:1.

Engel et al[13] identified abnormal lymphocyte profiles in a patient with CD and aggressive periodontitis. Furthermore, it is well known that Actinobacillus actinomycetemcomitans

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plays an important role in the pathogenesis of localized aggressive periodontitis, even in primary dentition.[14,15] Until now, however, there was no report about cases in which A actinomycetemcomitans had been detected in patients with an oral manifestation of CD.

With a diagnosis of periodontitis in the primary dentition, the dentist is faced with the question of whether a systemic disease should be ruled out by differential diagnosis.

CLINICAL AND LABORATORY FINDINGS

Swelling and reddening of the gums caused the parents of a 6-year-old boy to consult their family dentist. Further symptoms that had rather been neglected were a discrete swelling of the upper lip in the morning and bilateral commissural fissures (Figure 1). The dentist's therapy involved professional measures of oral hygiene and mouth rinsing, which were unsuccessful, resulting in the child being referred to the dental clinic of the University of Jena, Jena, Germany.

After a 3-month history of gingival pain, the boy presented with gingivitis and various periodontal lesions. The periodontal findings and detection of A actinomycetemcomitans in subgingival plaque initially led to the clinical diagnosis of a localized aggressive periodontitis. Supra- and subgingival plaque samples were taken with endodontal absorbent paper points (ISO 30, Dentalwerke, Munich), stored in the appropriate transport medium, and cultured immediately thereafter.[16]

The microbiological analysis detected A actinomycetemcomitans as well as Capnocytophaga species. The subgingival total count of anaerobic bacteria as well as the counts of A actinomycetemcomitans and Capnocytophaga species were higher than the supragingiva values (Table 1).

The entire maxillary gingiva was highly reddened and inflamed, whereas the lower jaw's gingiva showed no signs of inflammation. The maxillary anterior teeth were affected most severely. Here, due to the gingival edema, the probed pockets were especially deep. On average, the patient had a plaque index of 1.2,[17] sulcus bleeding index of 2.1,[18] and mean probing pocket depth of 4.2 mm (Figure 2). Most likely, pseudo-pockets caused by the edema contributed to a high share to the aforementioned mean probing pocket depth.[19] Radiographically however, no bone loss could be detected. Due to the clinical findings, the periodontitis was finally diagnosed to be a manifestation of a systemic disease.

Table 1. Microbiological Plaque Analysis

	Supragingival	Subgingival
Total anaerobic bacterial count	1.2×10^4	1.2×10^6
Actinobacillus actinomycetemcomitans	1.4×10^3	1.2×10^4
Capnocytophaga species	6.0×10^2	4.0×10^7

An immunological and metabolic analysis followed. The immunological analysis of the peripheral blood supplied normal values for immunoglobulin levels (IgG, IgM, and IgA), lymphocyte subpopulations (fluorescence-activated cell sorter), and the phagocytosis and chemotaxis functions of the polymorphonuclear neutrophil granulocytes.[16,20,21] Likewise, the values of the glyco- and lipometabolism were within normal range at that time.[21-23]

ATTEMPTED DENTAL THERAPY BEFORE INTESTINAL MANIFESTATION

The oral hygiene measures, which were carried out regularly at short intervals, were just as unsuccessful as the nonsurgical therapy. The latter was based on scaling and root planing[24] and an 8-day adjuvant antibiotic application with amoxicillin and metronidazole.[25] The antibiotics were administered after microbiological resistance determination. Professional cleaning of the mouth twice per week and daily chlorhexidine mouthrinses were administered, as the patient's pronounced sensitivity to touch restricted his ability to practice oral hygiene at home. Again, neither measure was suited to better the boy's condition significantly.

Three months after the periodontal manifestation, the boy developed massive intestinal symptoms, including diarrhaea, abdominal pain, and progressive weight loss. Clinically, this raised a suspicion of CD. Colonoscopy revealed

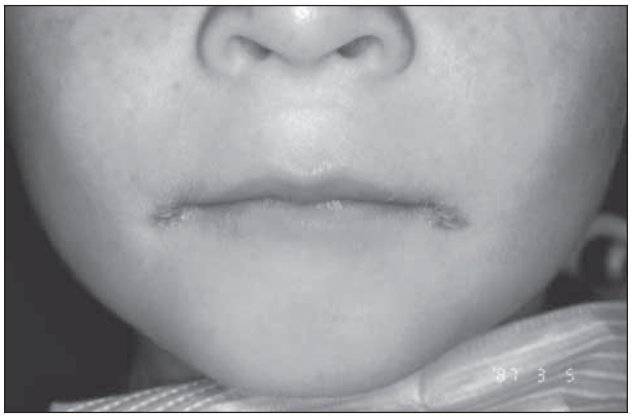

Figure 1. 6-year-old child with bilateral commissural fissures and slight upper lip swelling.

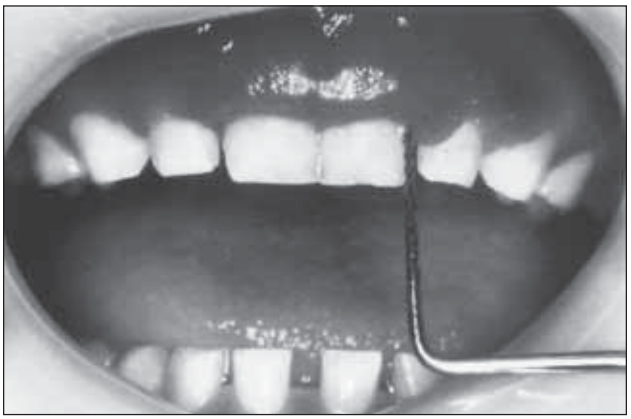

Figure 2. Clinical picture of the same patient, with suspected diagnosis of prepubertal periodontitis.

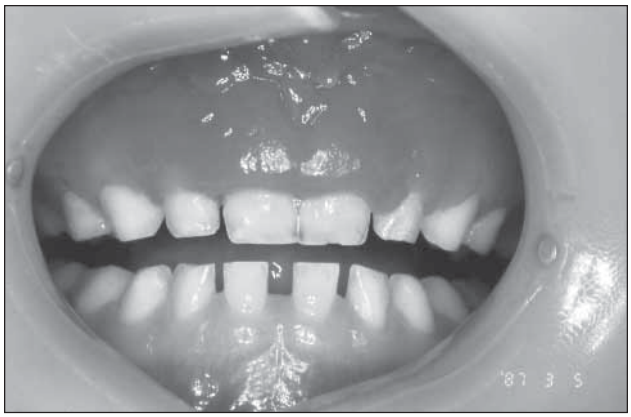

Figure 3. Two months later: Cobblestone relief of the gingiva after intestinal manifestation of Crohn's disease.

follicular hyperplasia of the terminal ileum, swelling, and fissural ulcers of the ileocecal valve. Histologic sections showed active colitis with masses of lymphocytes and noncaseating granulomas. The clinical picture of the gingiva had altered progressively and resulted in a typical cobblestone relief (Figure 3).

Chronic or aggressive childhood periodontitis is very rare.[26] As long as no general symptoms exist, it is difficult for clinicians to distinguish a special periodontitis form from oral manifestations of systemic disorders in which periodontitis is a concomitant affectation only. The present case report is mainly focused on this problem and the comprehensive differential diagnostic examination of periodontitis. Gingival swelling in children can be a manifestation of a number of systemic conditions, including CD.[27]

Plauth et al[12] reported about CD patients who developed oral symptoms simultaneously with intestinal ones. They describe lesions on the lips, buccal mucosa, vestibular sulcus, and gingiva. The most frequent symptoms were ulcerations and polypoid hyperplastic mucosa. If the gingiva is affected, the typical pavement relief, with cobblestone-like gingival alterations and a pronounced tissue thickening can be observed. This is a conspicuous symptom, with particular frequency in the terminal ileum and associated with isolated edemas of the mucosa and fissural ulcera.

In about 50% of the cases reported in the literature, complete remission of the oral symptoms under systemic or local application of steroids is described by Ellis.[28] The efficient drugs applied include, for example, prednisolone and azothioprine. Also described is the successful remission of oral symptoms after metronidazole therapy.[27] Metronidazole led to a marked improvement of both the gastrointestinal and gingival conditions.[29] In the present case, however, administering metronidazole and amoxicillin brought no relief from the symptoms.

In this context, it is particularly interesting to note that A actinomycetemcomitans, which is considered to be linked with localized aggressive periodontitis, was detected in the described case. Suzuki et al[15] also describe A actinomycetemcomitans in a case of localized aggressive periodontitis in primary dentition with radiographically proven bone loss. In spite of this, the presented case showed no alveolar bone loss. The author diagnosed preliminary periodontitis as a manifestation of an unknown systemic disease. Only after intestinal symptoms occurred was the diagnosis of CD made by the internist.

Conversely, Lamster et al[21] reported about a 28-year-old male with manifest CD who additionally presented with severe alveolar bone loss and an enhanced polymorphonuclear leukocytes (PMNL) function. In this case, periodontal breakdown was primarily caused by the bacterial infection.

Possibly, microbial colonization plays a more pathogenetic role in CD than had been previously believed. As early as 1986, van Dyke et al[30] wrote about the potential role of periodontopathogenic micro-organisms in the pathogenesis of chronic inflammatory diseases of the intestines. The connections currently reported between general diseases such as arteriosclerosis and certain periodontopathogenic bacteria should perhaps be discussed also for CD.[31] This is becoming even more important as the relationships between this disease and periodontitis have already been the subject of epidemiological studies.[28,32] Flemmig et al[32] reported in 1991 about the prevalence and degree of severity of periodontitis in 107 patients with CD and ulcerative colitis. In 30% of the patients, probing depths of 4 mm or greater were found in at least at site of each tooth. Compared with the health state of US adults, the periodontitis prevalence in patients with chronic inflammatory bowel disease is about 10% higher.

CONCLUSIONS

In summary, where periodontitis in primary dentition is clinically suspected, differential diagnosis should be applied to exclude CD and other systemic diseases that may have oral manifestations.

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