Periodontal Disease Associated With Oral-Facial-Digital Syndrome: A Case Report

Z Sllamniku-Dalipi, F Dragidella, M Disha, K Meqa, S Disha

Citation

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Abstract

The oral-facial-digital syndromes (OFDS) involve morphogenetic impairments that almost invariably affect the mouth, face, and digits and result in pleiotropic effects. This study is a clinical case report of a patient with periodontal disease and OFDS who underwent periodontal surgery.

A 24-year-old female patient, was referred to the Department of Periodontology and Oral Medicine in the University Clinical Dental Centre in Prishtina. Clinical periodontal screening and radiological examination revealed suprabony and infrabony pockets, gingival inflammation, tooth mobility, and pathologic tooth migration. We also observed bi-maxillary protrusion with protrusion of maxillary frontal teeth. Dysmorphic features affecting the head included facial asymmetry, micrognathia, broadened nasal ridge, and hemi-facial microsomia. Digital abnormalities affecting the hands and feet included brachydactyly.

After basic periodontal therapy and full-mouth scaling and root planing, periodontal surgery with a modified Widman flap in teeth 13-27 was performed. During the surgical intervention teeth.12, 11, 21, 22, 26 were extracted. Six weeks after periodontal surgery the patient underwent prosthetic rehabilitation.

Surgical periodontal treatment resulted in improved functionality and esthetics and had a satisfactory psychological impact on the patient with respect to oral health.

INTRODUCTION

Periodontitis is a chronic inflammatory disease that affects the supporting structures of the teeth. Chronic inflammatory periodontal disease is generally a painless, asymptomatic, and slowly progressive disorder that begins as inflammation of the marginal gingival in response to microbial colonization of the adjacent tooth surface (1). Increased inflammatory activity disrupts the normal balance of bone formation/ resorption and results in alveolar bone loss (2). Progression of periodontal disease ultimately leads to tooth loss (3).

Periodontal management includes a complete individual assessment of each patient. Medical and dental history, clinical and radiographic examinations, and an assessment of risk factors are all important factors in making an accurate diagnosis, predicting prognosis, and developing an optimal treatment plan (4). Conventional periodontal therapy includes both surgical and non-surgical approaches to treat the inflamed dento-gingival complex and improve overall periodontal health (5, 6).

A number of systemic genetic disorders increase both susceptibility to periodontal disease and the progression and aggressiveness of the disease. Alterations in the periodontal tissues may be a primary consequence of such systemic alterations or a secondary effect that either causes periodontal disease to progress without any apparent underlying cause or increases the severity of a previously established local condition (7). The principal causative agent of periodontal disease is bacterial plaque, which induces progressive tissue damage. However, the role of bacterial plaque in the presence of susceptibility to periodontal disease due to a systemic condition is debatable. In such cases, the development and evolution of periodontal disease is largely dependent on the host immune response, the integrity of the tissues, humoral and cellular immunity, and certain genetic, endocrine, and nutritional factors (8).

The oral-facial-digital syndromes (OFDS) are a heterogeneous group of developmental disorders that are characterized by an X-linked dominant mode of inheritance with lethality in males (9). This syndrome is characterized

by malformations of the face, oral cavity, and digits. In the literature there is little data reporting the effects of OFDS on the periodontal tissues. Clinical features include facial dysmorphism with oral, tooth, and distal abnormalities, polycystic kidney disease, central nervous system malformations, and abnormalities of the fingers (10).

This article presents the clinical case of a patient with severe periodontal disease and OFDS who underwent periodontal surgery, teeth extraction, and prosthetic rehabilitation.

CASE REPORT

A 24-year-old female was referred to the Department of Periodontology and Oral Medicine in the University Clinical Dental Centre in Prishtina. A detailed dental, medical and social history was obtained from the patient. She had complaints of a functional and esthetic nature, including inability to chew, lip incompatibility, gum bleeding, itchiness in the gums, and changes in the position of the teeth. Psychosocial discomfort due to incorrect positioning of the teeth was associated with a speech impediment.

A full-mouth periodontal examination was conducted and the following variables were determined: gingival index (Loe-Sillness), plaque index (Sillness –Loe), calculus index (Löe and Silness), probing depth (PD),clinical attachment level(CAL) and bleeding on probing(BOP). Clinical periodontal parameters were measured at six sites per tooth.

The gingival index (Löe-Silness) was recorded for all present teeth as follows: 0-normal gingival; 1-mild inflammation, slight change in colour, slight oedema and no bleeding on palpation; 2-moderate inflammation, moderate redness, moderate oedema, and moderate ulceration and the tendency for spontaneous bleeding; and 3-severe inflammation, marked redness, marked oedema, and marked ulceration and the tendency for spontaneous bleeding (11).

The plaque index (Silness and Löe) was recorded for all present teeth as follows: 0—no plaque in the gingival area; 1—a film of plaque adhering to the free gingival margin and adjacent areas of the tooth, with plaque recognised only by running a probe across the tooth surface; 2—a moderate accumulation of soft deposits within the gingival pocket, on the gingival margin, and on the adjacent tooth surface that was observable by the naked eye; and 3—an abundance of soft deposits within the gingival pocket and/or in the gingival margin as well as on the adjacent tooth surface(12).

Calculus index (Greene-Vermilion) was recorded for all present teeth as follows: 0 – no calculus; 1 – calculus covering up to 1/3 of the tooth surface; 2 - calculus covering from 1/3 up to 2/3 of the tooth surface; 3 – calculus covering more than 2/3 of the tooth surface (13).

The clinical attachment level (CAL) was measured using the cement-enamel junction as a reference point, specifically from the cement-enamel junction to a probable pocket depth. The probing pocket depth was measured from the crest of the gingival margin to a probable pocket depth. The probing depth and attachment level measurements were made at the mesio-buccal, buccal, disto-buccal, disto-lingual, lingual and mesio-lingual positions of every tooth, with the exception of third molars.

Bleeding on probing was assessed on the 6 sites at which PD was determined and deemed positive if it occurred within 15 seconds after probing. BOP was expressed as the percentage of sites that showed bleeding (14).

The mean values were with regard to plaque index: 1.9 ±0.8; calculus index: 1.1±0.3; bleeding index: 85.2± 15.0; gingival index: 2.0 ±0.6; periodontal pocket depth: 4.7±0.9 and clinical attachment level: 5.3± 0.7.

Clinical periodontal screening and radiological examination revealed combined suprabony and infrabony pockets, gingival inflammation, tooth mobility, and pathologic tooth migration. We also observed bi-maxillary protrusion with protrusion of maxillary frontal teeth, malocclusion, and gothic palatum (Fig. 1, 2).

Figure 1

Pretreatment photographs showing protrusion of maxillary frontal teeth, gingival hyperplasia and inflammation and pathologic tooth migration

Figure 2

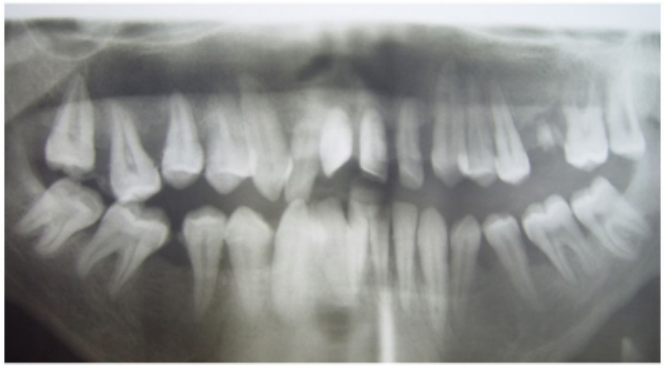
Pretreatment intraoral photographs showing bi-maxillary protrusion and gothic palatum.

Figure 3

Pretreatment lip incompatibility

Figure 4

Pretreatment lip incompatibility



A clinical extraoral examination showed dysmorphic features affecting the head, facial asymmetry, down-turned eyes, micrognathia, broadened nasal ridge, and hemi-facial microsomia. The digital abnormities affecting the hands and feet included brachydactyly (Fig. 5,6).

Figure 5

Brachydactyly and clinodactyly of the fingers

Figure 6

The patient's small feet

After basic periodontal therapy and full-mouth scaling and root planing, routine laboratory test: blood evaluation for the number of platelets, red and white blood cells, glycemia, sedimentation of erythrocytes, and bleeding and coagulation times were performed. Laboratory test results were within normal values. Periodontal surgery with a modified Widman flap in the region of teeth 13-27 was performed under local anesthesia. This technique offers the possibility of establishing an intimate postoperative adaptation of healthy collagenous connective tissue to tooth surfaces and provides access for adequate instrumentation of the root surfaces and immediate closure of the area. The initial incision was internal bevel incision to the alveolar crest

starting 0.5 to 1 mm away from the gingival margin. Scalloping followed the gingival margin. The papilla was left with a thickness similar to that of the remaining. The gingiva was reflected with a periosteal elevator. A crevicular incision was made from the bottom of the pocket to the bone, circumscribing the triangular wedge of tissue containing the pocket lining. After the flap was reflected, a third incision was made in the interdental spaces coronal to the bone with interproximal knife, and the gingival collar is removed. During the surgical intervention teeth 12, 11, 21, 22, and 26 were extracted (Fig. 7,8).Tissue tags and granulation tissue are removed with a curette. The root surfaces are checked, and then scaled and planed. Every effort is made to adapt the facial and palatinal interproximal tissue adjacent to each other in such a way that no interproximal bone remains exposed at the time of suturing. Interrupted direct sutures are placed in each interdental space.

Post operative antibiotics (amoxiclav tablets 1g, BID for 5 days) and analgesics (ibuprofen capsules 400 mg as needed) were described. Six weeks after periodontal surgery the patient received prosthetic rehabilitation (Fig. 10,11). The patient also underwent digital surgery (arthrodhesis contracture in proximal interphalangeal joint of digit IV and arthrodhesis contracture in distal interphalangeal joint of digit V).

DISCUSSION

Patients with OFDS are considered to have special needs. It is necessary to recognize the oral, facial, and digital characteristics of this specific group of patients in order to provide them with the best treatment (15). The case presented here involves a patient with OFDS associated with advanced periodontal disease. The patient presented with general characteristics of facial asymmetry, down-turned eyes, micrognathia, broadened nasal ridge, and brachydactyly.

Periodontal and oral examination of the patient revealed suprabony and infrabony pockets, gingival inflammation, tooth mobility, pathologic tooth migration, bi-maxillary protrusion, malocclusion, and gothic palatum. A periodontal surgical procedure was performed because of exacerbation of periodontal disease.

CONCLUSIONS

Surgical periodontal treatment resulted in improved functionality and esthetics, and had a satisfactory psychological impact on the patient with respect to oral health and quality of life. As maintenance treatment, the patient was advised on good tooth brushing technique, and regular plaque removal and rigorous oral examination was recommended to maintain a healthy and stable periodontal tissue status.

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Author Information

Zana Sllamniku-Dalipi
Department of Periodontology and Oral Medicine, Dentistry School, Medical Faculty, University of Prishtina
Prishtina, Kosova

Fatmir Dragidella
Department of Periodontology and Oral Medicine, Dentistry School, Medical Faculty, University of Prishtina
Prishtina, Kosova

Metush Disha
Department of Periodontology and Oral Medicine, Dentistry School, Medical Faculty, University of Prishtina
Prishtina, Kosova

Kastriot Meqa
Department of Periodontology and Oral Medicine, Dentistry School, Medical Faculty, University of Prishtina
Prishtina, Kosova

Saranda Disha
Department of Periodontology and Oral Medicine, Dentistry School, Medical Faculty, University of Prishtina
Prishtina, Kosova