

Dental Implants in a Young Patient with Papillon-Lefevre Syndrome: A Case Report

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Papillon-Lefevre Syndrome is characterized by generalized rapid destruction of the dental alveolar supporting bone and diffused palmoplantar hyperkeratosis. The disorder manifests itself as an autosomal recessive disease with an occurrence of about one to four cases per million.¹ It affects both the primary and secondary dentition. The periodontal changes usually appear before the age of 4 years. Inflammatory response in the periodontium leads to rapid bone loss and exfoliation of teeth. Because both sets of dentitions are affected, these patients are usually edentulous and wearing complete dentures by their teen years.

The exact immunologic abnormality of Papillon-Lefevre Syndrome is unknown. It has been reported that the disease may be associated with diminished neutrophil activity.² Microscopic changes include marked chronic inflammation with predominant plasma-cell infiltration, osteoclastic activity, and lack of osteoblastic activity.³ The bacterial flora in this disease is similar to those found in adult periodontitis with a prevalence of gram negative cocci, rods, and spirochetes.⁴

Because conventional periodontal treatment usually fails to arrest the rapid progression of periodontitis, se-

A case is reported of dental implant placement in a 13-year-old patient diagnosed with Papillon-Lefevre Syndrome. Two titanium dental implants were placed in the mandible for an implant-retained denture after the patient complained of having an unstable prosthesis. Follow-up radiographs showed suc-

cessful osseointegration and preservation of alveolar bone 1 year after implant placement and the continual wearing of a functional dental prosthesis. (Implant Dent 2003;12:140-144)

Key Words: *alveolar bone preservation, implant-retained overdenture, early edentulism*

vere loss of alveolar bone is often the result.^{2,5,6} Early extractions of all permanent teeth has been considered as the treatment of choice to preserve the remaining supporting bone.⁷

CASE REPORT

A 13-year-old male diagnosed with Papillon-Lefevre syndrome was presented at the Los Angeles County/University of Southern California (LAC/USC) Medical Center outpatient dental clinic in April 1999. The patient was also seen by the dermatology and ophthalmology departments. The patient displayed the classic signs of diffused palmoplantar hyperkeratosis (Fig. 1). Intraoral examination revealed class III hypermobility in all remaining dentition with severe gingival inflammation. A panoramic radiograph showed generalized advanced bone loss with both an atrophic maxilla and mandible (Fig. 2). It was determined that all erupted teeth were nonrestorable and required extractions.

Upon physical examination, the patient showed no other abnormalities. Intravenous sedation was scheduled because the patient was apprehensive to having dental extractions. The combination of Brevital (Eli Lilly and Co., Indianapolis, IN), Midazolam (Ben Venue Labs Inc., Bedford, OH), and

Fentanyl (Abbott Labs, Chicago, IL) were used. A total of 11 teeth were extracted. Tooth 1 and 16 were spared because they were unerupted and would not interfere with the dental prosthesis. The patient was given antibiotics and analgesics postoperatively. The follow-up took place with the general dentist at the dental clinic. Complete upper and lower dentures were fabricated for the patient. One year later, the patient complained about the instability of the lower denture.

He was consulted for dental implant placement at the oral and maxillofacial department of the LAC/USC Medical Center. The treatment plan was to place two dental implants and fabricate an implant-retained overdenture for his mandible. A preoperative Panorex was taken before implant placement (Fig. 3). Two 4.0 × 13 mm Branemark system titanium implants (Nobel Biocare, Goteborg, Sweden) were placed in the left and right canine areas of the mandible. The implant placement surgery was done in the operating room under general anesthesia. An immediate postoperative Panorex was taken (Fig. 4). The patient was placed on an antibiotic regimen (tetracycline 500 mg) and Peridex (Alpharma USPD Inc., Baltimore, MD) rinse for 2 weeks. He was followed-up

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Fig. 1. Hyperkeratosis in both palms.

weekly for the first month and then at regular intervals by the oral surgery department.

The postsurgical recovery period was unremarkable with no chief complaint or complications. The lower complete denture was first relined periodically with Viscogel (Dentsply, Munich, Germany) to avoid immediate loading of the implant fixtures during osseointegration. The implants were subsequently uncovered 4 months after their placements into the mandible. They were clinically and radiographically determined to be osseointegrated successfully. The criteria for success were the absence of mobility, the absence of radiographic gap in the bone-implant interface, and the absence of pain or infection at the periimplant area. The lower overdenture was then modified with the O-ring-type of attachments and stabilized through these implants. A follow-up panoramic radiograph was taken at week 23 showing preservation of supporting bone (Figs. 5). One year follow-up appointment showed continued success of the treatment with no further bone loss. Periapical radiographs and clinical photographs of the implants were taken (Figs. 6 and 7).

DISCUSSION

Papillon-Lefevre Syndrome is a devastating disease process characterized by rapid destruction of the dental alveolar complex. It starts affecting the individual during childhood and poses both physical and psychological challenges to these patients. Rapid bone loss and exfoliation of teeth often lead to early edentulism and the need to wear removable dental prostheses.

In this case study, the patient was referred to the LAC/USC Medical

Center outpatient dental clinic for dental extractions because of his oral condition of Papillon-Lefevre Syndrome. The fabrication of upper and lower complete dentures restored form and function for this patient. However, because of the inadequate and continual loss of bone support in the mandible, the stability of the lower denture was compromised. The treatment option of an implant-retained overdenture was then deemed appropriate. This study has shown that the successful outcome of implant treatment in patients with Papillon-Lefevre Syndrome is achievable. Not only did the two titanium implants successfully osseointegrate, but the supporting alveolar bone was also preserved. These implants helped increase the retention and stability of the mandibular denture through their attachments and by preserving the underlying bone structure. These results concurred with the findings of Ullbro et al.⁸

Dental implants function much

like ankylosed teeth.⁹ It has been stated that because of this characteristic they are contraindicated in growing individuals because they may result in the infrapositioning of implants.¹⁰ It is speculated that as bone growth occurs, the implant fixture would remain at its original position, resulting in a new but inferior position of the implant relative to the alveolar crest, thus termed infrapositioning. According to Behrendts,¹¹ the apposition of alveolar bone and the increase of alveolar height are completed during the early teen years. Whether infrapositioning will be of significance as the patient ages remains to be seen. Our successful implant placement in this 13-year-old patient certainly allows us to follow his growth, the prognosis, and the positions of these integrated implants in the near future. Nonetheless, the treatment approach in this case has shown initial success and has enhanced the therapeutic options in pa-

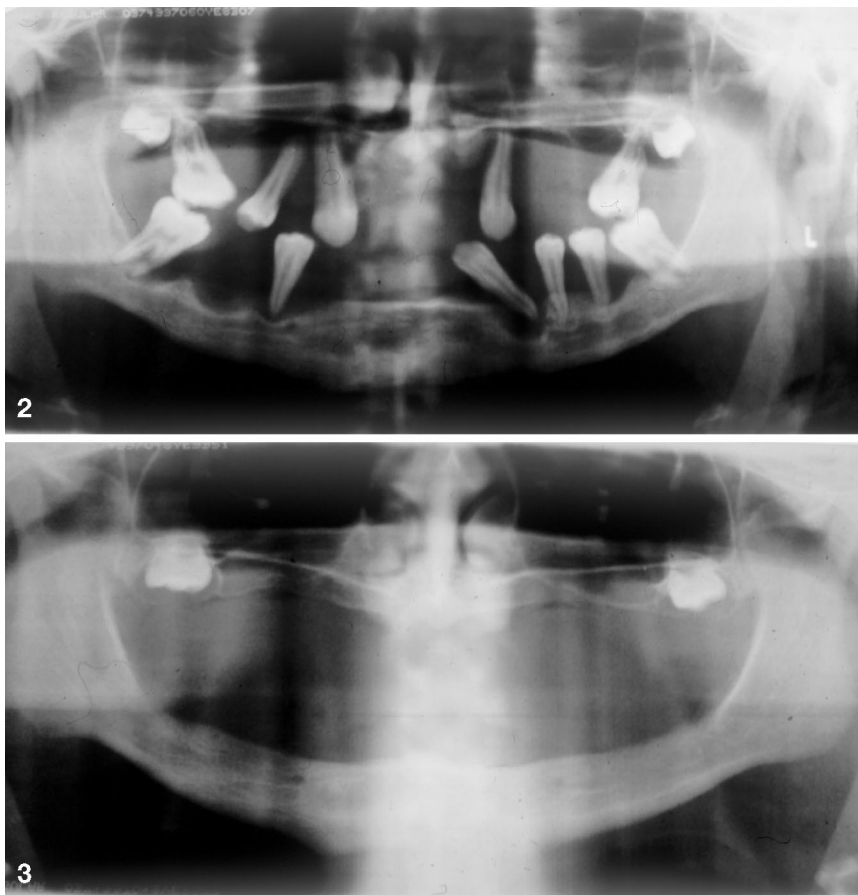


Fig. 2. Generalized advanced bone loss with hopeless dentition at the initial visit.

Fig. 3. Panorex of oral condition before implant placement.

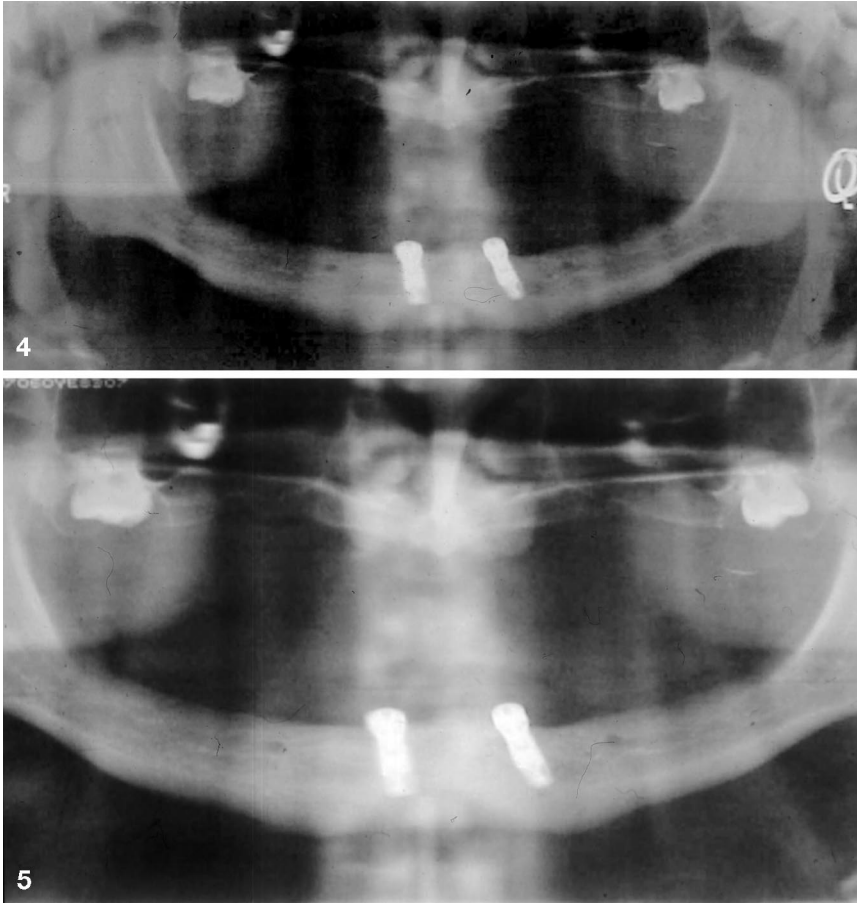


Fig. 6. Periapical radiograph of the dental implants at 1-year follow-up.

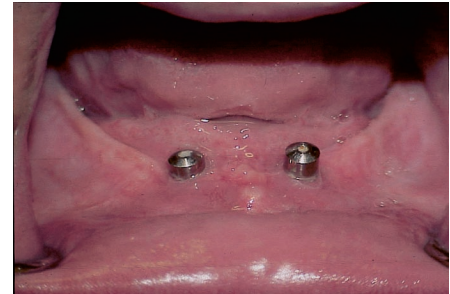


Fig. 7. Clinical picture of osseointegrated implants at 1-year follow-up.

Fig. 4. Immediate postimplant placement Panorex.

Fig. 5. Panorex at week 23 showing preservation of the alveolar bone.

tients with Papillon-Lefevre Syndrome.

CONCLUSION

This report has shown successful 1-year follow-up of implant osseointegration and alveolar bone preservation in a Papillon-Lefevre Syndrome patient wearing a functional implant-retained overdenture. This result provides a viable option for restoring edentulism in young individuals suffering from this disorder.

Disclosure

The authors claim to have no financial interest in any company or any of the products mentioned in this article.

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ZUSSAMENFASSUNG: Innerhalb des vorliegenden Artikels wird der Fall eines 13 Jahre alten, am Papillon-Lefèvre-Syndrom erkrankten Patienten geschildert, der einer Implantationsbehandlung unterzogen wurde. Nachdem der Patient über den mangelnden Sitz seines bisherigen Zahnersatzes geklagt hatte, wurden im Unterkiefer zwei Titanimplantate zur Befestigung einer implantatfixierten Prothese eingepflanzt. Bei der Nachuntersuchung, die ein Jahr nach erfolgter Implantation und dem ständigen Tragen der funktionalen Zahnprothese vorgenommen wurde, wurden Röntgenaufnahmen gemacht. Diese erwiesen eine erfolgreiche Integration der Implantate in das umliegende Knochengewebe sowie die vollständige Erhaltung des vorhandenen Alveolarknochens.

SCHLÜSSELWÖRTER: Erhaltung des Alveolarknochens, implantatfixierte Deckprothese, frühzeitiger Zahnverlust

ABSTRACTO: Se informa el caso de un informe dental colocado en un paciente de 13 años diagnosticado con el síndrome de Papillon-Lefevre. Se colocaron dos implantes dentales de titanio en la mandíbula de una dentadura retenida por implantes después de que el paciente se quejó de tener una prótesis inestable. Las radiografías de seguimiento demuestran una exitosa oseointegración y preservación del hueso alveolar un año después de la colocación del implante y el uso continuo de una prótesis dental funcional.

PALABRAS CLAVES: preservación del hueso alveolar, sobredentadura retenida con implantes, edentulismo temprano

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SINOPSE: registrou-se um relatório de caso de uma colocação de um implante odontológico em um paciente de 13 anos de idade diagnosticado com a Síndrome de Papillon-Lefevre. Dois implantes odontológicos de titânio foram colocados na mandíbula para uma dentadura fixada por implante após reclamação do paciente a respeito de instabilidade da prótese. Um ano após a colocação do implante e o desgaste contínuo de uma prótese odontológica funcional, as radiografias de acompanhamento exibiram osseointegração e preservação do osso alveolar bem sucedidas.

PALAVRAS-CHAVES: preservação óssea alveolar, sobredentadura fixada por implante, edentulismo prematuro

パピヨン・ルフェーブル症候群の若年患者におけるデンタルインプラント：症例報告

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概要：パピヨン・ルフェーブル症候群と診断された13歳の患者におけるデンタルインプラント設置の症例が報告される。患者の補綴不安定との訴えを受けて、インプラント支持デンチャーのために2本のチタンデンタルインプラントが下顎に設置された。インプラント設置と歯科補綴の機能的装着の1年後に継続管理のために撮影されたX線写真では、良好な骨統合と歯槽骨保存が認められた。

キーワード：歯槽骨保存、インプラント支持オーバーデンチャー、初期無歯症状

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