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ORAL SURGERY

CASE REPORT

Dental implantology and fibrous dysplasia: A 6-year follow-up case report and a literature review

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Abstract

Background and Aim: It is unclear if fibrous dysplasia (FD) represents a contraindication for implant borne rehabilitation, and only two successful cases are reported in the literature with a 4–2-year follow-up. The present paper discusses this issue reporting a full-arch maxillary rehabilitation with a >5-year follow-up.

Materials and Methods: A 79-year-old woman complained of a progressive asymmetry of the medial facial third with inter-arch occlusal alterations, reduced mandibular movements, upper right dental dislocation and mobility and toothache in the upper posterior jaw. The X-ray supported monostotic FD diagnosis was followed by a remodelling intervention in general anaesthesia, with the extraction of the hopeless teeth. After that and the failed rehabilitation with a removable prosthetic device, the patient underwent implant placement procedure for a fixed 'Toronto-bridge' prosthesis.

Results: After 6 years from implant loading, a posterior vestibular swelling of the affected maxilla was recorded, and one implant in 1.1 position had to be removed for peri-implantitis, without compromising the rehabilitation.

Conclusions: The acceptable results obtained in this case should promote the dental implantology practice in FD affected jaws, evaluating the proper clinical situations and the more adequate technological solutions.

KEYWORDS
case report, dental implants, fibrous dysplasia, maxilla

INTRODUCTION

The term fibro-osseous lesions (FOLs) categorize a group of bone disorders including neoplastic, dysplastic and developmental processes characterized by the replacement of normal bone with varying amounts of fibrous and immature mineralized tissue.[1,2] FOLs typically include ossifying fibroma (OF), cemento-osseous dysplasia (COD) and fibrous dysplasia (FD), sharing overlapping microscopic features.

Fibrous dysplasia represents 2.5% of all bone diseases and 7% of benign bone tumours. It is related to the mutation of the GNAS1 gene on chromosome 20q13, encoding the Gs alpha subunit of the heterotrimeric G protein complex. With the activation of adenylate cyclase and consequently increase in intracellular cyclic adenosine monophosphate, this genetic alteration determines an abnormal osteoblast differentiation with immature cellular and a stromal component of the bone.[1,3,4] The clinical seriousness of the disease depends on when the mutation occurred during embryonic development[5] and, as a consequence, the level of differentiation of the primary involved pluripotent cells with the affection of osteoblasts or fibroblast only, or melanocytes and endocrine

cells as well.[2] Polyostotic FD occurs in childhood and involves multiple bones, configuring the McCune–Albright syndrome in association with skin macules and precocious puberty. The monostotic form represents 70%–80% of the cases and involves the jawbones, particularly the posterior maxilla, often extending to the other cranial bones, configuring the 'craniofacial' form.[1]

The clinical scenario can be asymptomatic, with minimal medullary structure alteration, up to relevant deformities and problems related to the compression and dislocation of the organs and vascular–nervous structures inside the skeleton. Involving orbit, paranasal sinuses and the skull base can cause headaches, hearing and smelling loss and eye proptosis.[1,6] The typical radiologic 'ground-glass' or 'orange-peel' appearance, without a continuity solution with the healthy bone and no difference between spongious and cortical bone, reflects the histologic status: a fibrous stroma, with blood vessels and fusiform fibroblasts, associated with the immature woven bone are microscopically evident, with the so-called 'Chinese-letters' or 'alphabet soup' pattern. The therapy, consisting of bone remodelling in case of severe internal structures compression or deformity, should be delayed to a complete remission of the disease progression. With polyostotic and craniofacial FD, lesions cannot arrest even when osseous maturity is reached.[1,6,7] Moreover, a disease's reactivation in adults is possible.[7,8]

The alteration of the alveolar bone contour, dental resorption or dislocation, the anomalous inter-arch correlation, the dysplastic bone structure and its remodelling on time pose enormous difficulties in oral function rehabilitating for such patients. Dental implant borne fixed rehabilitation of FD affected patients is particularly challenging since the quality of the bone, too soft or sclerotic, can compromise the primary stability and osseointegration of the fixtures associated with the risk of osteomyelitis. Moreover, the persistence of an anomalous bone remodelling and the possibility of a disease reactivation can reduce the reliability of the prosthesis in time. Apart from a case report in 2008 with a 4-year follow-up,[9] no other cases have been published except for a recent case of posterior mandibular edentulism after 2 years of functional loading.[10]

The present paper reports a case of FD affecting a right upper jaw in a 79 old with a complete dental alteration, rehabilitated with fixed implant borne prosthesis and re-evaluated 6 years after functional loading.

CASE PRESENTATION

A 79-year-old woman was referred to Bologna's Oral and Maxillofacial Department (University of Bologna, Italy) to manage a suspected monostotic FD. The patient noted the onset and progressive slow increase in painless hard swelling of the right maxilla about 10 years before. No other bones were affected, without skin macules or endocrinologic disturbances. The medical consultation at that time, after a radiologic evaluation, expressed the diagnosis of a monostotic FD, and no therapeutic intervention was proposed. Since the scenario worsened, a new medical evaluation at our surgical division was required.

The patient complained about an evident asymmetry of the medial facial third with relevant masticatory problems and toothache in the upper posterior jaw.

These problems were due to evident hard tumefaction of the right maxillary alveolar process and inter-arch occlusal alteration, reduced mandibular movements, upper right dental dislocation and mobility. No systemic problems were referred, apart from controlled blood hypertension.

The extra-oral inspection recorded a hard swelling of the upper right labial and infra-orbit regions with difficulty in a relaxed closure of the labial rim and a downward lifting of the inferior contour of the right nostril (Figure 1). Intra-orally, an enlarged profile of the right alveolar process and a right hemi-palate swelling was evident; the teeth-borne full-arch fixed prosthesis had been removed a few weeks before to extract the #18 dental element by the dentist; the residual teeth stumps of the posterior and anterior maxilla appeared covered by the overgrowth of the alveolar bone and gingiva (Figure 2). The panoramic X-ray, taken before the full-arch fixed bridge removal (Figure 3) and CT (Figures 4 and 5), showed a radiopaque area involving the entire right upper alveolar process resulting in a thickened and irregular shape. The typical homogeneous ground-glass appearance with no distinction between cortical and spongious portions confirmed the monostotic FD diagnosis; the residual dental element appeared severely affected by periodontitis and care processes.

The maxillo-facial surgeon's team performed a first remodelling intervention in general anaesthesia, with the extraction of the teeth except for the upper left canine and second molar.

The clinical scenario improved with better mandibular excursion and enhanced function. In the meantime, about 2 months after the surgery, the patient came back asking for a fixed dental rehabilitation. Since no case of FD had been rehabilitated with implant insertion till then, considering the histologic and morphologic features of the dysplastic bone, the patient was informed about the risks of osseointegration failure and signed a specific, informed consent form before the intervention.

FIGURE 1 Inferior third of the patient's face.

FIGURE 2 Intra-oral frontal view before intervention.

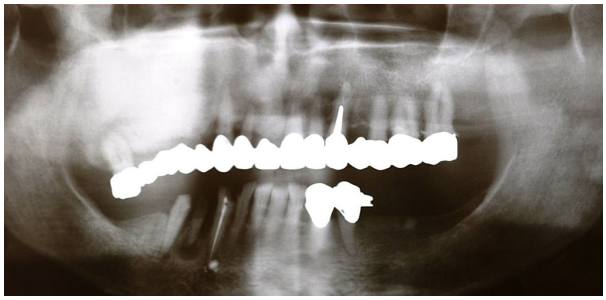

FIGURE 3 Pre-remodelling panoramic X-ray.

The implant surgery, performed in local anaesthesia, started with the raise of a full-thickness vestibular flap with a palatal para-crestal incision and the extraction of the root of the left canine. Seven implants were placed with the aid of a surgical guide, printed after a wax-up of the diagnostic models, for the initial drill osteotomy; the fixtures, 13/15 mm long and 3.8/4.5 mm large, were seated in #16, #14, #13, #11, #21, #24, #26 teeth location with a typical sequence of drilling under constant irrigation; the implant insertion torque was ever ≥30 N/cm without the necessity of under-preparation or tapering (Figure 6). The flap was repositioned and sutured over the fixtures, wholly submerged for 8 months. After a month of healing, the patient wore a denture only for aesthetic purposes for the subsequent 3 months, adequately relined to avoid dehiscence of the flap, and started progressively to chew soft food. A 'Toronto-Bridge' screw-retained prosthesis was connected to the fixtures after the healing screw and the abutments positioning. The mandible had been rehabilitated in the molar location with a partial dental prosthesis with a metallic framework supported by the residual teeth in the premolar and anterior area. (Figure 7). Our team controlled the patient monthly for the first 6 months. After that, the patient relied on her dentist for check-up appointments since she lived pretty away from our dental clinic.

After 6 years, the patient came back complaining about pain on palpation in the #11 site. No variation about her general health conditions was reported. According to the patient anamnesis, a significant vestibular overgrowth of hard tissue covered by normal-trophic oral mucosa was noted in the right upper jaw, slowly growing in the last 2 years; the prosthetic structure resulted overcome by the swelling with almost complete hiding of the artificial gingiva and the crowns (Figure 8). After the unscrewing of the prosthesis, the #11 position implant showed a >8 mm circumferential probing depth with bleeding; the endo-oral X-ray evidenced a loss of peri-implant bone >50% of the entire implant length (Figure 9). The #11 position's implant was easily removed and the prosthesis reconnected to the other fixtures without any alteration of the rehabilitation. No further remodelling was performed since the patient, 84 years old, expressed the will not to undergo any further surgical procedure. Meanwhile, the patient had been rehabilitated in the mandible with a removable denture for the loss of inferior teeth.

DISCUSSION

The osseous quality is considered crucial for the osseointegration of dental implants.[11] The literature reported about 80% of implant survival in irradiated and reconstructed jaw bone segments at long-term follow-ups,[12–14] and some information is available about implant results in medical-related osteonecrosis of the jaws.[15]

FD is a poorly known bone alteration, with a wide range of clinical manifestations, from syndromic systemic scenarios to minimal localization and different structural and radiological stadia of maturation. The dental implant placement for fixed rehabilitation could improve the oral functions in case of deformities, very difficult to manage with removable dentures for a low level of retention and stability.[9,16,17]

The jaws' most frequent fibro-osseous lesion is the cemento-osseous dysplasia (COD),[18] so-called for the affection of the alveolar bone only with closeness to the dental roots. This alteration can be sometimes histologically distinguished from FD for a globular distribution of the woven bone lamellae.[1,2,19] Periapical or focal COD, not associated with genetic alterations, is a self-limiting disease with rare deformities. The 'florid' variant presents extended and more critical scenarios with familiarity and specific chromosomic patterns. A few authors underlined the risks for implantology in COD affected patients for the too mineralized matrix of the dysplastic bone with a limited vascularization.[2,20] A total of seven fixtures were placed in jawbones affected by COD,[21–25] with the loss of two screws at 2[22] and 13[23] years of prosthetic loading. Two osteomyelitis with the sequestration of the peri-implant bone tissue were reported in two of the COD affected bone.[23,25]

Unlike COD, FD affects even the basal bone and is not circumscribable by the rest of the osseous contest. No particular problems were reported after dental extractions, during orthodontic treatments[17] and after orthognathic surgeries in

FIGURE 4 Multi-slice CT of the maxilla before surgery: Axial views.

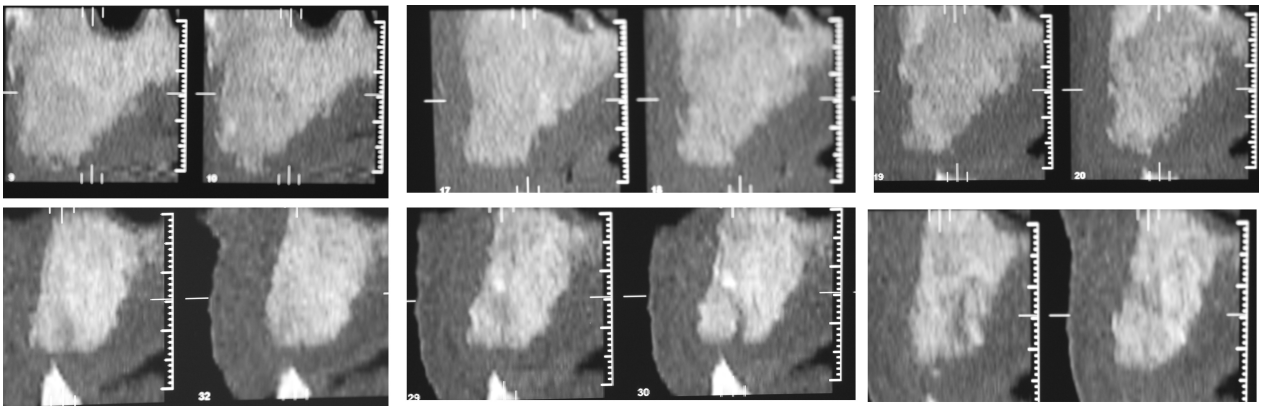

FIGURE 5 Multi-slice CT of the maxilla before surgery: Para-axial elaborations.

FD cases. Cheung et al.[26] found a not statistically inferior contact between the fixation screws and plates and dysplastic fibrous bone or normal one.

Bajwa et al.[9] first reported a bi-maxillary dental implant borne rehabilitation in a case of McCune–Albright syndrome, accomplished after 3 years of bone alterations' quiescence, with good results after 4 years of loading. Most recently, Adnot et al.[10] described a bilateral posterior mandibular fixed dental rehabilitation on implants, with one side affected by monostotic FD, in a 64-year-old patient. No difference between the two sides was found during the surgery and post-loading controls until the 2 years of follow-up. A different rationale was applied for the dental rehabilitation in another case, in 2014. Here the authors treated a craniofacial dysplasia with resection and free flaps reconstruction of the maxillary basal bone, zygoma and orbits and mandibular anterior portion; after the integration of the iliac crest flap in the dysplastic mandibular bone, dental implants were placed in the site of the graft to bear a fixed prosthesis, with a good 2-year follow-up outcomes.[3]

The titanium screws' osseointegration can depend on the status of maturation of the dysplastic tissue. According

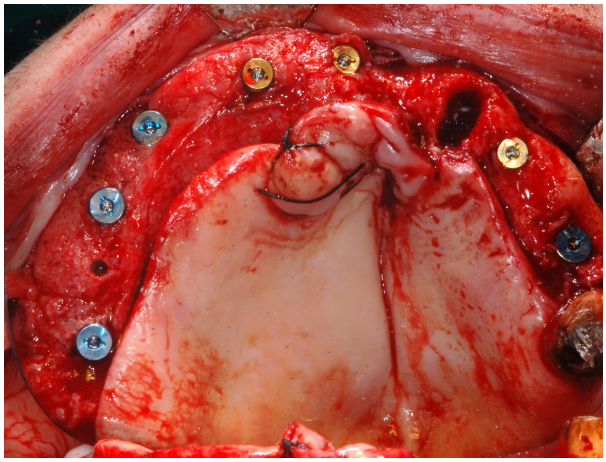

FIGURE 6 Intra-operative views at time of implant placement after the surgical remodelling.

FIGURE 7 Final view after the prosthetic connection.

FIGURE 8 Intra-oral view at 6-year follow-up.

to Panda et al.,[27] the structural evolution of the fibrous disease is radiographically evident as lucent zones, progressively becoming opaque through mixed appearances. Three main patterns were attributed to FD, the pagetoid, the cystic and the sclerotic. The sclerotic/ground-glass form permits good primary stability.[9,10] The monostotic FD, the most frequent form in craniofacial dysplasia, is more indicated for implant insertion since the quiescent variant represents almost all forms 50% of the cases, differently from the aggressive and

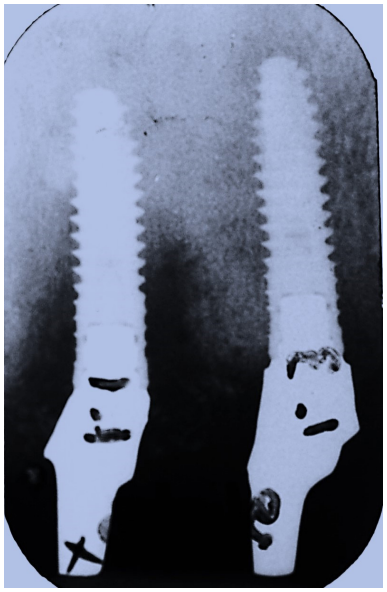

FIGURE 9 Intra-oral X-ray showing the loss of bone around the implant in #11 location.

slow-growing ones.[5] The quantitative ratio between fibrous and woven bone is quite regular in a lesion. This microscopic homogeneity, radiographically evident, enables the clinician to seat the implants in the correct prosthetic position without being conditioned by too immature radiolucent bone zones.

There is no sureness about the disease's reactivation, and new overgrowth can be observed even a long time after bone maturity.[2] A prevalence of an aggressive form might be possibly about 5%.[28] Eighteen per cent of reactivated cases were reported with the most extended follow-up of 9 years.[7] According to McDonald, most monostotic cases remain undetected for minimal clinical manifestations, becoming active or reactivated later.[7] The present case, with a typical craniofacial monostotic involvement of the mono-lateral maxilla, rendered in old age, presented a new over-growth 6 years after implant loading. This deformity compromised the aesthetic and the cleaning management of the prosthetic superstructure, but it would have been corrected simply with a new minimal bone remodelling approach if the patient had adhered to this proposed treatment. The peri-implantitis, responsible for one implant loss and considerable bone resorption around the closest fixture, involved normal bone without any correlation with the area of vestibular overgrowth. Careful bone drilling and abundant irrigation were recommended to prevent the bone tissue from necrosis and subsequent infection.[9,10] The >35 N/cm insertion torque reported by the other authors, and confirmed in the present case, is a good prerequisite for attaining the primary stability. Provided that the 'bone to implant contact' (BIC) is a requisite for dental implantology, the surface interaction between titanium with bone or other tissues needs major exploration.[11,23] The dynamic of peri-implant bone healing involves the resorption of the bone initially responsible for the primary stability, affected by the osteotomy-related trauma; after that, a pattern of intramembranous bone formation, maturation and remodelling renders.[11] Park et al.

removed an osteosclerotic sequestrum of cemento-osseous-dysplastic tissue perfectly embedded to a titanium fixture, with a high BIC value, after 16 years of functional loading.[23] This complication, associated with a minimal peri-implantitis in the coronal zone, could be explained, as considered above, by the low vascularity of the cemento-osseous dysplastic tissue. The dysplastic bone in FD appears as a less-mature bone with a good vascular supply and cellularity. In the present case, as long as possible implants were placed, waiting a healing period of 5 months before loading. The decision to unite the fixtures seated in ill and normal bone with a unique suprastructure made us more confident in a longer term result. The patient could have been better followed, but the old age with limited compliance of the patient and the distance from home prevent us from constant monitoring.

A few studies underlined the usefulness of further analyses, as histomorphometry or micro-CT, to better understand the characteristics of the jawbones[29,30] in terms of microarchitecture, density and cellularity. In particular, micro-CT is used to measure several variables, including bone volume, trabecular thickness, trabecular numbers and trabecular separation.[31] Hence, an investigation on a small biopsy before implant insertion could help identify the level of bone maturation, adapting the technique for implant bed preparation to the specific situation.

The advent of short implants with particular surfaces properties and a more rational acknowledgement of immediate loading could be considered in treating similar dysplastic hard tissues. Piezo-surgical devices could be helpful in reducing bone tissue damaging, favouring a less invasive implant seating.[32] With static and dynamic navigation systems, useful in the resection or remodelling phase,[33] the computerized technology can help customize the rehabilitation project according to the prosthetic demands and the bone structural features.[12]

CONCLUSIONS

This case report would promote the dental implantology practice in FD affected jaws, considering the similarity of the bone with the normal osseous tissue in terms of density, cellularity and process of remodelling and the new technology options available today. The treated patient did not suffer any complications related to implant placement, and prosthetically loaded, apart from peri-implantitis with the loss of a fixture, up to 6 years of follow-up.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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