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Fibrous Dysplasia in the Mandible with Class III Malocclusion: Surgical and Orthognathic Management Combined with Bone Graft and Surgical Dental Implants Placements: A Case Report

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1. Abstract

Fibrous dysplasia is a non-neoplastic developmental progression that disturb the craniofacial bones, identified by painless enlargement as a result of bone exchanged by abnormal fibrous tissue. Treating such conditions involves surgical, and implant-prosthetic rehabilitation is often necessary and can result in cosmetic, functional, and psychological impairment that greatly affects the patient's quality of life. This is a report of a 25-year-old man affected by fibrous dysplasia and class III malocclusion who required rehabilitation that was quite challenging. A multidisciplinary team was necessary for comprehensive care and optimal post-with proper excision of skeletal disorder. Later, the patient underwent reconstruction associated with orthognathic surgery, bone grafting to obtain the correct bone volume, and three dental implants inserted on the right side of the posterior mandible to support prosthetic restorations with reliable and logical treatment solutions

2. Introduction

Fibrous dysplasia (FD) is a skeletal disorder distinguished by the massive propagation of fibrous tissue in bone marrow, leading to osteolytic lesions, fractures, and deformations [1]. It may be divided into three categories: monostotic (74%), polyostotic (13%), and craniofacial (13%) [2]. Monostotic involves a single bone; polyostotic, having multiple lesions involving multiple bones; and McCune Albright syndrome, a polyostotic form of fibrous dysplasia that involves endocrine abnormalities. Malignant degeneration occurs in less than 1% of causes of fibrous dysplasia. Malignancies are almost exclusively osteosarcoma. Pain, the rapid growth of a lesion, and dramatic elevation of alkaline phosphatase may herald malignant transformation Treatment of mandibular tumors may include defects from surgical resection of the mandible, tongue, floor of mouth and associated structures [3]. Based on the amount of resection or extent of bone loss, mandibular defects can be classified as continuity and discontinuity defects [4]. Loss of mandibular continuity if not re-established alters the symmetry of the mandible, leading to altered mandibular movement and deviation of the residual mandible towards the affected side [5]. A free flap is an autogenous vascularized transplant, which involves the harvesting and detachment of bony and muscle tissue with its blood, and nerve supply and re-establishment by anastomosis to suitable recipient site vessels [6, 7]. Many treatment approaches can be found in the literature regarding orthopedic, orthodontic treatment or even surgery for class III malocclusions which is characterized by a variety of skeletal and dental components, including a large or protrusive mandible, retrusive maxilla, protrusive mandibular dentition, retrusive maxillary dentition, and combinations of these

components [8] Normal occlusion and improved facial esthetics of skeletal class III malocclusion can be achieved by growth modification [9], orthodontic camouflage, or orthognathic surgery [10]. The age of the patient, the severity of the malocclusion, the patient's chief complaint, clinical examinations, and cephalometric analysis will delineate the treatment of choice [11]. Rehabilitation of such patients is quite challenging and requires a multidisciplinary team for comprehensive care and optimal post-treatment functional outcomes [12–17]. Implants placed in the free flap reconstructed bone perform the same as those placed in native bone [16, 17].

The present case illustrates fibrous dysplasia in the mandible with class III malocclusion, managed with reliable and logical treatment solutions.

3. Case Presentation

A 25-year-old gentleman presented to our dental department complaining from tiredness associated with mild and transient pain, and forwardly placed lower front teeth, and difficulty in chewing. The patient is not aware of any medical problems or allergies, has never been hospitalized, and takes no medications. There was no family history of the patients following the findings.

A general physical examination revealed an asthenic body type with satisfactory vital signs. Clinical extra-oral examination revealed that the patient has a symmetrical face with an asymmetrical chin, prognathic mandible, convex facial profile, and increased lower facial height. Minimal incisal shows upon smiling with the lower incisors showing more, since they are in an anterior relationship to the upper incisor. Intra-oral examination revealed a class III molar and canine relationship bilaterally, anterior crossbite with a reverse overjet and overbite about -3 and -2 respectively (Figure 1). Preoperative panoramic radiograph shows a radiolucent lesion measuring around 1.5 X 2 cm related to teeth number 36 and 37. The lesion was round with no sclerotic borders causing root resorption of adjacent teeth (Figure 2). The teeth clinically had no periodontal problems and were not tender to percussion. There is no adjacent abnormal lymph node enlargement in the neck.

The patient signed a written informed consent form after the case presentation, treatment planning, and before the treatments.

Figure 1: Pretreatment frontal view revealed that the patient has anterior cross bite, a reverse overjet and overbite about -3 mm and -2 mm respectively.

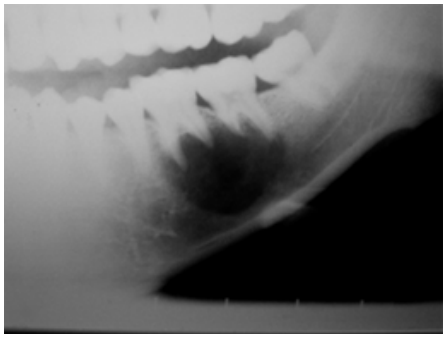


Figure 2: Preoperative panoramic radiograph showing a round radiolucent lesion with no sclerotic borders causing root resorption of adjacent teeth.

A decision was made to schedule the patient for a fine needle aspiration (FNA) biopsy. Cytology report stated that the smear shows hyper-cellularity with foamy macrophages lymphocytes and polymorph. No dysplastic cells are seen. Consistent with a benign tumor. At this point, the patient was scheduled for excisional biopsy in which the left side of the mandible was resected under general anesthesia. During the surgery the buccal mucoperiosteal flap was elevated buccally with distal release and the lingual mucoperiosteal flap was also elevated. Surgical removal of tooth numbers 36 and 37 was done and the lesion was identified. The entire lingual cortex was resorbed and the tumor had penetrated through the buccal cortex. The lesion was sent for a frozen section and was reported to be a sarcomatous lesion with very high cellularity and atypical cells. Which is extremely aggressive in nature. Multiple frozen sections were sent to the histopathologist, and he advised a resection of the area with at least 1 cm of safety margins anteriorly and posteriorly. In view of the aggressive nature of the tumor, it was decided to do a resection of the affected segment with a 1 cm margin. It was also decided to obtain a full clearance of the medial and lateral tissue. The segment of the mandible was resected anteriorly to tooth number 35 and distal to tooth number 38. The inferior alveolar neurovascular bundle was ligated and sacrificed. Frozen sections were sent again, and margins were found to be clear. A 9-hole reconstruction plate was placed in situ and secured with screws (Figure 3). A second plate was placed above the original plate and was fixed with eight screws of about 10mm in length (Figure 3). The patient had a class III occlusion preoperatively and the same occlusion was obtained postoperatively. The patient was followed up regularly and had an uneventful recovery. After a period of 4 months, the patient went for his second surgery which was a mandibular reconstruction with an autogenous graft harvested from the right iliac crest. During this phase of treatment, the patient underwent an orthognathic workup. Impressions were taken and upper and lower study casts were mounted in a semi-adjustable articulator before mock surgery was performed on the articulated model, as illustrated from the left (Figure 4a) and right sides (Figure 4b). Mock surgery was performed on the articulated model

to construct a stage surgical splint, as illustrated from left (Figure 5a) and right sides (Figure 5b). Extra oral submandibular approach was used associated with intraoral approach graft was about 3.5 x 2cm and was secured using the preexisting mandibular reconstruction plate using three 12mm screws. Drains were placed and closure was done in both the donor site and surgical site in layers. The patient was followed up periodically, healing was excellent with no complications.

The teeth were leveled and aligned to achieve dentoalveolar decompensation, and 0.019X0.025 Stainless Steel wire was placed in both arches. Facebow transfer was done with a slide metric facebow and the orientation of the maxilla in relation to the cranial base was recorded and transferred to a semi-adjustable articulator using a mounting Jig. The Mandible cast was articulated with the maxillary cast using an occlusal wax bite record. The surgery took place about 1 year after the previous procedure which involved Bilateral sagittal split osteotomy (BSSO) bone grafting from the left iliac crest to provide more vertical and horizontal bone for implants. During this procedure, the BSSO went down with no complications and with minimal modifications. Removal of the reconstruction plates was essential for the mandibular setback and was achieved using a submandibular transcutaneous approach. The occlusion was good and found to sit passively with no interference. The surgical acrylic splint was used for the intermaxillary fixation and the mandible was fixed using a Lei-binger plating system three 12mm lag screws were used on the right while another three on the left were secured furthermore with a 2.0 plate (Figure 6). Attention was fulfilled to harvest the bone from the iliac crest and graft it in the defected aspect of the mandibular alveolus. after 6 months, a mandible surgical stent with metal referring points was fabricated to determine the position of the implants (Figure 7), A panoramic radiograph was taken with the surgical stent for radiographic evaluation during treatment planning for implant placement and during surgical procedures to locate optimal implant placement sites (Figure 8). Three 12-mm-long Straumann dental implants were placed in the area of the lower left 2nd premolar with a diameter of 4.1 mm, and 1st molar, and 2nd molar with a diameter of 4.8 mm respectively (Figure 9). Laboratory procedures as well as fabricated solid abutments (Figure 10), were inserted on the implants, radiographs were taken for demonstrating the marginal bone level and the three single fabricated abutments were secured to the implants in fully seated positions (Figure 11). Delivery of the final restorations included confirmation that proximal contacts allowed the patient to perform normal oral hygiene procedures using dental floss and the occlusal scheme for all restorations was evaluated to ensure a firm-centric contact. A postoperative photograph (Figure 12) and radiograph (Figure 13) were taken after cementation to confirm the seating of the 3 implants supported restorations at the time of delivery. Oral hygiene instructions were reinforced, and the importance of periodic recall visits was emphasized. At the checkup after prosthesis insertion, the patient is satisfied and comfortable with the treatment modalities and no longer expressed any concerns regarding chewing, oral hygiene situation, and pain in the temporomandibular joint. The patient reports an improvement in functional occlusion.

Two years post-loading, CT scans and a three-dimensional study were performed, confirming the amount of bone around implants at the area of the lower left 2nd premolar (Figure 14a), 1st molar (Figure 14b), and 2nd molar (Figure 14c).

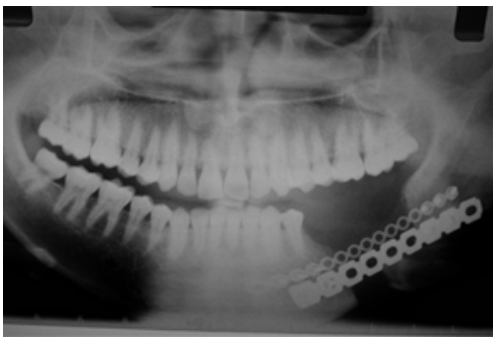
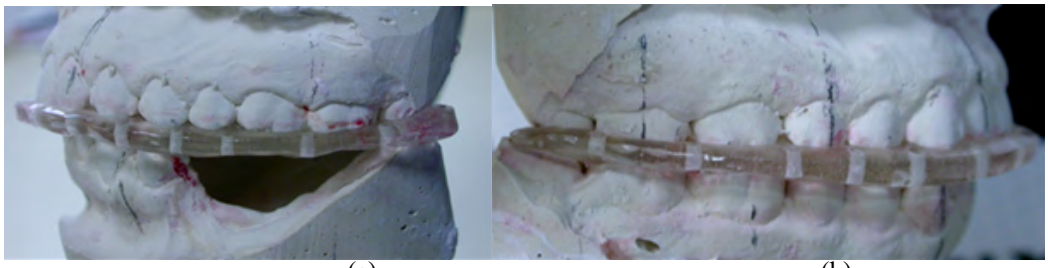


Figure 3: Panoramic radiograph showing segment of the mandible was resected anteriorly with two plates was placed in situ and secured with screws secured.

(a) (b)

Figure 4: Articulation for the maxillary and mandibular casts from both sides before mock surgery was performed. (a) Left side. (b) Right side.



(a) (b)

Figure 5: Mock surgery was performed on the articulated models and a stage surgical splint was fabricated from both sides. (a) Left side. (b) right side.

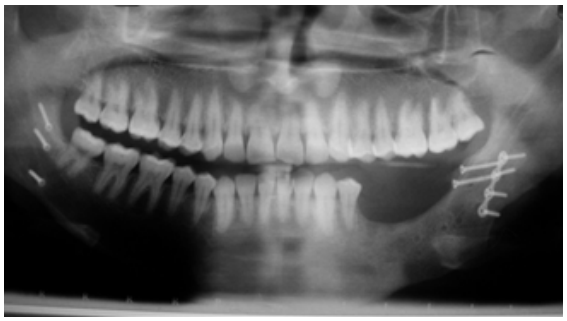


Figure 6: Panoramic radiograph showing 2nd surgery after orthognathic surgery and bilateral sagittal split osteotomy (BSSO) bone grafting from the iliac crest to provide more vertical and horizontal bone in the left side of the mandible which was fixed.

Figure 7: Mandible surgical stent with metal referring points to determine the position of the implants.

Figure 8: Panoramic radiograph was taken with the surgical stent for radiographic evaluation during treatment planning for implant placement and during surgical procedures to locate optimal implant placement sites.

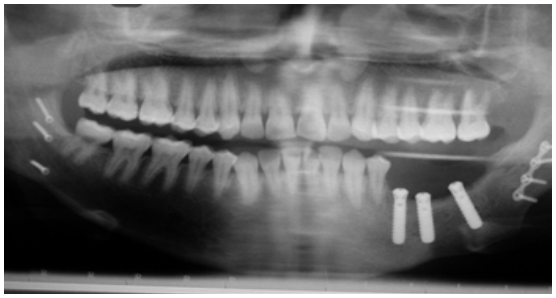


Figure 9: Panoramic radiograph demonstrating three dental implants were placed in the area of lower left 2nd premolar, 1st molar, and 2nd molar respectively.

Figure 10: Laboratory procedures for the fabricated solid abutments.

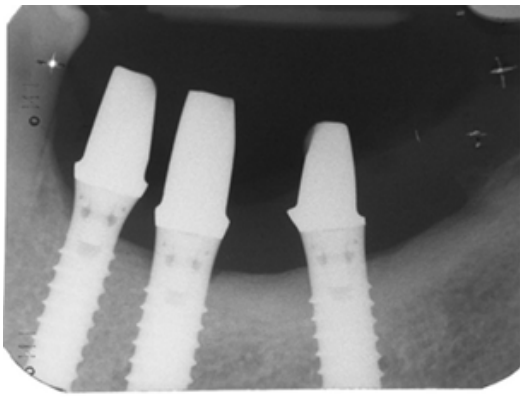


Figure 11: Radiograph demonstrating the marginal bone level and the three single fabricated abutments were secured to the implants in fully seated positions.

Figure 12: postoperative photograph showing the prosthesis after cementation to confirm the seating of the three implants supported restorations at the time of delivery.

Figure 13: Postoperative panoramic radiograph showing cemented crowns, the good quality and quantity of the bone, and marginal bone level for the three dental implants areas.

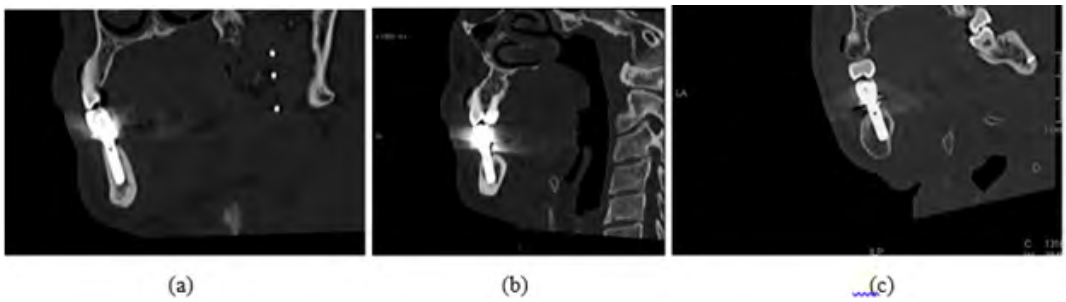


Figure 14: Cross-sectional CT scans showing supporting bone around the three implants 2 years post-loading. (a) Lower left 2nd premolar. (b) Lower left 1st molar. (c) Lower left 2nd molar.

4. Discussion

Current reconstructive techniques for fibrous dysplasia allow for achieving adequate aesthetic and functional results. Position the plate back onto the mandible in the exact position and will be used as temporarily bridge continuity defect without bone graft, but only with pending secondary reconstruction [18]. Plate fracture is possible when a plate bears the entire functional load for an extended period [19]. Implantation of bone graft, immediately or at a later stage, is necessary to support the construct [20]. In this study, the autogenous graft harvested from the right iliac crest was used in designing bone grafts for mandibular reconstruction followed by dental implantation [21].

The patient in this case also required orthognathic surgery to correct a concurrent malocclusion or facial/dental canting [22]. There is no documented contraindication for orthognathic surgery as long as the lesions are quiescent. Bone healing appears to be normal with conventional rigid fixation [22]. Regular follow-up with the surgeon is necessary to determine that there is no recurrence or further deformity.

Amit Mendiratta et al in 2013 reported in severe cases of class III malocclusion that, orthodontics alone is not possible, and an ortho-surgical approach becomes inevitable to improve occlusion, masticatory function, and more importantly aesthetic and facial balance [23]. The addition of the occlusal extension to the stent provided the surgeons with a reliable and consistent way to position and achieve the guide so that the reduction would mirror the surgical plan. The clinical success for the patient in this case report after orthognathic therapy can be defined as a combination of the following factors: patient satisfaction, correct static and functional occlusion, patient comfort, chewing, absence of pain in the temporomandibular joint and stability of the patient's status. However, isolated surgery of the mandible for prognathic lower jaw has long been the most applied procedure for Class III correction [24].

In 2014, Petrocelli and Kretchmer reported conservative treatment and implant rehabilitation of the mandible in a case of craniofacial fibrous dysplasia. The authors presented a case of implant rehabilitation of the mandible in a young patient affected by craniofacial fibrous dysplasia [25]. In this clinical case, surgical implant placements were performed in the bone-grafted area and restored to obtain excellent functional and aesthetic results.

Several factors influence implant survival, especially when the patient undergoes surgical removal of the fibrous dysplasia, and the mandible is resected. Without a doubt, the experience of the surgeon, bone quality, and technical aspects such as implant length, diameter, and primary stability each plays essential roles [26, 27]. As involved in the methods for this case, we used Buser et al's strict parameters for evaluating implant system success and long-term clinical trial results [28].

5. Conclusions

This clinical case with its natural progression, the components of the diagnostic evaluation, and the multi-disciplinary management and approach could be the best way to manage difficult cases of fibrous dysplasia in the mandible with class III malocclusion combined with orthognathic, bone graft, and implant-prosthetic rehabilitation. The combination of these benefits also provided the patient with a more thorough understanding of the nature and goals of the procedure, thereby improving the process of informed consent. Ultimately, this leads to increased satisfaction with the final functional and aesthetic result, which will significantly impact the patient's quality of life.

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