Microvascularized Fibular Bone Grafting for the Treatment of Mandibular Expansive Osseous Dysplasia - A Case Report

Camila de Oliveira Barbeiro, Mario Henrique Arruda Verzola[1], Roberto Henrique Barbeiro, Wilson Tadao Tachibana[2], Jorge Esquiche León[3], Andreia Bufalino

Department of Diagnosis and Surgery, São Paulo State University (UNESP), School of Dentistry, Araraquara, [1]Department of Surgery, Central Paulista University Center – UNICEP, [2]Orthopedics and Traumatology Surgeon, Private Practice, São Carlos, [3]Department of Stomatology, Public Oral Health and Forensic Dentistry, Ribeirão Preto Dental School, University of São Paulo (FORP/USP), Ribeirão Preto, SP, Brazil

Abstract

Rationale: Osseous dysplasia (OD) is a benign fibro-osseous lesion classified as periapical, focal or florid with some rare cases being diagnosed as expansive OD. **Patient Concerns:** A 43-year-old female presented with gross mandible expansion and tooth displacement. **Diagnosis:** Imaging scans revealed an expansive lesion in the anterior mandible, with varying opacity in the central region, and other smaller lesions in the region of teeth #37 and #47, consistent with expansive OD. **Treatment:** Surgical resection followed by immediate reconstruction of the mandibular defect using a microvascularized fibular graft. **Outcomes:** The patient had a 4-year follow-up, with adequate mandibular bone continuity, mastication, swallowing, and speaking ability reestablished. **Take-away Lessons:** Immediate reconstruction after large surgical resection is required as tissues retract over time, hampering late reconstructions. Microvascularized fibular graft aims at adequate and functional rehabilitation with osseointegrated implants and long-term follow-up is needed as florid OD may evolve into expansive OD.

Keywords: Bone dysplasias, case report, florid cemento-osseous dysplasia, mandibular reconstruction, vascular grafting

Introduction

Osseous dysplasia (OD) is a benign fibro-osseous lesion that affects tooth-bearing areas, being classified as focal, periapical, or florid. Florid OD involves more than one quadrant of the jaw and is more common in middle-aged black women.[1,2] Usually asymptomatic, OD is often discovered through routine imaging exams. On the other hand, expansive variants of OD require surgical resection due to their significant impact on jaw structure and function.[3,4] Cosmetic recontouring alone is ineffective, leaving a substantial defect.[3-6] This case report describes a case of expansive OD treated by surgical resection and microvascularized fibular bone grafting.

Case Report

A 43-year-old Afro-Brazilian female presented with mandibular enlargement and altered tooth position, having been diagnosed with florid OD a decade earlier [Figure 1]. Extraoral examination confirmed significant enlargement of the mandible. Intraoral examination revealed an asymptomatic, expansive lesion in the anterior mandible with firm consistency upon palpation, involving buccal and lingual mucosa and causing tooth displacement. Cone-beam computed tomography (CBCT) imaging showed a predominantly hypodense lesion with hyperdense areas, located from the distal region of tooth #35 to the distal region of tooth #45. Resorption and displacement of the teeth, as well as expansion of the buccal and lingual cortical plates, were observed. Two smaller lesions were identified at the level of teeth #38 and #48. After incisional biopsy, histopathological analysis revealed fusocellular mesenchymal proliferation with numerous foci of irregular calcification. The clinical, microscopical and radiographic findings were

Access this article online

Quick Response Code:

Website: https://journals.lww.com/aoms

DOI: 10.4103/ams.ams_63_23

Address for correspondence: Dr. Andreia Bufalino, Rua Humaitá, 1680, Araraquara, São Paulo 14801-903, Brazil. E-mail: andreia.bufalino@unesp.br

Received: 01-04-2023 **Last Revised:** 02-10-2023
Accepted: 17-10-2023 **Published:** 19-12-2023

How to cite this article: Barbeiro CO, Verzola MH, Barbeiro RH, Tachibana WT, León JE, Bufalino A. Microvascularized fibular bone grafting for the treatment of mandibular expansive osseous dysplasia - A case report. Ann Maxillofac Surg 2023;13:236-9.

Figure 1: Initial panoramic radiograph with multiquadrant radiolucent lesions, radiopaque halo around tooth 37 and near tooth 34. Radiolucent lesion with radiopaque areas near teeth 11, 21, 22 and 23

consistent with the diagnosis of expansive OD [Figure 2]. Despite its benign nature, it is associated with significant morbidity due to its expansive behavior even after biopsy. Thus, surgical resection via partial mandibulectomy followed by microvascularized fibular bone grafting was planned with a multidisciplinary team. Surgical planning was carefully explained to the patient so that she was aware of the risks and benefits of the procedure.

Surgical approach

Based on the CBCT scan, a three-dimensional (3D) prototype of the mandible was created to model the reconstruction plate (2.4 mm Locking-NEOORTO) and determine the mandible cutting sites. Surgical approach to the mandible was performed by transcervical access, with a focus on preserving the facial artery and jugular veins. The fibula was accessed by a mid-lateral incision over the fibula. Skin flaps were elevated in a suprafascial plane, the posterior intermuscular septum was identified between peroneus longus and soleus, the common peroneal nerve was identified and preserved, and the peroneal muscles were dissected off the anterior surface of the fibula, reflecting the peroneus longus and brevis muscles anteriorly. The anterior intermuscular septum was incised to gain access to the anterior compartment, and the extensor digitorum and extensor hallucis longus muscles were dissected off the fibula until the interosseous membrane was identified. A 3 mm cuff of lateral soleus, flexor hallucis longus, and tibialis posterior muscle were elevated with the fibula to preserve the periosteal and nutrient bone circulation. The peroneal vessels were identified proximally and distally and isolated, ligated distally, and protected proximally as the osteotomy of the fibula was performed with a Gigli. Further osteotomies and fixation to the pre-modeled plate were performed before the peroneal vessels were ligated proximally. After preparation of the graft, the peroneal vessels were marked and ligated with microvascular clamps.[7] The set plate, bone tissue, periosteum, and vessels were transferred to the recipient site of the mandible and properly fixed to the proximal stumps bilaterally, allowing stability. An end-to-end arterial anastomosis was performed on the facial artery and an end-to-end venous anastomosis was performed at a tributary vein of the external jugular vein.

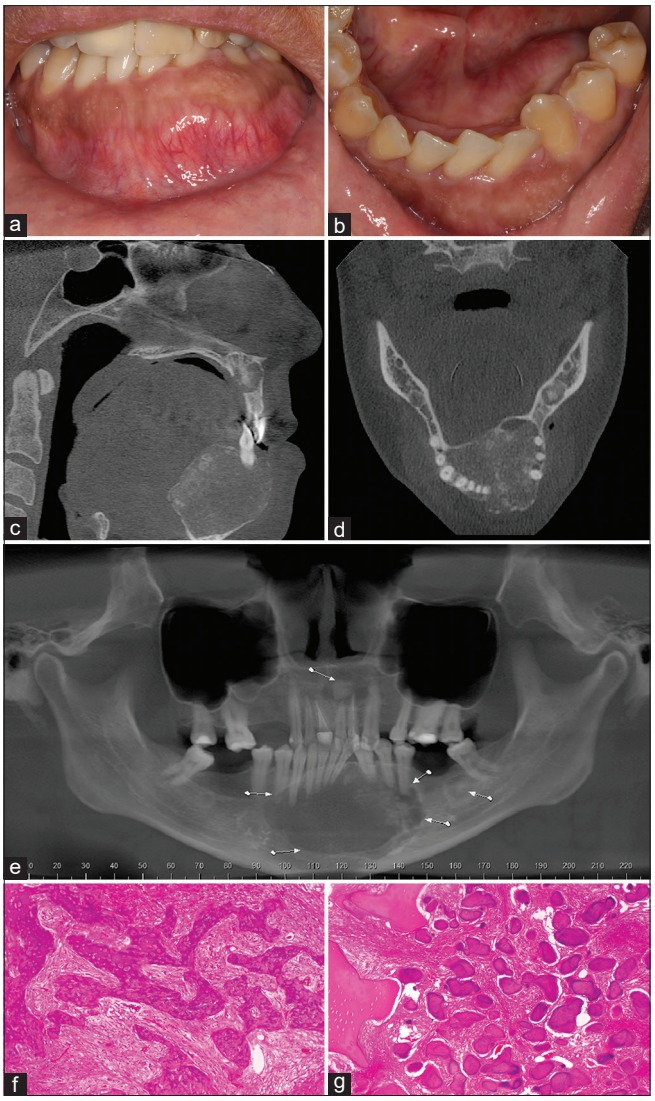

Figure 2: (a) Intraoral expansive lesion, (b) Buccal and lingual mucosal involvement and teeth displacement, (c) Sagittal reconstruction, (d) Coronal reconstruction, (e) Panoramic reconstruction of hypodense lesion with hyperdense areas from tooth #35 to #45, and two lesions in the mandibular body near teeth #38 and #48, (f) Irregularly shaped trabeculae of medullary bone in a fibrous stroma, (g) Psammomatoid bodies in fibroblastic stroma (H and E, ×10)

Figure 3 shows all the surgical procedures. The surgical sites were properly sutured and the patient was transferred to the intensive care unit while still intubated through a tracheostomy. After a 4-year follow-up, the patient showed no recurrence and exhibited adequate mandibular bone continuity, along with the restoration of oral functions, enabling rehabilitation planning with dental implants [Figure 4].

Discussion

OD is a benign fibro-osseous lesion that affects tooth-bearing areas of jaws in middle-aged black women. OD can be classified as focal, periapical, or florid based on its anatomical location. This condition is usually asymptomatic and is therefore

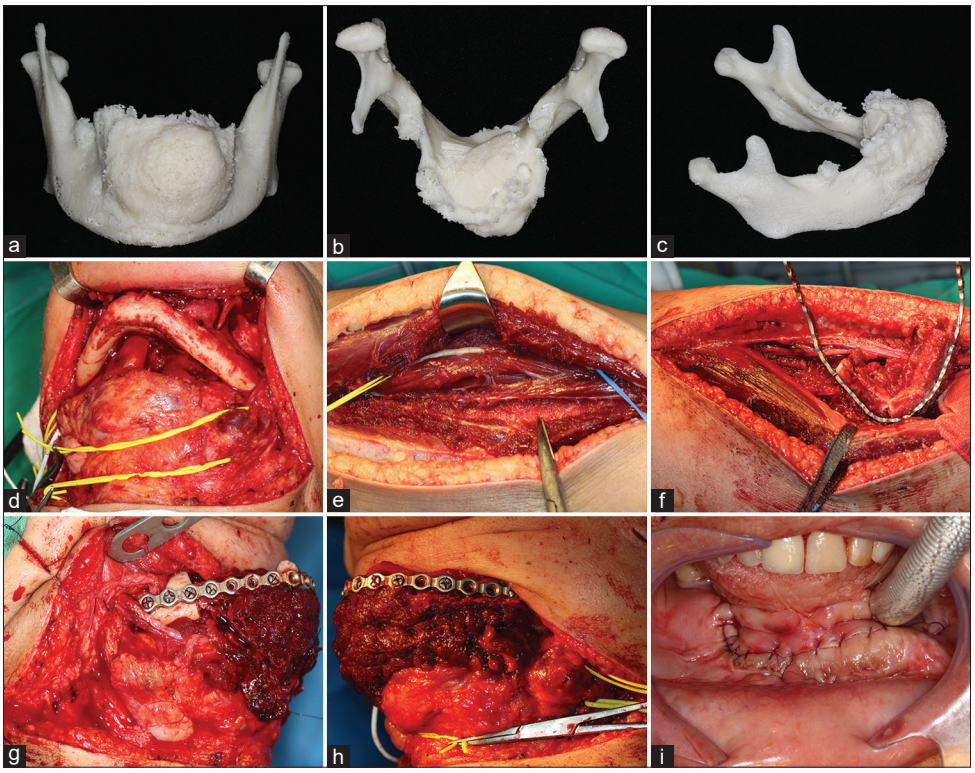

Figure 3: Surgical procedures. (a-c) Previous prototyping, (d) Transcervical access with special attention to the facial artery and jugular veins, (e) Mid-lateral incision over the fibula, (f) Fixation of the fibula to the pre-modeled plate, (g and h) Placement of the reconstruction plate, bone tissue, periosteum, and vessels in the mandible recipient site, (i) Immediate postoperative aspect

discovered on routine imaging exams, except when there is a secondary infection or expansion.[1,2] Radiographically, the lesion may be radiolucent, mixed radiolucent and radiopaque, or completely radiopaque in different stages. Histologically, OD reveals unencapsulated and fragmented lesions that show a fibrous stroma with variable cellularity and loose collagen, as well as mineralizing tissue composed of osteoid, bone, and cementum-like material.[1-6] Therefore, correlation with clinical and radiographic information is essential for the final diagnosis.

Expansive OD affects frequently the anterior mandible, whereas maxilla affectation is uncommon (25% of cases). This condition usually causes facial deformity due to their persistent growth that causes expansion of the cortical bones.[3-6] In these cases, cosmetic recontouring is not adequate, and surgical resection of the lesion is required. It is worth highlighting that expansive OD can share some clinical, radiographic, and histologic findings with a florid OD subset, which should be carefully evaluated to differentiate these conditions. This case emphasizes the need for long-term follow-up of patients diagnosed with florid OD, given the possibility of progression to expansive OD and the requirement for reconstructive surgery.

Large lesions usually require extensive removal of the involved bone tissue, leaving a significant bone defect. These sites usually do not have good regenerative potential because critical defects tend to repair themselves by fibrous connection. Thus, autogenous bone grafts removed from extraoral sites have been proposed for the treatment of this type of defect. This biomaterial remains the gold standard for reconstructions and subsequent rehabilitation with oral implants, although some limitations such as non-integration of the bone graft in the recipient area may occur due to the devitalization of part of the graft.[5,6,8] In this regard, using microvascularized bone grafts, such as the microvascularized fibular graft, increases the chance of bone stump healing, and decreases the rate of bone resorption and fracture when compared to non-vascularized bone grafts.[8]

Recognizing the limited vascularization in cases of extensive bone defects, microvascularized grafts have emerged as the preferred option, despite the challenges in graft size and configuration. Reconstruction of large bone defects utilizing microvascularized fibular graft usually provides solid and reliable bone continuity leading to a framework suitable for implantating osseointegrated implants to address aesthetic and functional issues. Improvements in microvascularized fibular graft reconstruction can be achieved through precise surgical techniques, advanced technology, and personalized patient-specific planning, including the use of 3DCT scans for graft customization and the use of prototypes. Once adequate bone continuity has been achieved with the graft, the patient can be rehabilitated with osseointegrated implants and a Misch FP3 fixed prosthesis after approximately six months postoperatively[9,10]

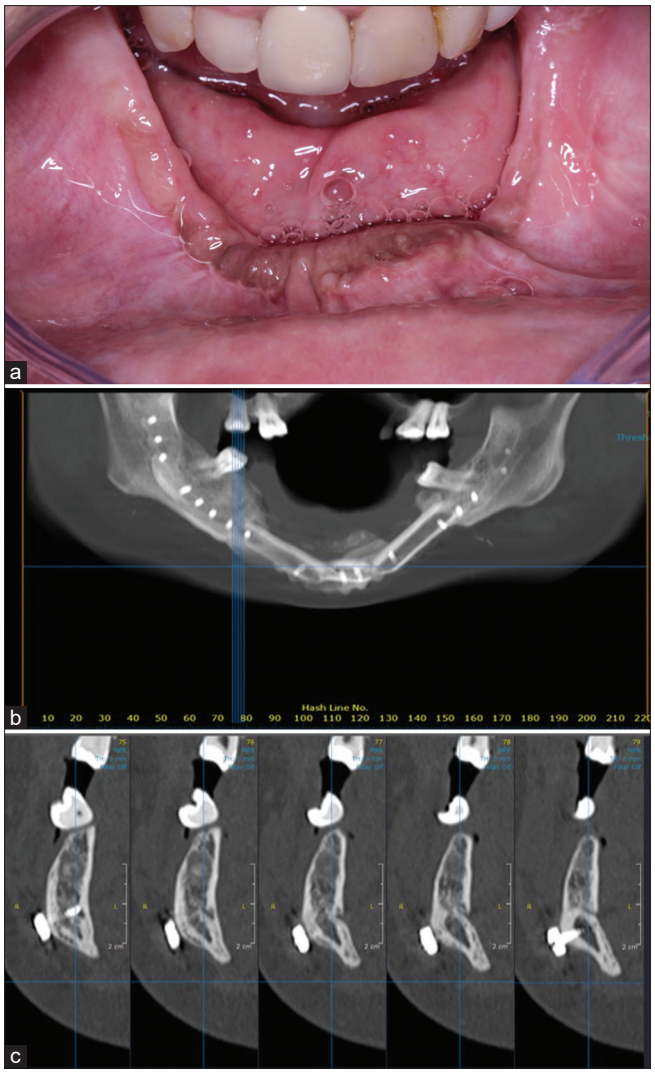

Figure 4: Follow-up at 4 years. (a) Clinical aspect, (b) Panoramic reconstruction, (c) Cross-sectional

Conclusion

The diagnosis of benign fibro-osseous lesions usually does not require microscopic investigations, but expansive lesions demand complete surgical removal. Patients diagnosed with florid OD may evolve into expansive OD, thus long-term follow-up is necessary. This case demonstrates that a well-planned surgical excision followed by a microvascularized fibular graft can restore adequate function and aesthetics. Microvascularized autograft from fibula is an adequate option for large bone defects arising from benign lesions and may allow for the installation of osseointegrated implants in the future.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Acknowledgments

We wish to acknowledge Dr. André Luiz Luchini Predin, general and oncologic surgeon, for being part of the surgical team and helping in the surgical procedures. He contributed in an essential way to the multidisciplinary team for the success of the case presented here.

Financial support and sponsorship

This study was financed in part by the Coordenação de Aperfeiçoamento de Pessoal de Nível Superior - Brasil (CAPES) - Finance Code 001.

Conflicts of interest

There are no conflicts of interest.

References

1. El Naggar AK, Chan JC, Grandis JR, Takata T, Slootweg PJ. WHO Classification of Head and Neck Tumors. 4th ed. Lyon: IARC Press; 2017.
2. Fenerty S, Shaw W, Verma R, Syed AB, Kuklani R, Yang J, *et al*. Florid cemento-osseous dysplasia: Review of an uncommon fibro-osseous lesion of the jaw with important clinical implications. Skeletal Radiol 2017;46:581-90.
3. Prasad C, Kumar KA, Balaji J, Arulmozhi M, Jayanandhini S, Priyadharshini R. A family of familial gigantiform cementoma: Clinical study. J Maxillofac Oral Surg 2022;21:44-50.
4. Noffke CE, Raubenheimer EJ. Expansive osseous dysplasia: Report of 9 lesions in an African population sample and a review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111:e35-41.
5. Raubenheimer EJ, Noffke CE, Boy SC. Osseous dysplasia with gross jaw expansion: A review of 18 lesions. Head Neck Pathol 2016;10:437-43.
6. Nel C, Yakoob Z, Schouwstra CM, van Heerden WF. Familial florid cemento-osseous dysplasia: A report of three cases and review of the literature. Dentomaxillofac Radiol 2021;50:20190486.
7. Wei FC, Mardini S. Flaps and Reconstructive Surgery. 2nd ed. London: Elsevier Health Sciences; 2016.
8. Chen J, Yin P, Li N, Wu L, Jian X, Jiang C. Functional mandibular reconstruction with double-barrel fibular flap and primary osseointegrated dental implants improve facial esthetic outcome. J Oral Maxillofac Surg 2019;77:218-25.
9. Manju V, Krishnapriya VN, Babu AS, Krishnadas A, Subash P, Iyer S. Prosthetic rehabilitation options in post-ablative maxillomandibular microvascular reconstructions. J Maxillofac Oral Surg 2023;22:10-9.
10. Urban IA, Montero E, Amerio E, Palombo D, Monje A. Techniques on vertical ridge augmentation: Indications and effectiveness. Periodontol 2000 2023. p. 1-30. [doi: 10.1111/prd.12471].