Original Article
pISSN 2383-5389 / eISSN 2383-8116
Journal of International Society for Simulation Surgery 2016;3(2):60-63
https://doi.org/10.18204/JISSiS.2016.3.2.060

ORCID
Dong-Young Kim: orcid.org/0000-0002-2772-2519
Myoung-Sang You: orcid.org/0000-0002-9285-0189
Kang-Min Ahn: orcid.org/0000-0003-1215-5643

Maxillo-mandibular Contouring Surgery in Monostotic Fibrous Dysplasia Patients using Simulation Surgery

Dong-Young Kim, D.D.S., Myoung-Sang You, D.D.S., M.S.D., Kang-Min Ahn, D.D.S., M.S.D., Ph.D

Department of Oral and Maxillofacial Surgery, College of Medicine, University of Ulsan, Asan Medical Center, Seoul, Korea

Fibrous dysplasia is quite a rare disease usually involving maxilla and mandible. Because of its benign clinical course, conservative contouring surgery has been recommended for facial deformity. 3D rapid prototype (RP) model gives a lot of informations before operation such as depth of drilling, area of resection and important anatomic structure. The purpose of this study was to report maxilla-mandibular contouring surgery in fibrous dysplasia patients. A total of 14 consecutive patients were included for surgical and esthetic evaluation. Among 14 patients, RP model study was performed in two patients with severe facial deformity. The other patients underwent contouring surgery under conventional methods. Surgical evaluation was performed with computed tomography scan before and after operation. Surgical resection was successful and patients were satisfied with the surgical results.

Key Words 3D RP · Simulation surgery · Fibrous dysplasia · Contouring surgery · Jaw bone.

Received: November 13, 2016 / **Revised:** November 14, 2016 / **Accepted:** November 18, 2016
Address for correspondence: Kang-Min Ahn, D.D.S., M.S.D., Ph.D.
Department of Oral and Maxillofacial Surgery, College of Medicine, University of Ulsan, Asan Medical Center, 88 Olympic-ro, 43-gil, Songpa-gu, Seoul 05505, Korea
Tel: 82-2-3010-5901, **Fax:** 82-2-3010-6967, **E-mail:** ahnkangmin@hanmail.net

Introduction

Fibrous dysplasia (FD) is a benign lesion usually involving jaw bones, rib bones and femur (1, 2). The incidence of craniofacial lesion has been reported between 10% and 25% in the monostotic form (3). Maxilla is the most common site for FD in craniofacial regions and bulging of the lesion requires grinding and/or reconstruction. Monostotic form of FD is not heritable and sporadic. The cause of FD is a gene mutation that prevents the differentiation of osteoblastic cells (4). Mutation of the GNAS-1 gene located in chromosome 20 was known for FD (5). Clinically, it is manifested clearly in the computed tomography (CT) scan, bone scan or bone spect images.

Because of benign entity and slow growing pattern of the lesion, aggressive treatment such as maxillectomy, mandibulectomy and flap reconstruction are not the choice of treatment. Contouring surgery has been recommended for treating FD in patients with esthetic problem (6). Exact amount of bone reduction in the maxilla and mandible is quite difficult because of the complex anatomical structure. The inferior orbital and mental nerves make it difficult to grind the lesion.

One of the efficient way to perform precise surgery is using virtual simulation surgery with 3-dimensional (3D) model (7). The 3-dimensional (3D) rapid prototype (RP) model made with CT scan data is helpful for evaluating lesions that should be removed (8-11). 1:1 size of the 3D RP model gives detailed information about the lesions and important anatomical structures (12).

In this study, 14 consecutive patients who underwent contouring surgery or mass enucleation for FD were reviewed ret-

rospectively. The postoperative results of contouring surgery were reviewed using CT scan data. Patients' satisfaction was evaluated by visual analog scale.

Materials and Methods

A total of 14 consecutive patients who underwent contouring surgery for FD between July 2008 and September 2016 were included in this study. Male to female patients was 3:11 and average age was 38.9±14.5 years old (13-58). Unaesthetic facial bulging was found in four patients and the other patient reported mild swelling of the maxilla and mandible. Preoperative CT scan was taken and 3D RP model was fabricated. Amount of resection was calculated in the CT scan data. Surgical margin was marked on the 3D RP model using pencil. Depth of removal was calculated in the CT scan data. Immediate postoperative CT was taken for evaluation of the precise operation. Institutional review board from our hospital issued an exemption to this study because of the use of collected existing data in such a manner that subjects cannot be identified directly or indirectly.

Results

Patients' demographic data and sites of the lesions are listed in Table 1. Female patients were 13 compared with three male. Female dominance was found in this study. Maxilla was involved in four patients and mandible lesions were found in ten patients. Patients with both sides of the jaw bone involvement were excluded in this study. The mandible bone was more common than the maxilla bone. Operation was performed under general anesthesia in nine patients. 3D RP model was fabricated in two patients with maxillary lesions. Comparison of preoperative and postoperative con-beam CT scans showed excellent symmetry.

A Case Report

A 17-year-old girl was referred from local dental clinic for evaluating maxillary swelling in August 2016. Patient noticed swelling of the right maxilla 6 months ago. There was no bone pain and discomfort during eating. Panoramic radiograph was taken for initial work-up (Fig. 1). From the panoramic radiograph, right maxilla was filled with radio-opaque materials and "ground-glass" like haziness was found. The radio-opaque lesion involved total right maxilla from lateral nasal cavity to posterior maxillary sinus cavity. It was extended in the inferior border of the orbit. Cone-beam CT scan was taken for further evaluation (Fig. 2). About 15 mm of bony swelling was found

Fig. 1. Initial panoramic radiograph showing radio-opaque lesion in the right maxilla.

Table 1. Demographic data of the patients and location of the lesions

No.	Age	Sex	Op. date	Site
1	54	M	2008-07-04	Lt. Mandible
2	18	F	2010-08-03	Rt. Mandible
3	41	F	2011-06-13	Lt. Maxilla
4	36	F	2013-05-27	Rt. Mandible
5	48	F	2013-07-08	Lt. Maxilla
6	42	M	2013-07-08	Rt. Mandible
7	52	F	2013-12-30	Rt. Mandible
8	13	F	2014-07-21	Lt. Mandible
9	58	M	2014-11-17	Lt. Mandible
10	52	F	2015-10-19	Lt. Maxilla
11	32	F	2016-05-23	Lt. Mandible
12	46	F	2016-05-30	Rt. Mandible
13	35	F	2016-06-27	Rt. Mandible
14	17	F	2016-09-26	Rt. Maxilla
Average	38.9			

Op: operation, M: male, F: female, Lt: left, Rt: right

Fig. 2. Buccal swelling of the maxilla in the lateral aspect of the maxillary sinus.

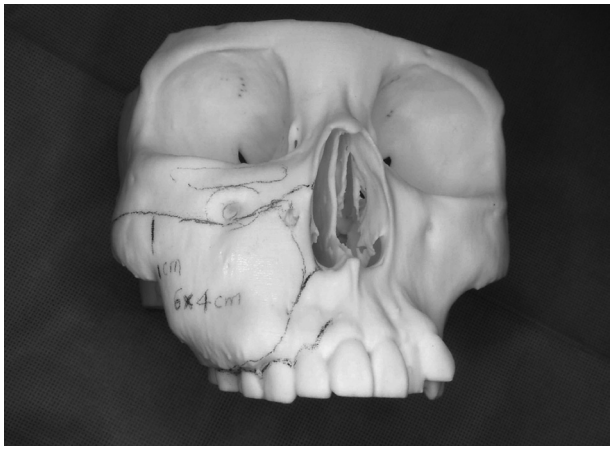

Fig. 3. 3-dimensional rapid prototype (RP) model showing 6x4 cm swelling of the right maxillary bone.

Fig. 4. Indentation of the lesion with 2 mm round bur.

in the lateral aspect of the maxillary sinus. Tentative diagnosis of FD was made with clinical and radiographic data.

3D RP model was fabricated based on 3D CT scan data (Fig. 3). 6×4 cm of bony swelling was found in the right maxillary lateral sinus wall. Inferior orbital nerve was involved in the FD lesion. Operation was performed under general anesthesia. Crevicular incision was performed for buccal flap elevation. Lesion was marked with sterile pencil. Depth of surgical margin was made with 2 mm round bur (Fig. 4). Depth of indentation was confirmed with metal ruler. Postoperative con-beam CT scan was taken the day after operation. Symmetrical maxillary lateral wall was found (Fig. 5). Postoperative recovery was uneventful and patient was satisfied with operation. Postoperative swelling was remained for 3 months. Patient could do usual activity two weeks after operation.

Fig. 5. Immediate postoperative con-beam CT scan showing symmetrical maxilla.

Discussion

Successful treatment of FD depends on an accurate diagnosis, radiographic assessment, surgical plan, surgeon's experience, surgical skill, hospital facility and patient's expectation (13-15). There is no effective medical therapy for FD. Only surgical approach is possible to correct bulging mass due to FD. During operation, grinding and contouring surgery are performed. When there is no information about the exact amount of bone reduction, operation is usually performed by surgeon's instinct and experiences. Postoperative asymmetrical facial appearance is caused by inappropriate surgical plan and bone reduction. Therefore, it is mandatory to make a plan for symmetrical bony reduction in FD patient. 3D RP model is easiest and non-invasive methods for surgical planning. It gives a lot of information about patient condition such as size and location of the lesion, asymmetry of facial skeleton, important anatomical structure and occlusion. In our study, a large maxillary lesion which caused

facial asymmetry could be successfully treated with 3D RP model mock surgery.

Craniofacial lesion involving maxilla is hard to eradicate because maxillary lesion invade into the maxillary sinus cavity. Total removal of the lesion requires aggressive surgery such as partial or total maxillectomy. It has not been recommended because of FD is benign entity with self-limiting progression. Usually there is no occlusal disturbance in FD patient because of slow growing characteristics. In our study, there was no patient who reported occlusal disturbance. Mass in the oral cavity did not make discomfort even though it had grown in the buccal side of the maxilla. The purpose of operation was truly esthetic concern in our case series.

Comparison of CT scans before and after operation was helpful to evaluate surgical results. In our study, con-beam CT was used for evaluation. The advantages of con-beam CT is less radiation dose, easily accessible, fast for image taking and usage of diverse software program for 3D analysis. Amount of bone reduction could be subtracted from 3D RP model, however, the depth of bone reduction was easily calculated from CT scan image.

Conclusion

Simulation surgery using 3D RP model is the easiest way to simulate real operation situation. In this study, 14 patients underwent contouring surgery with successful results. Large sized FD lesion with involving inferior orbital or mental nerve is easily found in the 3D RP model. Precise reduction of the lesion was confirmed with con-beam CT scan.

References

1. Eachempati P, Aggarwal H, Shenoy V, Baliga M. Multidisciplinary approach for management of a patient with fibrous dysplasia of maxilla. BMJ case Reports 2015;2015
2. Kruse A, Pieles U, Riener MO, Zunker C, Bredell MG, Gratz KW. Craniomaxillofacial fibrous dysplasia: A 10-year database 1996-2006. The British Journal of Oral & Maxillofacial Surgery 2009;47: 302-305
3. Valentini V, Cassoni A, Marianetti TM, Terenzi V, Fadda MT, Iannetti G. Craniomaxillofacial fibrous dysplasia: Conservative treatment or radical surgery? A retrospective study on 68 patients. Plastic and Reconstructive Surgery 2009;123:653-660
4. Ricalde P, Magliocca KR, Lee JS. Craniofacial fibrous dysplasia. Oral and Maxillofacial Surgery Clinics of North America 2012; 24:427-441
5. Chapurlat RD, Orcel P. Fibrous dysplasia of bone and mccune-albright syndrome. Best Practice & Research. Clinical Rheumatology 2008;22:55-69
6. Damm DD. Expansion of the maxilla. Craniofacial fibrous dysplasia. General Dentistry 2012;60:436-437
7. Villar-Puchades R, Ramos-Medina B. Virtual surgical planning for extensive fibrous dysplasia in the mandible. Aesthetic Plastic Surgery 2014;38:941-945
8. Ahn KM, Kim JJ. Maxillary reconstruction with free fibular flap using 3d rp model. J Int Soc Simul Surg 2014;1:32-36
9. Kim DH, Cha HS, Ahn KM. Mandibular reconstruction with free fibular flap and dental implant after ablative oral cancer surgery. J Int Soc Simul Surg 2014;1:90-94
10. Kim HJ, Hwang JH, Ahn KM. Fibula free flap for mandibular reconstruction using simulation surgery in bisphosphonate related osteonecrosis of the jaw. J Int Soc Simul Surg 2015;2:1-6
11. Park TJ, Kim HJ, Ahn KM. Mandibular reconstruction using simulation surgery with 3d rp model in osteoradionecrosis patient: A case report. J Int Soc Simul Surg 2015;2:76-79
12. Kang SJ, Oh MJ, Jeon SP. A novel and easy approach for contouring surgery in patients with craniofacial fibrous dysplasia. The Journal of Craniofacial Surgery 2015;26:1977-1978
13. Wang R, Li G, Liu C, Jia C, Han Y. Three-dimensional printing of reduction template in the contouring of craniofacial fibrous dysplasia. The Journal of Craniofacial Surgery 2016;27:1792-1794
14. Burke AB, Collins MT, Boyce AM. Fibrous dysplasia of bone: Craniofacial and dental implications. Oral diseases;2016
15. Unal Erzurumlu Z, Celenk P, Bulut E, Baris YS. Ct imaging of craniofacial fibrous dysplasia. Case Reports in Dentistry 2015;2015: 134123