

## Free Vascularized Dermofat Flap For Contouring Maxilla In Fibrous Dysplasia Patients

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**Background:** Fibrous dysplasia is a disease that causes bone thinning and growth of lesions in one or more bones and leads to bone weakness and scar formation within the bones. Especially when involving the skull or facial bones, the lesions can cause externally visible deformities.

**Methods:** We reported one case of fibrous dysplasia of the maxilla reconstructed in our Plastic Surgery Division of Cipto Mangunkusumo Hospital. Data was taken from the medical and surgery records.

**Results:** In this case, we used the Antero Lateral Thigh (ALT) flap to reconstruct the maxilla and facial contour in 26 years old girl with fibrous dysplasia. After the operation we had daily observation to evaluate the blood flow to the flap and it had satisfactory result without any complication.

**Summary:** Ultimately, free vascularized dermofat flap with ALT seems to be suitable for craniofacial contouring surgery, in this case for contouring maxilla. Facial contour could further be improved after secondary reshaping of the healed flaps. This led to nearly perfect long-term facial symmetry in all cases. The ALT proved to be a reliable donor site, providing enough well-vascularized fatty tissue for facial contour augmentation.

**Keywords:** *facial fibrous dysplasia, perforator-based anterolateral thigh (ALT) dermofat flap*

**Latar Belakang:** Fibrous dysplasia adalah suatu kelainan yang menyebabkan penipisan satu atau beberapa tulang yang mengakibatkan kelemahan dan parut dalam tulang. Jika hal ini melibatkan tulang wajah maka deformitas akan semakin jelas terlihat.

**Metodologi:** Kami melaporkan satu kasus fibrous dysplasia tulang maksila di Divisi Bedah Plastik dan Rekonstruksi RSCM

**Hasil:** Pada kasus ini, digunakan flap ALT untuk rekonstruksi maksila dan kontur wajah. Setelah operasi dilakukan evaluasi vitalitas flap dan didapatkan hasil yang baik tanpa komplikasi.

**Ringkasan:** Didapatkan bahwa free vascularized dermofat flap dengan ALT merupakan tindakan pembedahan yang sesuai untuk koreksi kontur wajah. Kontur wajah dapat diperbaiki lebih lanjut menggunakan prosedur sekunder pada flap yang telah sembuh sempurna. Hal ini mengakibatkan hasil simetri wajah yang permanen dalam semua kasus dan ALT merupakan daerah donor yang cukup baik untuk rekonstruksi kontur wajah.

**Kata kunci:** *facial fibrous dysplasia, perforator-based anterolateral thigh (ALT) dermofat flap*

**F**ibrous dysplasia (FD) is a benign disease of bone that was originally described by Lichtenstein more than sixty years ago<sup>1,2</sup>. FD is a developmental dysplastic disorder of bone in which the normal bone matrix is replaced by fibroblastic proliferation. Some believe that the immature woven bone is formed directly from abnormal fibrous connective tissue that is unable to form mature lamellar bone, hence the term dysplasia. Others believe that there is underlying abnormal fibroblast proliferation that results in the replacement of normal can-

cellous bone with an immature fibrous tissue that is poorly mineralize<sup>3</sup>. FD is a genetically-based sporadic disease of the bone, its "Mutations in the gene (GNAS I) encoding for the a-subunit of a signal transducing G protein (Gs-a) lead to increased c-AMP production affecting proliferation and differentiation of preosteoblasts<sup>4</sup>. The diagnosis of fibrous dysplasia is often made in infancy and childhood.

The maxilla or mandible may be involved but a predominance of the maxilla has been documented. Males are less often affected than

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females. The deformity of the jaw results from a progressively slow-growing painless swelling, but growth often slows or become arrested at a time coinciding with the onset of puberty<sup>5</sup>. FD presents in three forms – monostotic (including craniofacial), polyostotic and polyostotic with endocrinopathies. Although the term monostotic can be readily applied to cases of FD affecting the mandible alone, this may not be so for FD affecting the maxilla. There, FD can affect contiguous bones such as the zygoma. These cases have been called “craniofacial” FD<sup>6</sup>.

Clinically, a painless enlargement of the affected bone is the most common presenting symptom. Bulging of the canine fossa and hyper prominence of the zygomatic process of the maxilla are usual features of maxillary lesions with frequent involvement of the maxillary sinus. In the mandible, a swelling is most often found at the angle of the jaw<sup>7</sup>. Fibrous dysplasia of the maxilla or mandible is usually unilateral. The lesion grows very slowly, causing expansion of the involved bone and giving a non-tender facial asymmetry of variable degree<sup>8</sup>.

Treatment for fibrous dysplasia is almost always surgical intervention. If the lesion is extensive and presents with malocclusion and jaw disproportion, conservative recontouring surgery and repositioning of the affected jaw bone aiming at aesthetic correction or functional improvement are recommended<sup>9</sup>. Radiotherapy in the treatment of fibrous dysplasia is contraindicated because of the possibility of radiation induced sarcomas<sup>10</sup>.

Any patient who undergoes a maxillectomy for radical treatment of a maxillary tumor faces serious problems with mastication, swallowing, speech, and esthetics. Reconstruction is of paramount importance to these individuals, but this is a major challenge. There are several reconstructive techniques that involve the use of vascularized or non vascularized autogenous material or prosthetic devices and zygoma implants<sup>11</sup>. Pedicled transplants that do or do not require microvascular reanastomosis have also been reported<sup>12</sup>. This leads to multistage reconstruction.

## PATIENT AND METHODS

A 26 years old woman presented to our hospital with chief complain asymmetry of the facial bone. She had a defect on the maxilla after several reconstruction. Sixteen years ago (at age 10 years old), she had her right face painless swelling with hard consistency. She had difficulty to masticate, swallowing, speech, and esthetics. She was admitted to district hospital and had proper examination for her complain. She was diagnosed with facial tumor and suggested to have the tumor removed but she ignored it. On January 2007, she went to oncologist at our hospital, was done a biopsy and diagnosed as fibrous dysplasia. Multistage reconstructive operation was planned.

On first stage operation, the oncologist decided to do a surgery to remove the mass. They did a right hemimaxillectomy and hemimandibulectomy, and inserted a titanium implant to substitute the jaw. Two months later, she undergone an operation for fixation of the previous implant but failed and the implant was removed.

One year later, she was admitted to Plastic Surgery Division of our hospital and had a mandible reconstruction with free fibula transfer flap. On the second stage operation, we reconstructed the maxilla using one of her right rib and a titanium implant. Two weeks later, we reconstructed the palate with radial forearm free flap. After several months, we had serious problem of infection. We evacuated the pus and debris, and maintained the existing tissue transfer. We evaluated the outcome, the infection had been controlled and patient was then discharged. One year after, the patient went back to our clinic with a complaint of difficulty to swallowing, so we decided to do a shaving of the palate.

At third stage, we planned a surgery for maxilla contouring. The patient had defect on her maxilla after previous reconstruction (Figure 1). Two years after the first surgery, we had vascularized perforator-based ALT dermofat flap to close the defect of the maxilla. The flap was deepithelialized. Femoral descendens lateral circumflex artery and vein were used to be attached with superficial temporal artery and comitantes vein (Figure 2).



**Figure 1.** Twenty six year old woman admit to Plastic Surgery Division after several reconstruction, right maxillectomy and mandibulectomy. Chief complain is defect of right maxilla.



**Figure 2.** During operation, reconstruction with free vascularized dermofat flap. The flap is elevated (above, right), the perforator is safely removed. The recipient site is well prepared and the flap was attached (below, left). Immediate postoperative view (below, center and right).

After the operation, we had daily observation with Doppler to evaluate the blood flow to the flap and it had satisfactory result without any complication (Figure 3).

### **DISCUSSION**

Reconstructive surgery for defects resulting from head and neck cancer allows tumor

resection while maintaining quality of life. Various treatment procedures involving fat grafts and local or microvascular free flaps have been used for correction of facial contour deformities resulting from congenital anomalies or acquired deformities. Free flaps have been in clinical use for nearly three decades. In that time, their survival rates have improved as refinements have





**Figure 3.** From left to right, 1) before surgery, 2) 1 month after surgery, 3) 2 month after surgery. Maxilla contouring with free vascularized dermofat flap. It may show reduction of tissue amount because of fat tissue absorption but it has significant change in appearance.

been made in surgical technique and instrumentation. In this case, FD, the lesions are tumor-like growth that consist of replacement of the medullary bone with fibrous tissue, causing the expansion and weakening of the areas of bone involved. Especially when involving the skull or facial bones, the lesions can cause externally visible deformities. This leads to severe deformity of the facial contour, but still its etiology is unclear<sup>11</sup>.

There have been various attempts to surgically manage fibrous dysplasia especially in facial bones. Artificial implants like silicone or hydroxyapatite were used to correct the defect on the face. Autologous fat injection or dermofat graft, bone graft are also commonly used surgical methods. Recently, perforator-based ALT dermofat flap is being widely applied to correct the asymmetry of the face<sup>12</sup>.

Microvascular tissue transfer, this technique began to be used to cover defects in the oral and maxillofacial area in the early 1980's<sup>13,14</sup>. Currently, many different types of flap are used, with varying characteristics. Each type of flap has a spectrum of indications that may be broad or narrow; for special types of defect, multiple flaps can be used. Finding the optimal solution to a given reconstructive problem involves a determination of the most suitable tissue, the most favorable donor site, and the physiologically least stressful operation for the patient<sup>14, 15</sup>. Because this method leaves the superfluous fatty and muscular tissue at the

donor site rather than taking it along with the flap, a very thin flap results that is highly suitable for coverage of the usually flat defects of the oral cavity or facial skin<sup>16</sup>.

Mao et al, analyze the feasibility and reliability of free tissue flap transfers in the head and neck region. One thousand five hundred and eighty-four consecutive free flap transfers performed in 1,501 patients were reviewed. There were ten kinds of free flaps in this group, with the free fibula flap most commonly used, followed by free radial forearm flap, rectus abdominis flap, free jejunum flap, anterolateral thigh free flap, iliac crest flap, scapular flap, latissimus dorsi flap, lateral arm flap, and ileocecal flap. The overall success rate of flaps was 98.1% (1,554/1,584)<sup>17</sup>.

In this case, patient had several stage of operations. In particular order, patient had done free tissue flap transfers to fulfilled the mandibula and maxilla defect with free fibula flap, free radial form arm flap, and at last, anterolateral thigh free flap. It indicates that multiple stage reconstruction must be done.

The ALT flap, first described by Song et al, has gained popularity in head and neck reconstructive surgery. The ALT flap is a versatile soft-tissue flap in which thickness and volume can be adjusted for the extend of the defect. In our experience, flap elevation is relatively easy, although surgeon should be familiarized with the anatomy of donor site. It has a consistent anatomy of the main pedicle (desc-

ending branch of the lateral femoral circumflex vessels)<sup>13</sup>.

The anterolateral thigh (ALT) and the proximal lateral calf have been described as particularly suitable donor sites for reconstruction in oral and maxillofacial surgery<sup>15,18,19</sup>. The widest experience to date has been with ALT perforator-based flaps up to 8 × 25 cm in size, which are currently used routinely by some authors as an alternative to radial flaps because of the very low morbidity at the donor site, and which have a wide spectrum of indications. In contrast, soleus perforator-based flaps are usually used only for smaller defects, because the donor site in the proximal lateral thigh can only be primarily closed if the flap is 5 cm wide or less. Preservation of the cutaneous vessels of the calf keeps the morbidity of flap harvesting to a minimum<sup>20</sup>.

In this case, by deepithelializing the ALT flap, as dermofat flap, no loss in the bulk of the flap was observed and flap perfusion was not affected. The dermofat flap allows easy creation of a subcutaneous pocket under the cheek through the available upper incision line without extra scarring<sup>21</sup>. No publication reporting use of the ALT dermofat flap for maxilla contouring in the literature. The major advantages of the ALT dermofat flap were as follows: provision of a constant and safe pedicle, adequate bulk, a low risk of atrophy and infection by means of vascularized tissue, and well-hidden scar at the donor site.

### SUMMARY

Ultimately, free vascularized dermofat flap with ALT seems to be suitable for craniofacial contouring surgery<sup>23</sup>. In this case for contouring maxilla, although proper evaluation must be held to measure the absorption of fat tissue we had plant. For refinement, minor contour irregularities and absorption may be evaluated after 6 months<sup>22</sup>. Any further surgery may be required to restore the contour of the maxilla to be optimized, achieve the anatomically and functionally goal and gain the satisfaction of the patient<sup>23</sup>. Facial contour could further be improved after secondary reshaping of the healed flaps. This led to nearly perfect long-term facial symmetry in all cases. The ALT

proved to be a reliable donor site, providing enough well-vascularized fatty tissue for facial contour augmentation<sup>22</sup>.

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