Preoperative planning for advanced modelling of anterolateral thigh flaps in the treatment of severe haemifacial atrophy in Parry–Romberg and Goldenhar syndrome

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Abstract

Background: Technological advancement in medical science is constantly innovating solutions to the varied and complex challenges of surgery. Digital diagnostics and prospective microsurgery are rapidly evolving. Three-dimensional (3-D) imagery and computed tomography (CT) scanning can determine accurate dimensions of many defects. Subsequently, a thorough understanding of microvasculature and application of microsurgical techniques allows modelling of flaps to obtain an accurate transplant resulting in an aesthetic outcome following the very first operation.

Methods: Two patients with Parry–Romberg syndrome and one patient with haemifacial microsomia (Goldenhar syndrome) were treated with anterolateral thigh (ALT) flaps to restore facial volume, contour, and symmetry. In each case, a different approach in planning and performing the intervention was applied:

- The patient in the first case had a full-thickness ALT flap transplant with significant overcorrection.
- The patient in the second case had reconstruction with a partially thinned ALT flap guided by a clinically formed template made per manual measurements.
The patient in the third case had reconstruction with a precise primary thinned ALT flap with a template made according to data obtained from superimposed 3-D photographs and CT scans.

Results: All flaps survived. In cases 1 and 2, a corrective intervention was required to achieve acceptable facial symmetry. In case 3, a very good aesthetic result was achieved immediately after the first operation.

Conclusions: Digital methods of 3-D analysis offer great opportunities in creating a precise operative plan, and modern surgical techniques make it feasible to implement it intra-operatively. Overall, these methods shortened the rehabilitation time by avoiding further revision surgeries.

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Introduction

The pioneer microsurgeons who made eminent contributions to plastic surgery would admire contemporary plastic surgeons, who not only use perforator flaps in the free style form from any area of the body, but also perform thinning of flaps to acquire the required thickness and thereby improve results of reconstruction at the very first surgery.

Since 1984, when Song first demonstrated the use of the anterolateral thigh (ALT) flap, the possibility of microsurgical auto-transplantation has increased. Currently, microsurgical auto-transplantation is a standard approach in the treatment of patients with severe facial asymmetry, such as those affected by Parry–Romberg syndrome.

The treatment of patients with Parry–Romberg syndrome types 3 and 4 (Guerrerosantos et al.) and severe cases of Goldenhar syndrome with hemifacial microsomia, involves a comprehensive approach including correction of bony deformities and volume-contouring of tissue defects.

This paper illustrates the authors’ experience in treating 3 patients with severe hemifacial microsomia and hemiatrophy, with emphasis on the features of soft tissue flap transplantation. However, scrutiny of the adjuvant stages such as: orthognathic surgery and silicone implantation were not done.

Materials and methods

Patients

From 2015 to 2017 at the Central Research Institute of Dentistry and Maxillofacial Surgery Moscow, three patients aged between 25 and 32 years were treated. Two patients were diagnosed with Parry–Romberg syndrome (type 4), and one patient was diagnosed with Goldenhar syndrome (hemifacial microsomia). All patients had presented with a history of unsuccessful surgical corrections (e.g., lipofilling, silicone implants, and polyacrylamide gel injections) at several other institutions.

Preoperative assessment

According to the methods of Zhou et al., Kimata et al., and Xu et al. the perforator vessels on the thigh were identified using a hand-held audio Doppler scanner. In addition, colour duplex Doppler scanning of the thigh and neck was performed.
The methods of planning and intraoperative techniques evolved in technical precision from the first case to the third.

Surgical technique

An incision was made anterior to the tragus in the parotid-masseteric region in the subcutaneous layer above the superficial muscular aponeurotic system (SMAS) to avoid damage to the facial nerve branches. The skin within the atrophied zone was mobilised to form a pocket for flap placement. The recipient vessels were identified using the submandibular approach.

In all cases, the ALT flap was raised from the distal to the proximal end. During flap mobilisation, the perforators were identified as they exit through the fascia lata. When a thinned flap was required it was raised within the superficial fascial layer; and, the easiest way to identify perforators was in between the smaller superficial fat lobules and large deep fat lobules.10

After the flap was isolated on the identified perforating vessels, the fascia lata was incised, and further dissection continued in the subfascial space. The perforators led proximally to a common source, and dissection of the vascular pedicle continued until the required length was achieved.

The flap was then transferred to the face, and stabilised with holding sutures until the microsurgical anastomosis was completed.

The precision of the flap sculpting gradually improved from the first to the third case. Individual differences are described for each patient.

Observations

The atrophied area ranged from a minimum of $15.0 \times 6.0$ cm to a maximum of $21.5 \times 11.0$ cm. In the first and third case, the flap was raised on musculocutaneous perforators, whereas in the second, it was on the septocutaneous perforator.

The vascular pedicle was between 5 and 8 cm in length. The operating time varied according to the complexity of flap modelling (ranging from 6 h in the first case, up to 11 h in the third). Features of the surgeries are provided in Table 1.

| Patient | Type of Flap | Flap Size (cm) | Number and Type of Perforators | Flap modelling Type | Operation Time (H:Min) | Recipient Vessels | Post-Op Complications | Reconstructed Areas |
|---------|--------------|---------------|--------------------------------|--------------------|------------------------|-------------------|----------------------|---------------------|
| 1       | Fasciocutaneous | 15.0 × 7.0 | 1 Musculocutaneous perforator | None               | 5:40                  | Facial artery and vein | Early: Haematoma, inflammation, wound dehiscence | Zygomatic, parotid-masseteric, buccal, submandibular |
| 2       | Adipocutaneous | 19.0 × 7.0 | 1 Septocutaneous perforator | Fat excision        | 9:20                  | Facial artery and vein | Late: Displacement of anterior flap margin | Temporal, zygomatic, parotid-masseteric, buccal, submandibular  |
| 3       | Adipocutaneous | 21.5 × 11.0 | 2 Musculocutaneous perforators | Primarily modelled flap | 11:20                  | Lingual artery, facial & retromandibular veins | None | Frontal, temporal, zygomatic, parotid-masseteric, buccal, submandibular |
Results

Postoperative monitoring was done with standard clinical methods, tissue oximetry, and hand-held Doppler. Photographic documentation, 3-D photographs, and CT scans with radiographic contrast markers were repeated.

Cases

Patient one

In 2015, Ms. S, a 25-year-old woman, presented with right-sided haemifacial microsomia manifested as: anotia, malocclusion class 2 (Angle’s classification), asymmetric mandibular deformation, zygomatic bone hypoplasia, and haemifacial atrophy. She was diagnosed with Goldenhar syndrome. The treatment was performed in two phases. During the first phase, in March 2015, a series of operations were performed to correct the malocclusion (distraction osteogenesis in the right mandibular ramus, median palatal expansion, and mandibular bilateral split sagittal osteotomy and maxillary Le Fort 1 osteotomy) (Figure 1).

During the second phase, in September 2015, an auto-transplantation with a free adipocutaneous ALT flap that was 15.0 × 7.0 cm in size was performed. While creating a pocket at the recipient site, pigmented atrophic areas of skin with low degree of elasticity were excised. Of the harvested flap, an epidermal area of 14.0 × 3.0 cm was retained. The postoperative period was complicated with haematoma formation under the flap, wound dehiscence, and suppuration. The infection was controlled with incision and drainage, and antibiotics. Secondary sutures were applied at the site of wound dehiscence. The outcome was unsatisfactory with overcorrection (Figure 2).
In May 2016, a revision surgery was done for flap adjustment, thereby restoring the facial symmetry (Figure 3).

Patient two

In 2016, Ms. L, a 28-year-old woman, presented with progressive left haemifacial atrophy. The patient had disease onset at 4 years, and the atrophy progressed until the age of 15, when she was...
diagnosed with Parry–Romberg syndrome. In 2007, on stabilisation of the syndrome, lipofilling was done to restore the facial symmetry; however, fat resorption occurred (Figure 4).

In March 2016, the left haemifacial contour was reconstructed at our hospital using the ALT perforator flap that was $19.0 \times 7.0$ cm in size. Before the surgery, a personalised template was clinically made using a ruler to measure the discrepancy in soft tissue between the affected and contralateral normal side. On harvest, flap thinning was done per the pre-formed template. During de-epithelisation, prior to anastomosis, a skin paddle of $13.0 \times 2.5$ cm was kept to avoid tension at the suture site. The postoperative period was uneventful. The transcutaneous fixation sutures were removed on the fifth postoperative day (Figure 5).

Figure 4. Second patient, preoperative frontal view.

Figure 5. Second patient, intraoperative left lateral view of the face. Extended preauricular incision with an individually modelled de-epithelised adipocutaneous ALT flap with skin paddle placed over the affected region.
At a routine follow-up at 3 months, we discovered that the patient had an unsatisfactory outcome due to displacement of the anterior flap margin. During the revision surgery at 6 months, the position of the flap was corrected by suturing it to the periosteum of the mandible, zygomatic bone, and deep temporal fascia. In addition, the skin island was excised and lipofilling was done. The postoperative result was excellent (Figure 6).

Patient three

In 2017, Ms. L, 32-year-old woman, presented with right haemiatrophy, hypoplasia of the right zygomatic bone, asymmetric deformation of the jaws, and malocclusion class 2 (Angle’s classification). She was diagnosed with Parry–Romberg syndrome (type 4). Atrophy began when the patient was 6 years of age, and reached peak manifestation at the age of 13 years, stabilising at the age of 21 years. At the age of 20, in another hospital, she underwent silicone implants in the right zygomatic and temporal areas. Following an inflammatory response, the implants were removed and, subsequently, multiple corrective interventions were performed at other institutions, but they were ineffective. In 2017, at the age of 32, she came to our hospital, the Central Research Institute of Dentistry and Maxillofacial Surgery (CRID), Moscow (Figure 7).
Preoperatively, a 3-D photograph and CT scan were done with radiographic contrast markers. Nemotech Studio© software was used for flap planning. A detailed description of template preparation for flap harvest is elaborated in the Discussion section (Figure 8).

The template was placed according to the pre-located perforating vessels of the thigh. A flap size of 21.5 × 11.0 cm was harvested in the required layers to provide the necessary shape and thickness.
A flap based on two perforators was harvested, and a precise pre-modelling was done (Figure 9).

Figure 9. Third patient, intraoperative varying thickness of ALT flap harvest. Harvest of a primary-thinned adipocutaneous ALT flap in progress. Ruler and forceps demonstrating the varying thickness in different areas of the ALT flap as per the template (see Figure 8).

The flap was then transferred to the face. Post anastomosis, de-epithelisation was done, and the flap was fixed to immobile structures. Further details are provided in the Discussion section (Figure 10).

Figure 10. Third patient, intraoperative right lateral view of the face with flap in situ. The primary-thinned and modelled ALT flap placed over the recipient site after revascularisation (prior to de-epithelialisation). The varying thickness of the ALT flap is evident, as it matches the natural contour of the affected region.

At the recipient site, active drains were placed above and below the flap; and at the donor site, drains were placed between the vastus lateralis and rectus femoris muscles. The donor site wound was closed in layers. At the same time, lipofilling was performed in both lips and in the lower eyelid. Facial sutures were removed on the tenth postoperative day, and thigh sutures were removed on the fifteenth postoperative day.
The inclusion of two perforating vessels and the harvest of a precise pre-modelled adipocutaneous ALT flap prolonged the total operating time to 11 h. Consequently, a secondary revision was not necessary, as an excellent aesthetic result was achieved after the first operation (Figure 11).

Figure 11. Third patient, postoperative frontal view.

Features of the rehabilitation of all patients are presented in Table 2.

Table 2
Features of rehabilitation of all patients.

| Patient | Orthognathic Surgery | Implant | Lipofilling | Microsurgery | Outcome After 1st Surgery | Revision Surgery | Outcome After 2nd Surgery |
|---------|----------------------|---------|-------------|--------------|---------------------------|------------------|---------------------------|
| 1       | +                    | −       | −           | +            | Unsatisfactory            | +                | Good                      |
| 2       | −                    | −       | +           | +            | Unsatisfactory            | +                | Excellent                  |
| 3       | −                    | +(removed) | +       | +            | Excellent                  | −                | −                         |

+, performed; −, not performed.

Discussion

Anatomy and thinning

ALT flaps have a high level of variability in the source and type of perforating vessels. However, the anatomical variations did not prevent the ALT flap from gaining popularity for soft-tissue defect reconstruction.
The blood supply to the skin of the lateral thigh is provided by perforators that typically arise from the descending branch of the lateral circumflex femoral artery (LCFA). Depending on their relationship with the vastus lateralis and rectus femoris muscles, these perforators may be either septo- or musculocutaneous.

The principal role of the vascular plexuses in the blood supply of skin flaps has been recognised.\textsuperscript{18,19} Alkureishi L.W. et al.\textsuperscript{20} reported that the communicating vessels between the plexuses do play a significant role in the blood supply of the flap. Further refinement was demonstrated by Saint-Cyr M. et al.\textsuperscript{21,22} proving that the viability of the flap remained high even after radical thinning.

There are two methods of obtaining a thin flap: one is the thinning of the flap after harvest by excision of fat, and the other is harvest of a primary-thinned flap.\textsuperscript{12} Each method has its own advantages and limitations. After harvest, thinning of the flap is technically easier because, while raising the flap over its fascia lata, the perforating vessels become more apparent. However, proficiency in the harvest of a primary-thinned flap with desired thickness at the shortest operating time is technically challenging.

The latest techniques on “hot and cold spots” of “perforator distribution”\textsuperscript{10,23} are designed to simplify and accelerate the method of obtaining a primary-thinned flap. With some experience, this method allows the surgeon to raise the flap faster and minimise damage to the donor site by preserving the lateral cutaneous nerve of the thigh and its branches.

### Adipofascial and adipocutaneous ALT flaps

Gang Chai et al., Loubin Si et al., and Li Teng et al.\textsuperscript{13–15} mentioned the advantages of an adipofascial flap in eliminating asymmetry of facial contours, leaving an acceptable scar at the donor site with the possibility of complete skin preservation. Also, a harvest of de-epithelised flap preserves the fascia lata and lateral femoral cutaneous nerve. Likewise, Ji, Y. et al.\textsuperscript{24} also mentioned the feasibility of transfixing the fascia lata to the underlying immobile structures at the recipient site. However, because of the inherent mobility of the deep subcutaneous fat over the fascia, the adipofascial flap is not adherent to the pocket created at the recipient site and, thus, results in flap instability and sagging. Also, in adipofascial flaps, postoperative shrinkage may reach up to 25%.\textsuperscript{14} With such a high degree of variability, the feasibility of an accurate simulation in the third case would not be possible.

Adipocutaneous flaps consist of a superficial fatty layer with small fat lobules, which has a strong affinity to the dermis. Because of this strong bonding, fixation of the dermis to bony structures and temporal fascia appears more stable for flap fixation. Also, these fat lobules are more compact and dense, resulting in less postoperative shrinkage. This was one of the reasons why we chose an adipocutaneous flap for reconstruction. As the skin overlying the atrophy zone is of poor quality, it should be excised and, thus, the skin component of the adipocutaneous flap is necessary to replace the formed defect. However, the best aesthetic result was achieved with full de-epithelisation of the adipocutaneous flap. A major drawback of an adipocutaneous flap is a larger defect at the donor site.

### Flap planning

Using modern methods of 3-D visualisation, detailed analysis of defects can be planned for a better restorative outcome. Gang Chai et al.\textsuperscript{14} demonstrated the advantages of laser scanning in comparison to 3-D photographs.

Clinically it is important to maintain the head in a natural head position (NHP)\textsuperscript{25} to have an accurate assessment of the soft-tissue volume in the submandibular and submental regions. For a precise measurement, CT scans and 3-D photographs should also be performed in NHP. We performed a thorough marking of the atrophied facial zone and pasted radiographic contrast markers (Gutta-Percha and Radiological Markers, IZI Medical Product) onto the patient’s face. Then, 3-D photographs and CT scans of the patient’s head were taken, and the acquired images were superimposed using the Nemotech Studio\textsuperscript{©} software (Screen dump 1).
The centre of symmetry between the right and left sides was identified, and using calipers (in Nemotech Studio©), the dimensions of soft-tissue contour discrepancy at three different points of the affected region to the contralateral normal side were compared. The resulting data are presented in Table 3. Accordingly, an individualised template was created, and the values were noted on it (Screen dump 2).

### Table 3
Data obtained from digital analysis (of third patient).

| Zone                  | Normal Side (mm) | Affected Side (mm) | Discrepancy Between Healthy and Affected Sides (mm) |
|-----------------------|------------------|-------------------|---------------------------------------------------|
| 1. Temporal           | 68.8             | 56.8              | 12                                                |
| 2. Parotid-masseteric | 69.4             | 62.8              | 6.6                                               |
| 3. Submandibular      | 61.2             | 36.7              | 24.5                                              |

[Screen dump 2. Superimposed 3-D Photos and CAT Images in Nemotech Studio (green – affected side measurements; pink – healthy side measurements).]
Flap fixation and drainage at the recipient site

In the second patient, a complication of flap displacement on removal of the transcutaneous sutures during the postoperative period led to improvisation of the technique. Accordingly, in the subsequent case, the flap was fixed at five points: 1. the cartilage of the tragus, 2. temporal fascia, 3. periosteum of the zygomatic bone, 4. periosteum of the mandible, and 5. subcutaneous tissue of the submandibular region.

Given the large area of mobilisation and detachment, the placement of an active drainage below the flap gives the best results by preventing haematoma formation. Nonetheless, an additional drainage above the flap was established to reduce the mobility of the overlying skin, thus avoiding the wobbling effect.

Lipofilling and silicone implants

In cases with minor soft-tissue asymmetry and deformities, lipofilling is considered the treatment of choice due to its relative simplicity, low rate of donor site morbidity, and minimal complications. However, in cases of severe haemifacial atrophy and microsomia, the poor vascularity and microcirculation of superficial tissues is a major hindrance for lipofilling, as it often results in resorption of a significant amount of injected fat. In the second patient, two lipofilling procedures prior to flap transplantation resulted in an almost 100% resorption. However, lipofilling procedures performed post flap transplantation in the second and third patients showed retention of a major portion of the injected fat, as observed at 1-year post procedure. The reason for this phenomenon was the improved blood supply to the recipient area due to the transplanted vascularised tissue.

Likewise, the advantages of silicone implants are the feasibility of precise sculpted implants and absence of donor site morbidity. However, the implants do not match the softness and texture of the subcutaneous tissues. Because it is securely fixed to the bone, the rationale would be to use it in restoring symmetry of bony deformities, as it often provides better results than osteotomy and tissue transplantation do. When placing implants in the area of atrophy, especially in Parry–Romberg syndrome (type 4), one of the major problems observed was the atrophied skin and subcutaneous structures with poor elasticity, thus making it quite challenging to create an adequate pocket for the implant with its probable consequence of inflammation and extrusion.

Conclusions

Modern 3-D visualisation techniques, together with the feasibility of selective flap thinning, can create favourable conditions for an ideal outcome in soft-tissue atrophy and haemifacial microsomia. It was found that the conjunction of CT scan with 3-D photographs remained the most informative tool in analysing the differences in volume between the affected and healthy sides. Also, an in-depth analysis of the advantages and disadvantages of adipofascial and adipocutaneous flaps in choosing the preferred method of reconstruction was deemed necessary.

A positive aesthetic result of soft-tissue flap transplantation in volumetric contouring was found mostly dependent on the variables such as: determination of deficit in the affected zones, the use of flap modelling techniques, preplanning of fixation points to the underlying fixed structures at the recipient site, and possibility of maximum de-epidermisation of the applied adipocutaneous flap.

Successful treatment of patients affected with severe forms of haemifacial microsomia and progressive haemiatrophy (types 3 and 4) was feasible with a planned comprehensive approach. It was also ascertained that lipofilling and silicone implants were most efficiently applied post microsurgical transplantation, and not prior to the transplantation.

Conflict of interest

None.
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