

Reconstruction of Near-Total Loss of the Upper and Lower Lips due to Purpura Fulminans with Local Tissue and a Dual-Skin Paddled Anterolateral Thigh Flap

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Summary: It is difficult to totally reconstruct the lip, achieving good functional and aesthetic results. There have been few reports of reconstructing complete lip defects. Moreover, upper and lower lip necrosis by purpura fulminans has not been reported. We present a case of a 60-year-old male purpura fulminans patient with upper and lower lip necrosis. Fortunately, our patient had retained his oral commissure function. We reconstructed this defect with an orbicularis oris muscle-skin-mucosal pedicled flap derived from the region between the nasolabial folds for upper lip; a similar bipedicled flap for the lower lip and the donor site was closed with a dual-skin paddled anterolateral thigh flap. Postoperative results were satisfactory, that is, no lip tightness or aperture restriction was seen, and symmetry had been achieved. The new lips exhibited complete sensory recovery. Drooling was minimal during rest and feeding. We could select a method that combined the advantages of local and free flaps. We consider our method for this defect is superior to those described in previous studies, in that the restoration of lip sensation and oral sphincter function can be achieved to some extent in 1 stage while preserving the oral commissure function. (*Plast Reconstr Surg Glob Open* 2017;5:e1505; doi: 10.1097/GOX.0000000000001505; Published online 22 September 2017.)

Purpura fulminans (PF) is a rapidly progressive thrombotic disorder involving skin necrosis and disseminated intravascular coagulation. It mainly affects neonates and children and is associated with deficiency of the natural anticoagulants proteins C and S. It can progress to multiorgan failure caused by thrombotic occlusion of small and medium-sized blood vessels. Early recognition and treatment are essential for reducing the risk of mortality.^{1,2} PF can also occur in adults with severe infections. The absence of the spleen is known to be a predisposing factor for PF. Meningococcus, varicella, and pneumococcus are common causative bacteria of PF. In acute infectious PF, symmetrical purpura lesions appear on the face and periphery of the limbs and gradually progress to dry necrosis. Amputation is required in 19% of PF cases.^{2,3} However, reports about lip necrosis caused by this disease are rare.⁴ We report the case of a PF patient with upper and lower lip necrosis.

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PATIENT

The patient was a 60-year-old man.

After developing a sudden fever and diarrhea, he was diagnosed with septic shock and disseminated intravascular coagulation and admitted to the intensive care unit.

Streptococcus pneumoniae was detected in a blood culture. Splenic hypoplasia was seen on computed tomography. The day after the patient's admission, a purpuric rash was evident on his extremities and face, and the purpuric area subsequently became necrotic. Based on these symptoms, he was diagnosed with PF. In addition to his toes and 4 fingers (distal to the proximal interphalangeal joint), the upper and lower lips and the left side of the nasal root became necrotic (Fig. 1).

About 40 days later, when the patient's general condition had improved, amputation of the affected digits and debridement and reconstruction of the lips were performed.

After debriding the lips, we designed an orbicularis oris muscle-skin-mucosal pedicled flap derived from the region between the nasolabial folds for upper lip reconstruction, which preserved the oral commissure. In addition, we designed a similar bipedicled flap for the lower lip.

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Fig. 1. After developing purpura fluminans, in addition to his toes and fingers, the upper and lower lips and the left side of the nasal root became necrotic.

The upper and lower lip flaps were transferred. This approach allowed natural mouth closure to be achieved (Fig. 2). For the secondary defects, we used a dual-skin paddled anterolateral thigh flap,⁵ which contained 2 thinned flaps with 2 independent perforators. The pedicle was anastomosed to the facial artery and vein (Fig. 3).

After 1 year, the results were satisfactory, that is, no lip tightness or aperture restriction was seen, and symmetry had been achieved. The new lips exhibited complete sensory recovery. Drooling was minimal during rest and feed-



Fig. 2. We designed an orbicularis oris muscle-skin-mucosal pedicled flap derived from the region between the nasolabial folds for upper lip reconstruction, which preserved the oral commissure. In addition, we designed a similar bipedicled flap for the lower lip.



Fig. 3. For the secondary defects, we used a dual-skin paddled anterolateral thigh flap, which contained 2 thinned flaps with 2 independent perforators.

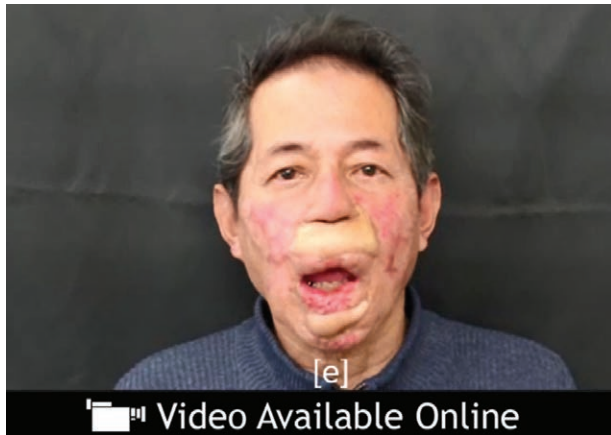
ing (Fig. 4; see video, **Supplemental Digital Content 1**, which shows a reconstructed oral sphincter function. Speech, ballooning of his cheeks, and smiling are the provided examples, <http://links.lww.com/PRSGO/A536>).

DISCUSSION

Reports of simultaneous upper and lower lip reconstruction are rare. Local flaps are useful for reconstructing sensate lips. However, they tend to result in tight lips



Fig. 4. After 1 year after operation. No lip tightness or aperture restriction was seen, and symmetry had been achieved. The new lips exhibited complete sensory recovery. Drooling was minimal during rest and feeding.



Video Graphic 1. See video, Supplemental Digital Content 1, which shows a reconstructed oral sphincter function. Speech, ballooning of his cheeks, and smiling are the provided examples, <http://links.lww.com/PRSGO/A536>.

due to a lack of tissue volume.^{6,7} For large defects, free flaps are usually selected.^{8–10} The use of a free radial forearm or anterolateral thigh flap combined with tendon or free muscle flap transfer might be the best reconstructive option for such cases at present.^{9,10} Fortunately, our patient had retained his oral commissure function. If it is possible to use an innervated local muscle, such as a gate flap,¹¹ to reconstruct the orbicularis oris muscle, oral competence might be preserved to a greater extent than can be achieved with previously reported methods.^{9,10} As reported previously, the cheek skin of PF patients is heavily scarred so local flaps are difficult to use because of the associated loss of elasticity.⁴

Therefore, we selected a method that combined the advantages of local and free flaps.

We consider that our method is superior to those described in previous studies, in that the restoration of lip sensation and oral sphincter function can be achieved to some extent in 1 stage while preserving the oral commissure (see video, Supplemental Digital Content 1, which shows a reconstructed oral sphincter function. Speech, ballooning of his cheeks, and smiling are the provided examples, <http://links.lww.com/PRSGO/A536>).

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PATIENT CONSENT

The patient provided written consent for the use of his image.

REFERENCES

1. Chalmers E, Cooper P, Forman K, et al. Purpura fulminans: recognition, diagnosis and management. *Arch Dis Child.* 2011;96:1066–1071.
2. Andreasen TJ, Green SD, Childers BJ. Massive infectious soft-tissue injury: diagnosis and management of necrotizing fasciitis and purpura fulminans. *Plast Reconstr Surg.* 2001;107:1025–1035.
3. Tanosaki M, Shimizu N, Lian CG, et al. Purpura fulminans managed with multi-limb amputation: substituted judgment and surrogate decision-making in the surgical management of necrotizing soft tissue infections. *Surg Infect (Larchmt).* 2014;15:853–856.
4. Urushidate S, Yokoi K, Higuma Y, et al. Nose and upper lip reconstruction for purpura fulminans. *J Plast Reconstr Aesthet Surg.* 2012;65:252–255.
5. Karonidis A, Yao SF. Chimeric anterolateral thigh free flap for head and neck reconstruction. *J Plast Reconstr Aesthet Surg.* 2009;62:e85–e86.
6. Madaree A, McGibbon IC, Morris WM. Reconstruction of both upper and lower lips. *J Craniomaxillofac Surg.* 1993;21:168–171.
7. Nath S, Jovic G. Total loss of upper and lower lips: challenges in reconstruction. *Br J Oral Maxillofac Surg.* 1998;36:460–461.
8. Jallali N, Malata CM. Reconstruction of concomitant total loss of the upper and lower lips with a free vertical rectus abdominis flap. *Microsurgery.* 2005;25:118–120.
9. Lai CL, Ou KW, Chiu WK, et al. Reconstruction of the complete loss of upper and lower lips with a chimeric anterolateral thigh flap: a case report. *Microsurgery.* 2012;32:60–63.
10. Ueda K, Oba S, Ohtani K, et al. Functional lower lip reconstruction with a forearm flap combined with a free gracilis muscle transfer. *J Plast Reconstr Aesthet Surg.* 2006;59:867–870.
11. Fujimori R. “Gate flap” for the total reconstruction of the lower lip. *Br J Plast Surg.* 1980;33:340–345.