Formosan Journal of Surgery (2016) 49, 27–30

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-fjs.com

CASE REPORT

Autologous fat grafting for treating lipoatrophy secondary to lupus erythematosus panniculitis

Hsiao-Peng Huang, Yung-Chia Huang, Yuan-Sheng Tzeng, Chih-Hsin Wang, Tim-Mo Chen, Shyi-Gen Chen*

Division of Plastic Surgery, Department of Surgery, Tri-Service General Hospital, National Defense Medical Center, Taipei, Taiwan

Received 16 February 2015; received in revised form 13 March 2015; accepted 31 August 2015
Available online 14 February 2016

KEYWORDS
lupus erythematosus panniculitis;
lupus profundus;
fat grafting

Abstract Lupus erythematosus panniculitis (LEP) is an uncommon variant of LE and accounts for only 1–3% of cutaneous LE cases. LEP lesions often heal with cutaneous scarring and lipoatrophy resulting in disfigurement. Studies regarding the treatment of permanent lipoatrophy and disfigurement caused by LEP are lacking in the literature. Here, we report our experience with a rare case of lipoatrophy caused by LEP that was treated using autologous fat grafting. A 41-year-old woman presented with indurated, flesh-colored, depressed plaques on both buttocks. After a series of investigations, LEP was diagnosed by a rheumatologist. Magnetic resonance imaging (MRI) showed a markedly decreased volume of the subcutaneous fatty layer and thickening over the covering cutis. After the inflammatory disease was controlled, we attempted to restore her body contour with autologous fat grafting. We arranged an MRI study after a follow-up period of 12 months. The volume of subcutaneous fat was calculated by integrating cross-sectional area data from consecutive images. The patient was satisfied with her body contour following the injection of 350 mL of fat into the right buttock and 50 mL into the left buttock in a two-stage procedure. No complications were observed following the procedure. Follow-up laboratory results were negative for anti-double-stranded DNA antibody and showed normal complement levels. After a follow-up period of 12 months, no nodules or disease reactivation was noted. The MRI showed a marked improvement in the volume of the subcutaneous fat of the buttock, and the survival ratio of transferred fat was calculated to be 65%. Lipoatrophy secondary to LEP is a rare disease that can cause distress to patients. Autologous fat grafting is a simple, fast, and effective method for alleviating depression deformities in

Conflicts of interest: None declared.

* Corresponding author. Division of Plastic Surgery, Department of Surgery, Tri-Service General Hospital, National Defense Medical Center, No. 325, Section 2, Chenggong Road, Neihu District, Taipei City 114, Taiwan.
E-mail address: rainscot@hotmail.com (S.-G. Chen).

http://dx.doi.org/10.1016/j.fjs.2015.12.002

patients with LEP. It has a potentially long-lasting effect in treating patients with permanent lipoatrophy and disfigurement caused by LEP but should be preferably performed only in patients with quiescent disease.

1. Introduction

Lupus erythematosus panniculitis (LEP) is an uncommon variant of LE and accounts for only 1—3% of cutaneous LE cases.[1—3] LEP is clinically characterized by erythematous or flesh-colored deep nodules in an inflammatory phase and generally occurs on the face, arms, legs, trunk, and abdominal regions. Although various medical treatments have been used for treating LEP,[4] disfigurement is frequently observed because of lipoatrophy.

Few reports regarding the treatment of permanent lipoatrophy and disfigurement caused by LEP currently exist in literature. Eastham et al[5] used hyaluronic acid and poly-L-lactic acid for soft tissue augmentation in cases of extensive LEP-induced facial atrophy. However, the effect was nonpermanent, and the material used in the augmentation was suspected to cause the reactivation of connective tissue disease.

The fat grafting technique is currently applied for both cosmetic and reconstructive purposes. The procedure is used for breast reconstruction, facial rejuvenation, body contouring, and correction of posttraumatic and postsurgical deformities.[6] However, data regarding fat grafting in LEP are limited. Valdatta et al[7] injected autologous fat into a patient with LEP with facial lipoatrophy in a three-stage procedure and reported satisfactory results during a follow-up period of 12 months. Gleeson et al[8] treated facial lipoatrophy in three patients with LEP by using fat grafting but reported that one of the patients experienced an acute fatal fat embolism following the procedure.

Here, we report our experience regarding a rare case of lipoatrophy caused by LEP treated using fat grafting.

2. Case report

A 41-year-old woman with a 6-month history of a depression over both buttocks caused by LEP was referred for treatment. Cutaneous examination revealed indurated, flesh-colored, depressed plaques with a severe hollow appearance of the right buttock (Figure 1). Magnetic resonance imaging (MRI) showed a markedly decreased volume of the subcutaneous fatty layer and thickening of the covering cutis (Figure 2). Physical examination and laboratory tests revealed arthritis, malar rash, oral ulcer, antineutrophil antibody (1:160, nucleolar pattern), and anti-double-stranded DNA antibody positivity (14.3 IU/mL; normal range, 0.0—10.0 IU/mL). A rheumatologist diagnosed LEP according to the 1997 revised criteria for classifying systemic LE. Inflammatory disease was controlled for 3 months by using a combination of sulfasalazine, prednisolone, and azathioprine. The patient requested restoration of body contour.

She underwent autologous fat grafting using the Coleman technique. In the first grafting attempt, we harvested subcutaneous fat from both lateral thighs by using aspiration cannulas 2 mm in diameter. The aspirated fluid was centrifuged at 604*g* (CN-820 simple type centrifuge, Hsiangtai Machinery Industry Co. Ltd., Taiwan) for 2 minutes. The fat tissue was then transferred to a 3-cm^3 Luer lock syringe (Terumo Syringe 5 cc; Medical Co.,Tokyo, Japan) for injection. We injected 200 mL of fat into the depressed area of the right buttock and 20 mL into the left buttock to correct the deformity. Six months later, the lesion showed marked improvement, particularly on the

Figure 1 Indurated, flesh-colored, depressed plaque with more severe depression on the right buttock in our patient.

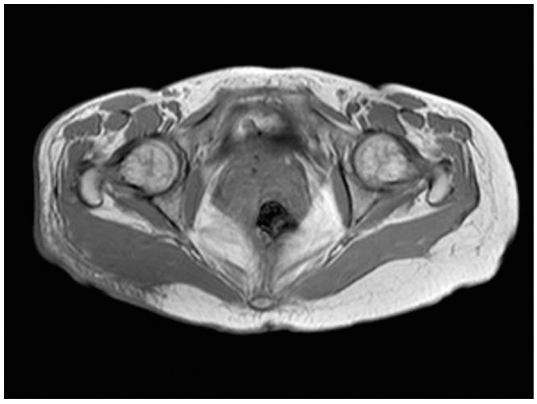

Figure 2 Magnetic resonance imaging showing significantly decreased volume of the subcutaneous fatty layer and thickening of the covering cutis.

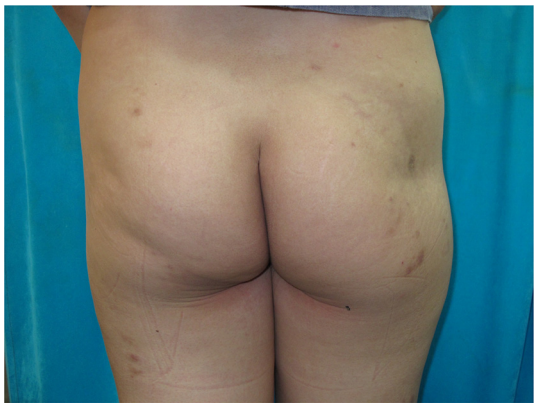

Figure 3 Six months after two fat grafting procedures.

right buttock. However, because a depressed body contour was still noted, she asked for a repeat procedure. We harvested fat from the bilateral flank and injected 150 mL into the right buttock and 30 mL into the left buttock. We arranged an MRI study after a follow-up period of 12 months. The volume of subcutaneous fat was calculated by integrating cross-sectional area data from consecutive images.

The patient was satisfied with her body contour after the injection of 350 mL of fat into the right buttock and 50 mL into the left buttock in a two-stage procedure. No complications were observed following the procedure. Follow-up laboratory examination results were negative for anti-double-stranded DNA antibody and showed normal complement levels. After a follow-up period of 6 months, no nodules or disease reactivation was noted (Figure 3). A significant improvement in the volume of the subcutaneous fat of the buttock was noted in the MRI taken 12 months after surgery (Figure 4), and the survival ratio of transferred fat was 65% by calculation.

3. Discussion

Compared with other LE forms, LEP is more frequent in women. The average age of onset (from late 30s to early 40s) of LEP is similar to that of other chronic cutaneous LE forms. LEP has a chronic relapsing clinical course. Patients develop tender subcutaneous nodules or plaques, most commonly on the proximal extremities (face, shoulders, upper arms, breasts, hips, and buttocks) but rarely on the distal extremities.[9] Following the nodular stage, these inflammatory lesions often heal with cutaneous scarring and lipoatrophy that lead to disfigurement. Chronic inflammation and hyaline necrosis of the subcutaneous tissue can be seen histologically.[10] An autoimmune etiology is confirmed by the presence of antinuclear antibodies and low complement levels. LEP with changes consistent with those in discoid LE is sometimes referred to as lupus profundus.[11] Other possible complications include alopecia, enophthalmos, central retinal artery occlusion, mastitis, thrombophlebitis, and proptosis.[12] Despite its benign characteristics, lipoatrophy can affect body shape and appearance, thus affecting the psychological aspect in daily life.

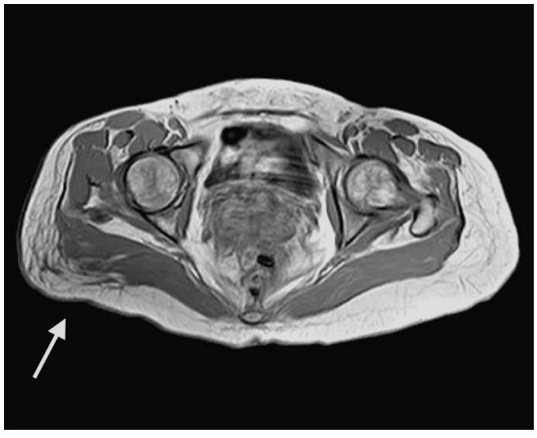

Figure 4 A distinct improvement is shown on magnetic resonance imaging 12 months after surgery. The volume of subcutaneous fat in the buttock has increased markedly.

Dermal fillers have been used for treating disfigurement caused by LEP,[5] but neither permanent nor nonpermanent fillers have shown promising results.[13] Problems exist regarding the use of dermal fillers in patients with connective tissue disease. Because of the theoretical risk of the reactivation of connective tissue disease as a result of antigenic stimulation,[14] complications such as granulomatous "rubberizing" reaction can occur, but are rare.[15]

Here we report on a patient with LPE who underwent a staged fat grafting procedure with a 35% loss of transferred fat after 12 months. Fat grafting to treat three-dimensional defects of lipoatrophy has the following advantages: a simple surgical procedure, shorter convalescence period, readily available source, and potentially long-lasting effect. However, a study including 10 participants reported a 50% loss of transferred fat within 3 months of the procedure, with an additional 6% loss noted between 3 months and 6 months after the procedure; no fat loss was reported thereafter.[16] Therefore, a staged fat grafting procedure may be required to achieve the target volume.

Complications of fat grafting, such as infection, bruising, and hematoma or seroma formation, are rare. Similarly, the risk of fat embolism is believed to be low, and the incidence of fat embolism for large abdominal liposuctions is estimated to be 1.3 per 100,000 population.[17,18] Liposuction with tumescent anesthesia and fat reinjection are generally regarded as safe. However, Gleeson et al[8] described a patient with LEP who died as a result of fulminant fat tissue embolism after the grafting of a small volume (35 mL) of autologous fat for facial lipoatrophy. It appears that in cases with substantial lipoatrophy and subcutaneous scarring because of LEP, fat injection into the surrounding fibrotic tissue can facilitate the entry of fat into the venous circulation. Furthermore, because LEP is an inflammatory disease, the risk of inflammatory reaction is higher when fat and vascular endothelium come in contact. Whether fat grafting for patients with LEP is related to a higher risk of fat embolism syndrome requires in-depth study.

Although fat embolism is a rare complication of the fat grafting technique, it is the most severe and lethal. To avoid fat embolism syndrome, using a large, blunt cannula rather than a small, sharp cannula can prevent the perforation of the vascular wall, thus avoiding entry of fat into

the venous circulation. Epinephrine at the injection site induces vasoconstriction, thus making it more difficult to cannulate the vessel.[19] In addition, the surgeon should ensure that only minimal force is exerted. Aspiration should be performed before fat is injected to avoid contact with veins and arteries.[20]

Treatment for sequelae of panniculitis, including surgical techniques and filler injections, remains controversial in the active inflammatory phase of the disease because trauma itself may be an inciting factor; the lesions have a chronic, remitting course that can be exacerbated by trauma.[9] Corrective techniques may be considered for stable, noninflammatory atrophic plaques. However, controlling the disease before initiation of such treatments is crucial.[4]

In conclusion, lipoatrophy secondary to LEP is a rare but potentially disfiguring disease that can cause distress to patients. Autologous fat grafting is a simple, fast, and effective procedure for alleviating depression deformities. It has a potentially long-lasting effect in treating permanent lipoatrophy and disfigurement caused by LEP but should be performed only in patients with quiescent disease.

References

1. Arai S, Katsuoka K. Clinical entity of lupus erythematosus panniculitis/lupus erythematosus profundus. *Autoimmun Rev*. 2009;8:449—452.
2. Crowson AN, Magro C. The cutaneous pathology of lupus erythematosus: a review. *J Cutan Pathol*. 2001;28:1—23.
3. Strober BE. Lupus panniculitis (lupus profundus). *Dermatol Online J*. 2001;7:20.
4. Braunstein I, Werth VP. Update on management of connective tissue panniculitides. *Dermatol Ther*. 2012;25:173—182.
5. Eastham AB, Liang CA, Femia AN, Lee TC, Vleugels RA, Merola JF. Lupus erythematosus panniculitis-induced facial atrophy: effective treatment with poly-L-lactic acid and hyaluronic acid dermal fillers. *J Am Acad Dermatol*. 2013;69:e260—e262.
6. Gir P, Brown SA, Oni G, Kashefi N, Mojallal A, Rohrich RJ. Fat grafting: evidence-based review on autologous fat harvesting, processing, reinjection, and storage. *Plast Reconstr Surg*. 2012;130:249—258.
7. Valdatta L, Cherubino M, Tamborini F, Pellegatta I, Maggiulli F. A case of facial lipoatrophy secondary to lupus profundus managed with lipofilling technique. *Case Rep Dermatol Med*. 2012;2012:720518.
8. Gleeson CM, Lucas S, Langrish CJ, Barlow RJ. Acute fatal fat tissue embolism after autologous fat transfer in a patient with lupus profundus. *Dermatol Surg*. 2011;37:111—115.
9. Martens PB, Moder KG, Ahmed I. Lupus panniculitis: clinical perspectives from a case series. *J Rheumatol*. 1999;26:68—72.
10. Marzano AV, Tanzi C, Caputo R, et al. Sclerodermic linear lupus panniculitis: report of two cases. *Dermatology*. 2005;210:329—332.
11. Costner MI, Sontheimer RD, Provost TT. *Lupus Erythematosus. Cutaneous Manifestations of Rheumatic Diseases*. 2nd ed. Philadelphia: Lippincott Williams & Wilkins; 2004:15—64.
12. Hansen CB, Callen JP. Connective tissue panniculitis: lupus panniculitis, dermatomyositis, morphea/scleroderma. *Dermatol Ther*. 2010;23:341—349.
13. Carvalho Costa IM, Salaro CP, Costa MC. Polymethylmethacrylate facial implant: a successful personal experience in Brazil for more than 9 years. *Dermatol Surg*. 2009;35:1221—1227.
14. Vera-Lastra O, Medina G, Cruz-Dominguez Mdel P, et al. Human adjuvant disease induced by foreign substances: a new model of ASIA (Shoenfeld's syndrome). *Lupus*. 2012;21:128—135.
15. Lemperle G, Morhenn V, Charrier U. Human histology and persistence of various injectable filler substances for soft tissue augmentation. *Aesthetic Plast Surg*. 2003;27:354—366.
16. Hoerl HW, Feller AM. *Autologous Fat Volume Retention Evaluation by Magnetic Resonance Imaging*. New York: Marcel Dekker; 2001:31—42.
17. Teimourian B, Rogers 3rd WB. A national survey of complications associated with suction lipectomy: a comparative study. *Plast Reconstr Surg*. 1989;84:628—631.
18. Christman KD. Death following suction lipectomy and abdominoplasty. *Plast Reconstr Surg*. 1986;78:428.
19. Coleman SR. Avoidance of arterial occlusion from injection of soft tissue fillers. *Aesthet Surg J*. 2002;22:555—557.
20. Wang DW, Yin YM, Yao YM. Internal and external carotid artery embolism following facial injection of autologous fat. *Aesthet Surg J*. 2014;34:NP83—NP87.