Photodiagnosis and Photodynamic Therapy 27 (2019) 449–451

Contents lists available at ScienceDirect

Photodiagnosis and Photodynamic Therapy

journal homepage: www.elsevier.com/locate/pdpdt

Case report

Combination of electrocoagulation and photodynamic therapy for angiolymphoid hyperplasia with eosinophilia in the external ear

Lisha Wu[1], Fangfang Li[1], Wei Shi, Juan Su, Mingliang Chen, Xiang Chen, Shuang Zhao*

Department of Dermatology, Xiangya Hospital, Central South University, 87 Xiangya Road, Changsha, Hunan, 41008, China

ARTICLE INFO

Keywords:
Angiolymphoid hyperplasia with eosinophilia
Photodynamic therapy
Electrocoagulation

ABSTRACT

Angiolymphoid hyperplasia with eosinophilia (ALHE) is an inflamed vascular tumefaction of uncertain pathogenesis and is characterized by higher recurrence rates after surgical excision or other approaches. In the present study, we performed a new approach by combining electrocoagulation with photodynamic therapy (PDT) for the treatment of two ALHE patients. They had multiple treatments in the past and had poor therapeutic effects with relapse each time. After informed consent was obtained, electrocoagulation was used to remove the superficial lesions of ALHE, and the first session of PDT was immediately applied to the lesion. A total of three sessions of PDT were applied to each patient with an interval of 7–10 days. Through more than 12 months of follow-up, the two patients showed complete regression, and no recurrence was observed. Meanwhile, patients were very satisfied with the cost and cosmetic outcomes of the combination treatment. Based on our results, we strongly recommend the combination of electrocoagulation with PDT as a favourable treatment for ALHE, especially in the external ear and other areas that are inconvenient for routine surgery.

1. Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon, benign vascular proliferation. The aetiology of ALHE remains unknown. Clinical classification also remains unclear. The diagnosis of ALHE is usually based on clinical features and histopathological examination. ALHE is characterized by a proliferation of blood vessels and histiocytoid endothelial cells with hobnail appearance and a mixed inflammatory infiltrate consisting of lymphocytes, plasma cells, and eosinophils [1]. However, there is still a nonevidence-based treatment approach for ALHE based on retrospective reports and case studies [2s]. To date, surgical excision is considered the most effective therapy for ALHE, and it still has a recurrence rate of up to 40.8% [2]. Meanwhile, many approaches to treat ALHE have been tried, including laser treatment, cryotherapy, local injection and so on. Regardless of the treatment procedures, recurrence and incomplete resolution of ALHE remain frequent. There is a recurrence rate of more than 40% [1]. In the particular location of the external ear, there is no good treatment [3].

Photodynamic therapy (PDT) has been widely used in a variety of cutaneous disorders, including actinic keratoses, viral warts, psoriasis and some non-melanoma skin cancers [4]. The advantages of PDT include its comparatively less invasiveness to surgical procedures, a low recurrence rate, and good cosmetic outcomes. However, the limitations for topical PDT might be inadequate penetration of topically applied photosensitizers through tissue or insufficient local bioavailability and, hence, decrease cellular uptake [5]. In the present study, the main purpose of pretreatment with electrocoagulation was to increase the topical penetration of PDT photosensitizers in ALHE lesions. The results showed that the combination treatment had an excellent effect and no recurrence after 12 months of follow-up. This is the first reported case using the combination of electrocoagulation with PDT for the treatment of ALHE in the external ear.

2. Case report

A 21-year-old woman and a 55-year-old man were referred with more than a one-year history of red itchy papules and nodules in the posterior auricular area and external auditory canal that had gradually increased in size and number. The diagnosis of patients was also biopsy proven as AHLE (Fig. 1). The patients have still complained of intense and recurrent itching, even though they had received a variety of treatments in the past, such as intralesional or topical corticosteroids, carbon-dioxide laser therapy and so on.

After routine disinfection, standard bipolar electrocoagulation (CV-2000A, Beijing Kangwei Electronics & Technology Co., Ltd., China) was first performed to remove visible lesions. Briefly, the energy settings

* Corresponding author.
E-mail address: shuangxy@csu.edu.cn (S. Zhao).
[1] Their contribution to this article is equal.

https://doi.org/10.1016/j.pdpdt.2019.07.020
Received 23 March 2019; Received in revised form 28 June 2019; Accepted 26 July 2019
Available online 27 July 2019

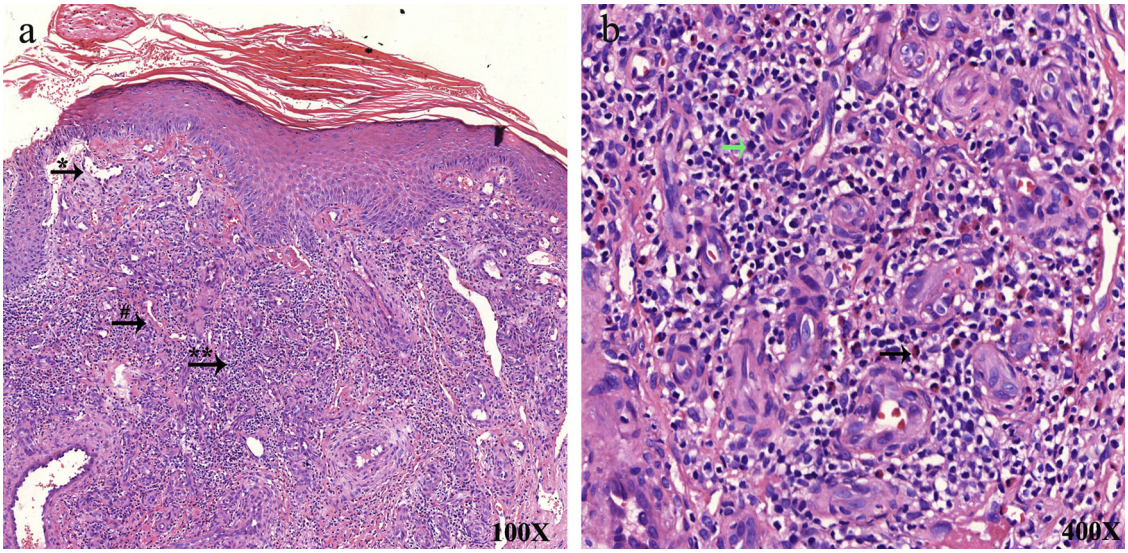

Fig. 1. Representative histological images stained with haematoxylin-eosin (HE) at original magnification × 100 (a) and × 400 (b). The results show a proliferation of blood vessels (#) and histiocytoid endothelial cells with a hobnail appearance (*). Meanwhile, the mixed inflammatory infiltrates consist of lymphocytes (**), plasma cells (as green arrow indicated) and eosinophils (as black arrow indicated).

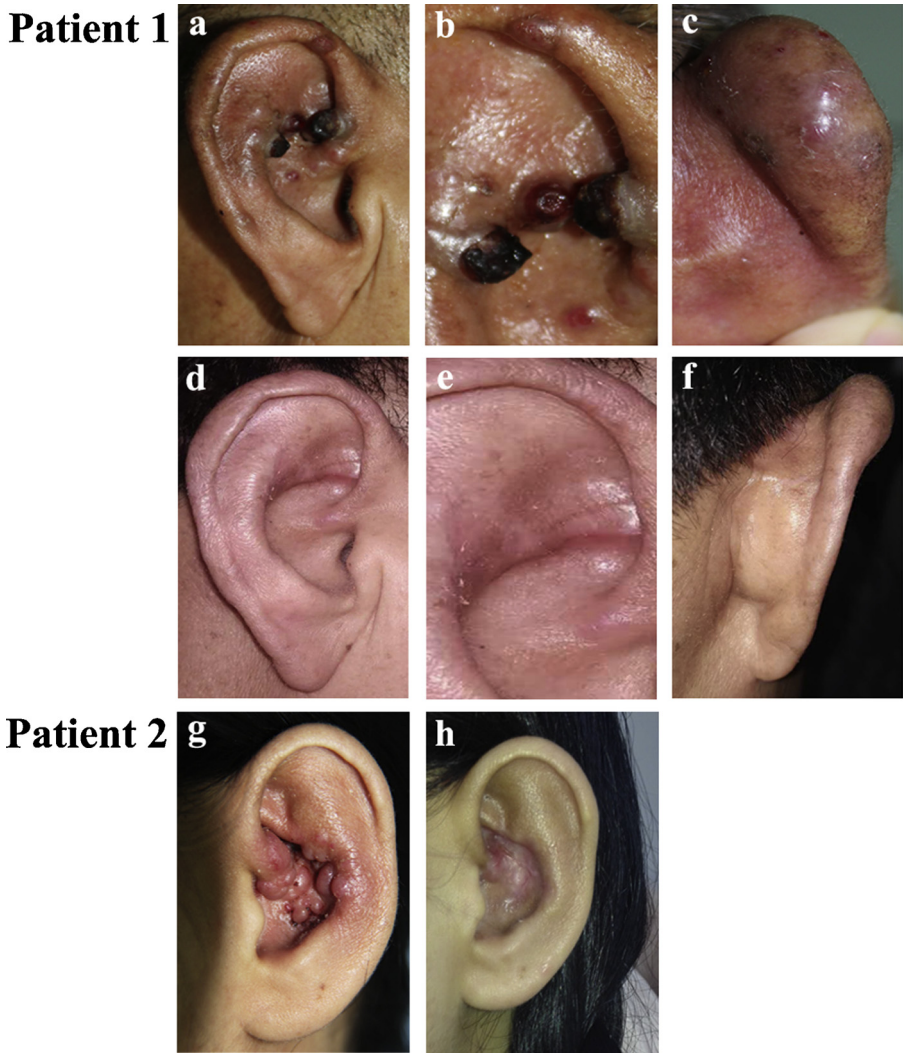

Fig. 2. The clinical presentation after pre-treatment with electrocoagulation. Multiple papules or nodules on the external ear (a) and periauricular area (c) are associated with mild erosions, exudation, and scabs (b). (d, e, f) The same positions presented after one-month treatment in a 55-year-old man. Clinical photos from a 21-year-old woman before (g) and after (h) the combined treatment.

were 15 W, the duration of therapy was 2–3 seconds once, and electrocoagulation was performed several times until all of the visible lesions were removed. The process of electrocoagulation should be executed softly and carefully, with removal of just the superficial lesion above the epidermis. Then, we cleaned all the scabs, and immediately, the first session of PDT was applied to the lesion. Briefly, the electrocoagulated wound was covered with gauze containing 20% 5-aminolevulinic acid (ALA, Shanghai Fudan-Zhangjiang Bio-Pharmaceutical Co., Ltd., China) in saline solution for 3 h, and the coverage area of ALA was extended by an approximately 1-cm margin compared with the electrocoagulation wound. After removing the gauze with ALA solution, we performed irradiation with a 635-nm **red**-light laser (80 mW/cm^2) for 20 min (I series 'power' LED-IB (cylinder), Wuhan Yage Optic and Electronic Technique Co., Ltd., China) (Fig. 2). A total of three sessions of PDT were conducted for each patient, each with a time interval of 7–10 days. Erythromycin ointment or iodine complex disinfection solution was used for routine wound management after the operation. The symptoms of itching were completely relieved, and no recurrence was observed after more than 12 months of follow-up. Meanwhile, the Dermatology Life Quality Index (DLQI) assessment showed scores of 23.5 before the surgery and 4.5 after 12 months of follow-up. Importantly, the two patients were very satisfied with the cost and cosmetic outcomes achieved with the combination treatment (Fig. 2).

3. Discussion

To date, many approaches have been used to treat ALHE, such as conventional therapies, including steroids, isotretinoin, cryotherapy, laser therapy or surgical excision, as well as therapies aimed to treat new targets, such as IL-5 (mepolizumab), VEGF-A (bevacizumab) and IFNα-2a [1,2,6,7]. However, the issues of recurrence and incomplete resolution of the disease remain unresolved regardless of the treatment used. According to previous studies [1,2], there was up to 100% failure rate for isotretinoin treatment, and the failure rates of topical, systemic or intralesional corticosteroid approaches were up to 98.2%, 87.8%, and 79.1%, respectively. Even cryotherapy therapy still has an 80.5% failure rate. Comparatively, a lower failure rate has been observed following surgical excision (40.8%) and pulsed dye laser treatment (50.0%). In addition, the most common location for ALHE was the head and neck region, especially the ear and periauricular area (36.3%) [2]. These locations are inconvenient for routine surgery, resulting in poor treatment effect, recurring repeatedly or possibly leading to poor cosmetic outcomes. Thus, the greatest difficulty is achieving complete remission for ALHE patients.

Both ALHE patients in our report had been previously treated with multiple methods, and they still suffered from repeated recurrence. Through the combination electrocoagulation with PDT, both patients achieved complete remission and good cosmetic outcomes. Consistent with our results, Sotiriou E et al. also believed that PDT could be an alternative therapeutic approach for AHLE or used as a neoadjuvant treatment to reduce AHLE lesion size, especially for cases in which the size or site of lesions limits the efficacy or acceptability of other treatments [8]. However, different from that previous report, we performed electrocoagulation before the first session of PDT. Two patients showed complete regression and good cosmetic outcomes through more than 12 months of follow-up. We considered that the procedure may reduce the thickness of ALHE lesions via electrocoagulation, and then the increased penetration ability of the photosensitizer could efficiently augment PDT depth and achieve an excellent therapeutic effect. PDT has additional benefits, including the elimination of potentially invisible lesions, promotion of wounding heals and reduction of scar formation. In addition, PDT can regulate the immune response, resulting in a low recurrence rate and fewer adverse effects [5,9]. Moreover, the procedure is comparatively simple, as bipolar electrocoagulation probes are common devices that are easy to operate. Future studies are needed to explore the exact mechanisms, as well as to expand the treatment to more ALHE patients.

This is the first study to treat ALHE patients by the combination of electrocoagulation with PDT. Based on our successful experience and the fact that this approach is a relatively simple procedure, we highly have recommended this combination treatment for ears and other body parts that are inconvenient for routine surgery.

Funding sources

The National Natural Science Foundation of China (Grant no. 81430075).

Declaration of Competing Interest

None to declare.

References

[1] R. Guo, A.C. Gavino, Angiolymphoid hyperplasia with eosinophilia, Archives of pathology & laboratory medicine 139 (5) (2015) 683–686.
[2] B.L. Adler, A.E. Krausz, A. Minuti, J.I. Silverberg, H. Lev-Tov, Epidemiology and treatment of angiolymphoid hyperplasia with eosinophilia (ALHE): a systematic review, J. Am. Acad. Dermatol. 74 (3) (2016) 506–512 e11.
[3] D. Yadav, S. Singh, N. Bhari, S. Gupta, Angiolymphoid Hyperplasia of External Ear Treated With Intralesional Radiofrequency Ablation, BMJ Case Reports 2018, (2018).
[4] X. Wen, Y. Li, M.R. Hamblin, Photodynamic therapy in dermatology beyond non-melanoma cancer: an update, Photodiagn. Photodyn. Ther. 19 (2017) 140–152.
[5] U. Keyal, A.K. Bhatta, G. Zhang, X.L. Wang, Present and future perspectives of photodynamic therapy for cutaneous squamous cell carcinoma, J. Am. Acad. Dermatol. 80 (3) (2019) 765–773.
[6] E. Bahloul, M. Amouri, S. Charfi, O. Boudawara, H. Mnif, T. Boudawara, H. Turki, Angiolymphoid hyperplasia with eosinophilia: report of nine cases, Int. J. Dermatol. 56 (12) (2017) 1373–1378.
[7] T. Isohisa, K. Masuda, N. Nakai, H. Takenaka, N. Katoh, Angiolymphoid hyperplasia with eosinophilia treated successfully with imiquimod, Int. J. Dermatol. 53 (1) (2014) e43–4.
[8] E. Sotiriou, Z. Apalla, A. Patsatsi, D.D. Panagiotidou, D. Ioannides, Angiolymphoid hyperplasia with eosinophilia: good response to photodynamic therapy, Clin. Exp. Dermatol. 34 (8) (2009) e629–31.
[9] S. Kwiatkowski, B. Knap, D. Przystupski, J. Saczko, E. Kedzierska, K. Knap-Czop, J. Kotlinska, O. Michel, K. Kotowski, J. Kulbacka, Photodynamic therapy - mechanisms, photosensitizers and combinations, Biomedicine & pharmacotherapy = Biomedecine & pharmacotherapie 106 (2018) 1098–1107.