

Spontaneous symmetrical giant keloids at the bilateral labia majora: a case report

Journal of International Medical Research

48(4) 1–6

© The Author(s) 2019

Article reuse guidelines:

sagepub.com/journals-permissions

DOI: 10.1177/0300060519891664

journals.sagepub.com/home/imr

WenChao Zhang¹, Xiaojun Wang¹,
Jiuzuo Huang¹, Wenfang Dong² and
Xiao Long¹ 

Abstract

Keloids are caused by an imbalance between collagen matrix decomposition and production during wound repair. Spontaneous keloids are a rare type of keloid that arise without a significant history of trauma or surgery. We herein report a case involving a 59-year-old woman with symmetric neoplasms at the bilateral labia majora that had persisted for about 3 years and a >10-year history of pruritus and pain at the bilateral labia majora with folliculitis. The bilateral labia majora gradually swelled and lost their normal physiological morphology. The patient was diagnosed with keloids. Under general anesthesia, the keloids were resected. The right incision was sutured directly and the left was closed using an inguinal flap. Adjuvant radiotherapy was administered after the surgery. No recurrence of the keloids was observed at the 1-year follow-up, and the patient was satisfied with the appearance of the vulvar region. This is the first report of spontaneous symmetrical giant keloids at the bilateral labia majora, and it emphasizes the importance of the effect of abnormal inflammation on keloid formation. Repairing genital defects using an inguinal flap has only a minor effect on the patient's postoperative movement, and an ideal vulvar appearance can be achieved.

Keywords

Keloid, perineal region, labia majora, surgery, radiotherapy, inguinal flap

Date received: 16 August 2019; accepted: 7 November 2019

Introduction

Keloids are caused by an imbalance between collagen matrix decomposition and production during wound repair.¹ Keloids mainly develop at locations with high regional tension and overactive sebaceous glands, such as the chest, back, or

¹Department of Plastic Surgery, Peking Union Medical College Hospital, Beijing, China

²Department of Plastic Surgery, The Twelfth Department of Plastic Surgery, Plastic Surgery Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing, China

Corresponding author:

Xiao Long, Department of Plastic Surgery, Peking Union Medical College Hospital, No. 1 Shuaifuyuan, Dongcheng District, Beijing 100730, China.

Email: longxiaopumch@163.com



Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative

Commons Attribution-NonCommercial 4.0 License (<https://creativecommons.org/licenses/by-nc/4.0/>) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (<https://us.sagepub.com/en-us/nam/open-access-at-sage>).

mandibular margins; however, they rarely invade mucosal tissue.² Spontaneous keloids are a rare type of keloid that develop without a significant history of trauma or surgery, and the main cause is thought to be closely related to micro-trauma or minimal cutaneous inflammation. According to previous reports, spontaneous keloids are not only associated with certain syndromes but can also develop in people with atopy and after letrozole and isotretinoin therapy.³ Previous studies have revealed that perineal keloids mainly occur at the mons pubis.⁴ We herein report a rare case of spontaneous symmetric giant keloids at the bilateral labia majora, which has not been reported previously.

Case report

A 59-year-old woman visited our hospital with the chief complaint of symmetric neoplasms on the bilateral labia majora that had been present for about 3 years and were occasionally accompanied by pain and itching. The patient also had a >10-year history of pruritus and pain at the bilateral labia majora with folliculitis, without a significant history of trauma or surgery. The protracted presence of these symptoms resulted in gradual swelling of the bilateral labia majora. Other than

antibiotic treatment, the patient received no other therapy for the keloids, such as laser treatment, intralesional steroid injection, or surgery. Physical examination revealed obvious swelling of the bilateral labia majora and loss of the normal physiological morphology, while the bilateral labia minora showed no obvious abnormalities (Figure 1). The patient had a history of two cesarean deliveries, and the original surgical incision scars were slightly hyperplastic but without keloid formation. Furthermore, the patient had no keloids anywhere else on the skin, and her family members had no history of keloids.

Considering that other treatment methods would need to be repeated several times, the patient decided to undergo surgery directly. Under general anesthesia, the keloids were totally resected from the bilateral labia majora. The right incision was sutured directly. However, the left wound was too large to be sutured directly; therefore, this wound was repaired using an inguinal flap. The postoperative pathologic examination showed that the vulvar lesion consisted of scar tissue with coarse collagen fibers and scattered abscesses (Figure 2).

Hypofractionated electron-beam radiation at 6 MeV was performed twice within 24 hours and 1 week after the operation with a dose of 9 Gy each time, and the



Figure 1. (a) Physical examination revealed obvious swelling of the bilateral labia majora and a loss of normal physiological morphology, while the bilateral labia minora showed no obvious abnormalities. (b, c) After resection of the keloids, the right incision was sutured directly and the left was repaired using an inguinal flap. The surgical scar was not obvious, and an ideal vulvar appearance was achieved.

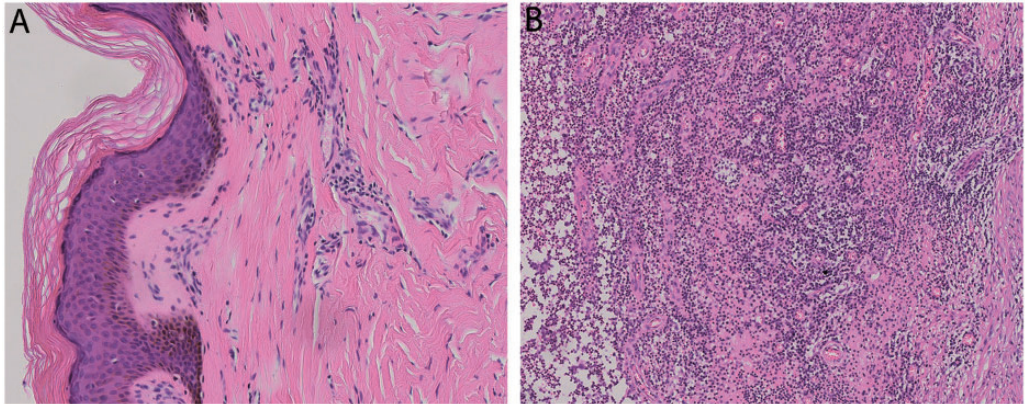


Figure 2. Hematoxylin and eosin staining. The postoperative pathological examination results showed that the vulvar lesions consisted of (a) scar tissue with coarse collagen fibers and (b) scattered abscesses.

margin of the radiation field was 1 cm around the treated area.

The patient had no postoperative limitation of movement, and all operative incisions healed well without effusion, hematoma formation, or wound dehiscence. The stitches were removed 14 days after the operation. The vulva was symmetric in appearance without obvious deformity, and the patient's symptoms of pruritus and pain were significantly relieved (Figure 1).

The patient's permission was obtained for the use of her photographs, and all identifying information has been excluded. Additionally, the patient was provided written informed consent for the publication of all anonymized data related to her treatment. No approval by an ethics committee or institutional review board was required because this article describes a rare case of a common disease, all treatments were routinely performed according to established guidelines, and no treatment trial was attempted.

Discussion

Keloids are caused by excessive proliferation of collagen matrix triggered by cutaneous injury, which commonly occurs after

trauma, burns, surgery, or inflammation.⁴ Keloids can not only be aesthetically displeasing but are also accompanied by symptoms of pain, itching, and functional disability, which significantly affect the patient's quality of life.⁵ Unlike hypertrophic scars, keloids grow beyond the boundaries of the original wound and do not spontaneously regress over time. Thicker eosinophilic (transparent) collagen bundles, also known as "keloid collagen," can be found in keloids through pathological examination.² Common predilection sites include the back, chest, auricle, and pubic area. Keloids rarely develop on soft skin and mucosal areas, such as the eyelids and labia. Jones et al.⁴ reported of a case of huge keloids on the perineum in 2006. However, spontaneous symmetric giant keloids on the bilateral labia majora have not been previously reported.

Proinflammatory factors, such as interleukin 1 α , interleukin 1 β , interleukin 6, and tumor necrosis factor α , are found at significantly higher levels in keloid tissue than normal tissue. Ogawa² reported that unlike other skin masses, keloids are associated with abnormal inflammatory responses in the skin, especially in the dermal reticular layer. The patient in the

present case had no history of trauma involving the vulva, and the pathological examination showed that the vulvar lesion consisted of scattered abscesses, further confirming that the onset of keloids is strongly associated with local recurrent folliculitis. The pathological results also supported the notion that the formation of keloids is associated with local abnormal inflammatory responses. Considering that the patient had a >10-year history of inguinal folliculitis, we could not overlook hidradenitis suppurativa (HS) in this case. HS is a chronic, relapsing, inflammatory skin disease that is often found in areas containing apocrine glands, such as the axillae, groin, perineum, and perianal region, and its clinical features include persistent or recurrent flares of inflamed painful nodules, sinus tracts, and irregular scar hyperplasia. Several cases of keloid formation in HS wounds have been reported.⁵

The treatment of keloids includes steroid injections, radiotherapy, cryotherapy, laser therapy, pressure therapy, and surgical resection. Intralesional steroid injection is the first-line treatment for early keloids, and triamcinolone is a commonly used steroid.⁶ The treatment interval is 4 to 6 weeks, and the recommended dose ranges from 10 to 40 mg/mL of triamcinolone.⁷ The effectiveness of the steroid injection ranges from 50% to 100% in different populations with a relatively high recurrence rate of 9% to 50%.⁸ Pulsed dye laser therapy was the first studied laser treatment method for keloids, and its mechanism mainly involves the induction of collagen remodeling, a decrease in cellular activity, and a reduction in the production of transforming growth factor.⁹ Multiple studies have proven that pulsed dye laser therapy for keloids can achieve an obvious reduction in erythema with approximately 50% to 70% improvement of keloids.¹⁰⁻¹³ No consensus has yet been reached on the best treatment method for keloids. Several

of the above-mentioned treatments must often be combined to achieve a better therapeutic effect according to the individual patient's specific condition. Bu et al.¹⁴ proved that surgical resection combined with postoperative radiotherapy can significantly reduce the volume and recurrence of keloids, while simultaneous application of postoperative photodynamic therapy can further improve the efficacy.

For large and refractory keloids, surgical resection combined with radiotherapy is a very effective and reliable method of treatment.¹⁵⁻¹⁸ Shen et al.¹⁹ retrospectively analyzed 834 cases of keloid lesions in 568 patients and reported that large-segment electron-beam radiation (18 Gy/2 fr, 1 week apart) postoperatively (within 48 hours) can reduce the recurrence rate of keloids to 11.75%. For the patient in the present case study, after total removal of the bilateral labia majora keloids, the right incision was sutured directly and the left was repaired using an inguinal flap. The patient had no limitation in movement, and the surgical scar was not obvious. Certainly, other reconstruction options such as skin grafting or anterolateral thigh flaps can also be used. However, considering the difficulty of surgery and the impact of postoperative radiotherapy, and to allow the patient to get up and move around as soon as possible, we chose to use an inguinal flap to repair the genital defect.

The present case is valuable because it highlights the following points:

1. Perineal keloids mainly occur on the mons pubis and rarely invade mucosal tissue. This is the first report of spontaneous symmetric giant keloids occurring on the bilateral labia majora.
2. The cause of the keloids in this patient was recurrent folliculitis on the bilateral labia majora, and the pathological results further support the notion that the formation of keloids is strongly

associated with local abnormal inflammation, especially in the dermal reticular layer.

3. Repairing genital defects using an inguinal flap barely affects patient postoperative movement, and an ideal vulvar appearance can be achieved. It is an effective treatment method for perineal defects.
4. Surgical resection combined with hypofractionated high-energy electron-beam radiotherapy can effectively reduce the recurrence rate of severe keloids that respond poorly to nonsurgical treatment.


Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

ORCID iD

Xiao Long  <https://orcid.org/0000-0002-3409-2113>

References

1. Lee H and Jang Y. Recent understandings of biology, prophylaxis and treatment strategies for hypertrophic scars and keloids. *Int J Mol Sci* 2018; 19: pii: E711.
2. Ogawa R. Keloid and hypertrophic scars are the result of chronic inflammation in the reticular dermis. *Int J Mol Sci* 2017; 18: pii: E606.
3. Jfri A and Alajmi A. Spontaneous keloids: a literature review. *Dermatology* 2018; 234: 127–130.
4. Jones K, Fuller CD, Luh JY, et al. Case report and summary of literature: giant perineal keloids treated with post-excisional radiotherapy. *BMC Dermatol* 2006; 6: 7.
5. Jfri A, O'Brien E, Alavi A, et al. Association of hidradenitis suppurativa and keloid formation: a therapeutic challenge. *JAAD Case Rep* 2019; 5: 675–678.
6. Gold MH, McGuire M, Mustoe TA, et al. Updated international clinical recommendations on scar management: part 2—algorithms for scar prevention and treatment. *Dermatol Surg* 2014; 40: 825–831.
7. Gauglitz GG and Kunte C. [Recommendations for the prevention and therapy of hypertrophic scars and keloids]. *Hautarzt* 2011; 62: 337–346.
8. Robles DT, Moore E, Draznin M, et al. Keloids: pathophysiology and management. *Dermatol Online J* 2007; 13: 9.
9. Park JH, Chun JY and Lee JH. Laser-assisted topical corticosteroid delivery for the treatment of keloids. *Lasers Med Sci* 2017; 32: 601–608.
10. Koike S, Akaishi S, Nagashima Y, et al. Nd:YAG laser treatment for keloids and hypertrophic scars: an analysis of 102 cases. *Plast Reconstr Surg Glob Open* 2015; 2: e272.
11. Al-Mohamady Ael-S, Ibrahim SM and Muhammad MM. Pulsed dye laser versus long-pulsed Nd:YAG laser in the treatment of hypertrophic scars and keloid: a comparative randomized split-scar trial. *J Cosmet Laser Ther* 2016; 18: 208–212.
12. Annabathula A, Sekar CS and Srinivas CR. Fractional carbon dioxide, long pulse Nd:YAG and pulsed dye laser in the management of keloids. *J Cutan Aesthet Surg* 2017; 10: 76–80.
13. Nouri K, Jimenez GP, Harrison-Balestra C, et al. 585-nm pulsed dye laser in the treatment of surgical scars starting on the suture removal day. *Dermatol Surg* 2003; 29: 65–73.
14. Bu W, Fang F, Zhang M, et al. Combination of 5-ALA photodynamic therapy, surgery and superficial X-ray for the treatment of keloid. *Photodermatol Photoimmunol Photomed* 2019; 1–3. doi: 10.1111/phpp.12506. [Epub ahead of print]
15. van Leeuwen MC, Stokmans SC, Bulstra AE, et al. Surgical excision with adjuvant irradiation for treatment of keloid scars: a systematic review. *Plast Reconstr Surg Glob Open* 2015; 3: e440.
16. Xu J, Yang E, Yu NZ, et al. Radiation therapy in keloids treatment: history, strategy,

- effectiveness, and complication. *Chin Med J (Engl)* 2017; 130: 1715–1721.
17. Hoang D, Reznik R, Orgel M, et al. Surgical excision and adjuvant brachytherapy vs external beam radiation for the effective treatment of keloids: 10-year institutional retrospective analysis. *Aesthet Surg J* 2017; 37: 212–225.
 18. Ogawa R, Mitsuhashi K, Hyakusoku H, et al. Postoperative electron-beam irradiation therapy for keloids and hypertrophic scars: retrospective study of 147 cases followed for more than 18 months. *Plast Reconstr Surg* 2003; 111: 547–553; discussion 54–55.
 19. Shen J, Lian X, Sun Y, et al. Hypofractionated electron-beam radiation therapy for keloids: retrospective study of 568 cases with 834 lesions. *J Radiat Res* 2015; 56: 811–817.