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CASE REPORT

Management Reconstruction of Eyelid Kissing Nevus

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Abstract

Introduction: An unusual type of congenital nevus known as "kissing nevus" develops on one eye's upper and lower lid and becomes enormous while the eyelids are closed. Reconstruction is complex since it creates both aesthetic and functional issues. **Case Presentation:** A 23-year-old female patient attended our ophthalmology outpatient clinic stating that she had a painless, non-progressing black nevus covering her left upper and lower eyelid since birth. The dark brown-black, 55 mm in size, with hypertrichosis that extended to the cheek and covered about two-thirds of the left upper and lower eyelid edges. A free supraclavicular skin transplant restored the complete nevus shortly after removal. We performed canthoplasty, lateral canthopexy, and full-thickness graft procedures. Three weeks following the procedures, the result was satisfactorily proved. Through histopathological examination, intradermal nevus pigmentosus was identified. **Conclusions:** A secure, trustworthy, and aesthetically pleasing option for head and neck deformities reconstruction involves a free supraclavicular graft.

Keywords: kissing nevus; divided nevus; reconstruction; skin graft; congenital

Introduction

Congenital divided melanocytic nevus, also known as kissing nevus, is a congenital compound that affects equal areas of the upper and lower eyelids.[1] This uncommon form of melanocytic nevus affects the upper and lower eyelids unilaterally. When the eyelids are closed, the nevus appears to be "kissing" or "divided" as it extends beyond the edge of the lid. Although it is typically recognized at birth, there were situations in which it manifested later in life.[1],[2]

Approximately 1% of all neonates have congenital nevi, and the literature has reported fewer than 150 cases of kissing nevus or divided nevi.[1] It was described first by Fuchs in 1919; they are frequently in whites, with fewer cases in Asians and none reported in African descent.[1] The majority of lesions were < 1.5 cm in size. The lids typically emerge as ectodermal protrusions during the sixth week of intrauterine pregnancy, develop together, and fuse during the twenty-fourth week.[2] Subsequently, they progressively drift apart. Melanoblasts are derived from the neural crest and migrate to the epidermis between weeks 12 and 24 of gestation. A split, divided, or kissing nevus results from a failure in melanocyte migration or a Schwann cell of neuroectodermal origin during nevus formation. This disease has many treatments, like dermabrasion, cryotherapy, primary closure after excision, and skin grafts. Dermabrasion and cryotherapy were recommended for obliterating congenital melanocytic nevus if the lesion was treated early in life in the superficial dermis.[3],[4] However, if the lesion has been attached to the deep dermis and subcutaneous tissue, the suggested treatment is full-thickness excision and skin graft.[1],[2],[3],[4]

In this case, we describe an uncommon case of a kissing nevus of the eyelid and explore the best course of action for reconstruction.

Case presentation

A 23-year-old woman attended our ophthalmology outpatient clinic stating that she had a painless, non-progressive black nevus covering her left upper

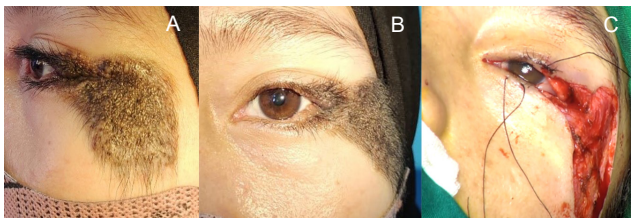

Figure 1. (A-B) A female with left kissing nevus of the eyelid; and (C) Intraoperative photo showing resection of the lesion and resulting defect.

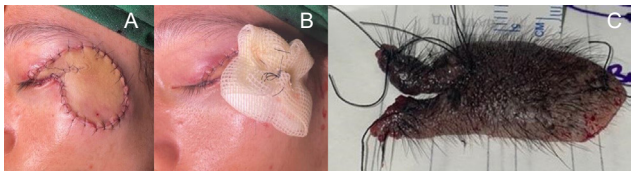

Figure 2. (A) Intraoperative photo showing reconstruction using free skin grafts; (B) Bolstering the skin graft; and (C) Entire excised nevus showed cystic lesion near lateral canthus and hypertrichosis.

and lower eyelid since birth. Firstly, the lesion in the upper and lower eyelid was small, however, had grown according to the patient's age and extended to the cheek. There was no complaint of blurred eye, ptosis, or pain. The patient had been performing laser two times, however, the complaint did not improve. The patient denied any trauma or surgery. There was no history of similar illness in the family and no history of congenital anomaly in the patient's family.

During the examination, we discovered a 55 x 30 mm dark brown-black nevus with hypertrichosis that covered about two-thirds of the left upper and lower eyelid edges and extended to the cheek. The ophthalmology examination showed visual acuity 5/5 on both eyes with 12 mmHg intraocular pressure (IOP). There was no pigmented cornea, sclera, and conjunctiva. The posterior segment was within the standard limit (Figure 1).

We planned to excise the nevus and perform reconstructive of the eyelid. After removal, a free supraclavicular skin graft was used to restore the complete nevus. We carried out canthoplasty, lateral canthopexy, and full-thickness graft procedures. During the operation, we discovered a 2 x 2 mm cyst (Figure 2). We immediately sent the lesion for histopathology, and the result showed that each of the lids was impaired by an intradermal compound nevus pigmentosus (Figure 3). Postoperatively, the graft survived well, and no complications were noted. Three weeks after the operation, satisfactory results were shown (Figure 4).

Discussion and conclusions

Nevi are frequently found on the eyelid margin. It can involve the upper and lower eyelid margin named kissing nevus. Its characteristic was molded to the ocular surface.[5] Asymptomatic benign nevi require no treatment, but malignant transformation of a junctional or compound nevus can occur in rare cases.[5],[6]

Kissing nevus is an uncommon congenital dermatological anomaly that develops on the edge of the upper and lower eyelids. It is also called divided nevus, split ocular nevus, and panda nevus.[1],[3],[4],[7] Melanocytic nevus is a benign proliferation of melanocytic cells arranged in the dermis, epidermis, or other tissue.[1],[3],[4],[7] It is a form of non-vascular epidermal nevus. Kissing nevus most frequently occurs on the eyelids; penis and fingers are less common sites.[1],[3],[4],[7]

Palpebral buds, or preliminary eyelids, develop above and below the cornea in the seventh week of pregnancy and measure 21 mm in length from top to bottom. The two eyelids meet at the ninth or tenth week of pregnancy (38-40 mm) when the epidermal layer merges without the mesenchymal layer. Lipids accumulate at the junctional zone during the 20th week when the eyelids experience a complete separation. They persist between the 28th and 30th weeks. Melanocytes from neural crest-derived melanoblasts migrate to the base of the embryonic epidermis of the scalp and face during weeks 12 and 14 of gestation.[3],[8],[9]

Fuchs (1919)[3] that he was the first to describe kissing nevus; it is more common in white people, with fewer cases in Asian people and none in those of African origin. The lesion was mostly less than 1.5 cm in size. It is unclear whether females are more likely than males to develop eyelid kissing nevi. Most lesions cause aesthetic concern from birth and expand slowly with age, becoming thicker, more darkly pigmented, and more similar to warts.[4]

Amblyopia due to deprivation may arise from a divided nevus of the eyelids when an infant reaches a particular size.[3],[4],[8],[9] The upper lid's mass covers the visual axis and induces mechanical ptosis. In addition, it may result in epicanthus, ectropion, and epiphora (from puncta compression). Patients with kissing nevus should be treated as soon as possible because of the side effects of divided nevus, which include ptosis and amblyopia. The first year or two of life is suggested for applying for the treatment. Patients who may have deprivation amblyopia need to start treatment earlier.[3],[4],[8],[9]

Corneal injury may result from the lesion's expansion outside the lid margin. The classification system for congenital nevus is according to the size of the nevus and histological type. The first classification based on the size

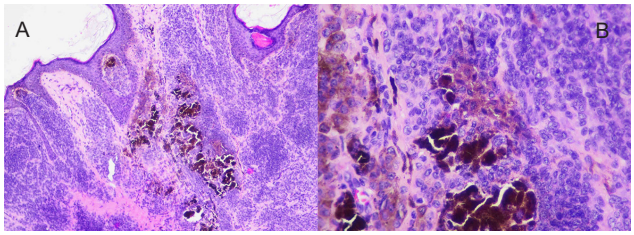

Figure 3. (A) Histopathology kissing nevus of the eyelid 10x magnification and (B) 40x magnification showed intradermal compound nevus pigmentosus.

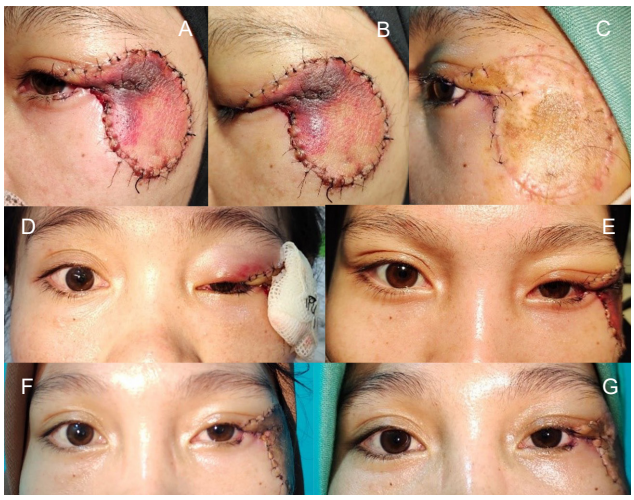

Figure 4. (A) One day post operative; (B) One week post operative; (C) Three weeks post operative after suture removal; (D) One day post operative patient; (E) One week post operative patient; (F) Two weeks post operative; and (G) Three weeks post operative.

is (a) small (less than 1.5 cm in diameter); (b) medium (1.5-20 cm in diameter); and (c) large (greater than 20 cm in diameter)—the second classification based on histological type (melanocytic, cellular or compound). The incidence of malignancy ranges from 0-42%, and the giant congenital melanocytic nevus is the only risk for malignant transformation as they invade the lower two-thirds of the dermis, subcutis, and surrounding nerves and adnexa.[8]

In order to avoid functional issues, including visual impairments, uncorrectable amblyopia (related to ptosis), and epiphora from puncta compression, the majority of experts suggest considering surgical correction as soon as possible. Also, there is a definite risk of malignant change in the nevus, which is best documented for giant nevi, but it is thought to exist for medium-sized nevi as well. Thus, early surgical treatment is recommended. Surgery for this lesion had been reported to be successful in two-month-old infants. Others have suggested that surgical correction be implemented at four to six (before the commencement of education). A significant issue was the reconstruction of the eyelid margin for corneal and ocular protection. The sensitive lid margins and thin skin structure make relatively significant local flap restoration inappropriate.[3],[10],[11]

Treatment has included non-surgical and surgical procedures, and various techniques have been documented. However, it is still debatable. In addition to removing the periorbital area, the primary purpose of treatment for kissing nevus is to make the proper interventions at the ciliary edge, including the area that forms eyelashes.[2],[3],[12] The components of the defect created after surgical excision of nevi need to be rebuilt with physiologically identical tissues. The most popular reconstruction method was a one- or two-stage skin graft. The donor sites for skin grafting are the upper arm, supraclavicular area, post-auricular, and contralateral eyelid.[2],[3],[12] It is crucial to match skin tone, with the contralateral eyelid typically making for the finest aesthetic match. Even though the contralateral or ipsilateral lid skin transplant is the most suitable tissue match, the availability of eyelid skin is restricted. In this patient, we choose the supraclavicular region for skin graft with consideration of skin color and defect size. We did not perform a skin graft from the contralateral eyelid because it was insufficient to cover the defect.[2],[3],[12]

The viability of free grafts depends on the vasculature in the recipient site, as they are harvested from distant locations.[4],[13] The supraclavicular region is a dependable donor option to cover significant head and neck defects, as it is similar in color, thickness, and flexibility. The selection of treatment management is based on the size and extent of the lesion, the patient's age, and the surgeon's preference.[4],[13]

The surgical approach is evaluated following the location, size, and extent of the lesion, as well as the age and involvement of the puncta. The subsequent principle was implemented during the development of reconstructive surgeries[3],[4]: (1) create minimal tissue injury, (2) maintain the integrity of the tarsal plate, particularly in the case of puncta involvement, unless corneal abrasions are present, and (3) if necessary to repair cheek defects, an advanced flap should be inserted above the skin graft using a two-stage procedure. The controversy surrounding the complete versus partial excision technique persists. A dermal nevus was incompletely excised at six, as Wu Chen et al.[13] described. However, the patient's verrucous and enlarged appearance, which was concerning for malignant transformation, was the result of significant regeneration of the lesion several years later.[3],[4]

If the nevus is treated early in life and still limited to the superficial dermis, dermabrasion is an appropriate technique for removing the congenital melanocytic nevus. However, the treatment should be a full-thickness excision followed by repair with a full-thickness skin graft or divided skin if the nevus involves deep dermis and subcutaneous tissue. According to various studies[3],[9], if the deeper dermis or subcutaneous tissue is involved, the lesion may recur as rapidly as four weeks after dermabrasion.

The other non-surgical management methods included cryotherapy and laser therapy. Ehlers in Alfano et al. (2007)[15] also characterized cryotherapy as a potential treatment for congenital melanocytic nevi, however, his research was limited to a few cases in the late 1960s. He reported that CO_2-freezing and diathermy coagulation were ineffective in an adult woman and a young child. Due to its high recurrence rate of up to 50% and the necessity of repeated therapy, laser treatment for congenital melanocytic nevi remains a topic of debate.[15] The use of Q-switch ruby laser (694 nm), Q-switched neodymium: yttrium-aluminum-garnet (Nd-

YAG) (532 and 1064 nm), and CO_2 laser for congenital melanocytic nevus of the head and neck has produced uncertain results in a variety of studies. Nevertheless, there is a theoretical possibility that laser irradiation could accelerate malignant transformation; however, this theory has never been empirically validated.[3],[15]

Functional and cosmetic complications, such as ptosis and visual field defects, may result from kissing nevus of the eyelids. If the nevus is medium to large, surgical excision and reconstruction are necessary for management. The primary reasons for surgical debulking of divided nevi are cosmetic or functional concerns. The supraclavicular graft is a reliable, secure, and aesthetically pleasing option for reconstructing head and neck defects. Lifelong clinical follow-up is recommended.[16]

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References

[1] Vora R, Shah A, Patel T. Congenital melanocytic kissing nevus on right eyelid: A rare phenomenon. Indian Dermatol Online J 2020;11:627. https://doi.org/10.4103/idoj.IDOJ_481_19.

[2] Cho H-J, Lee W, Jeon M-K, Park J-O, Yang E-J. Staged mosaic punching excision of a kissing nevus on the eyelid. Aesthetic Plast Surg 2019;43:652–657. https://doi.org/10.1007/s00266-019-01362-0.

[3] Desai SC, Walen S, Holds JB, Branham G. Divided nevus of the eyelid: Review of embryology, pathology and treatment. Am J Otolaryngol 2013;34:223–229. https://doi.org/10.1016/j.amjoto.2013.01.004.

[4] Jia R, Zhu H, Lin M, Li Z, Sun Y, Luo M, et al. Clinicopathological characteristics and surgical outcomes of divided nevus of the eyelids. Ann Plast Surg 2012;68:166–170. https://doi.org/10.1097/SAP.0b013e3182119155.

[5] Wahyudi D. Spontaneous regression of divided nevus of the eyelid evaluated by dermoscopy: Is it hypopigmentation after regression or vitiligo. J Gen Pro Derma & Venere Indo 2020;4:101–106. https://doi.org/10.19100/jdvi.v4i2.212.

[6] Quaranta LFM, Verity DH, Paridaens D, eds. Oculoplastic, Lacrimal and Orbital Surgery. Cham: Springer Nature Switzerland; 2024.

[7] Kharel RS, Bhatta S, Shrestha GB, Shrestha JK. Malignant transformation of kissing nevus: A rare entity. Nepalese J Ophthalmol 2012;4:329–332. https://doi.org/10.3126/nepjoph.v4i2.6555.

[8] Papadopoulos O, Chrisostomidis C, Konofaos P, Georgiou P, Frangoulis M, Betsi E, et al. Divided naevus of the eyelid, seven cases. J Plas Recons Aesth Surg 2007;60:260–265. https://doi.org/10.1016/j.bjps.2005.12.032.

[9] Rajput GC, Mahajan D, Chaudhary KP, Deewana V. Kissing naevus arising from neural crest cells presenting as upper and the lower lid mass. J Neurosci Rural Pract 2015;6:417–419. https://doi.org/10.4103/0976-3147.158796.

[10] Yildirim N, Sahin A, Erol N. The surgical treatment of congenital divided nevus of the eyelid: A case report and review of the literature. Int Ophthalmol 2009;29:45–47. https://doi.org/10.1007/s10792-007-9163-5.

[11] Bayramiçli M, Ersoy B, Sirinoglu H. Surgical management of a congenital panda nevus with preexpanded triple forehead flaps and temporal island flap. J Cranio Surg 2012;23:1396–1398. https://doi.org/10.1097/SCS.0b013e3182587a04.

[12] Bhattacharjee K, Singh M, Bhattacharjee H. Amniotic membrane graft to reconstruct divided nevi of eyelids. BMJ Case Rep 2015:bcr2014209020. https://doi.org/10.1136/bcr-2014-209020.

[13] Wu-Chen WY, Bernardino CR, Rubin PAD. The clinical evolution of a kissing naevus after incomplete excision. Br J Ophthalmol 2004;88:848–849. https://doi.org/10.1136/bjo.2003.035634.

[14] Vardaxi C, Karamitsou P, Skliris JP, Gougousis S, Poutoglidis A. Extended scalp reconstruction with a free supraclavicular graft: A case report. Cureus 2023. https://doi.org/10.7759/cureus.34324.

[15] Alfano C, Chiummariello S, De Gado F, Fioramonti P, Bistoni G, Scuderi N. Divided nevus of the eyelids: Three case studies. In Vivo 2007;21:137–139.

[16] Lemaître S, Gardrat S, Vincent-Salomon A, Galatoire O, Lévy-Gabriel C, Desjardins L. Malignant transformation of a multi-operated divided nevus of the eyelids. Ocul Oncol Pathol 2018;4:112–115. https://doi.org/10.1159/000479069.