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Case Report

Surgical treatment of a case of upper peripunctal intradermal nevus of the upper eye lid

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Abstract: Nevus around the upper lacrimal puncta is rare, and its excision affects both the appearance and tear drainage function of the tissue. Previously, no case of upper lacrimal punctal tumor has been reported. We managed a case of pigmented intradermal nevus around the upper lacrimal puncta. We resected the nevus around the lacrimal puncta and reconstructed the lacrimal duct. After the operation, the ostium of the lacrimal puncta was open and exhibited a crater-like appearance. Syringing of the lacrimal passage showed no blockage. The patient was satisfied with postoperative appearance. This case revealed that pigmented intradermal nevus also grows around the upper lacrimal puncta. We developed a unique surgical method used to achieve a safe and effective of treatment this case.

Keywords: Peripunctal neoplasm, ocular cosmetic surgery, intradermal nevus

Introduction

Peripunctal tumors are uncommon lesions. They often present as circle or ring-shaped swelling around the punctum. They are currently reported along with pigmented moles, epithelial invasive cysts, non-pigment complex moles, papillomastoma, granulocysis, and acidophilic adenomas, etc [1, 2]. Usually, they invade and squeeze the puncta, resulting in epiphora in some patients. Sometimes they cause symptoms of visual occlusion or foreign body sensation. Peripunctal tumors also affect the aesthetic appearance of the eye in some patients. These patients may require surgery. The location of peripunctal tumors is unique. Careless treatment of lacrimal punctal ostium after the removal of peripunctal tumors may lead to epiphora being aggravated and make the operation even more difficult. No prior case of an upper lacrimal punctal tumor has been described before. Earlier literature showed that peripunctal tumors could be removed via shaving excision with or without punctal stent placement [1-6]. Here, we present our report to illustrate one case of upper lacrimal punctal pigmented intradermal nevus and demonstrate the safety and effectiveness of our unique surgical method.

Case report

A 53-year-old female patient presented with a black neoformation at the right upper eyelid that had persisted for 3 years. On March 20, 2017, she visited our hospital. No abnormalities were identified in her medical history, including no history of hypertension or diabetes, no surgical or traumatic history, no personal history, and no genetic history.

A physical examination showed the following. Eyesight: Vou 1.0. Eye position was normal, and eye movement was good. A peripunctal wart-like brown-black neoplasm was observed on the right upper eyelid, which was approximately 1 mm×1.5 mm (**Figure 1**). The pigment was unevenly distributed, and the mouth of the lacrimal puncta protruded with a dome-like uplift. Lip swelling was evident, the lacrimal punctal ostium appeared linear, and the lacrimal duct was unobstructed. Her eyes presented no conjunctival congestion, with evidence of a trans-

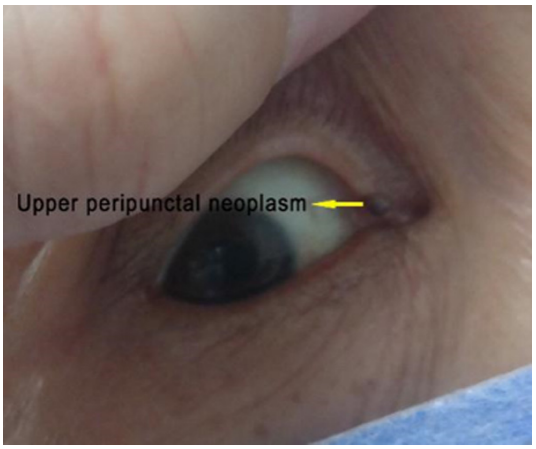

Figure 1. Preoperative appearance, showing upper peripunctal neoplasm (arrow).

parent cornea, KP (-), normal anterior chamber depth, clear iris texture, round pupils, d=3 mm, light reflex (+), a transparent binocular crystal, a fundus C/D of 0.3, and the presence of the foveal reflex.

Routine medical tests included, urine, feces, and biochemical tests, a blood examination for four items (hepatitis B, syphilis, hepatitis C, and AIDS), a chest position evaluation, and an electrocardiogram revealed no unusual results. Based on the patient's medical history, with the presence of a black neoplasm at the right upper eyelid for 3 years, and the appearance of a peripunctal wart-like brown-black neoplasm, we determined the diagnosis to be right eye upper eyelid neoplasm (considering the pigmented nevus).

The treatment methods used were as follows. (1) Preoperative examination and confirmation of the absence of surgical contraindications. On March 21, 2017 under local anesthesia, we performed right eye upper eyelid neoplasm resection using the following steps: 2% lidocaine and 0.75% bupivacaine mixed at equal volumes with 1:1000 epinephrine, was injected locally to achieve anesthesia. A plate was placed in the conjunctival sac of the upper eyelid. A sharp knife was used to make a 1-mm fusiform incision along the neoplasm. We removed the neoplasm and the wrapped lacrimal puncta. During the operation, we discovered that the brown-black tissue infiltrated the proximal canaliculus wall, so we trimmed the brown-black tissue in the proximal canaliculus with no remaining residue. We did not use sutures. The excised material was sent for a pathological examination. (2) Postoperative prevention of infection and nutritional support.

The results were as follows. The patient recovered well after the operation and did not complain of discomfort. On March 25, 2017 follow up revealed no patient complaint of pain or epiphora. An ocular examination showed that the right upper eyelid incision had healed, exhibiting no redness. The lacrimal puncta ostium was open and the upper eyelid showed no varus or valgus. Patient showed Vou 1.0, normal eye position and good eye movement. Her eyes presented with no conjunctival congestion, with evidence of a transparent cornea, KP (-), normal anterior chamber depth, clear iris texture, round pupils, d=3 mm, light reflex (+), a transparent binocular lens, a fundus C/D of 0.3, and the presence of the foveal reflex. The pathology results showed a pigmented intradermal nevus and a cut edge and base without mole organization (**Figure 2**). The patient was discharged. After six months, the right upper eyelid intradermal nevus around the lacrimal puncta did not recur (**Figure 3**), and no pain or epiphora was reported. The patient was satisfied with the postoperative appearance. Syringing of the lacrimal passage showed no blockage.

Discussion

Peripunctal tumors (such as a pigmented nevus, epithelial inclusion cysts, pigmented compound moles, squamous and verrucous tumors, pyogenic granuloma, and eosinophilic tumors) are uncommon and are reported only to account for approximately 6.3% of all lacrimal puncta abnormalities and 0.27% of all eye surgeries [1]. Scott KR [3] reported 6 cases of nevus around the lacrimal puncta, including 4 women and 2 men (age range 12-75 years), with the nevus located in the right lower lacrimal puncta in 3 cases and in the lower left region of the lacrimal puncta in the other 3 cases. All the tumors were present for many years. The eyes had no history of infection, or history of trauma, and showed no epiphora. The six patients showed a dome-like uplift, their lips were swollen, and the mouth of the lacrimal puncta appeared linear. The moles of 2 patients, who were Vietnamese and Filipino,

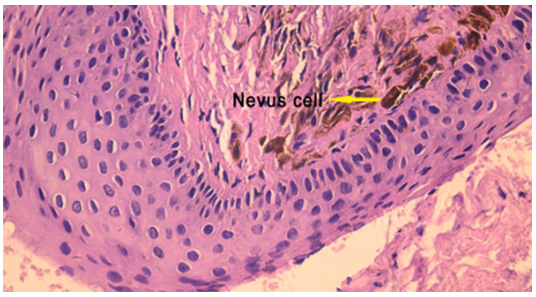

Figure 2. Pathological figure of excised material, showing nevus cell (arrow).

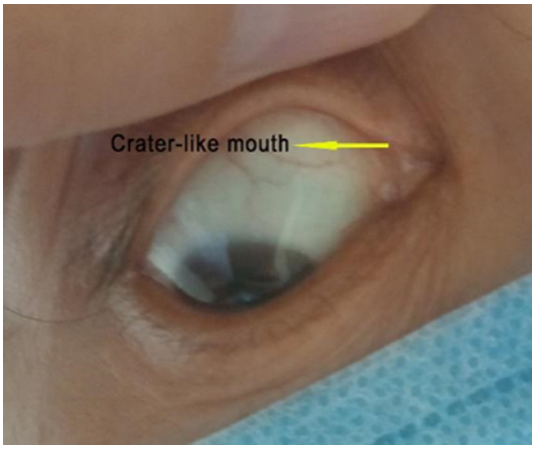

Figure 3. Postoperative appearance, showing crater-like mouth of the lacrimal puncta (arrow).

spread from the lacrimal puncta to the proximal canaliculus, while the moles of 3 Hispanic patients and 1 Caucasian patient did not involve the proximal canaliculus. Scott KR et al [3] found that the earliest cases of pigmented nevus involving the lacrimal puncta were reported in 1931, and only 4 other cases were reported until 1989. In addition, all previous case reports included limited clinical and pathological details, and all of the cases involved the lower lacrimal puncta. It has also been speculated that all cases involving the lower lacrimal puncta may be associated with a higher ultraviolet radiation dose because ultraviolet irradiation may play a role in the occurrence of a nevus. Frisch IB et al [4] reported 1 case of a neoplasm with constant growth. They also found that a neoplasm around the lacrimal puncta was first reported in 1931, and that 11 cases were reported in the literature. Hwang CS et al [5] also reported 1 case of lower eyelid neoplasm around the lacrimal puncta representing a melanin cell nevus. Currently, there are no reports of upper lacrimal puncta tumors.

All the characteristics of previous literature are shown in **Table 1**. Imaizumi M et al [6] proposed that the incidence of neoplasms around the lacrimal puncta is low because the lacrimal puncta is located at the junction of the eyelid and the lacrimal canaliculus. Lacrimal canaliculus consists of vertical part and horizontal part [7, 8]. Lacrimal canaliculus is lined with stratified squamous epithelium without keratinization, while eyelid is lined with keratinized squamous epithelium. Cells are smaller with high nuclear cytoplasm ratio in the basal layer of canaliculus epithelium. Lacrimal canaliculus epithelial stem cells (LCESCs) can be isolated from basal layer of canaliculus epithelium and have stem cell characteristics, similar to those of limbal stem cells (LSCs) [9]. These indicate that lacrimal canaliculus is apparently different from the eyelid.

Scott KR et al [3] suggested that a nevus around the lacrimal puncta must be distinguished from a nevus near the lacrimal puncta, as a mole near the lacrimal puncta may not completely encompass the lacrimal puncta. The nevus in our case was completely wrapped around the lacrimal puncta and some of the melanin tissue involved the opening of the inner wall of the proximal canaliculus. This is the first report of surgical treatment for this condition.

The lacrimal puncta is the starting point for a tear to flow into lacrimal drainage system. Abnormal position or closure of lacrimal puncta can cause lacrimal pumping failure and thus epiphora [10]. A neoplasm around the lacrimal puncta does not often cause epiphora [3, 5], and the reason may be related to that peripunctal neoplasm generally does not cause abnormal position or blocking of lacrimal puncta. The lower canaliculus plays the major role in normal tear drainage [11]. In this case of an intradermal nevus around the upper lacrimal puncta, no epiphora were observed, and the clinical manifestations were consistent with those of previous reports and anatomical physiological theory.

Lacrimal punctal neoplasm resection may affect facial features and tear drainage function [12], representing a major topic in ocular cosmetic surgery and lacrimal duct surgery. We strive to protect tear drainage function and to minimize the effect of surgery on the shape of the upper eyelid.

Table 1. Characteristics of previous literature

Author	Number of cases (M/F)	Location of swellings	Appearance of swellings	Characteristics of symptoms	Race of patients
Kevin 1989	6 (2/4)	Lower lacrimal puncta	Dome-shaped with swollen lips and a slit-like punctal ostium	No epiphora	2 Asian and 4 Caucasian
Jackson 1931	11	Lower lacrimal puncta	No information	No information	No information
Frisch 2003	1 (0/1)	Lower lacrimal puncta	Well-vascularized, non-pigmented tumor with a smooth, shiny surface	No epiphora	No information
Hwang 2011	Z (1/0)	Lower lacrimal puncta	an elevated, pigmented mass surrounding puncta	No epiphora	1 Asian

M: male, F: female.

The lacrimal puncta must be reconstructed after resection of a peripunctal neoplasm. Peripunctal neoplasm resection and reconstruction methods include 3-snips [13], trephining operation [14] and lacrimal duct drainage tube insertion [5, 15]. Lacrimal duct drainage tubes include epidural catheter and Crawford silicone tubes. Treatment of peripunctal neoplasm by cryotherapy can reduce the damage to the tear drainage system [16]. At present, the puncta reconstruction surgeries are not perfect. 3-snips makes the circular appearance of puncta into a crack. Lacrimal duct drainage tube insertion may hurt the lacrimal canaliculus, lacrimal sac and nasolacrimal duct mucosa. In our case, a lacrimal duct stent was not placed in the lacrimal duct because we trimmed the melanin tissue that had infiltrated into the proximal canaliculus and the lacrimal puncta opening was widened. After the operation, the mouth of the lacrimal puncta was open and exhibited a crater-like appearance. This report provides new insight into reconstruction of the lacrimal puncta.

Reconstruction of the eyelid is challenging because that eyelid provides a sophisticated interaction among anatomy, function, and aesthetics. The reconstruction approach of the eyelid varies depending on the size, location, and thickness of the defect [17]. The principle of when to use a graft, direct closure, a distant flap, or lid-sharing procedures is the key point of successful reconstruction [18]. A defect affecting less than 25% of the eyelid in most patients can be closed directly [19]. This operation only involved removal of the lacrimal puncta and 1 mm of surrounding tissue. Therefore, skin flap transplantation was not preferred. The patient's postoperative appearance was unaffected. Referring to the principle of Mohs operation [20], the postoperative paraffin section of the specimen confirmed that there was no residual nevus tissue at the edge of the tissue resection.

In sum, this paper reports a case of an uncommon pigmented intradermal nevus around the upper lacrimal puncta and details the unique surgical methods used to achieve sufficient safety and effectiveness of treatment.

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Disclosure of conflict of interest

None.

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