

PRIMARY OCULAR CARUNCULAR BASAL CELL CARCINOMA IN A CHINESE PATIENT

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Although basal cell carcinoma (BCC) is the most common eyelid neoplasm, BCC that originates from the lacrimal caruncle is extremely rare. To the best of our knowledge, only seven cases have been reported and here we report the first documented primary caruncular BCC in an Oriental patient. A 73-year-old Chinese man presented with a telangiectatic, multilobulated, pigmented tumor that measured 5×5 mm, which had arisen from the lacrimal caruncle of the left eye 3 months previously. The patient underwent tumor excision, and histopathological examination revealed BCC. He received adjuvant chemotherapy with intra-arterial methotrexate (30 mg/m²). A nodular pigmented BCC recurred in the bulbar conjunctiva close to the original tumor 3 months later, and he underwent a second excision. Bleomycin (8.5 mg/m² monthly) was added to the chemotherapy regimen, which was changed to fluorouracil (300 mg/m² monthly) 2 months later. The tumor did not recur during follow-up of 22 months. Malignant tumors of the caruncle are infrequent. BCC should be considered in the differential diagnosis of a pigmented caruncular lesion.

Key Words: basal cell carcinoma, caruncle, ocular tumor
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Basal cell carcinoma (BCC) is the most common malignant neoplasm of the skin, and accounts for 90–95% of malignant eyelid tumors. However, occurrence of BCC in the lacrimal caruncle is extremely rare. A search of the literature found only seven illustrated reports of primary caruncular BCCs [1–6]. Here, to the best of our knowledge, we report the first case of primary pigmented caruncular BCC in an Oriental patient and review the previous seven cases.

CASE PRESENTATION

A 73-year-old Chinese man presented with an isolated, multilobulated mass over the left caruncle, without a connection to the surrounding periorbital and eyelid skin (Figure 1). The lesion was brown–black, with a telangiectatic, irregular surface (5×5 mm), and the tumor had been growing painlessly without ulceration for 3 months. Other ophthalmic examination was normal, except for bilateral cataract. The patient was a mechanical designer, without excessive sun exposure or a family history of malignancy. A review of his medical history revealed that he had hypertension and coronary artery disease but no other malignancy.

Under local anesthesia, the tumor, along with the entire caruncle was excised using a “no touch” technique. The tumor did not invade deeply into the orbit and extended only to the subcutaneous tissue.



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Histopathology showed infiltrative islands of basaloïd cells, with characteristic retraction spaces between tumor islands and surrounding stroma. Elongated nuclei and scanty cytoplasm with peripheral nuclear palisades were consistent with BCC (Figure 2A). Melanin pigmentation was found in clumps (Figure 2B).

Owing to inadequate surgical margins in the pathological report, the patient was referred to an oncologist for adjuvant intra-arterial chemotherapy. After chemotherapy with methotrexate (30 mg/m² monthly) for 3 months, one small (2 × 1 mm) pigmented, nodular conjunctival tumor was noted close to the resected caruncle. A second tumor excision revealed recurrent

BCC. Another exploratory biopsy of pigmented lesions in the bulbar conjunctiva did not show any malignancy. Bleomycin (8.5 mg/m² monthly) was added to the chemotherapy regimen, which was changed to fluorouracil (300 mg/m² monthly) 2 months later. Follow-up examination over an additional 22 months showed no evidence of tumor recurrence.

DISCUSSION

The histogenesis of BCC is unclear. Either pluripotential germ cells in the deepest layer of the epidermis, or basal cells of pilosebaceous structures have been proposed to develop into carcinoma. The caruncle serves a transition zone between the skin and conjunctiva. It has a non-keratinized epithelial lining similar to the conjunctival epithelium, and harbors skin elements such as hair follicles, sebaceous glands, accessory lacrimal glands and sweat glands. Consequently, BCC is likely to occur in the caruncle.

Of the eight primary caruncular BCCs reviewed in the literature, including this present case and seven previous cases, male cases (*n* = 6) were more common than female cases (*n* = 2) (Table) [1–6]. Patient ages ranged from 24 to 82 years, with five being older than 60 years. All the previous seven cases were from North America or Europe. Our report is believed to be the first case of an Oriental patient with caruncular BCC. Of the seven previous cases, five of the tumors were described as pale, vascularized and lobulated

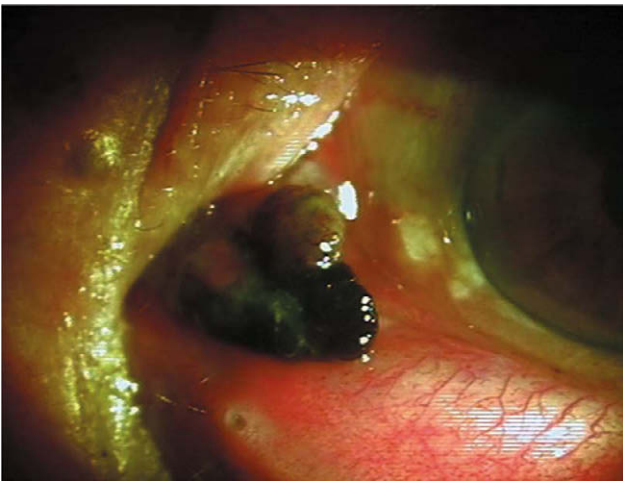


Figure 1. Darkly pigmented, multi-lobular, non-ulcerative tumor on left caruncle, 5 × 5 mm. Adjacent eyelid skin was free of tumor.

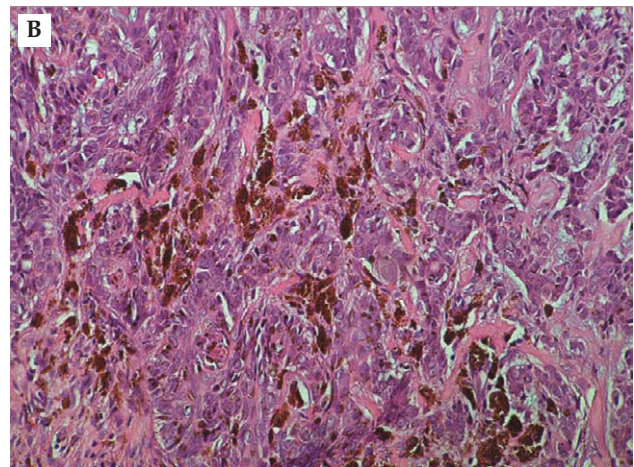
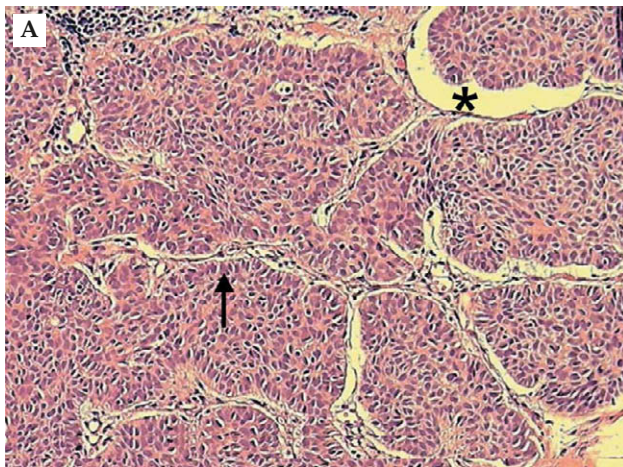


Figure 2. (A) Basaloid tumor islands with peripheral nuclear palisade (arrow) and typical retraction spaces (asterisk) (hematoxylin and eosin stain, 100×). (B) Pigment clumps between pleomorphic neoplastic cells. The diagnosis was a pigmented basal cell carcinoma (hematoxylin and eosin stain, 200×).

Table. Reported cases of primary caruncular basal cell carcinoma

Publication, year	Age (yr)/Sex	Clinical appearance	Treatment	Recurrence (follow-up)	Associated neoplasm	Reference
Poon et al, 1997	74/M	Multilobulated nodule, vascularized, pink	Excision	–	SCC, BCC	1
Meier et al, 1998	24/M	Nodule, vascularized, white, red center	Excision	No (14 mo)	No	2
Mencia-Gutierrez et al, 2005	80/M	Irregular, brown–black, vascularized	Excision	No (7 yr)	No	3
Ostergaard et al, 2005	60/F	Lobulated, cystic nodule, vascularized, pale, 3 × 4 mm	Excision	Yes (5.5 yr)	No	4
Rossmann et al, 2006	82/M	Nodule, pale	Excision, adjuvant radiotherapy	No (6 mo)	Periauricular skin cancer	5
Kaesler et al, 2006	72/F	Cyst	Excision	No (–)	–	6
Kaesler et al, 2006	52/M	Nodule, black	Excision	No (–)	–	6
Present case	73/M	Lobulated, brown–black	Excision, IA chemotherapy	Yes (22 mo)	No	

SCC=Squamous cell carcinoma; BCC=basal cell carcinoma; IA=intra-arterial; – Not stated.

nodules, whereas the other two were brown to black and similar to our tumor with dark pigmentation. Two patients had other skin cancers, including BCC and squamous cell carcinoma at sites distant from the eyelids. A majority of cases, in which eyelid BCC grows into caruncle, are not included in this series of reviews.

Complete excision of BCC with a tumor-free surgical margin is the principal primary treatment; however, radical excision with a wide margin in the caruncular area is difficult. The prognosis is worse once the tumor invades the orbit [4]. Ostergaard et al reported a case of caruncular BCC that recurred and extended into the orbit [4]. Rossmann et al reported a tumor that received adjuvant radiotherapy because of an uncertain tumor-free margin after surgical resection [5]. The patient in the present report received adjuvant intra-arterial chemotherapy. Intra-arterial infusion of chemotherapeutic drugs provides selective and increased perfusion of antineoplastic agents into the tumor, and local control of BCC using such chemotherapy has been reported [7]. Extensive BCCs on the wrist and thigh have been reported to respond to a combination of systemic and intra-arterial chemotherapy [6]. Large and unresectable BCC in the head and neck region had an 83% response rate when treated with intra-arterial chemotherapy using a combination of vincristine, bleomycin, methotrexate, and cisplatin [7]. Nevertheless, intra-arterial chemotherapy-induced stroke was a concern in this patient as he had cardiovascular disorders.

Radiotherapy is another optional adjuvant treatment of excised BCC in the medial canthal region, with a response rate better than 90% [8–10]. Our patient chose to receive intra-arterial chemotherapy after consultations with the surgical oncologist and therapeutic radiologist.

Various histopathological lesions have been reported from caruncles. The most common lesions are nevi and papillomas [11]. Other benign lesions include pyogenic granuloma, chronic inflammatory lesion, epidermoid cyst, dermoid cyst, epithelial inclusion cyst, oncocytoma, and sebaceous gland hyperplasia. Primary malignant tumors that arise from caruncles are unusual. BCC, Kaposi's sarcoma, keratoacanthoma, and sebaceous gland carcinoma have been reported [11,12]. The majority of malignant caruncular BCCs spread from the eyelid or medial canthus, but in our case, this was unlikely. If there had been any lesion in the adjacent skin, it would have grown to a remarkable size during follow-up of 22 months. In the first impression, this well-defined, darkly pigmented, caruncular nodule would have been considered to be a nevus. Nevertheless, its rapid growth suggested malignancy, and therefore, histopathological diagnosis was essential.

In conclusion, primary lacrimal caruncular BCC is rarely encountered. It has various presentations including pale, pink to darkly pigmented, cystic to lobulated, or nodular. Accurate diagnosis relies on photographically documented follow-up and timely

histopathological examination of suspicious lesions. The treatment of choice is complete excision with tumor-free surgical margins. If complete tumor resection is not achieved, adjuvant radiotherapy or chemotherapy should be considered.

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發生於中國病人之 原發性眼部淚阜基底細胞癌

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基底細胞癌 (BCC) 是眼瞼最常見的惡性腫瘤，但原發於淚阜 (lacrimal caruncle) 的 BCC 則極為罕見，至今僅有七例案例報告。本文為第一例東方人之原發性淚阜 BCC 病例報告。一位 73 歲台灣男性於左眼淚阜處發現一深色多葉狀腫塊，表面呈毛細血管擴張，於 3 個月內增長至 5 × 5 mm 大。該病患接受腫瘤切除之病理診斷為 BCC，術後並接受 methotrexat (30mg/m² monthly) 之輔助動脈內化療。三個月後於近原病灶之結膜處發生復發性 BCC。手術切除後加入 bleomycin (8.5mg/m² monthly) 及 fluoracil (300mg/m² monthly) 動脈內化療。追蹤至今共 22 個月，未有再次復發。原發自淚阜的惡性腫瘤相當罕見，對於淚阜處的深色病灶，BCC 須列入鑑別診斷之列。

關鍵詞：基底細胞癌，淚阜，眼腫瘤

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