

# Atypical Recurrent Presentation of Histologically Aggressive Type of Basal Cell Carcinoma with a Challenging Management- A Rare Case Report

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## ABSTRACT

Basal Cell Carcinoma (BCC) is the most common type of eyelid malignancy among white race individuals but is rare among blacks and Indians. The BCC predominantly affects the sun exposed parts like the neck, back and face (88-90%). These tumours are mostly located on the lower eyelid margin and the medial canthus as upper eyelids are protected from the Ultraviolet (UV) sunrays by the eyebrows and superior orbital margin. Although BCC seldom metastasizes, it is nonetheless considered malignant, since it can infiltrate adjacent tissues and cause substantial deformity and destruction. Here, authors have reported a rare case of 55-year-old female patient with recurrence of surgically treated BCC of upper eyelid involving eyebrow. Surgical excision is the most common employed treatment of choice in extensive tumours. Patient had extensive recurrent spread of BCC in left upper eyelid with multiple small ulcers in and around the left eye with pain and no Perception of Light (PL) vision. Exenteration of left eye was done followed by split skin autografting at the site. The BCC is usually not fatal, but early diagnosis and appropriate treatment promises better functionality and aesthetic outcome.

**Keywords:** Malignancy, Metastasis, Tumour

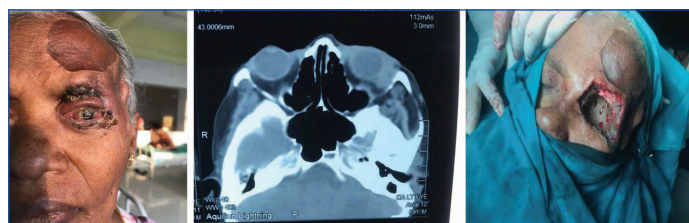
## CASE REPORT

A 55-year-old female patient, field worker by occupation, resident of South India, presented to the Ophthalmology Outpatient Department in January 2020 with an ulcerative lesion on the left upper eyelid. Patient first noticed a small ulcerative lesion near the left lateral canthus two years back which was diagnosed as BCC, following which wide local excision with a flap cover was done in 2018. Patient noticed a similar ulcerative lesion and complained of pain two months following the surgery at the same site which had gradually progressed to the present size measuring 3x0.5 cm in the upper eyelid in almost a year; associated with pain and had difficulty in opening the left eye. Patient also had history of severe itching and profuse foul smelling discharge. No history of any other systemic illness and family history of skin carcinoma or any other malignancy.

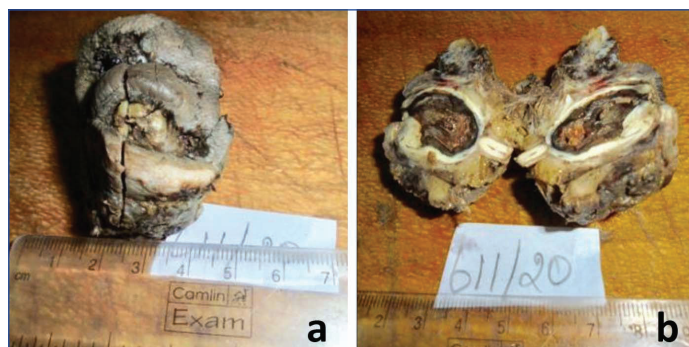
Ocular examination findings of left eye showed no PL. Left eyebrow and eyelid showed ulcerative lesion with rolled, beaded edges. Lid oedema and loss of upper lid eyelashes and sparse lower lid nasal eyelashes with profuse foul smelling purulent discharge was present. Multiple irregular shaped ulcers were noted on upper eyelid, Lower eyelid, below lateral canthus and eye measuring 3x0.5 cm, 0.5x0.5 cm, 3x1 cm and 4x1.5 cm, respectively with rolled, raised and beaded pigmented edge with ulceration in the floor [Table/Fig-1]. Tenderness was present. Bleeding was absent. Slit lamp examination of anterior segment was not possible to assess due to extensive spread of ulcerative lesion over left cornea and conjunctiva. Preoperative Contrast Enhanced Computed Tomography of Paranasal Sinuses (CECT-PNS) showed variable enhancing soft tissue thickening of size 2.1x2.0 cm at lateral canthus of eye involving the lateral rectus muscle and left eyelid with optic nerve thickening [Table/Fig-2]. The provisional diagnosis of recurrence of BCC of left upper eyelid was made based on the preoperative findings and investigations.

Since the patient had a history of BCC of left lateral canthus and presented now with similar ulcerative lesion of left upper eyelid and since the vision of the patient was completely compromised, no conservative management was planned, and we opted for surgical exenteration of left eye with Split Skin Grafting (SSG) [Table/Fig-3]. Periosteum along with entire orbital contents including the anterior

part of optic nerves, orbital fat, extra ocular muscles, eyelids and entire globe was removed. Patient's left eye was prepared for harvesting graft and a partial thickness SSG was placed over the recipient area of the eye socket. Exenterated specimen measured [Table/Fig-4a] 6x3.5x3.5 cm along with 1 cm long optic nerve. Cut section showed ulceroinfiltrative grey, white growth in the upper eyelid measuring 2.5x0.9x0.5 cm [Table/Fig-4b]. Another satellite lesion in the lateral canthus measuring 0.8x0.5x0.5 cm was also marked.

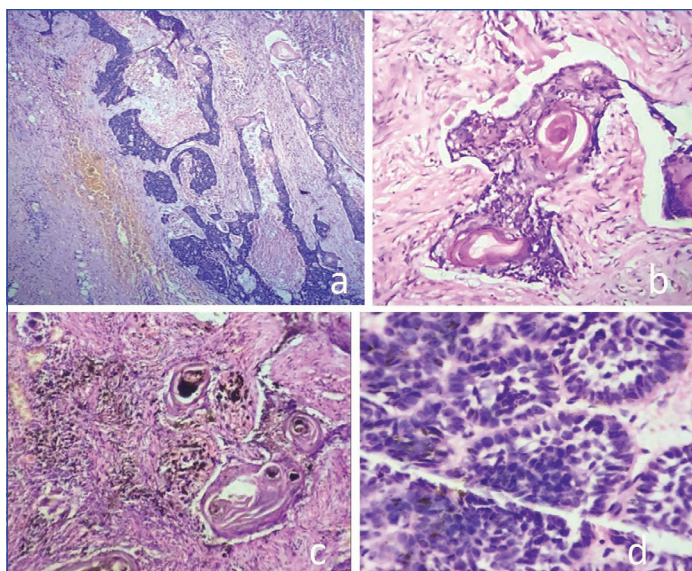


**[Table/Fig-1]:** Clinical presentation of patient showing the extensive basal cell carcinoma of left eye with hyperpigmentation of left forehead, which had been the site for previous skin flap. **[Table/Fig-2]:** Preoperative CECT-PNS showing variable enhancing soft tissue thickening of size 2.1x2.0 cm of lateral canthus of eye involving the lateral rectus muscle and left eyelid with optic nerve thickening. **[Table/Fig-3]:** Intraoperative picture of postgraft placement at the surgical site. (Images from left to right)



**[Table/Fig-4]:** a) Showing the exenterated eye specimen; b) Showing cut section of the specimen.

Histopathological report of intraoperative excised tissue showed features suggestive of metatypical (basosquamous) BCC admixed with areas of adenoid and pigmented BCC of left upper eyelid and lateral canthus [Table/Fig-5a-d] with tumour invasion in resected margin.



**[Table/Fig-5]:** Showing histopathological slide stained using haematoxylin and eosin stain suggestive of metatypical basal cell carcinoma with keratinisation, central necrosis, peripheral palisading and pigmentations. a) Nests and cords of tumour in scanner view (4x); b) Keratin pearl denoting keratinisation of tumour in high power view (40x); c) Abundant necrosis and pigmentation in low power view (4x); d) Peripheral palisading of tumour in low power view (40x).

Diagnosis was confirmed with histopathology report showing infiltrating malignant epithelial neoplasm in nests, cords, sheets and lace like pattern. Tumour nests with peripheral palisading, retraction artefact and peritumoural mucoid material deposition was observed with extensive areas of keratinisation, squamoid differentiation, pigmentation and necrosis.

Surgery went uneventful and the treatment was well tolerated. Patient was reviewed at postoperative day 14 [Table/Fig-6] and



**[Table/Fig-6]:** Patient after two weeks of surgery with split skin grafting on surgical site.

graft site was found healthy. Patient was kept for regular follow-up and prosthetic eye will be planned in the future.

## DISCUSSION

Basal cell carcinoma is the most common eyelid malignancy amongst the Caucasian population however, in India it is the second most common eyelid malignancy, first being squamous cell carcinoma [1-3]. The site distribution in this case is in concordance with the literature. The highest percentage (47%) of all BCC cases were marked to be located on the face, followed by the trunk, lower and upper limb, neck and hair free skin [4]. The trend observed in this case was confirmed in other studies [5]. Nodular BCCs were found to be the most frequent subtype of BCC for both males and females (69.74%) [4]. In a study conducted by Ghartimagar D et al., at Pokhara found that, the histological subtype predominant was nodular with 41 (53.94%) cases, followed by the 14 (18.42%) cases of pigmented and 10 (13.15%) cases of basosquamous subtype [6]. Few other studies conducted among South Asian population by Ullah O et al., Asif M et al., Laishram RS et al., Malhotra P et al., Chow VLY et al., Janjua OS and Qureshi SM, Chang JM and Gao XM had come to the similar conclusion of nodular variety being the most common type of BCC presentation [7-13]. The BCC being a rare presentation among South Asian population only few studies were conducted and details of studies done are mentioned [Table/Fig-7] [7-13].

The most frequent site of origin is lower eyelid and medial canthus. However, in the present case the lesion appeared in a much rarer site (left upper eyelid and lateral canthus). About 90% of BCC occurs among the age group of 55 to 70 years [14,15]. However, in this case the lesion had first appeared at the age of 50 years. After two months from the surgery the patient had a recurrence at the same site. The recurrent lesion gradually progressed in size in two years. The patient was an agricultural worker for almost 30 years, native of a hot weather village in South India, owing to prolonged high exposure to sunlight. The UV radiation, most specifically UV-B is currently believed to be the most crucial etiological factor for BCC. In a study conducted by Malhotra P et al., among BCC patients in North India, 91% patients had history of prolonged sunlight exposure [10].

The present case patient hails from a subtropical district in southern India a place with very high temperature. Patient is an agricultural field worker who previously used to work outdoors during morning hours regularly. In this case, environmental and occupational parameters would have played the key roles in the development of BCC.

Study	Sample size (N)	Duration and type of study	M:F	Age (years)	Sites	Subtype
Ullah O et al., 2008 [7]	100	4 years, prospective	0.8:1	Mean age: 56.3	—	24 pigmented nodular, 21 nonpigmented nodular, 30 ulcerative, and 6 lesions were of morphoic type.
Asif M et al., 2010 [8]	235	3 years, retrospective	1.2:1	32 to 90	Nose: 28.9% Eye: 24.7% Cheek: 20.4%	-
Laishram RS et al., 2010 [9]	30	5 years, retrospective	1:2	Median age: 70; Most common age group: 61 to 70 years	83.3% on head and neck, with predilection for face	Nodular subtype was the most frequent.
Malhotra P et al., 2011 [10]	34	3 years	1.6:1	Age range: 28 to 102 Majority (44%): 40 to 60	91.2% on head and neck, with most common site being medial/lateral canthus of eye	Most common histology subtype: nodular (64.7%); pigmented clinically (35.2%)
Chow VLY et al., 2011 [11]	225	10 years, retrospective	0.7	Age range: 22 to 100 Mean age: 73.1	Nose: 31.6% Cheek: 16.5%	Ulcer: 64.8% Nodule: 19.3%
Janjua OS and Qureshi SM, 2012 [12]	171	3 years, retrospective	1.4:1	Age range: 22 to 90 Mean±SD: 61.3±13.07	Most common site: nose (31.5%) followed by cheek (26.9%)	Nodular variety: 46.2% and pigmented type: 18.7%
Chang JM and Gao XM, 2013 [13]	243	8 years, retrospective	0.94:1	Mean±SD: 65.16±12.62	Head and neck region was the most common site (77.4%)	Nodular: 53.9% Superficial: 18.9% Infiltrative-morphoic: 18.5%

**[Table/Fig-7]:** Studies conducted on basal cell carcinoma in South Asia [7-13]. SD: Standard deviation

According to National Comprehensive Cancer Network (NCCN) guidelines 2019 for high risk BCC, Mohs micrographic surgery or an excision with 1 cm circumferential clearance is suggested [16]. According to study done by Ro KW et al., pigmented BCCs show lesser subclinical infiltration than non pigmented BCCs [17]. Since the patient presented with recurrence of BCC of left upper eyelid and since the vision of the patient was completely compromised, authors opted for exenteration of left eye with SSG with 1 cm circumferential clearance.

## CONCLUSION(S)

Basal cell carcinoma is an extremely complex disease, with many aetiological factors influencing its development. In this case report, the aetiological factors related to the development of this rare BCC presentation were older age and exposure to prolonged UV rays both environmental and in occupational form, with a clear impact on the patients' quality of life. This causes a significant burden to the health system, especially in such cases of invasive behaviour and recurrence after treatment. Educating the patients regarding all the risk factors and special attention to the diagnosis of minor lesions through self examination are crucial in prevention and improved prognosis, and faster diagnosis particularly in susceptible population.

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