

Sebaceous Carcinoma of Inner Canthus of the Eye: Report of a Case

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Abstract

Introduction: This is to report a rare case of sebaceous carcinoma of inner cantus of the eye in relation with rare skin lesions of epidermodysplasia verruciformis. Epidermodysplasia verruciformis is a rare skin disease which is caused by human papilloma virus.

Sebaceous carcinoma is a rare entity which is derived from sebaceous glands and to our best knowledge this is for the first time that we observe sebaceous carcinoma arising from lesions of epidermodysplasia verruciformis at the inner canthus of the eye.

We report a 50 years old male patient who admitted to our hospital 3 years ago.

Key words: Sebaceous carcinoma-Epidermodysplasia verruciformis – Inner cantus.

Introduction

Sebaceous carcinoma is an uncommon malignant tumor which is derived from sebaceous glands. It is extremely rare in eye lids and when occurs it arises from accessory lacrimal glands or meibomian glands (1).

Sebaceous carcinomas can occur in two locations ocular and extraocular, ocular sebaceous carcinomas are more aggressive than extra ocular (2).

Epidermodysplasia verruciformis is a rare skin disease which is caused by human papilloma virus (3).

Epidermodysplasia verruciformis is a flat wart which is caused by viral changes in superficial keratinocytes and subsequent changes in keratin production (4).we reported this case since the ocular type is rare and had not been reported previously. Although it had showed invasion and multiple recurrences during this 3 years, it did not cause blindness.

Case report

A 50 year old male patient attended to Besat hospital of Hamedan university of Medical Science Hamedan, Iran. Complaining of itching and bleeding skin lesion on the inner canthus of the eyes (Fig.1).

The patient was a taxi driver, so he was exposed to considerable amount of sunlight and he also had a usual habit of touching and cleaning the corner of the eye by his fingers.

Patient had brown flat pigmentations all over his body which were more pronounced on sun exposed areas and a bit depigmented on the hands and face because of frequent washing and rubbing.

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His father, his son and also his cousin have the same skin pigmentations. He was diagnosed for epidermodysplasia verruciformis when he was 20 year-old. He mentioned that he was suffering from the same mentioned warts on the Inner canthus of the left eye for several years and recently he felt that it had become itchy and started bleeding upon itching since last month (Fig.1A and B).

Prior to surgical treatment an incisional biopsy obtained in infirmary and histopathologic diagnosis of squamous cell carcinoma considered.

One week later the patient attended for surgical treatment considering Moh's micrographic surgery.

On frozen sections, diagnosis of sebaceous carcinoma had been proposed which had been confirmed later on permanent processing of obtained tissues (Fig.2).

There have been no bone invasion of lateral nasal and also no global invasion.

To further confirm the histopathologic diagnosis, Immunohistochemical examination with Epithelial membrane antigen(EMA), Carcinoembryonic antigen (CEA), CK7 and Androgen receptor obtained in which EMA, CK7, Androgen receptor were positive and CEA have been negative (Fig.3).



Fig 1. A and B patient 3 years ago. C: patient last month

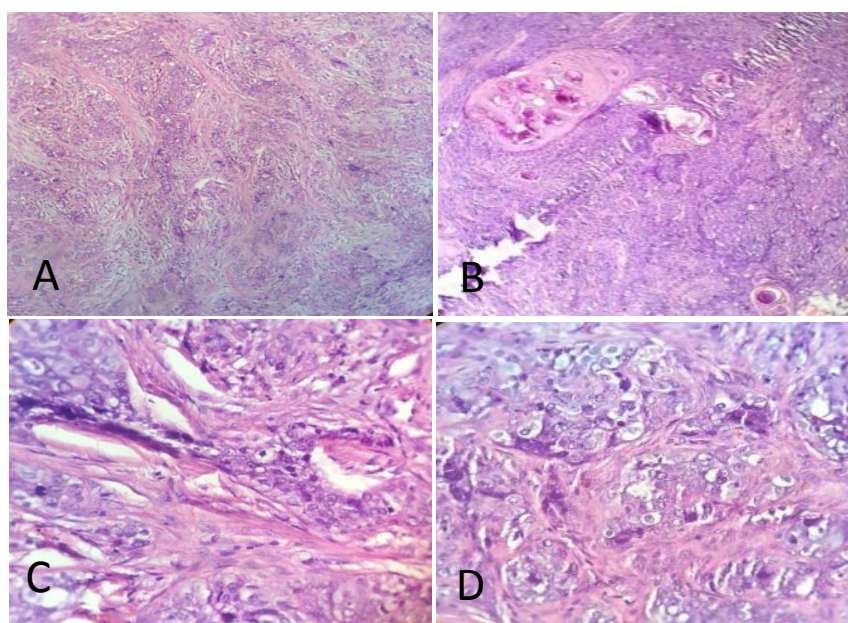


Fig 2. Hematoxylin and Eosin sections. A: x10. B: x40. C and D: x100

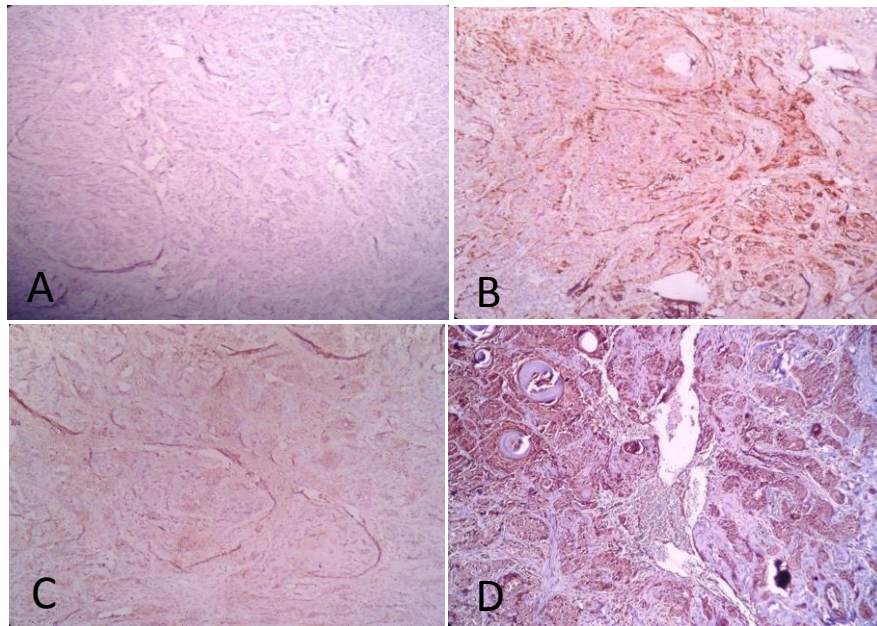


Fig 3. Immunohistochemical analysis shows,
 A: CEA negativity.
 B: EMA strong positivity C: CK7 weakly positive.
 D: shows strong positivity of tumor cells with androgen receptor

Discussion

Epidermodysplasia verruciformis is a cutaneous eruption which has two types, the first type caused by HPV-3 and 10 and the second type caused by HPV-5 and 8 which is more prone to malignancies; mostly SCC(3).

Epidermodysplasia verruciformis is transmitted in an autosomal recessive fashion and is due to mutation in epidermodysplasia verruciformis gene 1 and 2(5), Located on chromosome 17q25. Lewandowsky and Lutz reported it in the first time in 1922(6).Epidermodysplasia verruciformis is caused by defect in T-lymphocytes and subsequent defective innate immunity against certain strains of HPV virus (7).

Sebaceous carcinoma is a disease of elderly and is more common between Asians. Its clinical differential diagnosis are calazions, blepharitis, or conjunctivitis and also cicatrical pemphigoid, basal cell carcinoma and squamous cell carcinoma (2). Sebaceous carcinoma has two types; ocular and extraocular. Ocular type most frequently occurs on the eyelids. Typically originating from the meibomian glands and less commonly from the glands of zeis (8).

In microscopic examination, an irregular lobular aggregate of polyhedral cells, separated by fibrovascular septa, is observed. (Fig.2A)The cells

show clear or foamy cytoplasm and dark pyknotic nuclei. (Fig.2B)

Tumor shows variable degrees of cellular and nuclear pleomorphism and increases in nuclear to cytoplasmic ratio. . (Fig.2C and D) Some lobules show atypical keratinization and keratin pearl formation, like what is seen usually in SCC (8).

Patient follow up had been done and he had two recurrences. The first one was a year after initial surgery and simple excision performed. He had another recurrence recently, causing fixation of eye ball; so another excision of tumor with partial removal of eyelid performed together with 35 session of Radiotherapy, which were efficient and patient feels better since then(Fig.1,inset c).

Microscopic differential diagnoses of sebaceous carcinoma are basal cell carcinoma and squamous cell carcinoma.

In contrast to BCC, in sebaceous carcinoma undifferentiated cells have more eosinophilic cytoplasm. Atypical cytologic features are greater and tumor shows greater degree of invasiveness (8).

Squamous cell carcinoma with clear cell changes have dyskeratotic cells and keratin pearl formation (8) that we observed in our case. For this purpose, we used a battery of immunohistochemical markers to rule out SCC and BCC and to further confirm diagnosis of

sebaceous carcinoma. We used androgen receptor and it was positive.

In the study done by F.Asadi et al in 2010. They found that all the 19 cases of sebaceous carcinoma showed positive reaction with androgen receptor (9).

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