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ISOLATED LICHEN PLANUS OF THE CONJUNCTIVA WITH ASSOCIATED BOWEN'S DISEASE: A CASE REPORT

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ABSTRACT: Bowen's disease is an intraepithelial neoplasm involving the squamous epithelium, commonly occurring in the genital region. Rarely, it is also observed at skin in other anatomical location. Lichen planus is chronic dermatoses, which is commonly observed in the extremities and mucosa. In lichen planus, there will be dense inflammatory infiltrate in the sub-epidermal zone. Very rarely, Lichen planus involving the sub mucosa can undergo malignant transformation. Co-existing isolated lichen planus with the Bowen's involving the conjunctiva is a very rare event.

KEYWORDS: Bowen's disease conjunctiva, isolated lichen planus in the conjunctiva.

INTRODUCTION: Most of the carcinomas originate as an insitu lesion before becoming an invasive and metastasing lesion. The in situ lesions that occur in the squamous epithelium especially involving the genitals are Bowen's disease, Bowen's papillosis and Erythroplakia of Queyrat. Here with a report on isolated lichen planus with associated Bowen's disease involving the left conjunctiva near the limbus is being presented.

Pizzarello and Jakobiec¹ proposed designating intraepithelial tumor as conjunctival intraepithelial neoplasia (CIN) and classified the tumor into two types; mild dysplasia and severe dysplasia. In mild dysplasia, dysplastic cells occupy less than half the conjunctival epithelium and in severe dysplasia, more than half or the entire layer.¹ The World Health Organization (WHO) classifies carcinoma insitu with dysplasia and actinic keratosis as a CIN.²

When intraepithelial dysplastic cells, even partially, invade the basement membrane and infiltrate the sub epithelium of the conjunctiva, the condition is called squamous cell carcinoma (SCC). Conjunctival intraepithelial neoplasia and SCC are histopathologically well-defined conditions. However, it is difficult to determine the grading of dysplasia by clinical morphologic findings.

Conjunctival squamous cell carcinoma (intraepithelial and invasive) is the most common conjunctival malignancy in the United States.^{3,4} The incidence of this neoplasm is between 1 and 2.8 per 100 000 people per year and varies in different geographic locations.⁵ Epidemiological studies have shown that ultraviolet B radiation (290–320 nm) may be important in its pathophysiology.^{6,7} Ultraviolet B induced point mutations in the p53 tumour suppressor gene have been reported in other squamous cell carcinomas.⁸ Human papillomavirus (HPV) types 16 and 18 may also be important in tumour development.⁹⁻¹⁴

Intraepithelial squamous carcinoma (also known as conjunctival intraepithelial neoplasia, previously known as Bowen's disease) and invasive squamous cell carcinoma of the conjunctiva are histologically differentiated according to the invasion of epithelial basement membrane. In invasive squamous cell carcinoma cells infiltrate through the basement membrane and invade the substantia propria.³ Although intraepithelial squamous carcinoma may progress to invasive squamous cell carcinoma, the former lesions are localised in the epithelium.^{3,4} Clinically these tumours most

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commonly arise in the interpalpebral area of the perilimbal conjunctiva. The growth may be in a nodular, gelatinous, flat superficial leucoplakic, or diffuse invasive fashion. These tumors may present as localised slowly growing lesions that mimic benign conjunctival degenerations and sometimes coexist with pinguecula and pterygium.^{3,4}

CASE SUMMARY: A 45 years male was first seen in the ophthalmology clinic at Rajah Muthiah Medical College. He complained of impaired vision on the left eye for the past 3 years of duration. Neither he nor his relative's had noticed any other abnormality and his general health was good. On examination there was an elevated gelatinous lesion of size 2 x 3 cm size on the conjunctiva close to 1mm of the limbus in the left eye. The visual acuity was 6/12 on both eyes.

Slit lamp biomicroscopical examination suggested that the lesion was entirely confined to the epithelial layer. The anterior segment examination and fundi were normal except minimal nuclear sclerosis of crystalline lens. Other routine investigations all gave negative results. A provisional of intraepithelial carcinoma was made and wide excision was performed. Post-operative progress has been satisfactory but it is proposed to keep the patient under observation. Conjunctival growth was sent for histopathological examination.

MACROSCOPY: A single tiny grey white, grey brown tissue piece measuring 2 x 3 cms. Entire tissues was processed for histology.

MICROSCOPY: Section revealed squamous mucosal biopsy material up to sub mucosal level. There was expansion of squamous epithelium where the squamoid cells are present. There are dysplastic cells with rare mitotic figures. There are vacuolated squamous cells near the basement membrane. At places Max-Joseph space (art factual separation of the epithelium from the stroma). The basement membrane appears to be intact with foci of dense inflammatory infiltrate predominantly composed of lymphocytes. Occasionally they tend to form aggregate.

DISCUSSION: Bowen's disease is a form of intraepidermal SCC characterized by a persistent, non-elevated, red, scaly or crusted plaque with a small potential for invasive malignancy. It consists of a solitary lesion. It may occur on exposed or on unexposed skin. It may be caused on exposed skin by exposure to the sun and on unexposed skin by the ingestion of arsenic.

In 1912 Bowen described a skin lesion which he called precancerous dermatosis. It has since borne his name. The concept precancerous had been coined as early as 1896 by the French dermatologist Dubreuilh and implies that the lesion more or less regularly becomes carcinomatous. Jessner (1921)¹⁵ reported the first cases of Bowen's disease of mucous membrane (glans penis) and McGavie (1942)¹⁶ the first ones involving the cornea and conjunctiva. In the same year Ash & Wilder reviewed 93 cases of epithelial tumors at the limbus, 4 of them being classified as Bowen's disease.

Since then many more ocular cases have been recorded. In the belief, however, that a terminology based on the pathology of a lesion is preferable to an eponymous nomenclature, it would seem proper, in describing pre-invasive neoplasia of the conjunctival epithelium, to abandon the commonly applied ascription 'Bowen's disease' in favour of the more explicit term 'intraepithelial squamous cell carcinoma.'

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Conjunctival epithelial malignancies are more common in elderly and male patients, but may develop at a younger age especially in association with xeroderma pigmentosum or immunodeficiency.^{17,18-21}

Cutaneous and visceral malignancies in association with conjunctival epithelial tumours have been reported.^{3,17} Erie and associates reported 13% of patients had a history of malignant skin tumours. Twelve (20%) of our patients had skin malignancies; 11 were basal cell and one was an epidermoid carcinoma. Bowen's disease or in situ cutaneous carcinomas have been thought to be associated with internal cancer but recent population based data failed to show this association; thus, most investigators no longer use this terminology.²²

There are limited data on topical cytotoxic therapy. Frucht-Pery and Rozenman reported on three cases. Yeatts and associates treated six conjunctival intraepithelial neoplasia cases with topical 5-fluorouracil; five responded well but one progressed and required exenteration. Good short term conjunctival intraepithelial neoplasia response has been reported after topical 0.04% mitomycin therapy.

Surgical complications include corneal scarring, symblepharon, ocular hypotony, and iris atrophy and recurrence of tumor. Recurrence of conjunctival epithelial malignancies depends on the status of surgical margins. Simple excision of conjunctival intraepithelial or invasive neoplasia is associated with a 24–50% recurrence rate.

Several histological features are important for tumour recurrence; however, even in cases with milder degrees of atypia, careful assessment of histological margins is mandated. Tabin and associates excised intraepithelial tumours without intraoperative control of surgical margins; 25% had positive margins and incompletely excised cases developed twice as many recurrences as those with tumour free margins.

Erie and associates reported a recurrence rate of 41% in squamous cell carcinoma and 23.5% in conjunctival intraepithelial neoplasia after local excision with a follow up of 6 months to 39 years. Irvine reported a 10% local recurrence rate but half of the cases had either no, or less than 1 year, of follow up. Follow up examinations for longer than 12 months are necessary as the mean time to recurrence has been reported as between 8 and 22 months.

The definite diagnosis of Bowen's disease is invariably histological.

Bowen's disease of the eye is often localized to the limbal region. As a rule the lesion is described as gray or grayish red, pannus-like, gelatinous growth. It usually has a somewhat rough, granulated surface and a fairly firm consistence and is sharply delimited from healthy tissue. When present in the conjunctiva, the lesion is movable upon the sclera. Profuse vascularization of surrounding conjunctiva is often found.

In the present case a clinical diagnosis of Bowen's disease was sought but the Histopathological pictures showed additionally some features of lichen planus.

Typical lichen planus show vacuolar alteration of the basal layer and a band-like dermal lymphocytic infiltrate in close approximation to the epidermis, compact orthokeratosis, wedge-shaped hypergranulosis and irregular acanthosis. This constellation of findings is sufficiently diagnostic that a histologic diagnosis can be rendered in more than 90% of cases.

The entire excised specimen sent for Histopathological examination was further processed.

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The Histopathological study showed some areas with expansion of the squamous epithelium with dysplastic squamous cells and scattered mitotic figures. The basement membrane was intact without any breach in the continuity.

There are areas of inflammatory lymphocytic infiltrates in the sub epithelial zone. Some degree of basement membrane remodeling was noticed giving a vague saw tooth appearance. Hyperkeratosis which is characteristic of lichen planus was found. Art factual cleft (Max – Joseph) was seen between the basement membrane and the lichenoid infiltrate was observed in some foci. Vacuolar alterations of the basement membrane were also seen.

Here with isolated conjunctival lichen planus lesion with associated Bowen's disease is being presented for its rarity in view of the presentation and occurrence as far as the literature is concerned.

EQUIPMENT USED:

Nikon Coolpix-8400

x-denotes the power of objective

Stain used – (H & E)

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MICROSCOPIC PICTURES

Fig 1A & 1B: Inflammatory lymphocytic infiltrates in the sub epithelial zone and basement membrane remodeling. Max- Joseph space noticed (H & E) stained x4.

Fig. 1A

Fig. 1B

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Fig. 2A & 2B: Squamous epithelium with dysplastic squamoid cells, melanin pigmented cells. Basement membrane is intact (H & E) stained x10.

Fig. 2A

Fig. 2B

Fig. 3: Sub epidermal inflammatory infiltrate and Max- Joseph space.

Fig. 3

Fig. 4: (A-D): Expansion of Squamous epithelium. Squamoid dysplastic cells, rare mitotic figures and melanin pigmented cells

Fig. 4A

Fig. 4B

Fig. 4C

Fig. 4D

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Fig. 5: Basal cells showing vacuolations.

Fig. 5

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