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Case Series

A case series of Bowen's disease in a tertiary care centre in South India

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ABSTRACT

Bowen's disease is a rare premalignant condition with multifactorial etiology. Usually, Bowen's disease manifests as solitary, well defined erythematous plaque over sun exposed areas and less commonly over photo protected sites. Hereby we report eight patients with clinical diagnosis of Bowen's disease confirmed by dermoscopy and histopathology. Dermatological examination revealed presence of pigmented, hyperkeratotic plaques and classical morphology. Scales and glomerular vessels were the most common features in dermoscopy. Histopathology was characteristic with presence of atypical cells in all patients. Five patients were treated with surgical excision, two other patients with 5% imiquimod and 5-fluorouracil creams respectively. One patient was lost to follow up. Post excision biopsy revealed presence of invasive malignancy in two patients. Hence it is important to diagnose and treat Bowen's disease as early as possible.

1. Introduction

Bowen's disease is a rare squamous cell carcinoma in-situ of epidermis, initially described by John Templeton Bowen, an American dermatologist in 1912.[1] Clinically, Bowen's disease presents as solitary, well demarcated, asymptomatic, erythematous, scaly irregular plaque commonly over photo exposed sites such as head and neck, and extremities, although involvement of multiple sites are seen in 10-20% cases. Chronic ultraviolet radiation exposure, arsenic exposure, various HPV strains, chemical carcinogens, immunosuppression and chronic irritation are considered as some of the risk factors for development of Bowen's disease.[2] However the exact etiology is not clearly understood. Genital lesions (10%) and extra genital lesions (3-5%) can turn into malignancy if left untreated.[3] Hence early diagnosis and treatment is important as it can evolve into squamous cell carcinoma.

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2. Case Series

Here we report a series of eight patients with Bowen's disease who presented with various morphological patterns over a period of three years. Among the eight patients five were females and three were males, with age group ranging from fourth to eighth decade. All of them presented with asymptomatic solitary lesion over trunk and extremities with duration ranging from 6 months to 10 years. None of our patients had comorbidities or associations except one patient had oculocutaneous albinism. General and systemic examination found to be normal. Dermatological examination revealed presence of plaques with varied morphology such as classical type of Bowen's disease (back), hyperkeratotic type (left lower abdomen, right forearm and left index finger) and pigmented type (chest, abdomen, right thigh and right knee) (Table 1), (Figures 1, 2, 3, 4, 5, 6, 7 and 8). Diagnosis of Bowen's disease was made in six patients while differential diagnosis of TBVC / chromoblastomycosis was considered in one patient

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who had lesion over left index finger and in an other patient with oculocutaneous albinism, actinic keratosis was entertained. On dermoscopy, scales and glomerular vessels were the most common findings (87.5%) followed by homogeneous grey brown background (75%), brown and black globules, and homogeneous pink background (50%) (Table 2), (Figures 9, 10, 11, 12, 13 and 14). Dermoscopic and histopathological examination confirmed the diagnosis of Bowen's disease. Histopathological features are depicted in table 3 (Figures 16, 17, 18, 19 and 20).Five of our patients were treated with surgical excision. In two of them histopathology revealed presence of invasive squamous cell carcinoma. Two other patients were treated with 5% imiquimod and 5-fluorouracil creams respectively. One patient was lost to follow up.

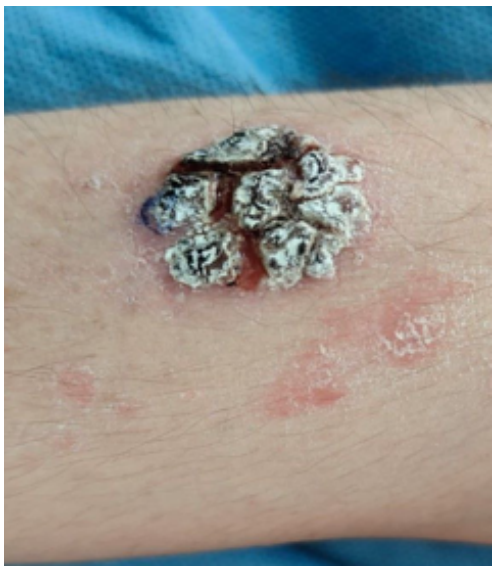

Figure 1: (Case1) Hyperkeratotic plaque over right forearm.

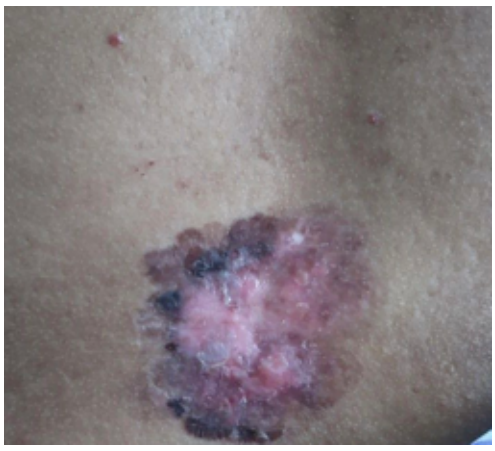

Figure 2: (Case 2) Erythematous plaque with fine scaling and crusting over lumbosacral area.

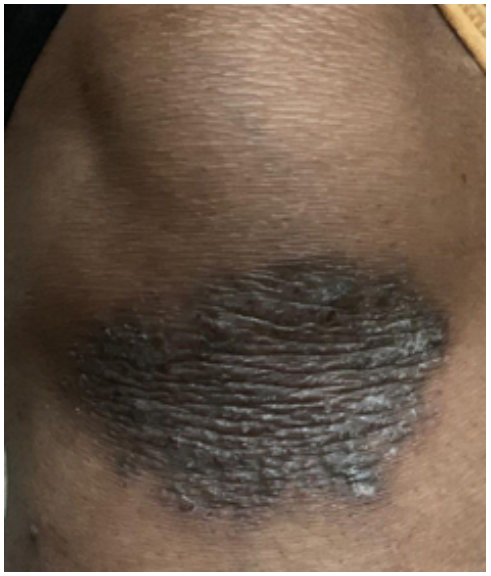

Figure 3: (Case 3) Pigmented plaque over right knee.

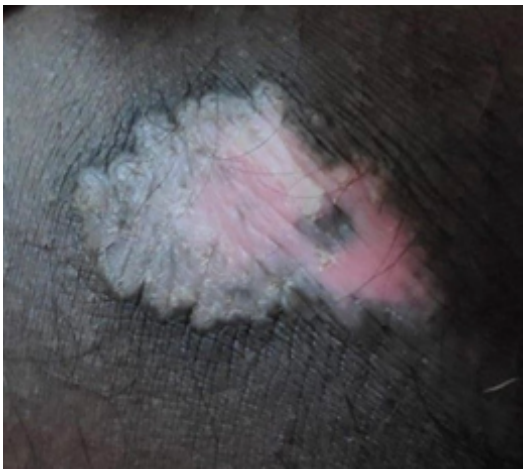

Figure 4: (Case 4) Erythematous plaque with peripheral rim of pigmentation over inner aspect of right thigh.

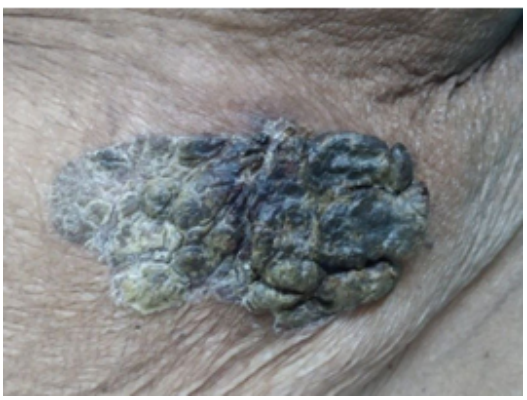

Figure 5: (Case 5) Pigmented verrucous hyperkeratotic plaque over left lower abdomen.

Table 1: Demographic details

Case	Age (yrs)	Sex	Occupation	Site	Duration	Ulceration	Clinical variant	Treatment given	Malignancy
1	34	F	Housewife	Right forearm	5 years	fissures+	Hyperkeratotic	Excision	-
2	44	F	Vendor	Back	4 years	-	Classical	Excision	-
3	50	F	Tailor	Right knee	1 year	-	Pigmented	5 fluorouracil cream	-
4	52	M	Farmer	Right thigh	5 years	-	Partially Pigmented	Lost to follow up	Not known
5	65	F	Housewife	Left lower Abdomen	6 months	-	Partially Pigmented/Hyperkeratotic	Excision	-
6	68	F	Housewife	Left lower Abdomen	10 years	+	Hyperkeratotic/ Pigmented	Excision	Malignant (SCC)
7	68	M	Retired worker	Left index finger	2 years	+	Verrucous /Hyperkeratotic /Pigmented	Excision	Malignant (SCC)
8	78	M	Retired officer	Chest	2 years	-	Partially Pigmented	5% imiquimod cream	-

Table 2: Comparison of dermoscopic findings of Bowen's disease in our study with previous studies

	Total cases (n=8)	Our study	Mun et al	Zalaudek et al	Bugatti et al
Scales	7	87.5%	96%	90%	64%
Dotted vessels	4	50%	12%	-	87%
Glomerular vessels	7	87.5%	77%	90%	-
Linear vessels	4	50%	12%	-	87%
Arborising vessels	1	12.5%	4%	-	-
Brown globules	4	50%	80%	90%	64%
Black globules	4	50%	30%	-	-
Homogeneous pink background	4	50%	70%	-	-
Homogeneous grey-brown background	6	75%	-	80%	64%

Table 3: Various histopathological features of Bowen's disease.

	No. of cases (n=8)	Frequency
Epidermal Changes		
Full thickness involvement	4	50%
Atypical keratinocytes	8	100%
Crowding of nucleus in keratinocytes	4	50%
Hyperkeratosis	4	50%
Psoriasiform pattern	4	50%
Pagetoid cells	6	75%
Clear cells	2	25%
Basement membrane zone intact	6	75%
Basal layer pigment accentuation	6	75%
Neoplastic cells with breech in Basement membrane	2	25%
Dermal Changes		
Increased vascularity	8	100%
Lymphocytic infiltrates	8	100%

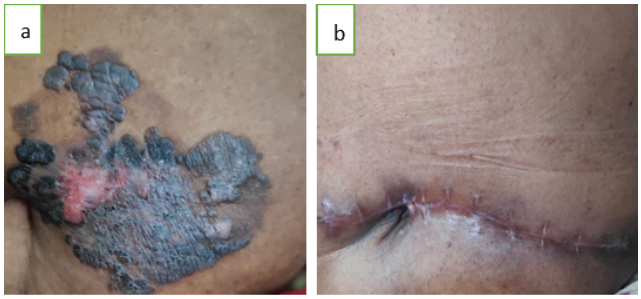

Figure 6: a: (Case 6) Pigmented plaque with ulcerated surface over left lower abdomen; **b:** (Case 6) Post excision

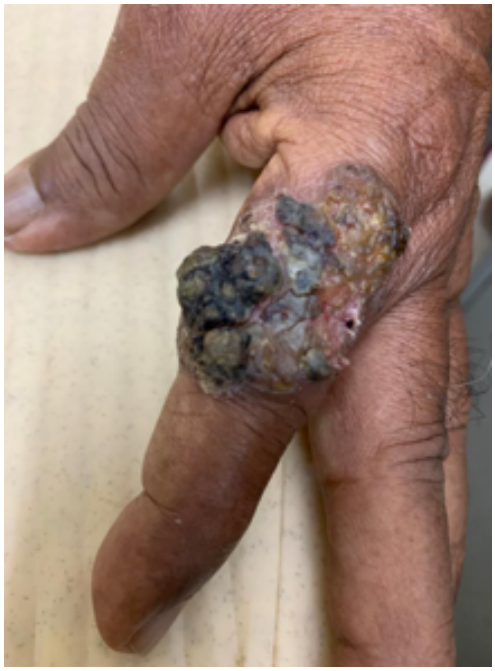

Figure 7: (Case 7) Pigmented verrucous plaque over left index finger

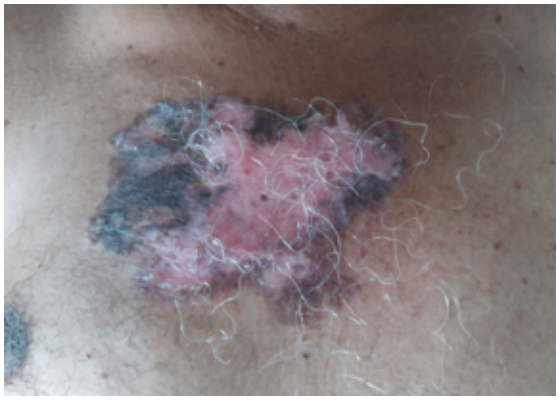

Figure 8: (Case 8) Erythematous and partially pigmented plaque over right side of anterior chest

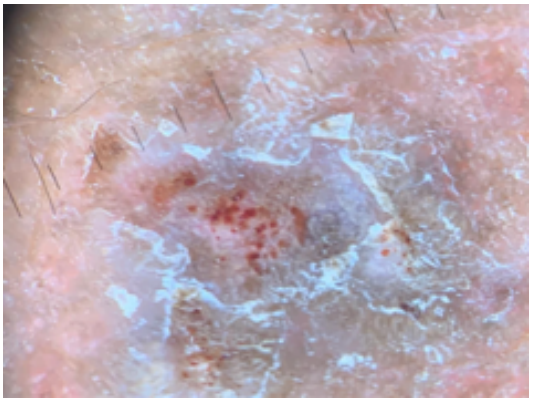

Figure 9: Scaling with Glomerular vessels

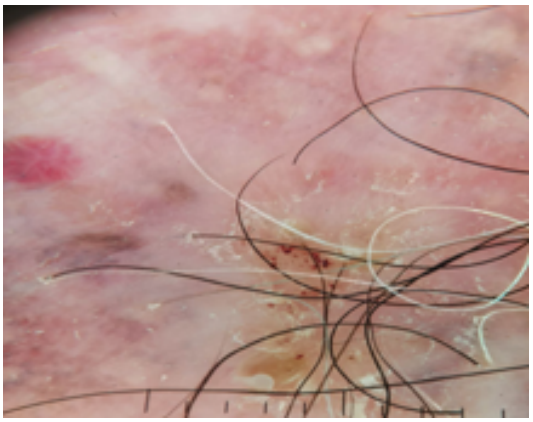

Figure 10: Glomerular vessels with brown globules

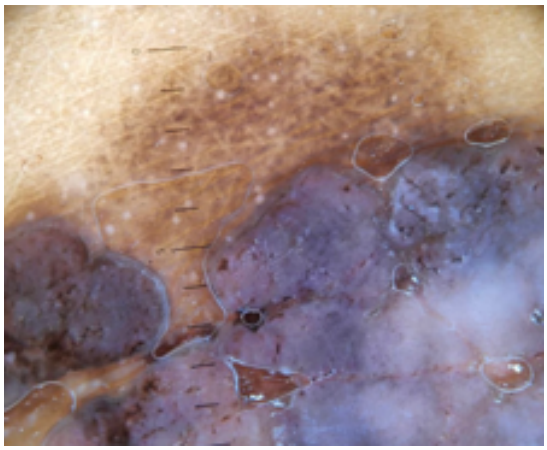

Figure 11: Brown globules with homogeneous grey brown background

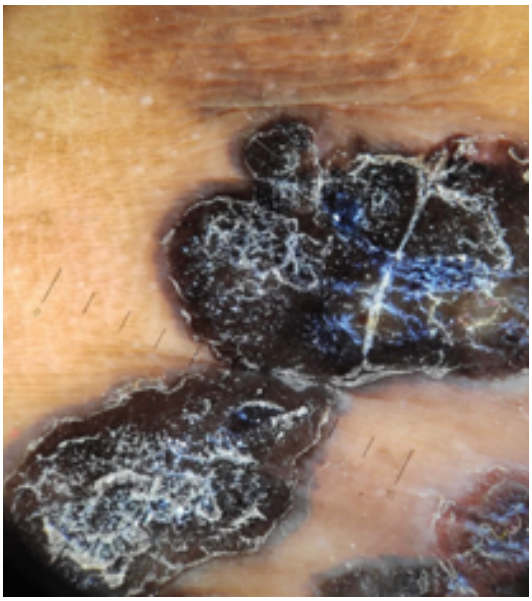

Figure 12: Scaling with homogeneous black background

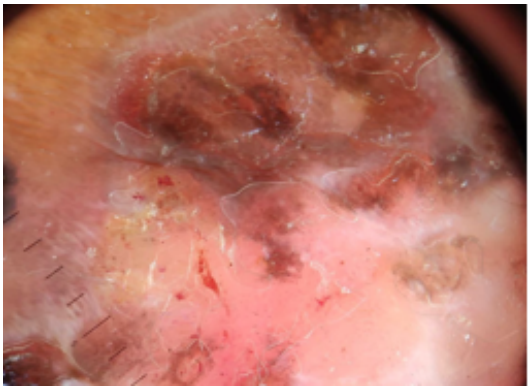

Figure 13: Linear vessels in homogeneous pink background

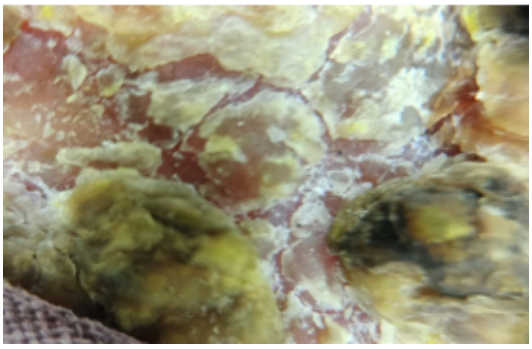

Figure 14: Yellowish hyperkeratotic scales

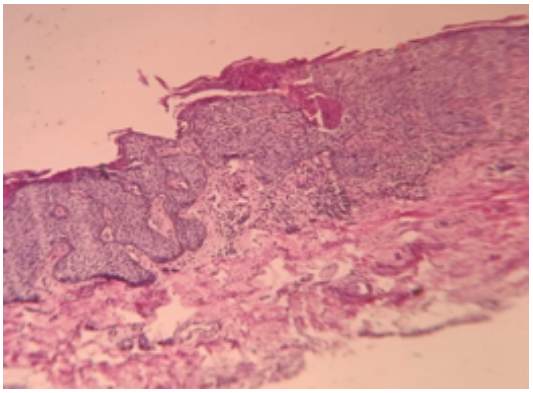

Figure 15: Scanner view (4x)showing hyperkeratosis and psoriasiform pattern

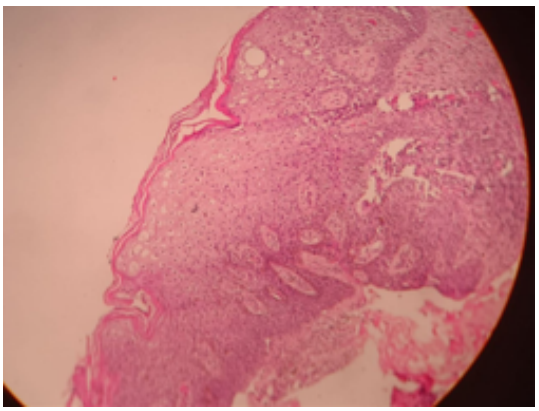

Figure 16: Scanner view (4x) showing atypical keratinocytes in windblown appearance

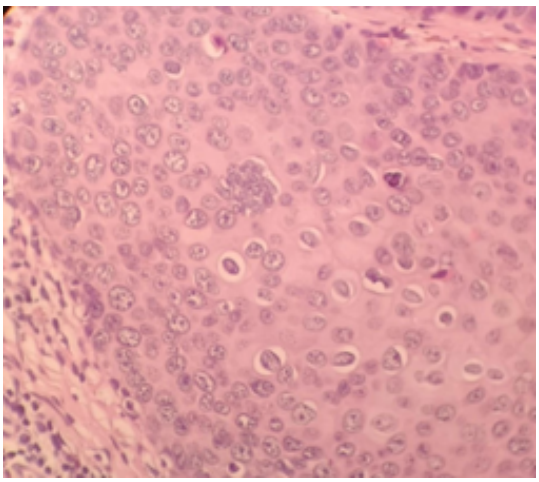

Figure 17: 40x magnification showing crowding of atypical keratinocytes with dyskeratotic cells and high mitotic activity

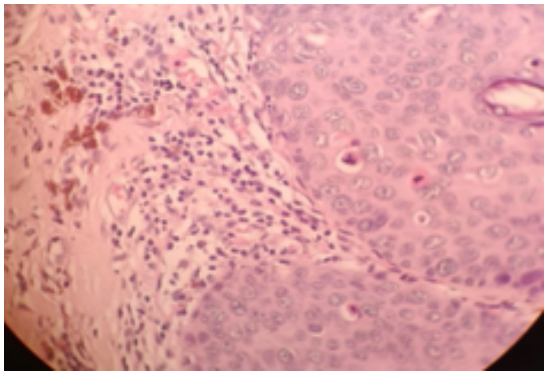

Figure 18: 40x magnification showing dermal lymphocytic infiltrates and pigment incontinence

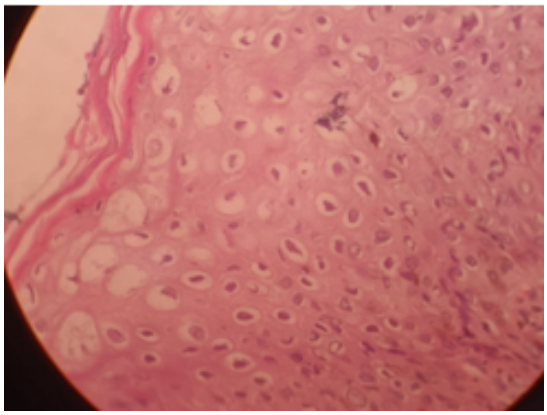

Figure 19: 40x magnification showing multiple clear vacuolated cells (Pagetoid cells)

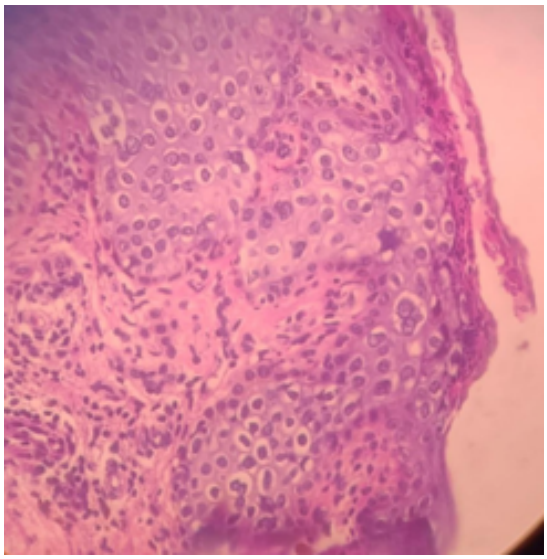

Figure 20: 40x magnification showing full thickness atypia with intact basement membrane

3. Discussion

Bowen's disease is a rare squamous cell carcinoma in-situ of epidermis which has the potential to turn into malignancy. It is seen commonly in elderly individuals between sixth to seventh decade of life with female preponderance, but can occur at any age group.[4] The true incidence of Bowen's disease in Indian population is not known. The incidence is high in Caucasians (1.42/1000).[5] Most of the lesions occur over photo exposed sites due to chronic exposure to ultraviolet radiation. In some patients, photo protected sites may be affected too and in these patients, exposure to arsenic and other chemicals and some of the HPV strains such as HPV 16, 31, 33, 56, 71 are implicated in the etiology.[6] Various studies had reported sixth to seventh decade of life to be the most common age group affected by Bowen's disease. But in this study only 37.5% belonged to sixth to seventh decade while 50% of patients belonged to fourth to sixth decade and there was one patient in eighth decade. A study done by Kossard and Rosen et al, reported head and neck (44%) as the most common site followed by lower limbs (29.8%), upper limbs (19.8%) and trunk (6.5%) in their study on 1001 patients with Bowen's disease.[7] In our study, in 50% of patients trunk was involved followed by upper limbs (25%) and lower limbs (25%).

Two of our patients developed Bowen's disease along the friction prone sites of the waistline which could be due to tight tying of skirts. Many waistline dermatoses due to wearing tight garments have been reported. One similar case was reported by Shankar et al in 2015.[8] Bowen's disease have been reported to arise from seborrheic keratosis, porokeratosis, Beckers nevus, erythema ab igne, small pox vaccination scar, outer sheet of epidermal and follicular cyst.[9] One of our patient had oculocutaneous albinism.

The classical lesions of Bowen's disease are asymptomatic, slow growing, well demarcated, erythematous scaly patch or plaque. It can range from dull pink to bright salmon red erythema. Ulceration is usually a sign of development of invasive carcinoma and may be delayed for many years after appearance of intraepidermal change. Some of the clinical variants described in the literature include intertriginous, verrucous, hyperkeratotic, pigmented, palmar, plantar, genital, periungual and subungual type.[10] Pigmented Bowen's disease which accounts for less than two percent of all is characterised by well defined hyperpigmented, flat, verrucous plaque with velvety surface.[11] Though this variant is rarely reported, in our study 75% of patients had pigmented Bowen's disease.

Dermoscopy has evolved as a quick, non-invasive tool that facilitates early diagnosis of Bowen's disease and helps in prompt initiation of treatment to reduce morbidity of patients. In addition it could be further used to monitor post treatment response of the patient. According to Zalaudek et al, glomerular vessels with scaly surface represent

specific dermoscopic criteria for diagnosis of Bowen's disease.[12] We found these findings in 87.5% of our patients respectively in concurrence with Zalaudek et al and Mun et al.[13] Glomerular vessels are variations of dotted vessels that are convoluted, tortuous capillaries mimicking glomerular apparatus of kidney. The dotted vessels which are characterised by regular or irregular distribution of red dots were seen in 50% of our patients as against 87% reported by Bugatti et al. Payavipapong et al classified Bowen's disease based on dermoscopy into three types.[14] They include 1. Classical Bowen's disease- consists of white scales, atypical vascular pattern, pinkish network. 2. Pigmented Bowen's disease- consists of small brown-black globules, structureless homogenous pigmentation, pigment streaks and networks. 3. Partially pigmented Bowen's disease- comprises both features of classical and pigmented type. Bugatti et al observed multicomponent pattern (64%) which includes black, brown globules and homogeneous grey brown background in addition to glomerular vessels and scales.[15]In our study we observed homogenous grey brown background in 75% of patients, and brown and black globules in 50% of patients.

Histopathological features of Bowen's disease include hyperkeratosis, parakeratosis, acanthosis, atypical keratinocytes with intense mitotic activity, hyperchromatic large pleomorphic nuclei and accompanying loss of polarity which gives an "windblown appearance" to the epidermis. The atypical cells can also extends into infundibula. Basement membrane remains intact and dermis show superficial perivascular mononuclear infiltrate and increased vascularity.[16]Several histopathological subtypes such as pagetoid, psoriasiform, pigmented, verrucous, hyperkeratotic, atrophic, acantholytic, epidermolytic, and orthokeratotic types had been reported.[9] Some of the histopathological variants of Bowen's disease reported by Palaniyappan et al were exhibited in our patients such as pagetoid (75%), hyperkeratotic (50%), psoriasiform (50%), and clear cell variant (25%). We observed that two of our patients progressed to develop invasive squamous cell carcinoma which is consistent with study done by Kao GF et al.[3] Progression to malignancy is seen in 10% of genital Bowen's disease and 3-5% of extragenital Bowen's disease. There is a risk of metastases in 13% of these invasive malignant cases and 10% mortality rate due to widespread dissemination.

Various treatment modalities are available for Bowen's disease depending upon the duration, size, number of lesions, location, comorbidities and immune status of the patient. They include topical 5% imiquimod cream, 5-fluorouracil cream, curettage, electrocautery, photodynamic therapy, Mohs-microscopic surgery, and surgical excision.[17]

4. Conclusion

We report this case series of Bowen's disease to highlight the significance of approaching each patient with high index of clinical suspicion, so that an early diagnosis and prompt initiation of appropriate treatment can be done. This is important because, Bowen's disease (squamous cell carcinoma in-situ of epidermis) may have a varied clinical presentations at unusual sites and progress into invasive squamous cell carcinoma and moreover there could be a plethora of histopathological features.

5. Source of Funding

None.

6. Conflict of Interest

None.

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