Case Report

Hyperkeratotic Bowen Disease-A Case Report

Ahmed SS[1], Ahmed SS[2], Haque, MM[3], Tabassum F[4], Reza AKM[5]

1. **Dr. Syed Shawkat Ahmed, Associate Professor, Department of Dermatology, Jahurul Islam Medical College, Bajitpur, Kishoreganj.*
2. *Dr. Syed Shair Ahmed, Assistant Registrar, Department of Dermatology, Z. H. Sikder Women's Medical College, Dhaka.*
3. *Prof. Md. Muzammal Haque, Professor and Head, Department of Dermatology, Jahurul Islam Medical College, Bajitpur, Kishoreganj.*
4. *Dr. Fahmida Tabassum, Medical officer, Department of Dermatology, Jahurul Islam Medical College, Bajitpur, Kishoreganj.*
5. *Dr. AKM Maruf Reza, Associate Professor, Department of Pathology, Jahurul Islam Medical College, Bajitpur, Kishoreganj.*

**** For correspondence***

Abstract

Bowen's disease is squamous cell carcinoma in situ of the epidermis. It commonly present as an asymptomatic, well circumscribed, solitary, erythematous scaly patch or plaque on sun-exposed site, typically on head, neck or lower limb. We present a case of a 50-year-old Bangladeshi women,with Fitzpatrick type IV skin type, presented with an well defined irregular, itchy,oozy, solitary, hyperkeratotic plaque on lateral aspect of right thigh of 10-years duration with histopathology consistent with Bowen's disease. We hereby reporting this case because of its varied clinical morphology, involvement of sun protected area, pagetoid histomorphology and without any evidence of arsenic exposure, irradiation or immunosuppression.

Keywords: *Bowen's disease, squamous cell carcinoma in-situ, sun-protected area.*

Received: *01.06.2021*

Accepted: *05.03.2022*

Introduction

Bowen's disease (BD) is a rare, progressive, intraepithelial carcinoma which was first described by Jhon Bowen in 1912[1]. It is very common in the Caucasian population with an incidence of 1.42 per 1000[2]. Classically, it present as an asymptomatic, well circumscribed, solitary, erythematous, scaly, patch or plaque on sun exposed site (typically head, neck or lower limb), however multiple lesions have been reported in 10 to 20% of patients especially with arsenic exposure[3]. The risk of transformation is to an invasive carcinoma is 3% to 5% in extragenital lesions and about 10% in genital lesions[4, 5]. Several etiological factors of Bangladesh have been reported and that includes irradiation (ultraviolet irradiation, radiotherapy, photochemotherapy), carcinogens (eg, arsenic), immunosuppression (eg, after organ transplantation, Aquired Immuno Deficiency Syndrome (AIDS). Viral (strong association of perianal and genital lesions with Human Papilloma Virus (HPV); 47% of acral and 24% of nonacral extragenital Bangladesh contain HPV genome) and some others like chronic injury or dermatoses[6, 7]. We hereby reporting this unusual case of hyperkeratotic type Bowen's disease presented to our rural based tertiary care teaching hospital.

Case report

A 50- year old Bangladeshi women, house wife, presented with a persistent occasionally itchy, progressively enlarging, irregularly shaped solitary plaque over lateral aspect of right thigh of 10-year duration in Jahurul Islam Medical College & Hospiatl dermatology outpatient department. Dermatological examination showed a large, single approximately 5×4 cm in size, well defined irregularly shaped hyperkeratotic plaque with slight oozing and ulceration [Fig: 1(A), 1(B)]. Examination of her nail and oral cavity were normal. There were no local regional lymphadenopathy or any other skin lesions elsewhere. Keeping the provisional possibilities of tuberculosis verrucosa cutis, chromoblastomycosis, hypertrophic lichen planus and superficial basal cell carcinoma, an incisional biopsy was performed from the plaque which showed profound hyperkeratosis, parakeratosis, acanthosis with presence of dyskeratoticpagetoid cells within the epidermis. Dermis showed perivascular lympho-histiocytic infiltrate in upper part with intact basement membrane, without any invasion. These histological features were consistent with squamous cell carcinoma in situ Bowen's disease [Fig:2(A),2(B)]. We reconfirmed our diagnosis by a second expert dermatopathologist. Routine investigations of her blood, urine, hair for arsenic and other biochemical tests were within normal limits. Her serological tests were also found negative and she had no history of exposure to arsenic, irradiation, organ transplantation or on any immunosuppressive agents.

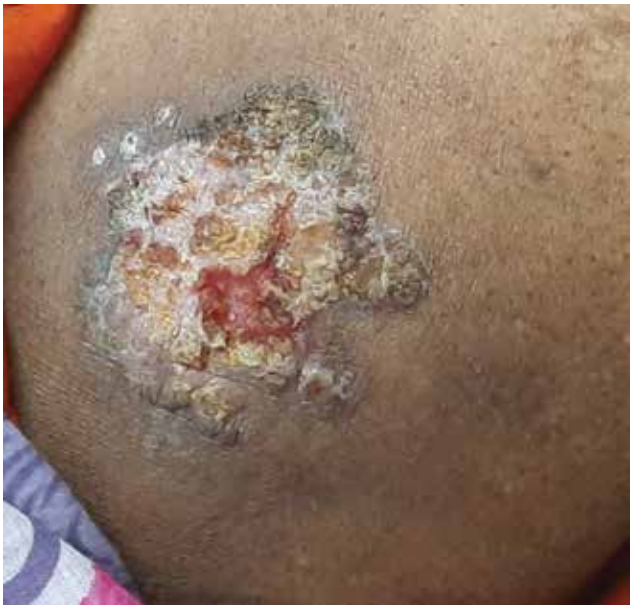

Fig: 1(A)

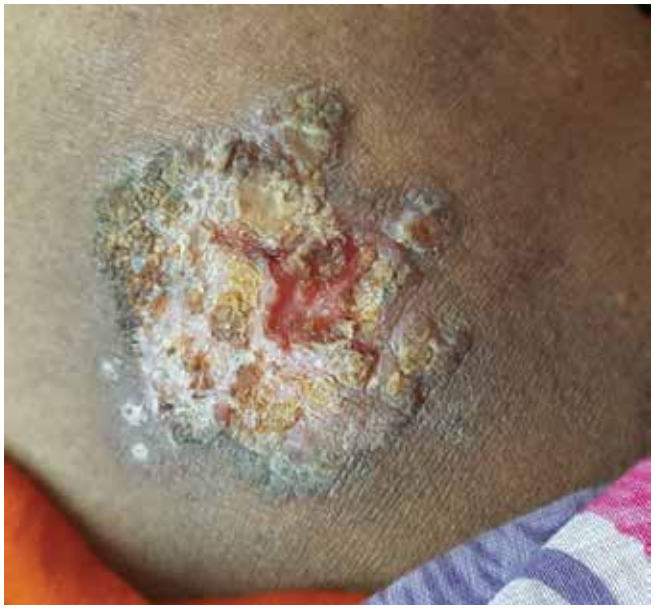

Fig: 1(B)

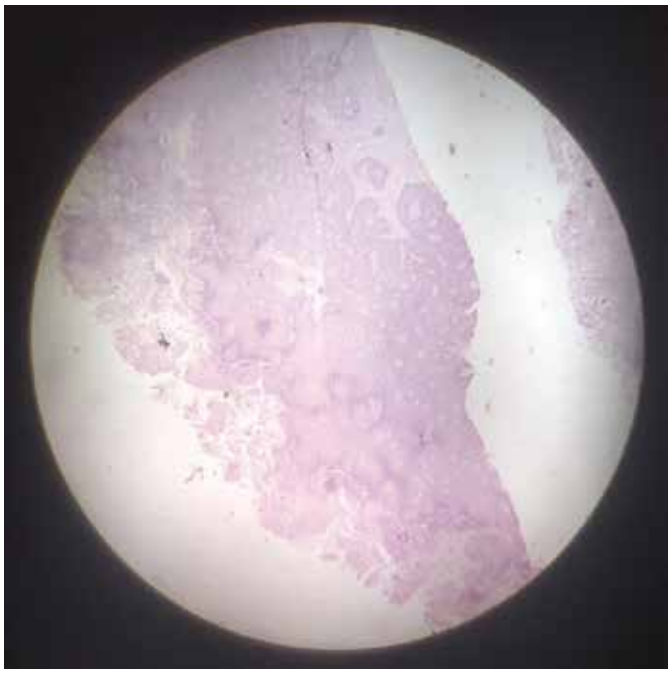

Fig 2 (A): Showing dysplastic epithelium

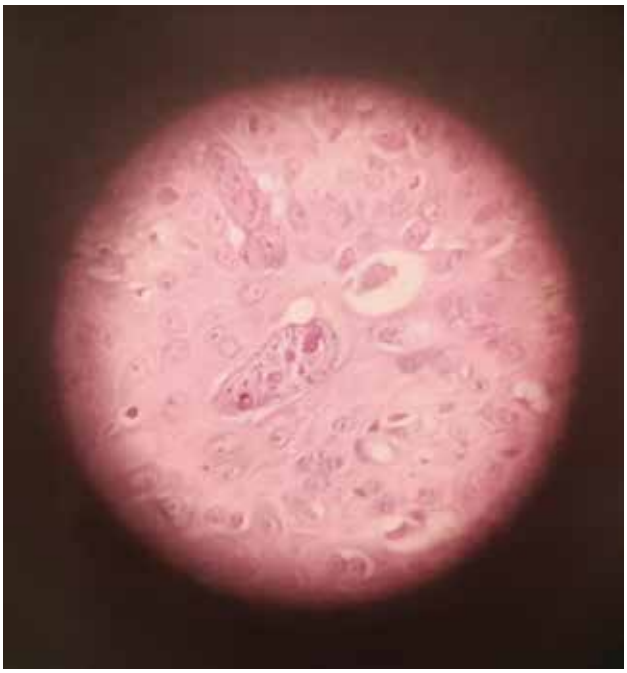

Fig 2 (B): Showing dyskeratotic squamous cells in the epidermis

Discussion

Bowen's disease is a rare premalignant progressive intra-epidermal carcinoma affecting skin and /or mucous membrane with a potential to progress to squamous cell carcinoma[1]. It can occur at any age but mostly seen in elderly Caucasian individuals over the age of 60, with male preponderance. Bowen's disease commonly presents as an asymptomatic, well circumscribed, solitary, erythematous, scaly patch or plaque, but multiple lesions are seen in 10-20% cases with history of arsenic exposure and lesions are then present on palms and soles or covered areas[3,8]. Our case was unusual for several reasons. First, the morphology of the lesion in the form of a solitary, itchy, oozy, irregular, hyprekeratoticplaque was atypical. In our country generally presents as an erythematous, scaly patch or plaque[1]. Other uncommon clinical variants are pigmented, verrucous, atrophic, intertriginous, palmar, planter, genital, periungual and subungual type[1,8]. Hyperkeratotic variant of Bowen's disease has less often been reported, Shyam B et al reported a case in Gujarat, India, and that was on face[8].

Secondly, the location of the lesion over thigh was also atypical. The common sites for Bowen's disease include the chronically photo-exposed sites like head and neck and dorsae of hands and lower legs[7,9]. Thestrup-Pedersen et al[10] described six hundred seventeen cases of bowen's disease out of which 73.5% cases occurred over photo-exposed sites (head and neck, and hands). Kossard et al[11] analyzed data of one thousand one histologically proven cases of Bowen's disease and found head and neck region to be the commonest site (44%) followed by lower leg (29.8%) arm (19.8%) and torso (6.5%). On the other hand, in the series of one hundred eight cases by Cox[12], lower legs were the commonest sites (75%), followed by face and scalp (13%) and hands and wrists (11%). But none of these studies give any details about lesions over thigh.

Thirdly, the histology of our patient's lesion showed profound hyperkeratosis, parakeratosis, acanthosis with presence of dyskeratotic pagetoid cells within the epidermis with intact basement membrane. No vacuolated atypical cells were seen, which are characteristic of arsenical Bowen's disease. Pagetoid

dyskeratosis (PD) is a rare histopathological finding, its incidence being reported as 5% among all cutaneous Bowen's biopsies done in a single institution[13].

Treatment modalities of Bowen's disease include surgical excision, Moh's micrographic surgery, cryotherapy, curettage with cautery, chemotherapy with topical 5-Fluorouracil, 5% Imiquimod cream, laser (CO_2 laser, Ex: YAG ablative fractional laser) radiotherapy and more recently photodynamic therapy (PDT) with topical photosensitizer ð aminolevulinic acid[1,14]. Surgical excision with leaving a safe margin may be a better option than all other modalities for solitary hyperkeratotic Bowen's disease and Moh's micrographic surgery may be considered for genital, perianal, periungual, subungual or recurrent lesion where tissue preservation is essential[9].

All therapeutic options have failures and recurrent rates at least in the order of 5-10%, and no treatment modalities appear to be superior in all clinical situations[9]. Hence a follow up for possible recurrence at 6-12 month is recommended[9], although few authors recommend that gold standard for cure rate in skin cancers (as for other cancers) should be 5 year-point after treatment[15]. A more practical approach is to follow such patients for at least 1 year post-treatment to detect a recurrence.

Conclusion

Although Bowen's disease is most frequently diagnosed in Caucasian population over the age of 60, with Fitzpatrick type 1 & 2 skin types and males are believed to be more frequently affected, it can also occur in population with other skin types. Our case is a 50-year old female with Fitzpatrick type IV skin type and we are reporting this unusual case because of its varied clinical morphology, involvement of sun-protected area, pagetoid histomorphology and no evidence of arsenic exposure, irradiation or immune-suppression.

References

1. Singh S, Khaitan BK, Sharma MC, Seenu V, Kumawat M, Chatterjee P. Bowen's disease on finger. A diagnostic and therapeutic challenge. Indian J DermatolVenerolLeprol 2013; 79: 227-30.
2. Reizner GT, Chuang TY, Elpern DJ, et al. Bowen's disease (Squamous cell carcinoma in situ) in Kaui, Hawaii. A population- based incidence report. J Am AcadDermatol 1994; 31: 596-600.
3. Swaroop MR, Manas SN, Basavaraj HB et. al.Bowen's disease: a series of cases. Int J Health Sci Res. 2015; 5(2): 488-491.
4. Kao GF. Carcinoma arising in Bowen's disease. Arch Dermatol.1986; 122: 1124-6.
5. Cox NH, Eedy DJ, Morton CA. Guidelines for management of Bowen's disease. Br J Dermatol.1999; 141: 633-41.
6. Clavel C, Pham-Huu V, Durlach A, et al.Mucosal oncogenic human papillomaviruses and extragenital Bowen's disease. Cancer. 1999; 86: 282.
7. Cox NH, Eedy DJ, Morton CA. Guidelines for management of Bowen's disease: 2006 update. Br J Dermatol.2007; 156: 11-21.
8. Shyam B, Verma D. Hyperkeratotic Bowen's disease- a case report. Dermat on line journal. 2008; 14(6): 24.
9. Duncan KO, Geisse JK, LeffellDJ. Epithelial precancerous lesions. In: Wolff K, Goldsmith LA, Katz SI, Gilchrest BA, Paller BS, Leffell DJ, editors. thed.New York: Mcgraw-Hill; 2008. P. 1007-27;>[Google Scholar] (https://scholar.google.com/ scholar? hI=en&as_sdt=0%2C5&q=Duncan% 20K).

10. Thestrup- Pedersen K, Ravnborg L, Reymann F. A description of disease in 617 patients. ActaDermVenereol 1988; 68: 236-9. [Google Scholar] (https://scholar.google.com/ scholar?hI =en&as_sdt=0%2C5&q=Thestrup-Pedersen% 20K, %20Ravnborg%20L, %20 Reymann%20).

11. Kossard S, Rosen R. Cutaneous Bowen's disease: An analysis of 1001 cases according to age, sex and site. J Am AcadDermatol 1992; 27: 406. [Google Scholar] (https://scholar.google.com/ scholar?hI =en&as_sdt=0%2C5&q=kossard%20S, %20Rosen%20R.%20Cutaneous%20Bowen).

12. Cox NH. Body site distribution of Bowen's disease.Br J Dermatol 1994; 130:714-6. [GoogleScholar](https://scholar.google.com/scholar?hI=en&as_sdt=0%2C5&q=Cox%20NH.%20Body%20site%20distribution%20of%20Bowen).

13. Strayer DS, Santa Cruz DJ. Carcinoma in situ of the skin: A review of histopathology. J CutanPathol 1980; 7:244-59. [GoogleScholar] (https://scholar.google.com/scholar?hI=en&as_sdt =0%2C5&q=Strayer%20DS.%20Santa%20Cruz% 20DJ.%20Carcinoma%20%).

14. PatelKB.Bowen's disease Treated with Imiquimod and Cryotherapy. Indian Journal of Dermatology 2012; 57(3):239-41.

15. Murphy ME, Brodland DG, Zitelli JA. Definitive surgical treatment of 24 skin cancers not cured by prior imiquimodtherapy. A case series. Derm [GoogleScholar](https://scholar.google.com/scholar?hI=en&as_sdt=0%2C5&q=Murphy%20ME,%20Brodland%20DG,%20Zitelli%20JA.%20Defi).