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Case Report

Oral isotretinoin induced giant lobular capillary haemangioma over scalp: a rare case report

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ABSTRACT

With the advent of isotretinoin in the last 2 decades, dermatotheraputics has reached a major milestone especially in treating moderate to severe acne. Oral isotretinoin is an isomer of all-trans retinoic acid and a metabolite of retinol (vitamin A). In addition to its excellent response in moderate to severe acne, there are a number of well-documented side effects related to the use of isotretinoin. Lobular capillary haemangioma is a very rare side effect. It is sometimes known as granuloma pyogenicum and it is an acquired, benign vascular tumor that arises in tissues such as the skin and mucous membranes. Herein, we report a case of isotretinoin-induced lobular capillary hemangioma over the scalp (an atypical site).

Keywords: Isotretinoin, Lobular capillary hemangioma, Acne

INTRODUCTION

Isotretinoin is an important oral systemic drug in the management of acne vulgaris since its inception. But just like a two faceted coin it is known to be associated with numerous cutaneous adverse effects including chelitis, facial dermatitis, xerosis, pruritus, conjunctivitis, and dry nasal mucosa.[1] Rarely, it also causes lobular capillary hemangiom usually around the nail sulci.[2] Normally, it presents as red papules, polyps, or nodules on the gingiva, fingers, lips, face and tongue of children and young adults, affecting both males and females. Most commonly they are associated with trauma, but systemic retinoids have rarely been implicated as a causative factor in their appearance. They are usually seen in periungual areas but here we report isotretinoin induced lobular capillary hemangioma over scalp, a very rare site.

CASE REPORT

A 27-years-old male presented to our outpatient department (OPD) with history of severe nodulocystic acne on his face and upper back for 4-5 years (Figure 1). He had no other significant systemic complaints. Patient took multiple short courses of oral doxycycline, topical benzoyl peroxide, tretinoin, clindamycin in past but had no clinically significant improvement. Before starting treatment all the required base line investigation was done. Patient was then started on isotretinoin 30 mg per day for the first month and dose was increased to 60 mg per day from the next month. He was responsive to the therapy and his acne improved. When he returned after completing the second month at a dose of 1 mg/kg (60 mg, total cumulative dose of 2700 mg), he reported a lesion on the back of the scalp. The appearance of lesion was acute in onset and it enlarged rapidly over a short period of time to a size of 3×3 cm. It was associated with pain and used to bleed easily on minor trauma. He reported that it began appearing one or two weeks after the increase of isotretinoin dose. He denied any history of trauma, infection or any other drug intake. Oral isotretinoin was then gradually tapered and stopped over the period of next 2 weeks. After stopping the drug, the size began to decrease to the current size of 2×3 cm. Local cutaneous

examination revealed a solitary skin colored pedunculated nodule of size (2×3 cm approximately) with matted hairs, crusting and tenderness (Figure 2). The excision was performed under local anesthesia keeping in mind the fact that complete excision is the preferred method of lesion removal because of risk of recurrence. Moreover, it provides an excellent specimen for histopathologic characterization. Specimen was sent to histopathology which showed flattened epidermis with dermis showing angiomatous tissue arranged in discrete lobules of dilated blood capillary network and poorly canalized vascular tufts (Figure 3).

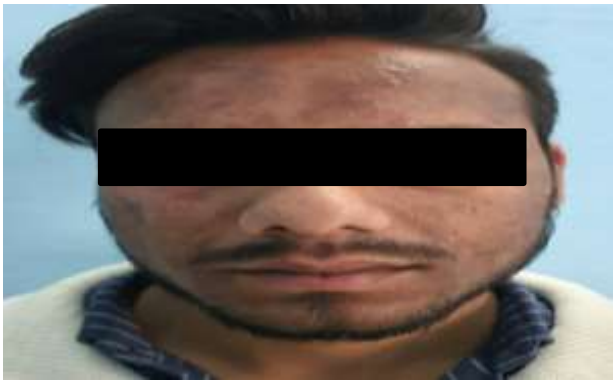

Figure 1: Clinical picture of patient showing nodulocystic acne on face.

Figure 2: Clinical picture showing pedunculated nodular lesion over scalp.

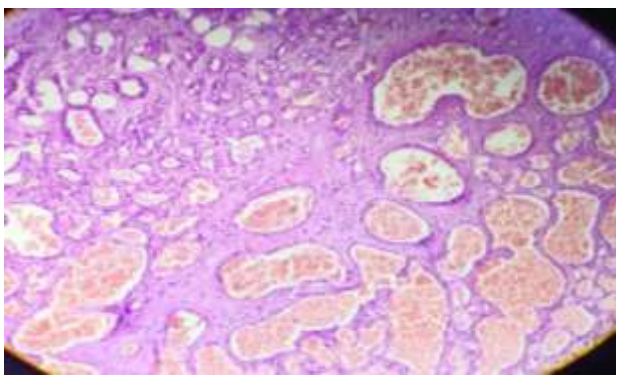

Figure 3: Histopathology: (40x view) shows multiple lobulated angiomatous tissue.

On the basis of clinical and histopathological findings a diagnosis of isotretinoin induced giant lobular capillary hemangioma was made. The dose of isotretinoin was tapered down to (0.5 mg/kg/day). Patient was followed up for 2 months, no recurrence was observed at that site.

DISCUSSION

Isotretinoin was first approved for the treatment of severe acne by the US food and drug administration (FDA) in 1982. It has the reputation of being the only drug that impacts all the etiological factors in the pathogenesis of acne vulgaris i.e., cellular differentiation, reduction in sebum production, influence on comedogenesis by decreasing hyperkeratinisation, altering the microenvironment within the duct making it less favourable for P. acne and also increases host defence mechanism which explain its anti-inflammatory properties. Adverse events of isotretinoin are well known but rarely patient have stimulation of granulation tissue, most commonly leading to the development of pyogenic granuloma eruptions in periungual area. A review of literature disclosed few reports of this unusual adverse reaction. The exact mechanism of development of granulation tissue is not well known but Baran et al first proposed that isotetinoin causes local dryness exfoliative dermatitis and increased skin fragility.[3] This not only increases the chances of trauma and infection in the nail folds but also causes accumulation of scales and debris which induces foreign body reaction. Piraccini et al hypothesized that retinoids cause onycholysis with subsequent desquamation in the proximal nail fold, leading to foreign body reaction.[4] This theory was widely accepted for the development of lobular capillary hemangioma during retinoid therapy. Retinoids are also reported to have angiogenic properties. The development of lobular capillary hemangioma might stem from the effects of angiogenic growth factors particularly vascular endothelial growth factor (VEGF). In addition to the severity and location, underlying cause of lobular capillary hemangioma must be taken into account, because subungual, periungual occur via mechanism different from that of classical PG. Most of the cases reported in the literature are of periungual PG although it can occur on the other sites as well, like scalp, chest, back, face and less frequently on genital area. Exner et al and Valentic et al first reported the formation of PG-like lesions as a side effect of isotretinoin therapy in 1983, followed by a report of eight cases by Campbell et al in the same year.[5-7] Barrales et al reported a case of an 18-years-old female taking 30 mg/day of oral isotretinoin for severe acne had multiple ulcerated exophytic dome shaped lesions prone to haemorrhage in the intergluteal fold and pubic region.[8] Diagnosis of pyogenic granuloma was supported by histopathology findings. This was observed again in a trial of 16 total patients of which three patients were reported to have developed proliferative granulation tissue at the sites of previous acne lesions. In a series of 49 patients with severe nodulocystic acne treated with isotretinoin, Exner et al described 3 male patients in whom

inflammatory hemorrhagic lesions had formed in preceding crusted acne nodules, with pyogenic granuloma developing in one patient on the thigh 2 weeks after discontinuation of isotretinoin (1 mg/kg/d).[5] Topical retinoids have also been reported to cause excessive granulation tissue formation. Dawkins et al first documented PG arising at the site of scalp psoriasis in a single patient being treated with 0.1% topical tazarotene gel in the same location.[9]

CONCLUSION

In our report we are highlighting the atypical presentation of pyogenic granuloma on scalp. The pathophysiology is still not clear but it may be due to increased skin fragility and increased vascular proliferation by the drug. A plausible explanation for our case of isotretinoin induced pyogenic granuloma could be due to increased chances of friction and rubbing of the occipital area during sleep. Further studies are needed to confirm this hypothesis and additional discussion is warranted regarding the possible link between pyogenic granuloma and their atypical presentation.

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