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Case Report

An enigmatic cutaneous presentation of plasma cell granuloma- A rare case report

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ABSTRACT

Plasma cell granuloma is a rare reactive tumour-like proliferations composed mainly of plasma cells, lymphocytes and innate immune cells like macrophages. Cutaneous plasma cell granuloma (CPCG) is extremely rare benign growth. A good immunophenotyping study is required to confirm the diagnosis. Surgical resection is the primary choice of treatment. We report a case of cutaneous plasma cell granuloma in an adult male.

1. Introduction

Plasma cells are medium sized round to oval cells with eccentrically placed nuclei. The nuclear chromatin is often arranged in cart wheel like or clock face pattern. Plasma cell granuloma has been known by different terms such as inflammatory myofibrohistiocytic proliferation, inflammatory pseudotumor, and xanthomatous pseudotumor.[1] Most common site of involvement is lung, although it may occur in any organ.[2] The aetiology of this lesion is still unknown. Various hypothesis has been suggested, that the pathogenesis has infectious, autoimmune, and vascular origins.[3]

2. Case Report

A 25-year-old male presented with single red raised lesion over right cheek for 3 years duration. It started as spontaneous small papule 3 years back, and after that following a blunt trauma, it increased in size for last 6 months. There was no history of evening rise of temperature, photosensitivity or any topical application over the lesion. Regional lymphadenopathy was not observed. On examination there were multiple erythematous papules and nodules coalescing to form a well circumscribed single plaque of size 2X2 cm over right cheek (Figure 1). On palpation, lesion was soft in consistency and non-tender. On diascopy lesion was not blanchable. Dermoscopy showed lobulated lesion separated by strands with crown of the lesion showing blood vessels over erythematous background (Figure 5). There was mild scaling, featureless white areas and few brown dots. Granuloma faciale, lupus vulgaris, Jessner lymphocytic infiltrate and sarcoidosis were considered in the differential diagnosis. Routine blood investigations were within normal limits. Serum immunoglobulin IgG4 level was 32mg/dl(normal). Mantoux test did not show erythema or induration. Punch biopsy was carried out. The haematoxylin and eosin-stained soft tissue section showed epidermis with mild spongiosis, dermis and subcutis showed sheets of plasma cells admixed with granuloma lymphocytes, histiocytes (Figure 2). The stroma contained hyalinized collagen fibres . No spindle cells were seen. Periodic Acid Schiff (PAS), Acid Fast Bacilli (AFB) and Fite Faraco (FF) stains were non-contributory. A provisional diagnosis of plasma cell granuloma was made and immunohistochemistry was carried out to confirm the diagnosis. Immunohistochemistry

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(IHC) study consisting of markers CD 138 and CD 20 which are specific for plasma cells and B-lymphocytes respectively was done. CD 138 showed diffuse strong membrane positivity (Figure 3) specific for mature plasma cells and CD 20 showed focal positivity (Figure 4) correlating with lymphocytes in germinal centre of granuloma. IHC study for Smooth Muscle Actin was negative. Based on the IHC findings, a diagnosis of Plasma cell granuloma was made.

The patient was referred to plastic surgery for complete excision of the lesion. No evidence of recurrence was observed one year after excision.

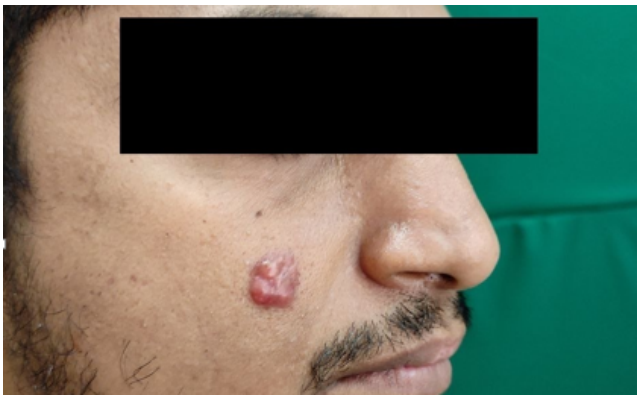

Figure 1: Multiple erythematous papules and nodules coalescing to form a well circum scribed single plaque of size 2×2 cm over right cheek.

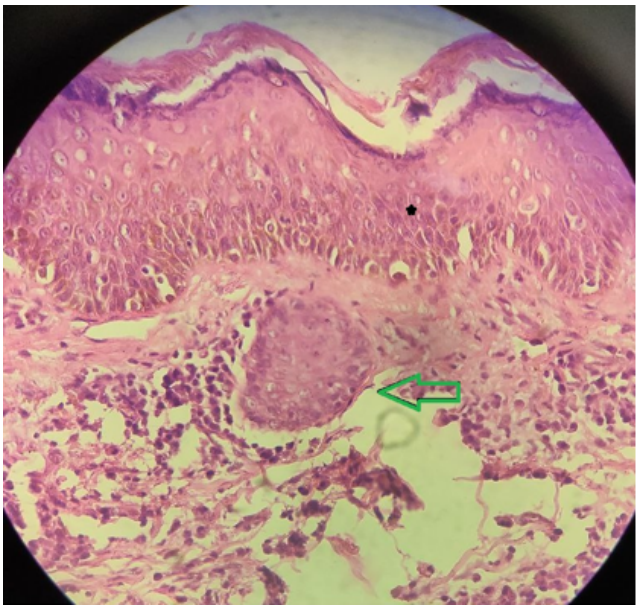

Figure 2: Epidermis with mild spongiosis (black star), dermis and subcutis showed sheets of plasma cells admixed with granuloma (green arrow) showing lymphocytes and histocytes.

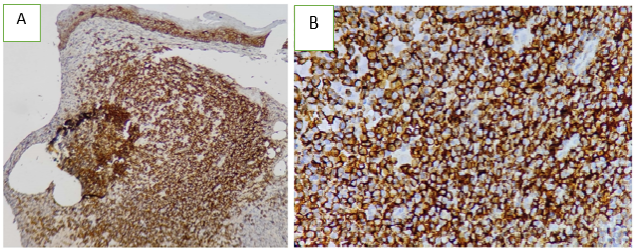

Figure 3: IHC showed CD 138 diffuse strong membrane positivity specific for mature plasma cells;**A:** IHC at 10X; **B:** IHC at 40X

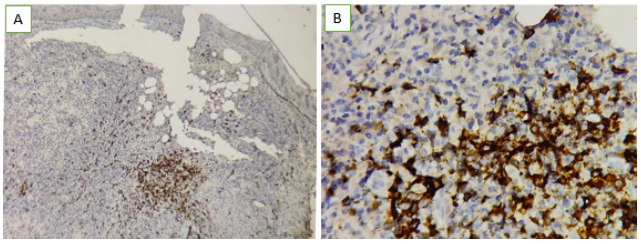

Figure 4:

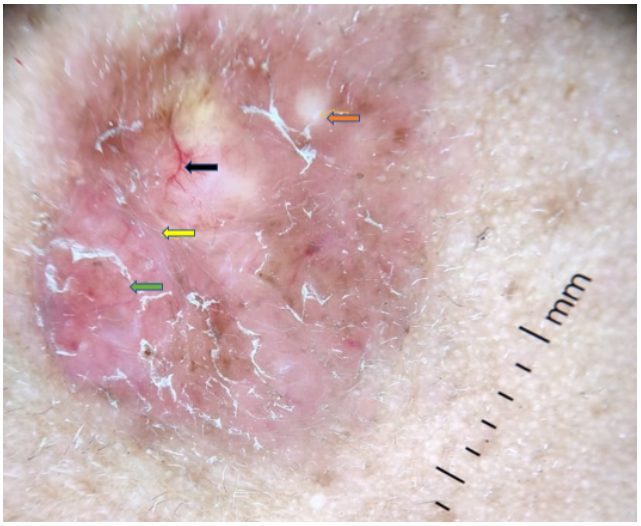

Figure 5: Polarised dermscopy (DERMLITE) shows lobulated lesion separated by strands (Yellow arrow) with crown of the lesion showing blood vessels (black arrow) over erythematous background with mild scaling (green arrow), white areas (orange arrow) and few brown dots.

3. Discussion

Plasma cell granuloma is also called inflammatory pseudotumor and inflammatory myofibroblastic tumor due to its heavy content of lymphoid and plasmocytic elements.[4] First reported by Hurt and Santa Cruz in 1990,[5] CPCG is a rare entity with an elusive aetiology. The presence of polyclonal plasma cells, lymphocytes, and histiocytes suggests an infectious or autoimmune origin.[5] PCG most commonly arises from lungs, although can arise from other

organs[3] like in our case which in uncommon. Though In the head and neck region, orbit and gingiva is the most common location[5] our case had cutaneous presentation over cheek. Plasma cell granulomas are usually solitary[6] as in our case but simultaneous multiple lesions can also occur.[6] This lesion has no sex predilection and may occur at any age.[1] A review of all CPCG cases reported previously,[6] together with ours, has revealed common histopathologic feature of plasma cell-rich infiltrate admixed with neutrophils and lymphocytes.[7] The plasma cells show eccentric nuclei with clear perinuclear h off. The stroma tends to be fibrotic with areas of reactive fibroblastic proliferation.[8] Immunohistochemistry studies show that plasma cells are positive for CD 138[8] and shows membrane positivity as in our case. Polyclonal nature of plasma cells is illustrated by positive reactivity for both kappa and lambda light chains by IHC or in situ hybridization.[8] Surgical excision of the lesion is the treatment of choice with good outcomes.[7] IgG4 disease is one of the very close differential diagnosis for cutaneous PCG which was ruled out in our case as no systemic involvement and regional lymphadenopathy was seen which is very commonly seen in former.[9] Based on histopathology characteristic features commonly seen in IgG4 disease for its diagnosis like storiform fibrosis, obliterative phlebitis and eosinophils were not seen in our case.[9] However confirmatory gamma and kappa light chain IHC study could not be performed due to financial constrains. In terms of prognosis, Plasma cell granuloma is generally benign, nonrecurring condition; but local aggressiveness, and recurrences may complicate the outcome of the disease.[10]

To conclude we report here a case of Plasma cell granuloma for its rare cutaneous presentation. It is infrequent benign lesion of uncertain etiology.[7] While relatively benign, PCG can simulate malignancy, can become symptomatic according to its size and location and thus may create a diagnostic challenge.[8]

4. Source of Funding

None.

5. Conflict of Interest

None.

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