DOI: 10.5958/2319-5886.2015.00089.2

International Journal of Medical Research & Health Sciences

www.ijmrhs.com Volume 4 Issue 2 Coden: IJMRHS Copyright @2015 ISSN: 2319-5886

Received: 7th Feb 2015 Revised: 20th Feb 2015 Accepted: 24th Feb 2015

Case report

DIAGNOSTIC DILEMMA IN A CASE OF GINGIVAL LESION PLASMA CELL GRANULOMA VERSUS EXTRAMEDULLARY PLASMACYTOMA- RESOLVED BY IMMUNOHISTOCHEMISTRY: A CASE STUDY

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ABSTRACT

Plasma cell granuloma is a rare reactive tumor- like lesion composed of polyclonal plasma cells. It primarily affects the lungs but occurs in other anatomic locations such as orbit, paranasal sinuses, larynx, tonsils, ears, tongue, lip, oral cavity and gingiva. A 65- year old female presented with the chief complaint of swelling over the right upper gingiva and mobility of right upper 2nd and 3rd molar teeth since 3-4 months At histopathology due to presence of uniform population of plasma cells a histopathological diagnosis of plasma cell rich lesion was made with a differential diagnosis of extramedullary plasmacytoma and plasma cell granuloma. However, immunohistochemical staining for kappa and lambda chains showed a polyclonal process and antibodies to CD138 were strongly positive, confirming the diagnosis of plasma cell granuloma. The case describes a rare condition of plasma cell granuloma occurring at an unusual site. Authors also emphasize the importance of immunohistochemistry in differential diagnosis of plasma cell rich lesions.

Key words: Gingiva, Immunohistochemistry, Plasma cell

INTRODUCTION

Plasma cell granuloma is a rare, reactive, non-neoplastic lesion composed of polyclonal plasma cells. This entity in the gingiva was first described in 1968 by Bhaskar, Levin and Firch. [1] This lesion does not have a sex predilection and may occur at any age. The exact incidence and etiopathogenesis is unclear. However, it may arise due to periodontitis orperiradicular inflammation due to a foreign body or an idiopathic antigen. Parasitic etiology has also been postulated.[2] It affects various organs like lungs, paranasal sinuses, reticuloendothelial system, orbit, ears, larynx, tonsils, lip, oral cavity and rarely gingiva.[3] In exceptional cases, synchronous and metachronous involvement has also been documented.[2] Histopathologically, it is composed of polyclonal population of plasma cells in a fibrovascular background.Russell and Dutcher bodies can be seen. It is important to distinguish it from plasmacytoma, since the later can be an early feature of multiple myeloma. [3] Immunohistochemistry helps in making this distinction. Treatment is complete resection of the mass. There are conflicting reports about the biological behavior and prognosis. [3]

The present report highlights the occurrence of plasma cell granuloma occurring at an unusual location i.e, gingival with emphasis on the need for distinguishing this tumor like lesion from the other plasma cell rich lesions like solitary bone and soft tissue plasmacytoma. The report also depict the role of immunohistochemistry in arriving at an accurate diagnosis.

CASE REPORT

A 65- year old female presented with the chief complaint of swelling over the right upper gingiva and mobility of right upper 2nd and 3rd molar teeth since 3-4 months. There was no history of rapid increase in the size of swelling. There was no history of trauma. On clinical examination, a solitary, well-defined swelling measuring 1.5 x 1cms, involving the upper free gingival margin and part of the attached margin was present. The swelling was mildly tender, had a smooth pink surface and was bleeding on probing the gingival crevices. There was no exudation of pus. Patient was not a case of diabetes mellitus. A provisional diagnosis of pyogenic granuloma was made. Excision biopsy was done and specimen sent for histopathologic examination.

Routine hematoxylin and eosin stain was done. Immunohistochemical staining for ki67, CD138, kappa and lambda immunoglobulin light chains was done.

Histopathological examination of the specimen revealed sub- epithelial sheets and clusters of plasma cells in perivascular location with many Russell and Dutcher bodies (Figure 1A and B) There was evidence of binucleation and multinucleation(Figure 1 C). At places few plasma cells showed coarse chromatin and prominent nucleoli (plasmablasts) (Figure 1 D). No other inflammatory cells were seen. Hence a histopathological diagnosis of plasma cell rich lesion was made with a differential diagnosis of extramedullary plasmacytoma and plasma cell granuloma.

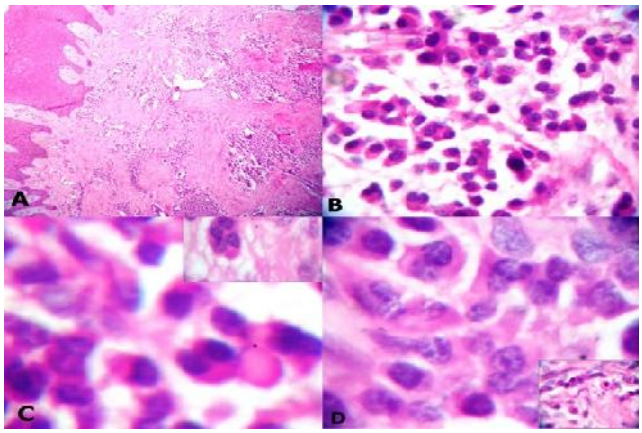

Fig 1: A: Section shows acanthotic epithelium with sub- epithelial sheets and clusters of plasma cells (H & E, × 40) B: Section showing clusters of plasma cells (H & E, × 1000) C : Binucleated plasma cells (single arrow) and Russell body (double arrow) (H&E, × 1000) Inset depicts multinucleated plasma cell (H & E, × 1000) D: Section shows mild pleomorphism with plasmablasts (H & E, × 1000). Inset depicts mott cell-Intracytoplasmic inclusions. (H & E, × 1000)

In view of these findings further work up done to rule out plasma cell dyscrasias. Whole body X ray, renal function test, serum protein electrophoresis, urine for Bence Jones proteins and serum calcium were within normal limits. Hematological profile was normal except for anemia of 9 g %.

Further, immunohistochemical staining for kappa and lambda chains showed a polyclonal process, antibodies to CD138 were strongly positive and immunostaining for ki-67 was negative, confirming the diagnosis of plasma cell granuloma (Figure 2).

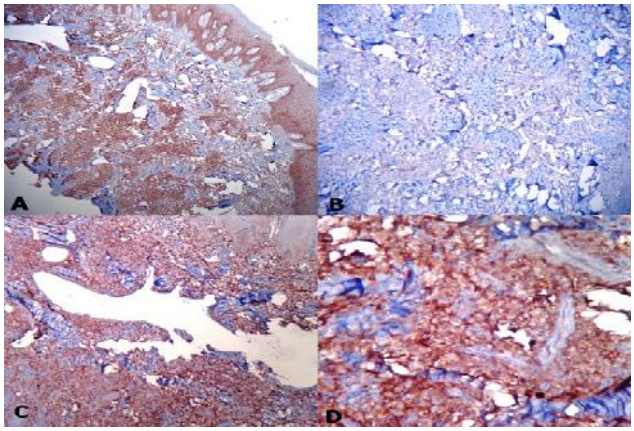

Fig 2: A: Immunohistochemical stain for CD 138 showing strong positivity (H & E, × 100), B: Negative immunostaining for ki- 67 (H & E, × 100), C: Immunohistochemical stain for kappa chains show strong positivity (H & E, × 400), D: Immunohistochemistry for lambda chains show strong positivity (H & E, × 400)

DISCUSSION

Plasma Cell Granuloma is an uncommon tumor like lesion characterized by proliferation of predominantly plasma cells admixed with other inflammatory cells, lymphocytes, histiocytes, mast cells and eosinophils. Myofibroblast has also been demonstrated in the lesion which shows a myxoid/collagenous stroma. The lesion imitates multiple myeloma or plasmacytoma histologically. [4]

This proliferative lesion has predilection for lungs. Other than this it is known to occur in brain, kidney, stomach, heart. When it occurs in the head and neck region, the common sites affected are oral mucosa,

tongue, lip, buccal mucosa, tonsil, paranasal sinuses and rarely gingiva. [3]

Different terminologies have been adopted to describe this lesion which include inflammatory myofibroblatic tumor, inflammatory pseudo tumor, inflammatory myofibrohistiocytic tumor, and so on. [4] In 2002, WHO included it under the intermediate category of fibro myofibroblastic tumors. [5] Varied nomenclature used to describe this lesion has led to a uncertainty over the exact incidence and biologic nature as to inflammatory or neoplastic.

Plasma cell granuloma need to distinguished from other plasma cell rich lesion like osseous solitary plasmacytoma, multiple myeloma, and soft tissue plasmacytoma. Histologically plasmacytoma shows monomorphic population of plasma cells with presence of plasmablast, bi and multinulcation and many Russell and dutcher bodies. In contrast, however, plasma cell granuloma though shows predominance of plasma cells, there will be intermingling of other inflammatory cells like lymphocytes, mast cells and eosinophils. [6]

However histological examination, at times, is misleading as in our case. The absence of inflammatory cells other than plasma cells and abundance of Russell and dutcher bodies accompanied by cells with plasmablastoid morphology lead us to consider plasmacytoma in the diagnosis. Likewise absence of any history of infection or trauma, habit of chewing tobacco or betel nuts furthered up to the diagnostic dilemma. Differentiation from plasma cell neoplastic lesions is imperative given that 14% of multiple myeloma show signs of oral manifestations and that 24% present as solitory plasmacytoma which eventually progress to multiple myeloma. Likewise soft tissue plasmacytoma has predilection for head and neck region. [7, 8]

Although the main contributing pathways for the pathogenesis are hard to pin down, many authors have favored an immunologic basis for the etiology of plasma cell granuloma. Data concerning the molecular mechanisms involved in the pathogenesis is unknown, Coffin et al have documented the finding of human herpervirus -8 DNA sequence and over expression of IL 6 and Cyclin D 1 in PCG. [9] Similar Kim et al in their study on cyclosporine induced plasma call rich gingival growth, have recognized the role of interleukin -6 and phospholipase C- γ1 [10]

Most of the time, complete surgical excision is curative. Controversy exists on role of radiotherapy or steroids in unrespectable cases. [5,6]

Regardless of the verity that PCG is a benign entity, cases showing aggressive behavior and recurrences are on record.

What was known: Presence of other inflammatory cells and Dutcher bodies favor a polyclonal, non-neoplastic process.

Novel insight: Even a pure plasma cell lesion (absence of other inflammatory cells and Dutcher bodies) does not imply a neoplastic process. Hence it is mandatory to evaluate with immunohistochemistry and proliferation markers to rule out a neoplastic process.

CONCLUSION

This case describes a rare condition of plasma cell granuloma of the gingiva. This case highlights the need to biopsy unusual lesions to rule out potential neoplasms. It emphasizes the need for histopathological examination of all excised tissue regardless of clinical diagnosis and the need for immunohistochemistry in the differential diagnosis of plasma cell rich lesions.

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