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CASE REPORT

ATYPICAL PLASMA CELL GINGIVOSTOMATITIS: A RARE CASE REPORT

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ABSTRACT

Background: Plasma cell gingivostomatitis belongs to a group of uncommon benign inflammatory conditions characterised by macular lesions that are bright red, velvety, sharply circumscribed, flat to slightly elevated in nature. Intra-oral lesions can manifest as dense band-like gingival enlargements with plasma-cystic infiltrate seen in the histo-pathological sections. The present case report highlights findings of atypical plasma cell gingivostomatitis further managed through clinical, radiological, histopathology, immunohistochemistry, hematological analysis and treated with surgical periodontal therapy.

Method: Intra-oral periodontal parameters were assessed followed by non-surgical periodontal therapy. In the later phase, internal bevel gingivectomy carried out and approximated with interrupted sutures. Furthermore, gingivoplasty with a 810nm diode laser, for better adaptation of gingival margins was performed. The patient was recalled for follow-up visits at regular intervals and results were maintained.

Result: Intra-oral histopathological sections revealed plasma cell gingivitis whereas hematological reports were suggestive of anemia. Gradual follow up showed reduced gingival inflammation. Post-surgical wound healing was satisfactory. Disappearance of extraoral induration was highlighting feature in the post-operative phase.Further patient was recalled for follow-up visits at regular intervals and results were found to be maintained.

Conclusion: The overall gingival condition was seen to be improving at the follow-up visits. The authors thus conclude that the case of plasma cell gingivitis treated by comprehensive periodontal therapy showed satisfactory results at 3 years follow-up and no recurrence in extra-oral findings was noted.

Introduction

Benign inflammatory conditions affecting the oral cavity are often commonly characterised by a bright red, velvety texture, sharply circumscribed, fixed to underlying tissues, blanching on application of pressure, bleeding on provocation, causing burning of oral mucosa. In 1952, one such immuno-inflammatory condition was described by Zoon et al as plasma cell infiltrate. Similarly, Plasma cell gingivostomatitis (PCGS) is a condition known to affect gingival tissues and the peri-orificial membrane and histo-pathological sections showing an exaggerated infiltrate compromising mainly of plasma cells[1-2].

Plasma cell gingivitis is also referred to as Plasma cell gingivostomatitis, Plasma cell infiltrate, Allergic gingivostomatitis, Idiopathic gingivostomatitis, Atypical gingivostomatitis and Plasma cell orificial mucositis[3-5]. Plasma cell mucositis more commonly is known to affect tongue and lips, conjunctiva, laryngeal region, epiglottis, nasal apertures, vulva. Allergens such as cinnamaldehyde, pipali, karpura, maricha, shunthi, haritaki, Kasni, tejovati, lavanga, pudina, shuddha gairika and sugandhit drava are causative agents for plasma cell gingivostomatitis. In 1995, Van der Kerkhof and Van Barr et al reported extraoral manifestations like oral cheilitis, plasma-acanthoma associated with this oral condition[6-8].

Anaemia of Chronic disease (ACD) is defined as the anaemia occurring in chronic inflammatory conditions, infections, or neoplastic disorders that are not due to marrow deficiencies or other diseases, and occurring despite the presence of adequate iron stores and vitamins[9-11]. The current concept states that the pro-inflammatory cytokines released in to the bloodstream in any chronic systemic disease tend to down regulate the process of erythropoiesis. The processes involved in the progression ACD could be attributed to these cytokines and interferons that further cause shortened red cell survival, blunted erythropoietin response to anaemia, impaired erythroid colony formation in response to erythropoietin and abnormal mobilization of reticulo-endothelial iron stores[12-14].

ACD is also been known to be associated with chronic periodontal diseases. The case we reported here was a case of atypical plasma cell gingivostomatitis with combined periodontal and extra-oral manifestations, most likely highlighting etiology of allergic origin since the patient had a history of using cinnamaldehyde containing tooth powder for maintenance of oral hygiene. Furthermore, the case report incorporates comprehensive management of patient including non-surgical and surgical periodontal therapy.

Case presentation:

A 45 years old female patient reported to the Department of Periodontology and Oral Implantology with chief complaint of swelling of gums in upper and lower front region of jaw since last 3-4 years (Fig No. 1,2). Patient reported no relevant medical history. She showed positive history of pain, discomfort, burning sensation with swelling, difficulty in chewing/mastication, and bleeding on provocation from the gums since last 3 years for which she did not seek any treatment. She used red colored tooth powder with horizontal scrub method for cleaning her teeth. She rubbed this tooth powder with

finger for 15-20 minutes roughly on all surfaces of her teeth and rinsed with water thereafter. She preferred cleaning her teeth only once a day and refrained using any other interdental aid.

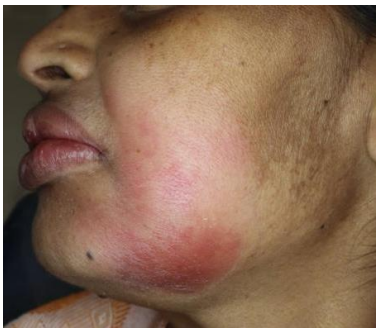


Figure no 1: Pre-operative extra-oral view

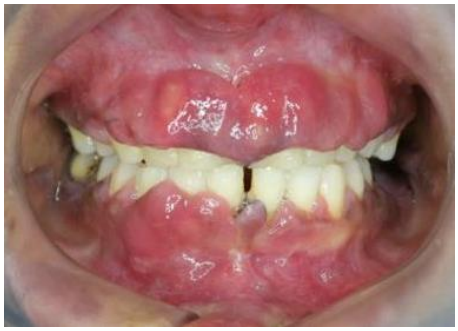


Figure no 2: Pre-operative intra-oral view

Extra-oral examination revealed swollen and crusted upper and lower lips with bright reddish hue on it along with angular cheilitis. (Fig No. 3) Simultaneous bilateral marked induration on both the cheeks was noticed along with dry scaly skin. There was presence of diffuse swelling over the angle of the mandible.

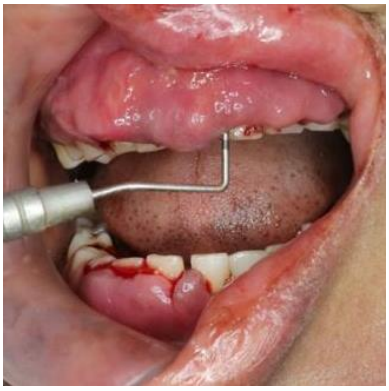


Figure no 3: Pre-operative assessment of periodontal parameters

Intraoral gingival examination showed bright reddish hue bleeding on provocation, soft edematous and friable in consistency with altered contour, absence of stippling. Diffuse erythematous gingival enlargement demarcated from mucogingival junction (MGJ) covering more than 2/3rd of the clinical crown and was generalised to both maxillary as well as mandibular dentition. Gingival contour showed loss of knife edge with blunting of interdental papillae. Minimal supragingival and subgingival deposits were present with marked halitosis classified under severe category by organoleptic rating. Generalized combined pockets extending from 7-9mm were seen with significant clinical loss of attachment.

Radiographic evaluation was advised to help assess bone levels (Fig No. 4). The hematological investigations carried out prior to surgical intervention revealed anemia (Hb count: 7.5gm/dl) with lymphocytosis. She was referred to a general physician for the same, wherein she was advised haematinics (Syrup Haemup) for 3 months. Following that, hematological investigations were repeated after 4 months and they revealed Hb: 10.7gm/dl.

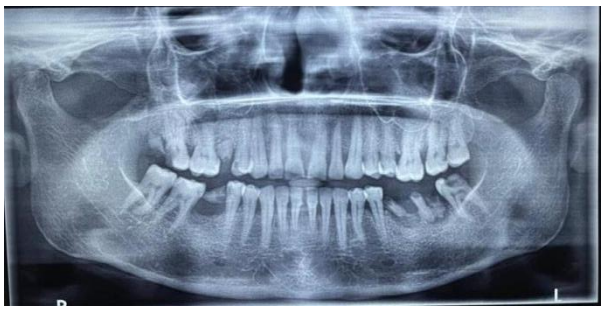


Figure no 4: Radiographic assessment: orthopantomogram

OTHER LABORATORY INVESTIGATIONS:

SERUM IgA: Levels of serum IgA < 5gm/dl are seen to be abnormal. While, the serum

IgA level in the given sample was 3gm/dl pre-operatively. Similarly, post- operatively the levels were as follows : Serum IgA was 90.68 mg/dL (normal range, 90–385 mg/dL), and Secretory IgA was 3.0 mg/dL (normal range, 2–20 mg/dL). Roman et al stated that plasma-cell gingivitis may be associated with low levels of Serum IgA and Secretory IgA, that further leads to localized, repetitive, sub-clinical infections and thus cause formation of plasma-cell infiltrate. As a result, in the post-operative phase, the levels showed a mild surge in both the levels.

The initial line of treatment was Phase I therapy included ultrasonic scaling and root planing followed by a 2 weeks of maintenance with appropriate brushing techniques and patient was advised oral mouth rinse of 0.12% chlorhexidine mouthwash 10ml twice a day for a week. Oral prophylaxis was repeated wherever required and a biopsy was carried out prior to initiation of surgical periodontal therapy. The results of which obtained were plasma cell gingivostomatitis. A written informed consent was obtained from the patient.

In surgical phase, flap technique for gingival enlargement i.e internal bevel gingivectomy/ undisplaced flap surgery was performed. Post debridement; the flaps were approximated using 3-0 black silk suture material with interrupted sutures. The patient was recalled after a week for suture removal. There were no complications seen post flap surgery. Gingivoplasty was carried out with diode laser to keep the margins well-defined. Further patient was recalled for follow-up at 1 week, 15 days, 1 month, 3 months, 6 months, 9 months. Subsequent follow-up visits showed gradual decrease in extra-oral lip swelling as well as a marked decline in gingival bleeding and improvement in periodontal parameters. Post-surgical wound healing was satisfactory. There was no recurrence observed during subsequent visits.

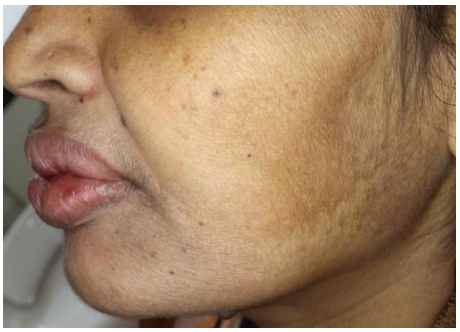


Figure no 5: Post-operative resolution of extra-oral inflammation

In the post-surgical phase (Fig No. 6) topical application of 0.1% triamcinolone acetate was advised thrice a day for a period of 3 months and was tapered gradually which resulted in decreased gingival inflammation, pain, burning and improvement in overall comfort. Following treatment, patient counselling was carried out for change in oral hygiene habits and to motivate patient for continuation of standard oral hygiene measures. Patient was recalled for follow-up almost for 3 years and was found to be well maintained. (Fig No. 7).

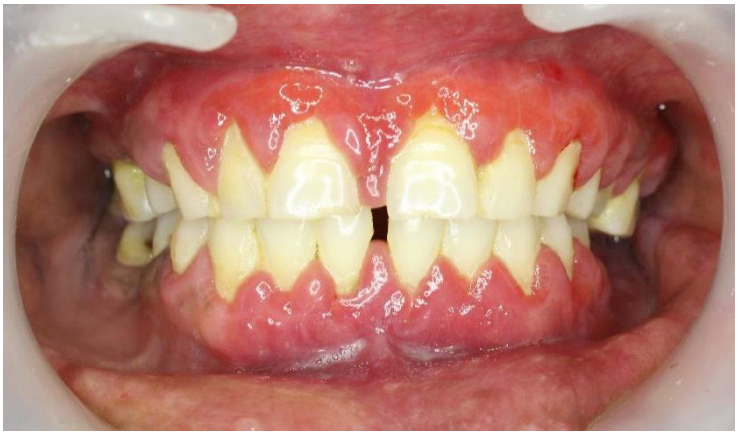


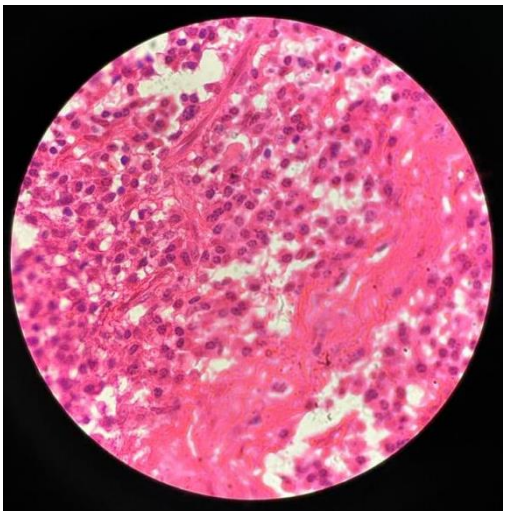
Figure no 6: A 3 year post-operative follow-up

PROVISIONAL DIAGNOSIS: Idiopathic gingivostomatitis

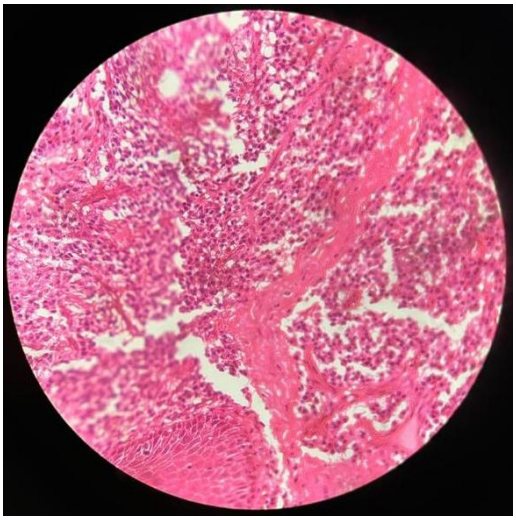
DIFFERENTIAL DIAGNOSIS: Discoid lupus erythematosus, lichen planus, Cicatricial pemphigoid, Leukaemia, Multiple Myeloma

HISTOPATHOLOGY: Histopathological presentation of plasma cell gingivitis.
(a) Spongiotic hyperplastic epithelium overlying inflamed stroma.
(b) Epithelium showing marked acanthosis and leucocytic exocytosis.
(c) Epithelium exhibiting hyperplasia with entrapped fibrovascular cores and spongiosis.
(d) Acanthosis and plasma pooling within epithelium.
(e) Stroma exhibiting intense inflammatory infiltrate with interspersed engorged blood vessels.
(f) High power showing predominantly plasma cells exhibiting eccentric nuclei. (Fig No. 7).

FIGURE NO 7: HISTOPATHOLOGICAL SECTIONS AT 10x AND 100x.

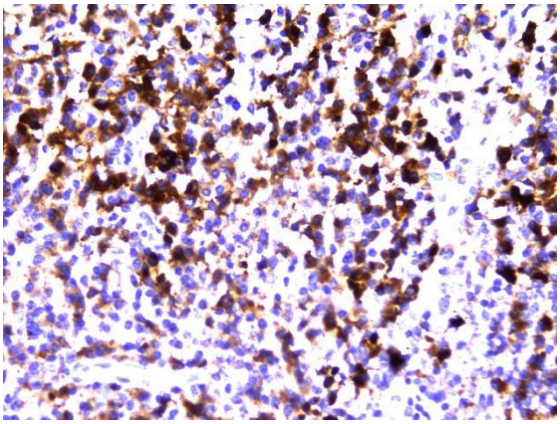


H&E (100x) Photomicrograph showing plasma cells with typical eccentrically placed nucleus exhibiting cart wheel appearance of nuclear chromatin.

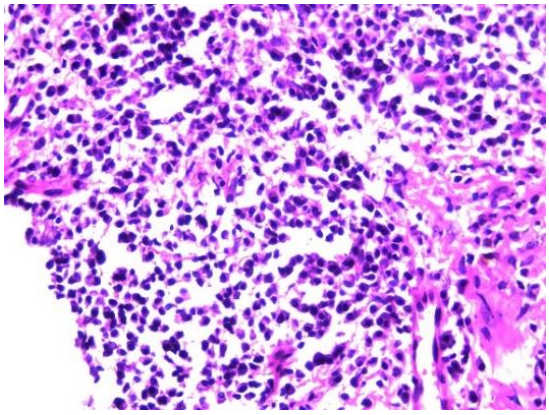


H & E(10x) Photomicrograph showing sub-epithelial aggregation of predominant plasma cells

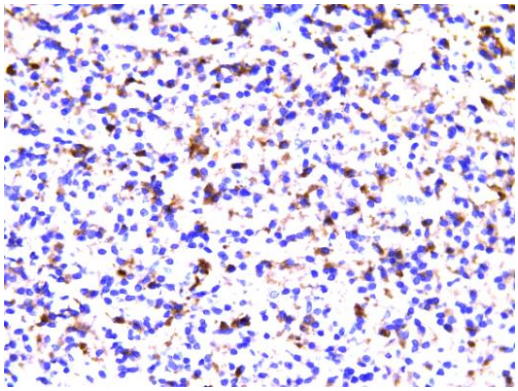
Figure no 8: Immunohistochemical markers kappa and lambda



ASSESSMENT: Histopathological differentials considered were PCG, IgG4 related disease (RD), and PCDs such as plasmacytoma and its multi-centric form-PCM. Immuno histochemically, plasma cells in all the cases showed mixed reactivity for both Kappa and lambda (kappa lambda ratio of 1:01) confirming a polyclonal nature. These cells were negative for CD117, CD56, PAS; immunopositive for CD138 and exhibited low to negative Ki67 proliferation index (1%–4%).



Immunohistochemistry: kappa sections



Immunohistochemistry: lambda sections

FINAL DIAGNOSIS: PLASMA CELL GINGIVOSTOMATITIS

Discussion:

In the present case of plasma cell gingivostomatitis, intra-orally the gingival enlargement was pinkish red in color characterized by velvety texture, sharply circumscribed lesion, fixed to underlying tissues, showed blanching on application of pressure accompanied by bleeding upon provocation and occasional burning of oral mucosa.

Bacteria and their products evoke an immuno-inflammatory reaction within the host tissues. Although this process is intended to eliminate the microbial challenge, it often results in damage[15]. The pro-inflammatory cytokines further interact with hepcidin (the iron regulating hormone) that leads to downregulation of erythropoiesis. A marked rise in WBC's owing to severe gingival inflammation was seen in the blood profile of the patient. A marked decline in erythropoiesis could be attributed to an increased demand to produce neutrophils and other granulocytes with few other immuno-competent cells in response to inflammation[16-18]. Thus, the cause of anaemia in the present case could be considered as ACD in response to the long standing peridontal inflammation. However, ACD being a precursor for long term infections showed a positive co-relation in the present case. In addition, prescribed haematinics along with surgical periodontal therapy aided not only marked surge in the levels of haemoglobin but also aided in betterment of periodontal health.

A true combination of gingivitis, cheilitis accompanied with glossitis is seen in plasma cell gingivostomatitis, as a result it was named as plasma cell gingivostomatitis syndrome by Silverman and Lozada. In 1995, Gargiulo et al classified PCGS as: i) PCGS caused by allergen; ii) neoplastic in origin; iii) unknown in origin[19]. Another case study carried out by Kerr et al 1971 reported presence of cinnamaldehyde in chewing gum with combined triad of plasma cell gingivitis, cheilitis and glossitis. They reported complete regression of the intra-oral swelling upon discontinuity of these chewing gums. Similarly, in the present case plasma cell gingivostomatitis showed decline in extraoral induration over cheeks and complete regression. Patient reported use of red coloured tooth powder and the ingredients of which were as follows cinnamaldehyde, pipali, karpura, maricha, shunthi, haritaki, Kasni, tejovati, lavanga, pudina, shuddha gairika and sugandhit drava. Upon complete discontinuation of this tooth cleaning powder,

favorable results were observed at regular visits which would indicate probable co-relation between the known allergen cinnamaldehyde and clinical condition. Similar results were found in a study carried out by Kerr and Kenneth in 1981[5].

Few studies were carried out to study this inflammatory lesion in detail. Farrier and Perkins et al 2008 reported a case of plasma cell cheilitis, that was diagnosed so due to slightly raised, eroded lip appearance. Differential diagnosis for the same could be cheilitis granulomatosa, dermatitis venenata, actinic cheilitis and plasma cell cheilitis. Furthermore, biopsy ruled out Granulomatous cheilitis as confirm diagnosis[20]. J S Prasanna 2016 stated significance of antigenic identification for appropriate diagnosis of this condition along with clinical, radiographic, haematological and histopathological screening. As a result, all of these investigations were performed along with Serum IgA and Secretory IgA. However, an incisional biopsy revealed a chronic inflammatory lesion[21]. Pulikari et al 2022 treated another case of plasma cell gingivitis with photo-bio-modulation and concluded 5 years follow-up showed no recurrence[22]. Similarly, in the present case, 810nm diode laser used for gingivoplasty resulted in similar additional benefits of photo-bio-modulation. The present case showed erosion and swelling with crusting over the lip surfaces. Intra-orally the gingival enlargement was seen covering anterior 2/3rds of the clinical crown. Thus diagnosis of dermatitis venenata was also considered since the lesion developed in the lip as well as gingiva and could be contact allergy due to shared gingival lesions[23].

Few antioxidants, antibiotics, multi-vitamins, haematinics wherever required, topical corticosteroids could be of help in comprehensive management of this condition. Some authors have reported that potent steroids (eg, clobetasol propionate), both topical and intra-lesional, have produced remission, whereas other authors have reported clear resistance to these steroids and have recommended surgical excision, radiotherapy, or cryotherapy for severe cases[24-25]. Morioka et al obtained spectacular results using intra-lesional injections in extraoral sites and same can be applied to intraoral sites too. Intra-lesional injection of interferon is another effective method used for management[26]. Tamaki et al obtained optimal results using oral griseofulvin in 2 cases of plasma cell cheilitis, one of which had been unsuccessfully treated with prednisolone and topical corticosteroids[27]. However, in the present case report, only use of topical steroids along with haematinics and antibiotics aided better outcomes.

Conclusion:

Careful case history taking prior to initiation of any treatment, hematological investigations, appropriate histo-pathological investigations are essential in order to differentiate other plasma cell infiltrate related infections from PCG. Although recurrences are quite common in this condition, no case till date has reported progression to malignancy, thus emphasizing its nature as benign inflammatory lesion. The present case has shown promising results following comprehensive management using a combined approach.

Conflict of Interest Statement:

None

Acknowledgement Statement:

None

Funding Statement:

None

Summary:

The present case was diagnosed for plasma cell gingivitis and was treated with surgical periodontal therapy and with adjunctive topical steroids. The anemia as well as gingival condition was seen to be improving at the follow-up visits. The authors thus conclude that the case of atypical plasma cell gingivitis treated by surgical intervention shows no recurrence at 5 years follow-up too.

ABBREVIATIONS:

PCG: Plasma cell gingivitis
PCGS: Plasma cell gingivostomatitis
Hb: Haemoglobin
MCG: Mucogingival condition
IHC: Immunohistochemistry
ACD: Anemia of Chronic Disease

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