Case Report

Desquamative gingivitis: A state of uncertainty

Ahila Dommy, Shyam Padmanabhan, Sanjeela Guru

Department of Periodontics, Vydehi Institute of Dental Sciences and Research Center, Bengaluru, Karnataka, India

ABSTRACT

"Desquamative gingivitis" can be a clinical manifestation of a variety of diseases. Definitive diagnoses of these lesions are difficult without the use of advanced diagnostic techniques. Even with the use of various diagnostic aids and techniques, sometimes, the clinician may reach a state of uncertainty. This case report describes one of such cases where the definitive diagnosis could not be established.

Key words: Desquamative gingivitis, diagnostic aids, pemphigus

INTRODUCTION

"Desquamative gingivitis" (DG) is a descriptive term, first introduced by Prinz in 1932, that indicates the presence of erythema, desquamation, erosion, and blistering of the attached and marginal gingiva.[1] The majority of disorders causing DG are systemic in nature, and their oral and extraoral manifestations can cause high morbidity and sometimes life-threatening complications.[2] "DG" can be a clinical manifestation of a variety of diseases, including a number of dermatoses. These dermatoses include pemphigus, cicatricial pemphigoid (benign mucous membrane pemphigoid [MMP]), and lichen planus. However, many other mucocutaneous autoimmune conditions such as bullous pemphigoid, pemphigus vulgaris, linear immunoglobulin A (IgA) disease, dermatitis herpetiformis, lupus erythematosus, and chronic ulcerative stomatitis can clinically manifest as DG.[3] Therefore, it is pivotal to identify the condition to establish the appropriate therapeutic approach. Despite using various advanced diagnostic aids, the cause of DG cannot be elucidated in one-third of cases.[4] The present article describes one of such cases where the definitive diagnosis of the disease could not be established.

CASE REPORT

A 62-year-old male patient reported to Vydehi Dental College and Hospital, Bengaluru, with the complaint of burning sensation associated with redness and bleeding from the lower front gums for 8 months. The burning sensation was aggravated during consumption of the spicy food. Initially, redness of the gingiva was localized in the mandibular anterior region which then gradually appeared in the other areas of oral mucosa. There were no other associated symptoms. Medical history was noncontributory. Personal history revealed that the patient was a nonsmoker, brushes his teeth using neem stick and no alteration in the regular dietary habit, all of which rule out the possibility of thermal burn or any other allergic reactions. The patient had visited a private dental clinic 3 months back for the same and was prescribed an ointment for topical application all over the lesions. Nature of the medication was not reported. There was no resolution of the burning sensation.

Address for correspondence:
Dr. Ahila Dommy,
Department of Periodontology, Vydehi Institute of Dental Sciences and Research, Bengaluru - 560 066, Karnataka, India.
E-Mail: ahiladommy@gmail.com

Access this article online	
Quick Response Code:	Website: www.jiadsr.org
	DOI: 10.4103/jiadsr.jiadsr_4_17

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Dommy A, Padmanabhan S, Guru S. Desquamative gingivitis: A state of uncertainty. J Indian Acad Dent Spec Res 2016;3:52-5.

Examination of the lesion

Intraoral examination revealed diffuse erythematous inflamed appearance throughout the maxillary and mandibular marginal, interdental, and attached gingival with bleeding on provocation [Figure 1]. Multiple bullae were present in the maxillary tuberosity, mandibular right molar region [Figure 2]. Periodontal findings include generalized loss of attachment and attrition. Because of burning sensation and pain, oral hygiene measures were difficult and discouraged for the patient. Based on the history and clinical appearance, a provisional diagnosis of pemphigoid was established. Careful oral prophylaxis was performed, and soft debris from the lesions was flushed out using normal saline. Topical application of 0.1% kenocart was advised for a week.

Investigation

The patient was referred to a dermatologist to rule out cutaneous involvement. Hematological examination was performed, which includes complete blood count, RBS (Random Blood Sugar), and HIV rapid (Tri-Dot method). Hematological examination revealed eosinophilia which gives a differential diagnosis of pemphigus, dermatitis herpetiformis, exfoliative dermatitis, and malignant neoplasms. Radiographic investigation showed generalized horizontal bone loss.

Second visit

On examination, lesions appeared same with no improvement in the clinical conditions. Nikolsky's sign was positive [Figure 3]. Since there was no improvement in the clinical conditions, biopsy and direct immunofluorescence (DIF) were advised and we decided to start with systemic steroids. The patient was advised to take tablet wysolone 40 mg twice daily for 10 days along with tablet betnesol forte.

Biopsy findings

Perilesional biopsy obtained from the attached gingiva of 35. 36 revealed tissue fragments lined by hyperplastic stratified squamous epithelium exhibiting ulceration and atypia of the lining epithelium. Inflammation induced and covered by inflammatory cells. Subepithelium shows dense mixed inflammatory cell infiltrate consisting of lymphocytes, neutrophils, and plasma cells. Proliferating blood vessels are noted. The biopsy was negative for viral inclusions, granulomas, or malignancy [Figure 4].

Direct immunofluorescence

DIF showed well-oriented stratified squamous epithelium which is negative for IgG, IgA, IgM, C3c, and C1q [Figure 5].

Third visit

The patient was reevaluated after 10 days; systemic administration of steroids led to improvement of clinical symptoms. Within 2 weeks, most of the lesions in the mouth had reduced. Since the lesions resolved after systemic steroids [Figures 6 and 7], the drug dosage was tapered. The patient was followed up for 6 months with intermittent recurrence of the lesions in spite of the use of topical steroids.

DISCUSSION

This paper describes the dilemmas of the clinician in arriving at a definitive diagnosis of certain oral conditions. The gingiva is one of the main targets of autoimmune diseases. In 1960 it was suggested that DG is not a specific disease entity but a gingival response associated with a variety of conditions.[3] Providing an exact diagnosis to desquamative lesion becomes very difficult due to various differential diagnostic lesions including chronic bacteria, viral, and fungal infection, as well as reaction to medications, mouthwashes, and chewing gum. Some less common, Crohn's disease, sarcoidosis, some leukemias also present clinically as desquamative lesion.[5] Therefore, it is mandatory to identify the disease responsible for DG. Management of oral symptoms includes removal of contributing/causative factors where possible and medication with topical and systemic immunosuppressants.[6]

Figure 1: Diffuse erythematous inflamed gingiva

Figure 2: Bullae irt maxillary tuberosity

Figure 3: Nikolsky's sign positive

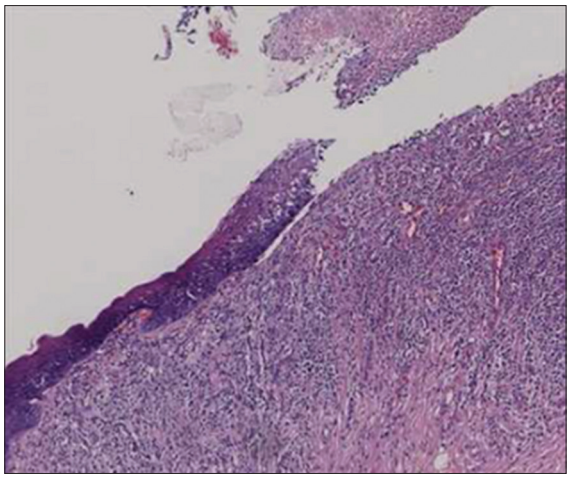

Figure 4: Histopathologic appearance of the lesion

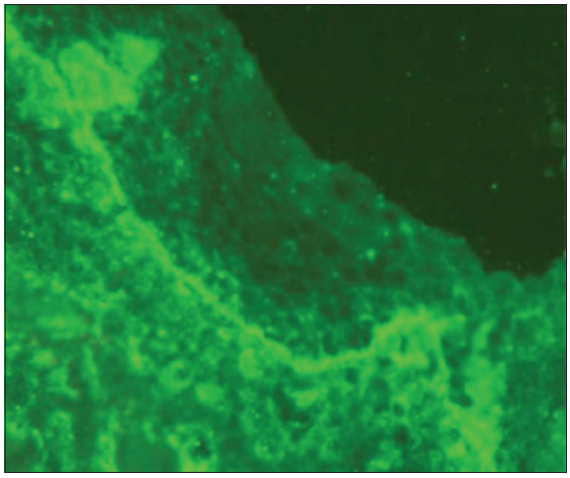

Figure 5: Well-oriented stratified squamous epithelium which is negative for immunoglobulin G, immunoglobulin A, immunoglobulin M, C3c and C1q

Figure 6: Resolution of lesions after systemic steroids therapy

At the first visit, history and clinical examination led us to the diagnosis of MMP. Since the characteristic lesions were restricted only to the mucosa, it was diagnosed as MMP clinically. Similar results were observed in the study done by Rogers *et al*., Laskaris *et al*., and Gallagher and Shklar.[7-9]

Diagnosis of these disorders on clinical grounds alone is difficult; a complete history, general and oral examinations, and biopsy with routine histologic examination are also required. However, to reach a definitive diagnosis biopsy with immunologic techniques, DIF and indirect immunofluorescence are now invariably important.

In our case, histopathologic features were inconclusive. Because of the discrepancies in clinical and histopathologic diagnosis, immunologic test was considered to arrive at a definitive diagnosis. DIF results were also negative for Igs IgG, IgA, IgM, complement C3c and C1q. Similarly, Galgali *et al*. in 2011 presented a case report where they could not achieve a definitive diagnosis. Based on the clinical findings, atrophic lichen planus was given as a provisional diagnosis and histopathologic features were suggestive of bullous pemphigoid. DIF results were negative for Igs IgG, IgA, IgM, complement C3c and C1q.[10]

Clinicians are likely to encounter similar cases in clinical practice where they could not achieve a proper diagnosis. DIF can have its own limitations such as precision in taking biopsy, transportation time, and processing errors. This can lead to poor results or misinterpretation of the processed tissue.[11] To establish a definitive diagnosis, further research is required to develop techniques which are more sensitive and cost-effective.

Furthermore, early diagnosis is important for the proper treatment of these disorders, which can prevent further progression of these diseases, thereby providing better quality of life to the patient. No standard treatment regimen is available to treat the lesion permanently as the main dilemma in diagnosis remains with defining the exact etiology of the condition.[6]

Figure 7: Disappearance of bullae irt maxillary tuberosity

CONCLUSION

Early diagnosis and treatment plan of DG will help us arrest the further progression of the disease. In few cases, due to limitation in the histopathological findings, definitive diagnosis cannot be achieved. Periodic evaluation is mandatory for the appropriate management of the recurrence.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Prinz H. Chronic diffuse desquamative gingivitis. Dent Cosm 1932;74:332-3.
2. American Academy of Periodontology. Oral features of mucocutaneous disorders (position paper). J Periodontol 2003;74:1545-56.
3. Newman MG, Takei HH, Klokkevolcl PR, Carranza FA. Carranza's Clinical Periodontology. 10th ed. Noida:Saunders, Reed Elsevier India Private Limited;2006. p. 411.
4. Rees TD. Adjunctive therapy. In: Proceedings of World Workshop in Clinical Periodontics. Chicago: American Academy of Periodontology; 1989.
5. Richards A. Desquamative gingivitis: Investigation, diagnosis and therapeutic management in practice. Perio 2005;2:183-90.
6. Rana M, Upadhayay P, Hooda A, Dahiya A, Kumar A. Desquamative gingivitis: A diagnostic dilemma. Int J Adv Case Rep 2015;2:1074-7.
7. Rogers RS 3rd, Seehafer JR, Perry HO. Treatment of cicatricial (benign mucous membrane) pemphigoid with dapsone. J Am Acad Dermatol 1982;6:215-23.
8. Laskaris G, Sklavounou A, Stratigos J. Bullous pemphigoid, cicatricial pemphigoid, and pemphigus vulgaris. A comparative clinical survey of 278 cases. Oral Surg Oral Med Oral Pathol 1982;54:656-62.
9. Gallagher G, Shklar G. Oral involvement in mucous membrane pemphigoid. Clin Dermatol 1987;5:18-27.
10. Galgali SR, Krishna S, Toshniwal N, Chaudhary A. Dilemma in diagnosis of mucocutaneous disorders. J Contemp Dent 2011;2:106-10.
11. Newman MG, Takei HH, Klokkevolcl PR, Carranza FA. Carranza's Clinical Periodontology. 10th ed. Noida:Saunders,Reed Elsevier India Private Limited;2006. p. 591.