Desquamative gingivitis: A state of uncertainty

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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. 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. “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. 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. 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.

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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]
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.\cite{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 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 IgG, IgA, IgM, complement C3c and C1q.\cite{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.\cite{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.\cite{6}
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.

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Conflicts of interest
There are no conflicts of interest.

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