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Gingival hyperplasia and conjunctival inflammatory nodule: a diagnostic pathway to sarcoidosis?

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Case report

A 14-year-old patient of South Asian descent presented to the clinic with a chief complaint of a persistent burning sensation in her gums for over 10 months, which has been progressively getting worse in the absence of any identified etiological factor. There was also a reddening of the inferior quadrant of the right eye (Figure 1) for two days resembling a rash. It was accompanied by itching, and irritation when closing the eyes. The split lamp examination yielded conjunctival hyperemia and chemosis. Both eyes exhibited 20/20 visual acuity and normal intraocular pressure (IOP).

Upon intraoral examination, there was generalized pebbly gingival hyperplasia of the maxillary and mandibular gingiva with profuse bleeding on probing and pain. The pain was a dull, continuous background, with an intensity on the Numerical Pain Scale of 5/10 (Figure 2).

A comprehensive blood analysis, which included a complete blood count with differential, clotting factor assessment, comprehensive metabolic, and vitamin panel, revealed elevated serum ACE levels above 100U/L (reference range: 8-65 U/L), elevated ESR levels of 83mm/hr, and Vitamin D levels of 14.9. The Mantoux test yielded a negative result and no abnormalities were observed in the Chest CT scan. The excised overgrown gingival tissue was examined under a microscope and revealed a high concentration of inflammatory cells including lymphocytes, plasma cells, macrophages, Langhans giant cells, and foreign body giant cells. Some areas showed the presence of solid granulomas without necrosis, with two polarizable inclusion bodies present in certain areas. No acid-fast bacilli were detected in the smears taken from the palate, and the culture conducted to isolate Mycobacteria sp. in the Löwenstein-Jensen medium yielded negative results. Therefore, after considering the patient's medical background, as well as the clinical and histological evidence, a conclusive diagnosis of 'Sarcoidosis' was determined.

Discussion

Sarcoidosis is a condition that affects several systems in the body and is characterized by the presence of noncaseating epithelioid cell granulomas in various organs (1). The underlying mechanisms are unclear. Nonetheless, it is believed that the development of this condition occurs as a result of an immunological reaction triggered by variables such as genetic predisposition, bacterial, viral, or fungal infections, as well as external environmental and occupational factors. These factors might ultimately contribute to the production of non-caseating granulomas (2).

The literature has reported a rare occurrence (about 2%) of isolated extra-pulmonary sarcoidosis, without any involvement of the lungs or heart (3). Schroff provided the initial account of oral sarcoidosis in 1942, whereas Poe published the corresponding histology image nearly a year thereafter. Oral manifestations typically manifest in individuals with chronic multisystem sarcoidosis and are rare during the acute phase. The oral lesions can exist as a single, numerous, or as a component of a widespread

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Figure 1. Inflammatory conjunctival nodule with limbally located phlyctenule at the inferior quadrant of the right eye.

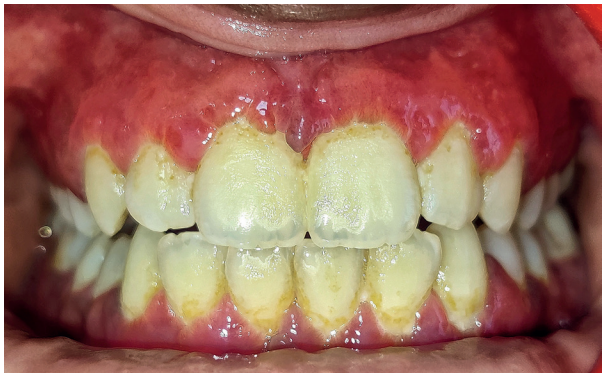

Figure 2. Generalized gingival hyperplasia with a pebbled texture, soft and spongy consistency, affecting both the maxillary and mandibular arches. The gingiva appears reddish-pink in color. Additionally, there is a loss of the normal scalloping pattern of the gingiva and blunting of the interdental papillae primarily observed in the maxillary arch.

disease (4). Oral involvement may be the initial or sole presentation of the disease in certain instances. Sarcoidosis can also cause granulomatous inflammation in any area of the eye and its surrounding structures, potentially leading to significant vision impairment and blindness. The ocular surface manifestations encompass the presence of conjunctival nodules, scleritis, and other related conditions (5). Calcific band keratopathy can occur as a consequence of sarcoidosis, characterized by persistent inflammation and hypercalcemia, which can lead to the accumulation of calcium beneath the epithelial layer (5,6).

Although previous reports have documented occurrences involving the sclera and mouth, the ambiguous nature of the symptoms and their similarity to other inflammatory disorders of the gums posed challenges in making an accurate diagnosis. The diagnosis was made by a process of elimination, particularly of other conditions that cause the formation of granulomas, and by comparing it with the patient's clinical symptoms.

The patient received systemic corticosteroids and hydroxychloroquine to treat systemic symptoms and hypercalcemia. However, to manage her oral condition, we planned to utilize low-level laser therapy to photobiomodulate the tissue. This non-pharmacological method has yielded excellent results thus far.

Conclusion

Diagnosing Oral Extrapulmonary sarcoidosis is difficult and perplexing, even for experienced dentists, because this condition is rare and dentists have limited exposure to it in their practice. Therefore, it is crucial to make a thorough diagnosis based on specific clinical characteristics after ruling out other possibilities to effectively manage the illness.

Conflict of Interest: The authors declare that we have no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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