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Orofacial Granulomatosis in Upper Lip : A Case Report

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Abstract

Introduction:

In 1985 Wiesenfeld *et al*., suggest the terminology of "Orofacial granulomatosis (OFG)" to explain a chronic granulomatous lesion that involving the orofacial area include the lip, face and oral cavity, the histopathological examination usually associated with noncaseating granulomas and multinucleated Langhans-type giant cells

Aim of Reporting This Case:

The aim of reportioning this case is highlighting the awareness of orofacial granulomatosis disease after excluding all diseases that has granulomatous lesion

Case presentation:

A 30-year-old male patient was referred to the oral medicine clinic due to swelling of the upper lip and redness of the gingiva. Clinical examination revealed a nodular lesion in the central area of the upper lip with a firm sensation upon palpation. Additionally, there was slight redness in the gingiva. The patient was medically fit, and our clinical differential diagnoses included Crohn's disease, Wegener's granulomatosis, sarcoidosis, and orofacial granulomatosis.

Lab tests requested for the patient included CBC, ACE, ESR, and CRP. All investigations yielded negative results.The patient was also referred to internal medicine for endoscopy.A biopsy was taken from the inner area of the upper lip under local anesthesia (1.8 mL of lidocaine with epinephrine) and sent for histopathological evaluation. The histopathology report showed non-caseating granulomas, confirming the final diagnosis of orofacial granulomatosis.

Intra-lesional injection of triamcinolone acetonide (40 mg/10 mL) was initiated. At the 2-week follow-up visit, the patient reported softening of the upper lip, though the redness persisted.

Conclusion:

Swelling in the upper lip can have various potential causes, including Crohn's disease, Wegener's granulomatosis, sarcoidosis, and orofacial granulomatosis. Greater awareness of the oral manifestations of these conditions is essential for accurate diagnosis and effective treatment.

Keywords: lip, granulomatosis, oral lesion

Introduction :

In 1985 Wiesenfeld *et al*., suggest the terminology of "Orofacial granulomatosis (OFG)" to explain a chronic granulomatous lesion that involving the orofacial area include the lip, face and oral cavity, the histopathological examination usually associated with noncaseating granulomas and multinucleated Langhans-type giant cells [1].

Tilakaratne *et al*. describe the terminology of "Idiopathic OFG" when lesions are restricted to the oral region without the identifiable granulomatous disease. The diagnosis should not be changed until the patient develops systemic manifestation of a specific granulomatous condition [2].

OFG is an uncommon recognized entity affecting the orofacial region . The age range mean in males is 23 years (range: 5–80 years) and for females 30 years (range: 6–84 years) with more of a female predilection [3].

The etiology of OFG could be associated with genetic, allergy due to various food substances/dental materials, immunological or infective causes [4].
The aim of this study is reportioning case of orofacial granulomatosis located in upper lip after excluding all diseases that has granulomatous lesion.

Case presentation:
A 30-year-old male patient was referred to the oral medicine clinic due to swelling of the upper lip and redness of the gingiva. Clinical examination revealed a nodular lesion in the central area of the upper lip with a firm sensation upon palpation. As shown in figure no.1 . Additionally, there was slight redness in the gingiva. The patient was medically fit, and our clinical differential diagnoses included Crohn's disease, Wegener's granulomatosis, sarcoidosis, and orofacial granulomatosis.
Lab tests requested for the patient included CBC, ACE, ESR, and CRP. All investigations yielded negative results. The patient was also referred to internal medicine for endoscopy. A biopsy was taken from the inner area of the upper lip under local anesthesia (1.8 mL of lidocaine with epinephrine) and sent for histopathological evaluation. The histopathology report showed non-caseating granulomas, confirming the final diagnosis of orofacial granulomatosis.
as shown in figure no.2
The intra-lesional injection of triamcinolone acetonide (40 mg/10 mL) was initiated. At the 2-week follow-up visit, the patient reported softening of the upper lip, though the redness persisted as shown in figure no.3

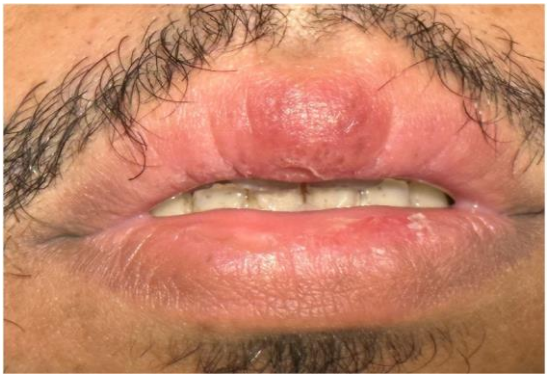

Figure 1: clinical picture shows nodular like lesion in central area of upper lip

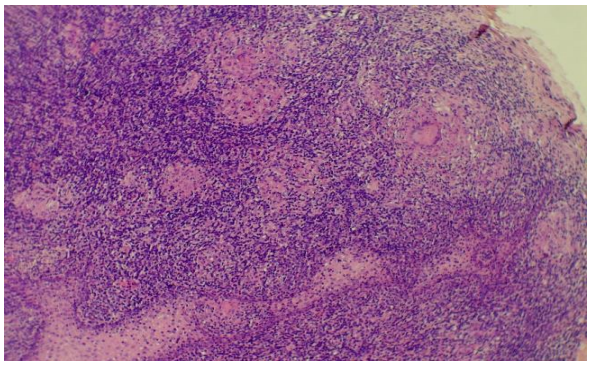

Figure 2: histopathological picture show multiple collection of macrophages surrounded by lymphocyte and clearly shows the multinucleated giant cells

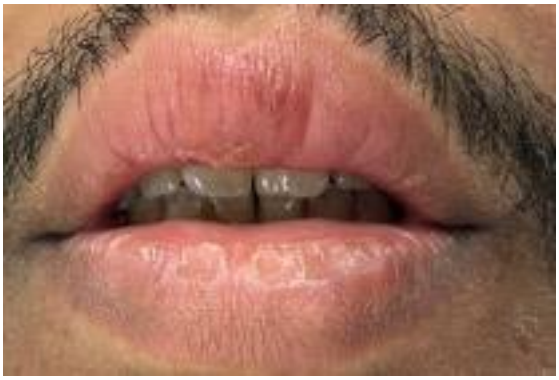

Figure 3: post injection of corticosteroid after 3 weeks patient shows headlining in upper lip and clinically is soft in palpation

Written and verbal consent were obtained from the patient to publish his clinical and histopathological images for scientific purposes. Additionally, IRB approval was granted by the Tabuk Institutional Review Board under protocol No. TU-077/024/266 for this case report.

Discussion:
Orofacial granulomatosis (OFG) can present either as a triad involving facial nerve, lip swelling, and fissured or furrowed tongue, referred to as Melkersson–Rosenthal syndrome (MRS), or as its monosymptomatic or oligosymptomatic forms, referred to as granulomatous cheilitis (GC). GC is a persistent relapsing-remitting, idiopathic, nontender swelling of one or both lips [5].
The etiology is unknown; they may association with granulomatous disease such as Crohn's disease (CD), sarcoidosis, and various infectious agents have been reported but the information is not valid [6].
Pharmacological agents include several medication that can be used to treat such lesion,

topical medication such as (corticosteroids and calcineurin inhibitors), intra lesion injection (corticosteroids), and systemic alternatives (corticosteroids, azathioprine, thalidomide, metronidazole and minocycline) [7,8].

Histopathology always shows a chronic inflammatory cells infiltration [9]. it is usually calcified a granulomatous lesion , less than 50% of patients have noncaseating granulomas, usually small and poorly defined, with lymphocytes surrounding epithelioid histiocytic cells.[9] Multinucleated giant cells can also be seen, with edema of the corium, lymphangiectasia and perivascular lymphocytic infiltration [10].

Idiopathic OFG is considered a diagnosis of exclusion [12]. Both physical examination and biopsy of oral lesions cannot be distinguish OFG, MRS or CD [4,10].

So, the differential diagnosis includes several diseases has same features such as Crohn's disease, mycobacterial infections, sarcoidosis and foreign body reactions [12].

Gastroenterology evaluation is advisable such as endoscopy with biopsy In additional, serum test of angiotensin converting enzyme levels, a tuberculin skin test, chest radiography, Ziehl-Neelsen stain and polarized light microscopy in histopathology [11].

Conclusion:

Swelling in the upper lip can have various potential causes, including Crohn's disease, Wegener's granulomatosis, sarcoidosis, and orofacial granulomatosis

Greater awareness of the oral manifestations of these conditions is essential for accurate diagnosis and effective treatment.

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