

Idiopathic orofacial granulomatosis in a pediatric patient: A case report

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Idiopathic orofacial granulomatosis (OFG) is a rare granulomatous disorder with variable oral manifestations. In children, recurrent gingival enlargement as the predominant manifestation can present a diagnostic challenge and requires careful histopathological and systemic evaluation. This case report presents an 11-year-old boy with recurrent diffuse gingival enlargement, gingival bleeding and difficulty in chewing. The gingival enlargement had recurred following previous surgical excision and was associated with increased probing depths but no evident alveolar bone loss. There was no history of food, medication or cosmetic allergy or relevant systemic illness. Histopathological examination showed non-necrotizing epithelioid granulomas, multinucleated giant cells and chronic inflammatory infiltrate. A complete work-up for secondary causes, including tuberculosis, sarcoidosis and Crohn's disease, was unremarkable, with no evidence of systemic disease, supporting a diagnosis of idiopathic OFG. Initial phase I periodontal therapy produced no appreciable improvement, following which the patient underwent full-mouth gingivectomy and gingivoplasty using electrocautery. The patient showed marked clinical improvement and remained asymptomatic without recurrence during 1 year of follow-up. This case highlights the importance of early recognition, biopsy and systematic evaluation for underlying systemic disease in children presenting with recurrent gingival enlargement and emphasizes the role of individualized management and long-term follow-up in idiopathic OFG.

Keywords: Orofacial granulomatosis, gingivectomy, gingival enlargement, granulomatosis, gingivoplasty

Introduction

Idiopathic orofacial granulomatosis (OFG) is an uncommon chronic granulomatous condition

characterized by persistent or recurrent orofacial swelling and histological evidence of non-necrotizing granulomatous inflammation. Clinically, gingival involvement may present as diffuse, non-tender enlargement that can interfere with oral hygiene, mastication and, in children, normal oral function. Because similar granulomatous changes may occur in systemic disorders, the diagnosis of idiopathic OFG is one of exclusion and requires careful clinical, laboratory and, when indicated, systemic evaluation. Reported associations include Crohn's disease, sarcoidosis, tuberculosis, leprosy, granulomatosis with polyangiitis and other granulomatous conditions¹⁻³. The pathogenesis is incompletely understood and is thought to involve interactions among genetic susceptibility, immune dysregulation and environmental or antigenic triggers^{1,2}. Although pediatric OFG is uncommon, recent reports indicate that children may present predominantly with gingival disease, sometimes without overt gastrointestinal or other systemic manifestations^{3,4,5}. These presentations can create a diagnostic challenge because recurrent gingival enlargement has a broad differential diagnosis, including inflammatory, drug-related, nutritional, infectious and systemic causes. Accordingly, the present case is clinically relevant because the patient presented with recurrent, gingiva-predominant disease, and the diagnosis was established only after histopathological confirmation and evaluation for important systemic mimics. The present case report highlights the challenges and considerations specific to pediatric OFG with gingival enlargement as the sole presenting symptom.

Case Report

An 11-year-old boy reported to the Unit of Pediatric and Preventive Dentistry with the chief complaint of swollen and bleeding gums and difficulty in chewing food for the past two years. The patient was treated in collaboration with the Unit of Periodontology. The patient visited a nearby dentist with the same complaint around 4 years ago. The dentist then performed surgical excision of the growth. After two years, he again developed similar symptoms along with enlargement, which increased gradually to the current size. A thorough history was

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Fig 1 — Intraoral view, (A) frontal view, (B) maxillary occlusal view, and (C) mandibular occlusal view, showing gingival overgrowth on the labial surface in the maxillary and mandibular anterior.

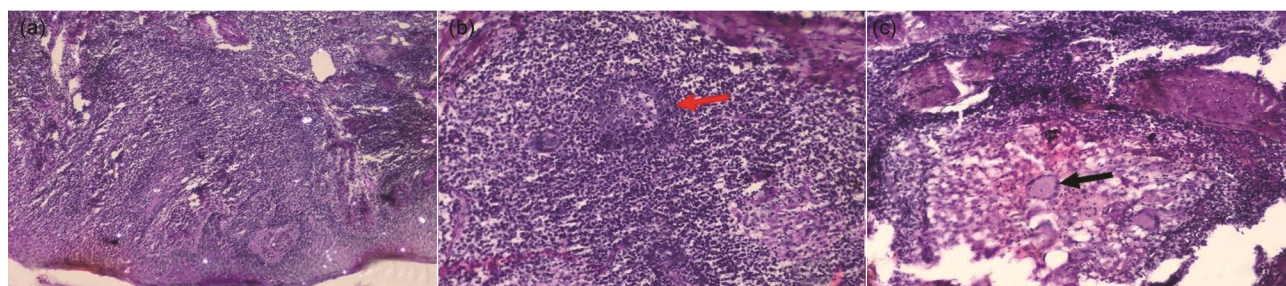


Fig 2 — Histopathological Images showing chronic inflammatory infiltrate with predominant cell type lymphocytes, (A) several noncaseating epithelioid granulomas (B)-denoted by red arrow with giant cells (C) denoted by black arrow.

taken. There was no history of food, medication, or cosmetic allergies, nor was there a family history of contributing factors. On examination, diffuse generalized gingival hyperplasia in the maxillary and mandibular anterior region extending up to premolars was seen (Fig 1).

The gingiva was red, smooth and shiny in appearance. It was mostly soft and oedematous with probing depth up to 10 mm. An orthopantomogram revealed no evident alveolar bone loss. Routine blood investigations and serological viral markers were advised and reportedly fell within normal limits. Vitamin C serum levels were 4.29 mg/L. After discussing the treatment plan with the parents, written informed consent for the procedure was obtained. Phase I therapy, which includes scaling and root planning, was carried out, and oral hygiene instructions were given. The child was advised to rinse twice daily with 0.2% chlorhexidine mouthwash (10 mL) at a 1:1 dilution for 2 weeks. When no improvement was observed with therapy, surgical excision was planned. Under all aseptic conditions, local anaesthesia was administered via infiltration. Crane-Kaplan tweezers were used to mark the bleeding points at the pocket depth. The external bevel incision was made using a Kirkland knife on the facial surface apical to the bleeding points at 45 degrees to recreate the normal festooning pattern of the gingiva. Following the bevel incision,

interdental incisions were given with the help of the Orbans knife. The remaining granulation connective tissue was removed with Gracey curettes, and the final trimming of the gingiva was done with fine Castroviejo scissors. Scaling and root planning were performed to remove any remaining calculus and achieve a smooth root surface for better gingival adaptation. Coe-pakTM (GC, Europe N.V.) periodontal dressing was given for 1 week. The excised tissue was sent for histopathological examination. Histopathological findings revealed hyperplastic stratified squamous epithelium, the stroma showed dense lymphocytic infiltrate and few non-necrotizing epithelioid cell granulomas and multinucleated giant cells were present, suggesting of granulomatous gingival enlargement (Fig 2).

Following this, the patient underwent a thorough investigation to rule out various granulomatous diseases. The patient reported no history of weight loss, appetite loss, or gastrointestinal symptoms, ruling out Crohn's disease. Both the Mantoux test and the sputum test were negative. Additionally, a chest X-ray showed no abnormalities, ruling out tuberculosis. Angiotensin-converting enzyme levels in the serum were assessed for sarcoidosis and were within the normal range. Thus, a final diagnosis of Idiopathic Orofacial Granulomatosis was made. Metrogyl ointment was prescribed for two weeks following surgery. Electrocautery was used to obtain



Fig 3. — Post-operative view, (A) frontal view, (B) maxillary occlusal view, and (C) mandibular occlusal view, showing reduced gingival enlargement after 1-year follow-up.

an optimal gingival margin contour. Follow-up assessments were conducted at 1 week, then at 1, 3, and 6 months, and finally at 1 year following completion of the full-mouth gingivectomy. There was a noticeable regression of gingival inflammation, with no recurrence after 1 year (Fig 3).

Discussion

The present case illustrates an uncommon pediatric presentation of OFG in which recurrent gingival enlargement was the predominant, and clinically apparent, manifestation. The diagnostic challenge in such patients is that gingival enlargement is nonspecific and may result from inflammatory periodontal disease, medications, nutritional abnormalities, hereditary conditions or systemic disease. In OFG, the finding of non-necrotizing granulomatous inflammation should therefore trigger a structured search for secondary causes rather than being interpreted in isolation^{1,6,7}. Granulomatous oral lesions may occur in association with Crohn's disease, sarcoidosis, tuberculosis and other systemic disorders. The absence of gastrointestinal symptoms in the present child reduced the clinical suspicion of Crohn's disease, while negative Mantoux and sputum testing, a normal chest radiograph and a normal serum angiotensin-converting enzyme level provided no supportive evidence for tuberculosis or sarcoidosis. Importantly, these investigations should be interpreted in the context of the clinical presentation; the diagnosis of idiopathic OFG remains a diagnosis of exclusion, and continued clinical surveillance is appropriate because systemic disease may become apparent later in some patients^{3-5,7}. The histopathological findings in this case—dense lymphocytic inflammation with non-necrotizing epithelioid granulomas and multinucleated giant cells—were central to establishing the granulomatous nature of the gingival lesion. Similar gingiva-

predominant presentations have been described in pediatric and adult reports, although the overall condition remains uncommon^{4,5,8,9}. Management of OFG is individualized according to the severity, extent and functional impact of disease. Reported approaches include observation, topical or intralesional corticosteroids, systemic corticosteroids, conventional surgical excision, electrosurgery and laser-based procedures^{7,10}. In the present patient, the enlargement was extensive, recurrent and associated with bleeding and impaired mastication. Initial periodontal therapy did not result in adequate regression; therefore, surgical reduction was selected to restore gingival contour and function. Gingivectomy followed by careful gingivoplasty and electrocautery produced a satisfactory clinical result, with no recurrence observed over 1 year. The favorable outcome also underscores the importance of postoperative plaque control and scheduled follow-up in pediatric patients undergoing surgical treatment. Although the follow-up period was limited to 1 year and the case represents a single patient, the report reinforces a practical clinical message: recurrent or disproportionate gingival enlargement in a child should not be managed solely as a local periodontal problem when histology demonstrates granulomatous inflammation. Histopathological confirmation, targeted exclusion of systemic disease and individualized multidisciplinary management are essential.

Conclusion

Pediatric OFG may present predominantly as recurrent gingival enlargement and can closely mimic other causes of generalized gingival overgrowth. In the present case, histopathological demonstration of non-necrotizing granulomas, together with the absence of an identifiable infectious, systemic, drug-related or allergic cause, supported a diagnosis of

idiopathic OFG. Full-mouth gingivectomy with gingivoplasty/electrocautery restored gingival form and function, and no clinical recurrence was observed during 1 year of follow-up. The case emphasizes the value of early biopsy, systematic exclusion of secondary granulomatous disorders and multidisciplinary follow-up when evaluating persistent or recurrent gingival enlargement in children.

Informed Consent

Written informed assent was obtained from the guardian of the patient. In the form, the guardian of the patient has given consent for his images and other clinical information to be reported in the journal. They understand that the name and initials of the patient will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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