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Long standing Idiopathic gingival hyperplasia of oral cavity with invasion of maxillary sinus: A rare case report

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ABSTRACT

INTRODUCTION: Idiopathic gingival hyperplasia is a rare entity (about one in 1,75,000 individuals). It is characterized by a slow progressive benign enlargement, affecting the attached gingiva, marginal gingiva, and interdental papilla.

PRESENTATION OF CASE: This case report highlights the management of an unusual case of long standing idiopathic gingival hyperplasia involving the right maxillary sinus.

DISCUSSION: Management of gingival hyperplasia depends on the severity of the condition. In this case, surgical excision was performed in both the arches, that resulted in the creation of an oroantral communication, which was protected with a prefabricated custom-made acrylic stent. Despite having a visible raw area of epithelialization evident on the 2nd post operative day, there were no significant signs of recurrence even at a follow-up of 2 years post surgery. Many authors advocate extraction of involved teeth, in addition to the gingival excision, in the presumption of a permanent cure.

CONCLUSION: The patient was satisfied with the resultant esthetic and functional outcome of the treatment. But, the possibility of recurrence cannot be ruled out, so the patient should be kept under close observation. She may also require subsequent surgeries, thus making psychological counseling mandatory.

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1. Introduction

Gingival hyperplasia is usually caused by an increase in submucosal connective tissue elements [1,2]. The propensity of gingival hyperplasia was found to be higher on the palatal rather than the labial side. The involvement of the palatal gingiva is less pronounced in the interdental region whereas the labial involvement appears to be markedly consistent, suggesting that the relative contribution of the connective tissue to the gingival enlargement varies from region to region [2].

The gingival hyperplasia can be seen either as an isolated disorder or as a part of genetic syndromes for instance Murray–Ramon syndrome, Zimmerman–Laband syndrome, Cross syndrome, Jones syndrome, Byars–Jirkiewicz syndrome, and Rutherford syndrome [3,4]. It is classified as iatrogenic, inherited, and idiopathic. The incidence of idiopathic gingival hyperplasia is rare (about one in 1,75,000 individuals) [2] characterized by a slowly progressive benign enlargement, which affects the attached gingiva, marginal gingiva, and interdental papilla [1,5,6]. The condition usually

appears consistent with the eruption of the permanent dentition and is rarely associated with primary dentition [3].

The most accepted mechanism responsible for Gingival hyperplasia is the SOS-1-RAS-MAPK pathway. The activation of this pathway surges the expression of type IV collagen, growth factors, and reduces the expression of matrix metalloproteinase [4,7].

There are various complications associated with gingival hyperplasia. Difficulty in mastication and swallowing, predisposes the patient to swallow partially masticated food that eventually results in gastric disturbances [8].

Early diagnosis and management of gingival hyperplasia are imperative for maintaining optimum gingival health, function, esthetics and also to reduce the adverse effect on systemic as well as psychological health. The usual treatment of minimal gingival overgrowth relies on maintaining proper oral hygiene while cases of advanced gingival hyperplasia require extensive surgical intervention to restore both function and esthetics [6].

This case report describes an unusual case of idiopathic gingival hyperplasia with the involvement of maxillary sinus which is an extremely rare finding. There are no recent case reports found in the literature with similar conditions to date. This work has been reported in line with the SCARE criteria [18].

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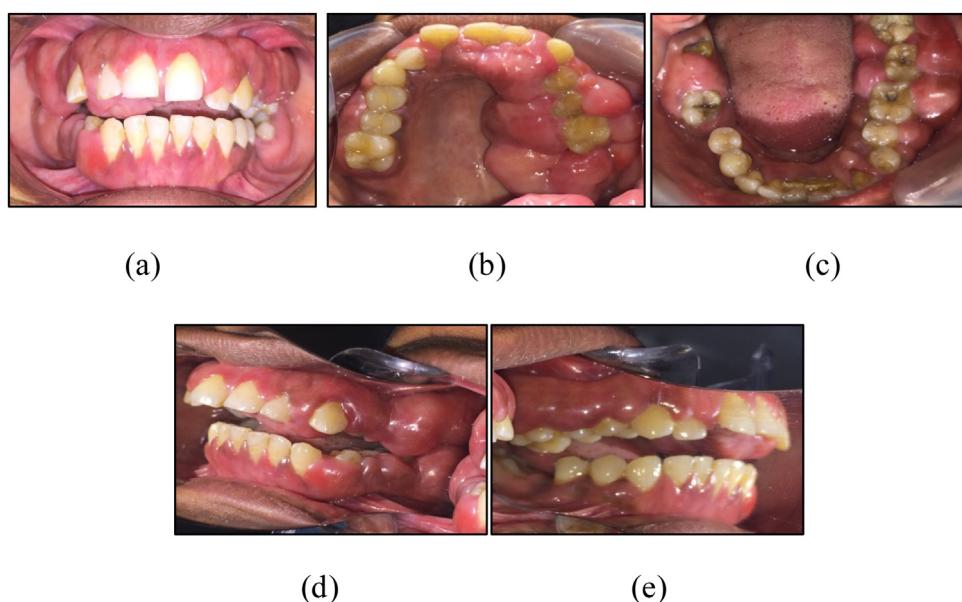
E-mail address: drkainatkhan01@gmail.com (K. Khan).



(a)

(b)

(c)

Fig. 1. Frontal (a) and lateral view of patient (b and c).**Fig. 2.** (a-e) preoperative photographs showing severe generalized diffused nodular gingival hyperplasia.

2. Case report

A 32-year-old female reported with a chief complaint of swelling in the gums of maxillary and mandibular jaw since 7–8 years, along with burning, occasional bleeding, difficulty in speaking, masticating and swallowing food. She was also concerned about the migration and drifting of the maxillary anterior teeth from their normal functional position. She reported to have received nonsurgical therapy for the same several times in the past 6–7 years with temporary symptomatic relief. Her personal, medical and family history was noncontributory.

Extraoral examination revealed a convex facial profile with bimaxillary protrusion and competent lips (**Fig. 1**). Intraoral findings disclosed significant diffused nodular gingival enlargement, on both buccal and palatal/lingual sides of maxillary as well as the mandibular arches. The gingivae were pinkish-red in color, firm and fibrous in consistency, with absence of the characteristic stippling. The enlargement was found to involve the marginal, attached gingiva and the interdental papillae of both arches, that covered almost all the surfaces of the teeth except incisal or occlusal surfaces. The gingival contour was also altered and projected into the

oral vestibule (**Fig. 2**). There was increased mobility and pathologic migration of both anterior and posterior teeth along with the presence of deep pseudopockets. The intermaxillary rest position was also increased. The orthopantomogram (OPG) and cone beam computed tomography (CBCT) scan showed severe generalized alveolar bone loss with floating teeth in both the arches. Significant bone loss causing erosion of the floor of the right maxillary sinus in the molar region was also noted (**Fig. 3**). The hematological investigations were within normal limits. So, an Incisional biopsy was performed and the histopathological examination revealed the features of inflammatory gingival hyperplasia. Based on all the findings available, a provisional diagnosis of idiopathic gingival hyperplasia was made.

Under general anesthesia surgical excision of Gingival overgrowth using scalpel was done arch wise on both the arches along with removal of floating teeth having poor prognosis (18,17,16,15,14,13,12,11,21,28,37,38,48) (**Fig. 4**). The teeth with good bone support were left in place. In the right maxilla, an iatrogenic oranoanal communication was created intraoperatively. For the protection of this fistula preoperatively fabricated custom-

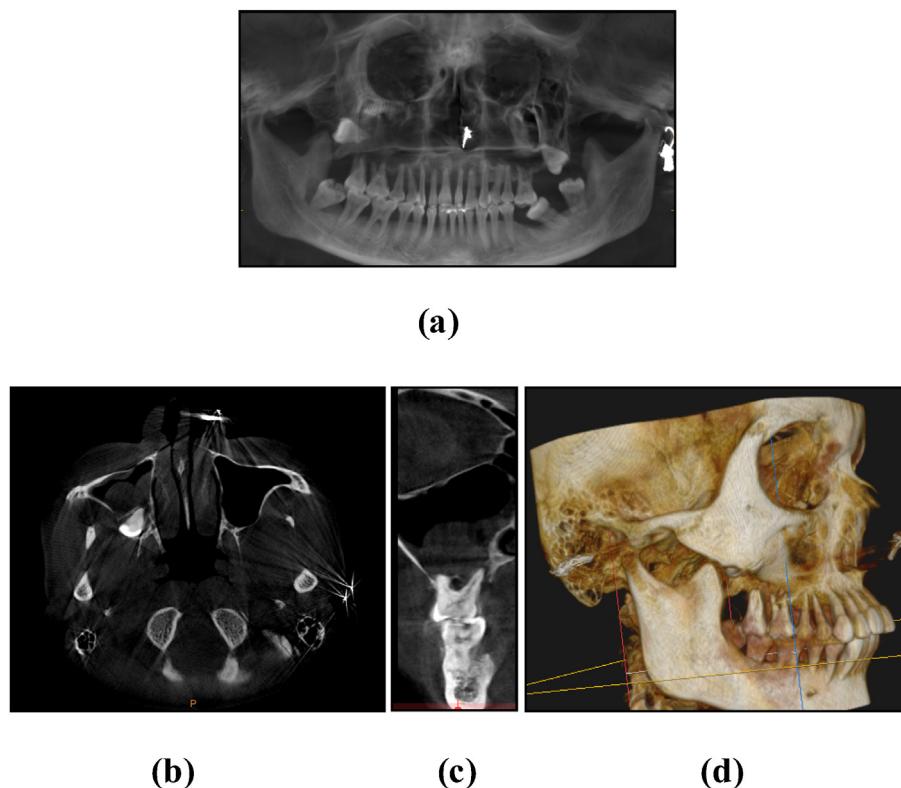
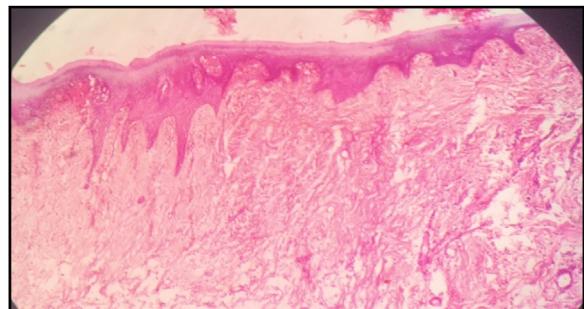


Fig. 3. (a) OPG showing excessive osseous destruction.

(b-d) CBCT scan reveals involvement of right maxillary sinus.



Fig. 4. Excised hyperplastic gingival tissue.



made palatal acrylic stent was placed. The patient's postoperative course was uneventful.

The final histopathological report of excised tissue revealed parakeratinized epithelium, with arcading pattern of rete ridges at places, and predominantly fibrous connective tissue, with collagen fiber bundles arranged haphazardly. The subepithelial chronic inflammatory cell infiltrate was consisting of plasma cells and lymphocytes, and a mild degree of vascularity. This further confirmed the diagnosis of fibrous gingival hyperplasia (**Fig. 5**).

The patient was recalled for follow-up every three months for 2 years, No significant recurrence was noted during this period.

3. Discussion

Gingival hyperplasia is the massive, bulbous, and abnormal enlargement of gingival tissues [1]. It shows a tendency to affect both maxillary and mandibular gingival tissues around the dentition [12].



Fig. 6. Progressive healing of orofistula at 6 month post operation.

Associated complications may include increased distal spacing, drifting of teeth, difficulty in maintaining oral hygiene, affected phonetics and esthetics, pseudopocketing,



Fig. 7. Postoperative photograph of patient at 2 year followup.

loss of masticatory ability, and difficulty in swallowing food [3,9–11].

Etiological factors and associated pathological changes used to classify types of gingival hyperplasia include inflammatory, drug-induced hyperplasia, associated with systemic diseases, neoplastic, and either idiopathic or hereditary gingival fibromatosis [1,13].

Idiopathic gingival hyperplasia is the rarest variety among all forms, with unknown etiology. It is clinically classified as—the symmetric form and the nodular form. The most common is the symmetric form characterized by uniform enlargement of the gingiva, whereas the nodular form shows the multiple enlargements in the gingivae [14]. In the present case, the gingival hyperplasia did not represent any relation to hereditary, syndromes, drugs, conditions, or endocrine problems; hence a diagnosis of idiopathic gingival hyperplasia was made.

In idiopathic gingival hyperplasia, the gingiva clinically appears to be pink, firm, fibrotic in consistency with a minutely pebbled surface [6]. Oral manifestation shows mild to severe enlargement. As a result, the teeth can even become buried, beneath the redundant hyperplastic tissues [1]. The condition may become painful if the tissue gets traumatized during mastication. In the present case too there were similar kinds of manifestations showing generalized nodular enlargement of gingival tissues affecting all the four quadrants of jaws making most of the teeth grade 2 and grade 3 mobile.

usually, the histopathological features consist of the normal overlying epithelium, rete pegs extending deep into connective tissues with some areas of hyperplasia, proliferating dense fibrous connective tissue with pronounced cellularity, reduced vascularity, coarse collagenous fiber bundles, and hyperkeratosis [15]. This case also revealed parakeratinized epithelium, predominantly fibrous connective tissue with an arcading pattern of rete ridges and reduced vascularity.

Management of gingival hyperplasia depends on the severity of the condition. The case reported here is different from other cases documented in literature as it showed the involvement of the right maxillary sinus as a result of excessive osseous destruction. So, looking at the severity of gingival enlargement, the Authors decided to opt for surgical intervention. Many authors advocate excision of the excess gingival tissue combined with the removal of involved teeth in severe and chronic cases because it seems that permanent cure could be obtained. The surgical therapy has been greatly recognized to improve patient's quality of life, as the excision of hyperplastic gingiva facilitates eating, speech, improves esthetics, and provides access for oral hygiene maintenance [16].

Surgical intervention can be carried out with the help of electrocautery, laser, or scalpel [6]. Advanced treatment modalities like laser and electrocautery have advantages in the ability to achieve hemostasis over the scalpel but there are also associ-

ated drawbacks like lateral heat damage, the skill of the operator involved, higher cost and delayed tissue healing. so, the conventional scalpel treatment is always considered to be a better approach in terms of precise incision line, faster wound healing, lower cost. Thus Authors decided to choose scalpel surgery [1,17].

Recurrence rates reports are always unpredictable, with several literature reports outlining no recurrence observed over a period of 2–5 years [6]. It has also been reported that a high rate of recurrence is observed after surgery, but even though the local and psychological benefits are temporary, they must not be underestimated and must outweigh the recurrence [8].

In the present case, initial re-epithelialization of the raw wound was seen immediately on the very next day of excision which is highly unusual and indicative of the faster rate of recurrence but the patient showed an only mild degree of gingival enlargement in the maxillary anterior region mesial to lateral incisor over one month. At the 6 months follow-up, after the procedure, progress in the healing of the oroantral fistula was noted (Fig. 6). The recurred gingival enlargement in the maxillary anterior region up till now at 2 years follow-up didn't show any progress further and also there were no signs of recurrence in the rest of the gingival region (Fig. 7).

4. Conclusion

The patient was satisfied with the resultant esthetic and functional outcome of the treatment. But, Since recurrence could be expected after surgery, the patient should be kept under close observation and may also require repeated surgical management of the recurred gingival hyperplasia thus making psychological counseling necessary for the patients.

Declaration of Competing Interest

The authors report no declarations of interest.

Source of funding

None.

Ethical approval

Ethical approval is exempted by the institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author's contribution

	Contributor 1	Contributor 2	Contributor 3	Contributor 4	Contributor 5	Contributor 6
Concepts	✓	✓	✓	✓	✓	✓
Design	✓	✓	✓			✓
Definition of Intellectual Content	✓	✓	✓	✓	✓	✓
Literature Search	✓	✓	✓	✓	✓	✓
Clinical Study	✓	✓	✓	✓	✓	
Experimental Study	✓	✓	✓		✓	
Data Acquisition	✓	✓	✓	✓		✓
Data Analysis	✓	✓	✓	✓	✓	✓
Statistical Analysis	✓	✓	✓	✓	✓	✓
Manuscript preparation	✓	✓	✓	✓	✓	✓
Manuscript editing	✓	✓	✓			
Manuscript review	✓	✓	✓	✓	✓	✓
Guarantor	✓	✓	✓	✓	✓	✓

Registration of research studies

Not a clinical trial, it's a case report.

Guarantor

Dr. Kainat Khan.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

- [1] V. Pant, S. Pandey, A rare case report of grade III gingival enlargement associated with chronic periodontitis: comparison of two treatment techniques, *Natl. J. Maxillofac. Surg.* 6 (1) (2015) 119.

- [2] J.P. Fletcher, Gingival abnormalities of genetic origin: a preliminary communication with special reference to hereditary generalized gingival fibromatosis, *J. Dent. Res.* 45 (3) (1966) 597–612, <http://dx.doi.org/10.1177/00220345660450032401>.
- [3] K. Kavvadia, E. Pepelassi, C. Alexandridis, A. Arkadopoulou, G. Polyzois, K. Tossios, Gingival fibromatosis and significant tooth eruption delay in an 11-year-old male: a 30-month follow-up, *Int. J. Paediatr. Dent.* 15 (4) (2005) 294–302.
- [4] M. Reenesh, C. Rath, L. Mukherjee, Different surgical management modalities for a case of idiopathic generalised gingival enlargement with chronic periodontitis, *J. Oral Maxillofac. Surg. Med. Pathol.* 26 (1) (2014) 57–60.
- [5] Khetarpal Shaleen, Pitale Unnati, Idiopathic gingival fibromatosis: the Pink elephant - a case report, *J. Pierre Fauchard Acad. (India Section)* 23 (3) (2009) 115–119.
- [6] G. Nagarale, S. Ravindra, S. Thakur, S. Setty, Long term follow up of idiopathic gingival enlargement associated with chronic periodontitis: a case report and review, *J. Indian Soc. Periodontol.* 17 (2) (2013) 242.
- [7] R.E. Stewart, G.H. Prescott, *Oral Facial Genetics*, The CV Mosby Co., St. Louis, 1976, pp. 596–597.
- [8] A. Shetty, H. Shah, M. Patil, K. Jhota, Idiopathic gingival enlargement and its management, *J. Indian Soc. Periodontol.* 14 (4) (2010) 263.
- [9] R. Chaturvedi, Idiopathic gingival fibromatosis associated with generalized aggressive periodontitis: a case report, *J. Can. Dent. Assoc.* 75 (4) (2009) 291–295.
- [10] N. Gagliano, C. Moscheni, C. Dellavia, et al., Morphological and molecular analysis of idiopathic gingival fibromatosis: a case report, *J. Clin. Periodontol.* 32 (10) (2005) 1116–1121.
- [11] A. Doufexi, M. Mina, E. Ioannidou, Gingival overgrowth in children: epidemiology, pathogenesis, and complications. A literature review, *J. Periodontol.* 76 (1) (2005) 3–10.
- [12] M. Jayachandran, S. Kapoor, R. Mahesh, Idiopathic gingival fibromatosis rehabilitation: a case report with two-year followup, *Case Rep. Dent.* 2013 (2013) 1–5.
- [13] Idiopathic Gingival Hyperplasia: A Case Report Merve Sirin, Meral Unur Department of Maxillofacial Surgery, Istanbul University, Turkey, 2020.
- [14] S. Bhatnagar, S. Galgali, G. Gontiya, U. Mohandas, Laser-assisted gingivectomy in pediatric patients: a novel alternative treatment, *J. Indian Soc. Pedodont. Prev. Dent.* 29 (3) (2011) 264.
- [15] B. Lai, J. Muenzer, M. Roberts, Idiopathic gingival hyperplasia: a case report with a 17-year followup, *Case Rep. Dent.* 2011 (2011) 1–5.
- [16] V. Yadav, S. Tewari, R. Sharma, S. Chakraborty, An unusual case of idiopathic gingival fibromatosis, *Contemp. Clin. Dent.* 4 (1) (2013) 102.
- [17] Funde, et al., Comparison between Laser, Electrocautery, and Scalpel in Drug-Induced Gingival Overgrowth, 2020.
- [18] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136.

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