

CASE REPORT

Nonsyndromic with Recurrent Idiopathic Gingival Fibromatosis: A Rare Case Report

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ABSTRACT

Idiopathic gingival fibromatosis is a genetic rare disorder, which is characterized by a progressive enlargement of the gingiva. Gingival enlargement is an overgrowth of the gingiva, which can be caused by various etiological factors such as poor oral hygiene, plaque accumulation, inadequate nutrition, hormonal stimulation, several blood dyscrasias, or long-term intake of certain drugs like phenytoin, nifedipine, or cyclosporine. A 14-year-old female patient reported to the Department of Periodontology, Mahatma Gandhi Dental College and Hospital, Jaipur, Rajasthan with her chief complaint of swollen gums in both upper and lower arches since 3 years, which was gradual in onset and increased in size since 4 months which covered almost half of the surface of each tooth. Also reported that 3 years ago, there was similar swelling for which surgical intervention in form of gingivectomy was carried out. The treatment plan for this case was followed by phase 1 therapy (scaling and root planing) and after completion of phase 1 therapy, the labial tissue from the mandibular anterior region was excised and sent for histopathological examination. Histopathological appearance revealed idiopathic gingival fibromatosis. Thereafter, the conventional gingivectomy under local anesthesia was performed to remove excess gingival overgrowth.

Keywords: Gingival enlargement, Gingival fibromatosis, Gingivectomy, Hereditary.

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BACKGROUND

Gingival fibromatosis is an unusual genetic disorder that causes emotional, functional, esthetic, and also difficulty during mastication. It is marked by an increased gingival enlargement.¹ Gingival overgrowth can be ascribed by various etiological factors such as inadequate oral hygiene, plaque accumulation, scanty nutrition, hormonal stimulation, several blood dyscrasias, or intakes of certain drugs like phenytoin, nifedipine, or cyclosporine for a long duration.^{2,3} The other terminologies of idiopathic gingival fibromatosis are gingival elephantiasis, hereditary gingival fibromatosis, and gingival hypertrophy. Males and females are equally affected and are seen in 1:175,000 people.⁴ Idiopathic gingival fibromatosis can also be associated with other syndromes like Zimmermann–Laband,⁵ Hypertrichosis,⁶ juvenile hyaline fibromatosis,¹ Rutherford,¹ Jones,¹ Ramon syndrome,¹ and tuberous sclerosis. Tuberous sclerosis is a triad of mental retardation, epilepsy, and cutaneous angiofibromas.⁷

The clinical features of gingival fibromatosis are marked by the normal gingival color with firm consistency that is asymptomatic and nonhemorrhagic.^{8,9} The gingival overgrowth can occur either in a generalized form (enlargement covers the gingiva throughout the mouth) or localized.^{9,10} Generally, idiopathic gingival fibromatosis occurs in a generalized form.¹ Idiopathic gingival fibromatosis condition involves an overgrowth of all the parts of the gingiva (interdental papilla, free gingiva, and attached gingiva) on both the maxilla and the mandible. Overgrowth of the gingiva is in the form of a large mass of firm, resilient, and dense fibrous tissue which expands over the teeth.

CASE DESCRIPTION

A 14-year-old female patient reported to the Department of Periodontology, Jaipur with a complaint of swollen gums on both the arches for 3 years, which was slow at the outset, and then the swelling started to dilate for 4 months. Due to swollen

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gums, she was facing a hurdle in speech, while chewing food, and during the cleaning of teeth with the brush and also complaint of bleeding gums and bad breath since last 4 months. The patient had also gone for surgical intervention for similar swollen gums 3 years ago.

The patient had no signs of mental retardation, hypertrichosis, or epilepsy and had no history of any medication that can be the reason for the occurrence of the overgrowth of the gingiva. The history of the family was noncontributory.

Extraoral examination of the patient revealed a convex profile and prominent lips. Intraoral examination exhibiting generalized overgrowth of the gingiva in both the arches. More than half of the crown was covered with gingival tissue (Fig. 1). The gingiva color was normal with dense consistency. Maxillary anterior teeth were proclined. There was spacing between the teeth and a deep bite was also present. Orthopantomogram (OPG) showed localized vertical bone loss with respect to 46 and horizontal bone loss 16 and 47 and except for the third molars, all the permanent teeth were present. The treatment plan was followed by phase 1 therapy (scaling and root planing) and after completion of phase 1 therapy, the labial tissue from the mandibular anterior region was excised and sent for histopathological examination. Histopathological



Fig. 1: Gingival overgrowth in anterior teeth region

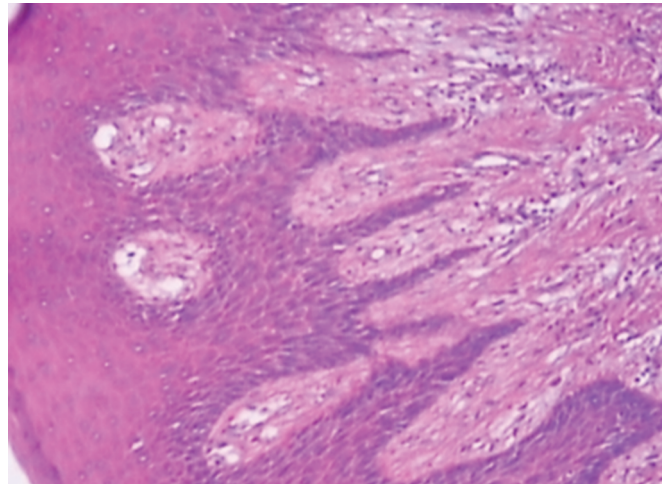


Fig. 2: Histopathological picture of the excised gingival mass



Fig. 3: Gingival overgrowth in the mandibular anterior region



Fig. 4: During the surgery



Fig. 5: Postoperative view of lower anterior teeth after 1 month



Fig. 6: Postoperative view of upper first quadrant after 1 month

appearance revealed idiopathic gingival fibromatosis which showed parakeratinized stratified squamous cell epithelium with elongated rete pegs. The connective tissue showed densely packed collagen bundles (Fig. 2). After the result of the biopsy, the external bevel gingivectomy in association with gingivoplasty was planned and performed under local anesthesia quadrant-wise (Fig. 3). And after the required crown lengthening was attained (Fig. 4) then the excised site was thoroughly irrigated and the periodontal dressing was applied for 7 days and the chlorhexidine mouthwash (0.2%)

was instructed to the patient, to rinse it twice a day and to maintain proper oral hygiene.

After 1-week dressing was removed and postoperative healing was uneventful. And after 1 week next quadrant was excised similarly. After 1 month of the recall, it was observed that there was no recurrence of the enlargement (Figs 5 and 6).

DISCUSSION

Gingival fibromatosis is an infrequent, benign condition that can be cropped up as an isolated disease, chromosomal abnormality, or can be associated with the syndromes.¹¹ It may be congenital or hereditary.¹ The pathogenesis behind the gingival fibromatosis is still not known but it has been observed that increased synthesis of collagen and proliferation of gingival fibroblast may be involved. The condition is genetically linked to chromosomes 2 and 5 at locus 2p21-p22 and 5q13-q22.^{12,13} And son of sevenless-1 (SOS-1) gene mutation has also been reported for the incidence of isolated gingival fibromatosis.¹⁴

It is rarely present at birth^{15,16} and is mostly initiated during permanent tooth eruption. It is less often seen in deciduous dentition. Both esthetic and functional problems resulting from gingival enlargement. Malocclusion impeded eruption, diastemas, incompetent lips, and prolonged eruption of primary dentition are the most common effect which has been seen in idiopathic gingival fibromatosis.¹⁷⁻¹⁹ Plaque accumulation and inadequate oral hygiene may develop gingival inflammation, periodontitis, resorption of the bone, and oral malodor. For the removal of gingival fibromatosis, surgical intervention in form of a gingivectomy is carried out either in a conventional form, electrocautery, or with laser but the most efficacious treatment is conventional gingivectomy.^{11,20}

For stabilization of the effective treatment, good oral hygiene must be maintained, as there is the possibility for the recurrence of the enlargement.¹ Delayed or minimal recurrence can be possible if the patient maintains good oral hygiene and come for a routine check-up and strictly follow the oral hygiene instructions.¹ However, genetic predisposition can also be one of the reasons for the recurrence.¹ Therefore, even in the well-maintained oral hygiene, long-term treatment effects cannot be predicted.¹

CONCLUSION

In children, gingival enlargement is a widespread disease, and for the accurate diagnosis and treatment of affected patients, consulting a specialist like a periodontist and pediatric dentist has a prime role in it. Esthetic correction and impaired function often demand surgical intervention. Routine follow-ups are required after treatment to analyze the overall maintenance of oral hygiene and periodontal treatment stability.

CLINICAL SIGNIFICANCE

Proper diagnosis and the known etiological factor can help in the future treatment plan associated with gingival enlargement.

REFERENCES

1. Coletta RD, Graner E. Hereditary gingival fibromatosis. A systematic review. *J Periodontol* 2006;77(5):753-764. DOI: 10.1902/jop.2006.050379.

2. Jadhav AS, Marathe SP, Recurrent idiopathic gingival fibromatosis with generalized aggressive periodontitis: A rare case report.
3. Lai B, Muenzer J, Roberts MW. Idiopathic gingival hyperplasia: a case report with a 17-year follow up. *Case Rep Dent* 2011;2011:986237. DOI: 10.1155/2011/986237.
4. Fletcher J. Gingival abnormalities of genetic origin: a preliminary communication with special reference to hereditary generalized gingival fibromatosis. *J Dent Res* 1966;45(3):597-612. DOI: 10.1177/00220345660450032401.
5. Holzhausen M, Goncalves D, Correa Fde O, et al. A case of Zimmermann-Laband syndrome with supernumerary teeth. *J Periodontol* 2003;74(8):1225-1230. DOI: 10.1902/jop.2003.74.8.1225.
6. Horning GM, Fisher JG, Barker BF, et al. Gingival fibromatosis with hypertrichosis: a case report. *J Periodontol* 1985;56(6):344-347. DOI: 10.1902/jop.1985.56.6.344.
7. Korol UB, Schoor R, Nanda V, et al. Gingival enlargement as a manifestation of tuberous sclerosis case report and periodontal management. *J Periodontol* 2008;79(4):759-763. DOI: 10.1902/jop.2008.070407.
8. Bozzo L, Machado MA, de Almeida OP, et al. Hereditary gingival fibromatosis: report of three cases. *J Clin Pediatr Dent* 2000;25(1):41-46. DOI: 10.17796/jcpd.25.1.e254616x22403280.
9. Bozzo L, de Almeida OP, Scully C, et al. Hereditary gingival fibromatosis: report of an extensive four-generation pedigree. *Oral Surg Oral Med Oral Pathol* 1994;78(4):452-454. DOI: 10.1016/0030-4220(94)90037-x.
10. Bittencourt LP, Campos V, Moliterno LF, et al. Hereditary gingival fibromatosis: review of the literature and a case report. *Quintessence Int* 2000;31(6):415-418.
11. Nayak PA, Nayak UA, Khandelwal V, et al. Idiopathic gingival fibromatosis. *Int J Clin Pediatr Dent* 2011;4(1):77-81. DOI: 10.5005/jp-journals-10005-1086.
12. Xiao S, Bu L, Zhu L, et al. A new locus for hereditary gingival fibromatosis (GINGF2) maps to 5q13-q22. *Genomics* 2001;74(2):180-185. DOI: 10.1006/geno.2001.6542.
13. Hart TC, Pallos D, Bowden DW, et al. Genetic linkage of hereditary gingival fibromatosis to chromosome 2p21. *Am J Hum Genet* 1998;62(4):876-883. DOI: 10.1086/301797.
14. Hart TC, Zhang Y, Gorry MC, et al. A mutation in the SOS1 gene causes hereditary gingival fibromatosis type 1. *Am J Hum Genet* 2002;70(4):943-954. DOI: 10.1086/339689.
15. Singer SL, Goldblatt J, Hallam LA, et al. Hereditary gingival fibromatosis with a recessive mode of inheritance: case reports. *Aust Dent J* 1993;38(6):427-432. DOI: 10.1111/j.1834-7819.1993.tb04755.x.
16. Anderson J, Cunliffe WJ, Roberts DF, et al. Hereditary gingival fibromatosis. *BMJ* 1969;3(5664):218-219. DOI: 10.1136/bmj.3.5664.218.
17. Danesh-Meyer MJ, Holborow DW. Familial gingival fibromatosis: a report of two patients. *N Z Dent J* 1993;89(398):119-122.
18. Clocheret K, Dekeyser C, Carels C, et al. Idiopathic gingival hyperplasia and orthodontic treatment: a case report. *J Orthod* 2003;30(1):13-19. DOI: 10.1093/ortho/30.1.13.
19. Kavvadia K, Pepelassi E, Alexandridis C, et al. Gingival fibromatosis and significant tooth eruption delay in an 11-year-old male: a 30-month follow-up. *Int J Paediatr Dent* 2005;15(4):294-302. DOI: 10.1111/j.1365-263X.2005.00646.x.
20. Ramer M, Marrone J, Stahl B, et al. Hereditary gingival fibromatosis; identification, treatment, control. *J Am Dent Assoc* 1996;127(4):493-495. DOI: 10.14219/jada.archive.1996.0242.