International Journal of Medical and Pharmaceutical Case Reports

11(1): 1-5, 2018; Article no.IJMPCR.40157
ISSN: 2394-109X, NLM ID: 101648033

Elephantiasis Gingivae – A Case Report and Its Management

P. Venkatalakshmi Aparna[1*], S. Leena Sankari[2], A. Priyadharshini[1] and M. Kasthuri[3]

[1]Department of Oral Medicine and Radiology, Ragas Dental College and Hospital, Uthandi, Chennai, India.
[2]Department of Oral Pathology and Microbiology, Sree Balaji Dental College and Hospital, Chennai, India.
[3]Department of Oral and Maxillofacial Pathology, Kasthuri Dental Clinic, Velacherry, Chennai, India.

Authors' contributions

This work was carried out in collaboration between all authors. Author PVA designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors SLS and AP managed the analyses of the study. Author MK managed the literature searches. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/IJMPCR/2018/40157
Editor(s):
(1) Manuel Marques Ferreira, Professor, Area of Dentistry, University of Coimbra, Portugal.
Reviewers:
(1) R. Karthikeyan, Surendera Dental College & Research Institute, Rajasthan University of Health Sciences, India.
(2) Mehmet Yaltirik, Oral Surgery Istanbul Universty, Turkiye.
(3) Lauritano Dorina, University of Milan-Bicocca, Italy.
Complete Peer review History: http://www.sciencedomain.org/review-history/23783

Case Report

Received 2nd January 2018
Accepted 9th March 2018
Published 23rd March 2018

ABSTRACT

Gingival fibromatosis is a rare, benign, slowly growing heterogeneous group of disorders that develop local or diffuse enlargements of interdental papilla or attached gingiva and marginal gingiva. In advanced cases, the growth of the tissue may cover the occlusal surface of the teeth, thus causing functional, aesthetic, and periodontal problems, such as bone loss and bleeding, due to the accumulation of plaque and the presence of pseudo pockets. It affects both sexes equally. This condition is usually transmitted as a genetic syndrome and rarely, an isolated condition. Here, we discuss the case on gingival fibromatosis along with a brief description of some characters on clinical, etiological, genetic and histopathological aspects.

**Corresponding author: E-mail: apriya.omdr@gmail.com;*

Keywords: Gingival fibromatosis; gingival enlargement; genetic syndrome; hereditary gingival hyperplasia; idiopathic fibromatosis and hypertrophied gingivae.

1. INTRODUCTION

Gingival fibromatosis is a rare, benign, slowly growing heterogeneous group of disorders that develop local or diffuse enlargements within marginal and attached gingiva or interdental papilla [1]. In advanced cases, the excess tissue may cover the occlusal surface of the teeth, thus causing masticatory, aesthetic, and periodontal problems, such as bleeding and alveolar bone loss, due to the presence of pseudo pockets and plaque accumulation [2]. It affects both sexes equally. This condition is usually transmitted as a genetic syndrome, non-syndromic (hereditary) gingival fibromatosis and rarely, an isolated condition. Gingival enlargement occurs at the time of deciduous tooth eruption and also affect permanent dentition [3,4]. In some cases, gingiva appears pink to red, firm, dense, with a nodular stippled surface and tendency to bleed [5]. Usually, the enlargement is symmetrical but may be unilateral. It occurs due to many factors including inflammation, leukemic infiltration and use of such medications like phenytoin, cyclosporine or nifidipine [6-8]. Gingival enlargement is associated with syndromes like Ramon, Cross, Jones, Rutherford and Laband syndrome [9-11]. Other features such as hearing loss, abnormal finger, nose, splenomegaly, cherubism and psychomotor retardation [12,13] We present a case with gingival fibromatosis and its management.

2. CASE REPORT

A 21-year-old man, with the complaint of pain and swelling in the gums for the past 7 years and visited our dental clinic. His parents were non-consanguineous. He had a younger sister. None of his family members was involved with the similar disease. The patient was not taking any anti-epileptic, antihypertensive or immunosuppressive medications that could contribute to the enlargement of gingiva. However, the patient had smoked up to 15 cigarettes a day for the past 5 years and drank alcohol about 3 times a week. The history revealed that the patient's gums had gradually enlarged over the last 3 to 5 years and that the patient had pain during mastication and increased tooth mobility, especially right-side of upper and lower molars. Intraoral examination revealed moderate-to-severe gingival overgrowth of a firm, dense, pigmented, nodular and fibrotic consistency that involved the right-side of both the maxillary and mandibular arches. Bleeding on probing is negative. The overgrowth caused diastema and mastication problems. The attached gingiva excised from the buccal and interdental areas during surgery immediately fixed in 10% buffered formaldehyde solution and sent for histopathologic examination. The final diagnosis of fibromatosis was made and gingivectomy was done. The microscopic feature of these sections revealed para-keratinized stratified squamous acanthotic epithelia with thin rete ridges extending into the connective tissue. The underlying connective tissue showed dense wavy bundles of collagen fibres containing numerous fibroblasts. Some sections of the connective tissue exhibited infiltration of chronic inflammatory cells and areas of neovascularization that had red blood corpuscles within the lumen of the blood vessels. The patient initially underwent phase 1 periodontal therapy that includes scaling and root planning, oral hygiene instructions, and prescribed systemic doxycycline (100 mg twice a day, on the first day, followed by 100 mg once a day for 2 weeks) [14,15]. Phase 2 therapy involved periodontal surgery on right side of upper and lower jaw (gingivectomy combined with open-flap debridement). The patient was monitored regularly; no recurrence of gingival enlargement was observed 6 months after the surgery.

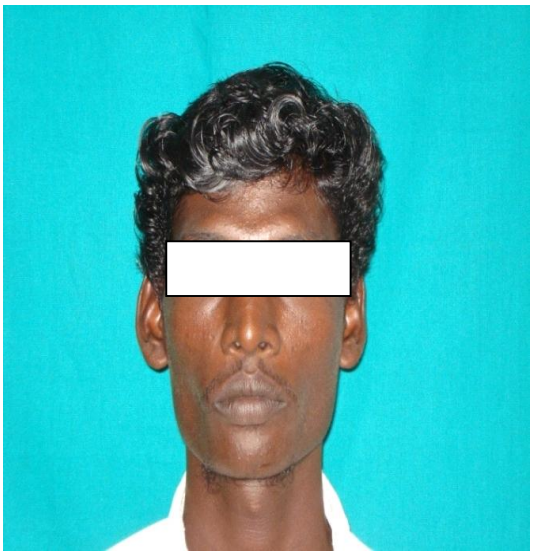

Fig. 1. Patient profile

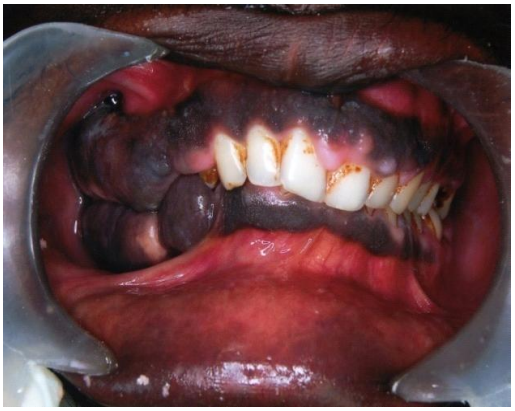

Fig. 2. Preoperative view of maxilla and mandible

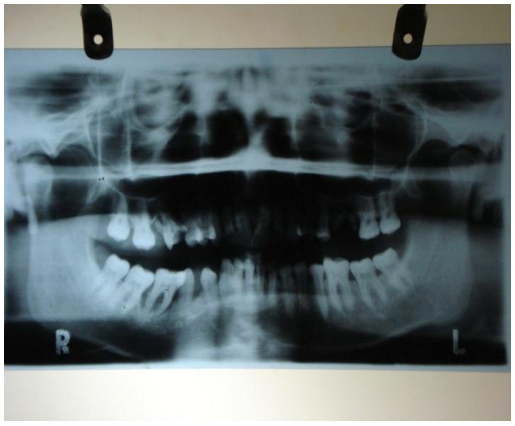

Fig. 3. Panoramic radiograph showed a coarse trabecular pattern in the alveolar bone, generalized horizontal bone loss

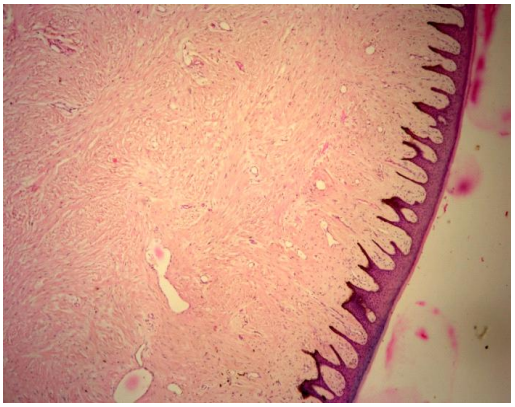

Fig. 4. Photomicrograph of the specimen illustrating the presence of a thickened acanthotic epithelium with elongated rete ridge and a densely fibrous connective tissue with numerous fibroblast (original magnification 10×)

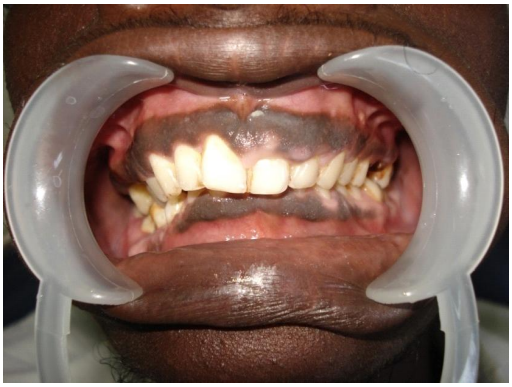

Fig. 5. Intraoral postoperative view of the maxillary and mandibular arches 6 months after surgery

3. DISCUSSION

Gingival fibromatosis is otherwise known as Elephantiasis gingivae, Fibromatosis gingivae, Hereditary gingival fibromatosis (HGF). In a majority of cases reported as hereditary, being transmitted through an autosomal dominant gene. Characterized by diffuse enlargement of the gingiva caused by a collagenous overgrowth of the gingival fibrous connective tissue. An estimated prevalence of GF is 1:750 [16,17] and is associated with various syndromes, such as Rutherford syndrome, Cross syndrome, Zimmerman-Laband syndrome, Costello syndrome, Prune belly syndrome, Ramon syndrome, Murray-Puretic-Drescher syndrome and Jones syndrome [10,13]. Recently GF has been identified a mutation in the Son-of-Sevenless-1 (SOS-1) gene that isolates the hereditary gingival fibromatosis phenotype [18,19]. Identification of the specific genetic basis should help to illustrate the pathogenic mechanisms that cause gingival enlargement in HGF. Clinically it affects one or both the jaws and it usually appears at the time of eruption of both deciduous or permanent teeth or present at the time of birth. Sometimes, gingival overgrowth does not occur until the eruption of deciduous or permanent dentition [20]. The overgrowth of gingival tissue can cover the entire or part of the crown that results in diastemas, teeth displacement, or retention of deciduous or impacted teeth, and may also cause masticatory, speech, psychological, and poor aesthetic problems [21]. The tissue is firm, normal or pale in colour, not usually inflamed, covered by smooth surface or stippled. In aged patients, the surface exhibits numerous papillary projections and the alveolar ridge appears into lobules.

Histopathologically consists of dense hypocellular, hypo vascular collagenous tissue, which shows numerous interlacing bundles are arranged in a parallel fashion. Surface epithelium exhibits long, thin rete ridges that extend deep into the underlying fibrous connective tissue [22]. Rarely shows scattered islands of odontogenic epithelium, areas of osseous metaplasia or small foci of dystrophic calcification and abundant neurovascular bundles may also be present. The overgrowth of gingival tissue shows new niches for the growth of microorganisms, deposition of plaque and formation of pseudo pocket that results in inflammatory cell infiltration of the gingival connective tissue [23]. Treatment of gingival fibromatosis is required because it causes functional problems. In case of gingival overgrowth with deep periodontal pockets and severe loss of alveolar bone, an internal bevel gingivectomy with open-flap debridement is necessary. This procedure was done for our patient to eliminate pockets, making to control plaque easier; to reduce the bulbous area of gingival tissue; and to promote the regeneration or repair of the alveolar bone defect. Our patient is being regularly monitored clinically as well as radiographically for improvement in his periodontal condition, and for any recurrence of gingival enlargement.

4. CONCLUSION

This case highlights the non- syndromic idiopathic gingival fibromatosis. Diagnosis is based on medical history, clinical examination, blood tests and histopathological evaluation of affected gingival tissue. The dental care professional should be able to diagnose the gingival fibromatosis especially in cases where the patients and parents needed for future genetic family planning.

CONSENT

As per international standard or university standard, patient's consent has been collected and preserved by the authors.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Rashi chaturvedi idiopathic gingival fibromatosis associated with generalized aggressive periodontitis: A case report. JCDA. 2009;75(4).
2. Gawron et al. Gingival fibromatosis: Clinical, molecular and therapeutic issues. Orphanet Journal of Rare Diseases. 2016; 11:9.
3. Baer PN, Benjamin SD. Fibrous hyperplasia of the gingival. In: Periodontal disease in children and adolescents. 2nd ed. JB Lippincott Company. 1974;81-84.
4. Ramer M, Marrone J, Stahl B, Burakoff R. Hereditary gingival fibromatosis: Identification, treatment, control. JADA. 1996;127:493-495.
5. Katz J, Chausho G, Navot G, Taicher S. Vigabatrin - induced gingival overgrowth. J Clin Periodontol. 1997;24:180-182.
6. Brightman VJ. Benign tumors of the oral cavity, including gingival enlargement. In: Burket's Oral Medicine. 9th ed. Lynch AM, Brightman VJ, Greenberg MS. Philadelphia: Lippincott-Raven. 1997;189-191.
7. Wynne SE, Aldred MJ, Bartold PM. Hereditary gingival fibromatosis associated withhearing loss and supernumerary teeth-a new syndrome. J Periodontol. 1995; 66:75-79.
8. Harrison M, Odell EW, Path MRC, Agrawal M, Saravanamuttu R, Longhurst P. Gingival fibromatosis with prune-belly syndrome. Oral Surg Oral Med Oral Pathol. 1998;86:304-307.
9. Coletta RD, Graner E. Hereditary gingival fibromatosis: A systematic review. J Periodontol. 2006;77(5):753–64.
10. Kelekis-Cholakis AK, Wiltshire WA, Birek C. Treatment and long-term follow-up of a patient with hereditary gingival fibromatosis: A case report. J Can Dent Assoc. 2002;68(5):290–4.
11. Baptista IP. Hereditary gingival fibromatosis: A case report. J Clin Periodontol. 2002;29(9):871–4.
12. Hereditary gingival fibromatosis associated with generalized aggressive periodontitis: A case report. J Periodontol. 2004; 75(5):770–8.
13. Gunhan O, Gardner DG, Bostanci H, Gunhan M. Familial gingival fibromatosis with unusual histological findings. J Periodontol. 1995;66(11):1008–11.

14. Tonetti M, Mombelli A. Early onset periodontitis. Ann Periodontol. 1999; 4(1):39–52.
15. Feres M, Haffajee AD, Gonclaves C, Allard KA, Som S, Smith C, Others. Systemic doxycycline administration in the treatment of periodontal infections. I. Effect on the subgingival microbiota. J Clin Periodontol. 1999;26(12):775–83.
16. Bittencourt LP, Campos V, Moliterno LF, Ribeiro DP, Sampaio RK. Hereditary gingival fibromatosis: Review of the literature and a case report. Quintessence Int. 2000;31:415–18.
17. Tiwana PS, De Kok IJ, Stoker DS, Cooper LF. Facial distortion secondary to idiopathic gingival hyperplasia: Surgical management and oral reconstruction with endosseous implants. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2005; 100:153–57.
18. Gawron et al. Gingival fibromatosis: Clinical, molecular and therapeutic issues. Orphanet Journal of Rare Diseases. 2016; 11:9.
19. Hart TC, Zhang Y, Gorry MC, Hart PS, Cooper M, Marazita ML, and others. A mutation in the SOS1 gene causes hereditary gingival fibromatosis type 1. Am J Hum Genet. 2002;70(4):943–54.
20. Skrinjari I, Bacic M. Hereditary gingival fibromatosis – report on three families & dermatoglyphic analysis. Journal of Periodontology. 1989;24:303–309.
21. Millet C, Rodier P, Farges JC, Labert N, Duprez JP. Surgical and prosthetic treatment in an elderly patient affected by unilateral idiopathic gingival fibromatosis: A case report. Gerodontology. 2012; 29:e1185–89.
22. Pernu HE, Oikarinen K, Hietanen J, Knuuttila M. Verapamil-induced gingival overgrowth: A clinical, histologic, and biochemic approach. J Oral Pathol Med. 1989;18:422–25.
23. Pasupuleti MK, Musalaiah SV, Nagasree M, Kumar PA. Combination of inflammatory and amlodipine induced gingival overgrowth in a patient with cardiovascular disease. Avicenna J Med. 2013;3:68–72.

Peer-review history:
The peer review history for this paper can be accessed here:
http://www.sciencedomain.org/review-history/23783