

## Idiopathic gingival fibromatosis

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يتحدث هذا التقرير عن طفلة تبلغ من العمر 5 سنوات تعاني من تضخم متزايد باللثة وذلك في كلا الفكين . ولقد أصبح هذا التضخم ملاحظاً أثناء بزوغ الأسنان اللبنية . وكان التضخم شديداً ومنتشراً شمل اللثة بجميع أقسامها ((الحنك العلوي والحنك السفلي والخيليات اللثوية بين السنية)) وغطى كل أسطح الأسنان تقريباً فيما عدا الحواف القاطعة للأسنان الأمامية أما الأسطح الطاحنة للظواحن فلقد تم تغطيتها بشكل جزئي . كان التضخم في الفك العلوي أشد منه في الفك السفلي . كانت القصة المرضية العائلية سالبة . بعد استئصال النسيج اللثوي المتضخم جراحياً تحت التخدير الكامل تم تشخيص الحالة كتضخم في اللثة غير معروف الأسباب وذلك بالنظر إلى القصة المرضية والفحص السريري وفحص الأسحة ونظراً لعودة التضخم مرة أخرى تم إزالته جراحياً للمرة الثانية .

A five year old girl presented with a slowly growing gingival enlargement of both arches which became pronounced during the eruption of deciduous teeth. A severe and generalized diffuse gingival hyperplasia involving the marginal, interdental and attached gingiva was observed covering almost all the surfaces of all the teeth except the incisal edges of the incisors. The occlusal surfaces of the molars were partially covered. The gingival hyperplasia was more severe in the maxilla than in the mandible. Familial history was negative. The case was diagnosed as idiopathic gingival hyperplasia based on the history, clinical features and histological findings. The gingival hyperplastic tissue was excised under general anesthesia. Recurrence necessitated repeated surgery.

### Introduction

Gingival enlargements are quite common and may be either inflammatory, non inflammatory or a combination of both. Idiopathic gingival hyperplasia is a rare condition of undetermined etiology described variously as fibromatosis gingivae, gingivaematosi,<sup>1</sup> hereditary gingival fibromatosis<sup>2</sup> idiopathic fibromatosis,<sup>3</sup> familial elephantiasis<sup>4</sup> and diffuse fibroma.<sup>5</sup> Diffuse gingival enlargement is also found to be associated with syndromes like Cross syndrome, Rutherford syndrome, Ramen syndrome, Zimmerman Laband syndrome and Juvenile hyaline syndrome. The purpose of this case report was to highlight the etiological factors and treatment.

### Case Report

A five year old girl was brought to the department of pedodontics with the chief complaint of a slow growing, nontender gingival enlargement since birth. The parent indicated that the enlargement became more pronounced at the time of eruption of the deciduous teeth. There was no history of epilepsy or long term medication for any ailments. Developmental milestones and other systems of the child were normal. This was an isolated case in their family according to the parents. There was a history of surgical exposure of maxillary deciduous anteriors, when the child was 3 year old. The other associated problems were difficulty in speech, mastication and swallowing.

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### Examination

Extra oral examination revealed a convex facial profile with a bimaxillary protrusion, incompetent lips and a broad nasal bridge. Intraorally, severe diffuse gingival enlargement involving the marginal, interdental and attached gingiva of both arches was observed, covering almost all the surfaces of the teeth. The enlargement covered partially the occlusal surfaces except the incisal edges of the primary anteriors (Fig. 2).

The gingival hyperplasia was more severe in the maxilla than the mandible, especially posteriorly. The gingival hyperplasia in the maxillary right posterior segment was almost projecting into the lower right vestibule (Fig. 1).



Fig. 1. Enlargement is more on the right than on the left side. Taken before surgery.

The color of the gingiva appeared normal with secondary inflammatory changes in the posterior segment, especially on the occlusal surfaces. Melanin hyperpigmentation was observed in most of the areas. On isolation and drying, the

gingival surface appeared pebbled with increased stippling. The hyperplastic tissue was firm in consistency and non tender on palpation. Due to the massive gingival enlargement, normal lip closure was prevented, but with force the child was able to approximate the lips closely.

The child experienced difficulty in mastication and had developed an altered tongue position with abnormal swallowing and open occlusal relationship. Drooling of saliva was also noticed. Based on the history and clinical features, the case was diagnosed as idiopathic gingival fibromatosis.



Fig. 2. Mandibular occlusal view before surgery.



Fig. 3. After surgery.

### Investigations

Full mouth periapical radiographs and orthopantomogram were taken to rule out bony changes, displacement and resorption of the teeth. Routine hematological investigations were also done. Findings from these investigations were within normal limits.

### Histopathological Findings

Sections showed a hyperparakeratinized hyperplastic stratified squamous epithelium with the underlying fibrous connective tissue showing bundles of collagen fibers, aggregates of chronic inflammatory cells, few blood vessels and extensive areas of hemorrhage.

### Treatment

After routine clinical and hematological examination, the maxillary and mandibular deciduous central and lateral incisors were extracted. The child was reviewed after 4 months to see if the hyperplastic tissue had regressed. Since the enlargement persisted, a single sitting full mouth gingivectomy procedure was done under general anesthesia. After a 4 year follow up, eruption of 11,12, 21,22,31 and 41 had occurred and recurrence of the gingival hyperplasia in the posterior segment of both arches was observed (Fig. 3), necessitating a second stage surgery which was done. The child is currently undergoing orthodontic treatment for the anterior open bite and a habit breaking appliance has been placed for the correction of tongue thrust. The child is under follow up observation.

### Discussion

Idiopathic gingival fibromatosis may be congenital or hereditary. Though the genetic mechanism is not well understood, the majority of the authors of reported cases attributed the condition to hereditary factors. The mode of transmission is mainly autosomal dominant.<sup>6,7</sup> The first polymorphic marker for hereditary gingival fibromatosis (HGF) phenotype in chromosome 2p21. Many cases are sporadic with no familial background.

Gingival hyperplasia can occur after therapy with drugs like phenytoin, cyclosporine, nifedipine and nitrendipine. Long term use of these drugs has to be ruled out. The incidence of gingival enlargement caused by phenytoin,<sup>8</sup> an anticonvulsant used in the treatment of epilepsy varies from 3 to 84.5%. whereas, cyclosporine a fairly potent immunosuppressive agent used to prevent organ transplant rejection and to treat several disease of autoimmune origin induced gingival enlargement in 30% of the cases.<sup>9</sup> Nifedipine, which is a calcium channel blocker used in the treatment of acute and chronic

coronary insufficiency, including angina pectoris and refractory hypertension and nitrendipine an analogue of nifedipine have also been reported to induced gingival enlargement.<sup>10,11</sup>

The condition may be associated with physical developmental retardation and hypertrichosis.<sup>12</sup> Although gingival tissue may appear normal at birth, hyperplastic gingival fibromatosis may become evident with the eruption of primary or permanent dentition, suggesting a trauma - induced tissue reaction during the eruption.<sup>13</sup>

Sometimes gingival enlargement does not occur until the eruption of the permanent dentition. Further enlargement does not occur once the growth of jaw is completed.<sup>14</sup> It has been suggested that gingival enlargement may be due to nutritional and hormonal factors, but these have not been completely substantiated. The constant increase in the tissue mass can result in delayed eruption and displacement of teeth, arch deformity, spacing and migration of teeth.<sup>15</sup> The condition is not painful until the tissue enlarges to partially cover the occlusal surface of the molars and become traumatized during mastication, which was observed in the present case. Due to massive gingival enlargement, an affected child usually develops abnormal swallowing pattern and experiences difficulty in speech and mastication. Along with these features, there may be some interference with the oral hygiene measures and normal mastication. All these will favor accumulation of materia alba and plaque, which further complicates the existing hyperplastic tissue. Maintenance of good oral hygiene is very important. It is not known if plaque control measures are effective in this condition, but it is good practice to maintain the plaque control following gingivectomy procedure.

Histologically, the gingival hyperplasia is mainly due to an increase and thickening of mature collagen bundles in the connective tissue stroma.<sup>16</sup> The nodular appearance can be attributed to the thickened para hyperkeratinized epithelium.<sup>17</sup> Various modalities of treatment had been proposed including radical treatment with extraction of the involved teeth, which was reported not to favor a recurrence of the growth.<sup>16</sup> The only treatment of choice in this condition was gingivectomy to satisfy the patient's esthetics. Though the tissue appeared to be pale and firm, the surgical procedure was complicated with excessive hemorrhage. Some have reported a case where apically positioned flap surgery and CO<sub>2</sub> laser evaporation were used to reduce the gingival tissue. Since recurrence could be

expected within a few months after surgery and may return to the original condition within few years, the patient may have to undergo repeated gingivectomy procedures. This often causes further increase in the patients and parents' psychological and emotional stress. Hence psychological counselling is a must for patients and parents.

### References

1. Ball EI. Case of gingivomatosis or elephantiasis of the gingiva. *J Periodontol* 1941;12: 26.
2. Laskin J, Weisberger D. Hereditary gingival fibromatosis. *J Oral Surg* 1961;14: 828.
3. Thukral PP. Idiopathic hyperplasia. *J Ind Dent Assoc* 1972; 44:109.
4. Raman Y, Berman, Bubbis JJ. Gingival fibromatosis combined with cherubism. *Oral Surg* 1967;24:455.
5. Buckner HJ. Diffuse fibroma of the gums. *J Am Dent Assoc* 1937;27:2003.
6. Emerson TG. Hereditary gingival hyperplasia. A family pedigree of four generations. *Oral Surg* 1965;19:1.
7. Jorgenson RJ, Cocker ME. Variations in the inheritance and expression of the gingival fibromatosis. *J Periodontol* 1974;45:472-477.
8. Angelopoulos AP, Goaz PW. Incidence of diphenylhydantoin gingival hyperplasia. *Oral Surg* 1972; 34: 898.
9. Seymour RA, Smith DG, Rogers SR. Comparative effect of azathioprine and cyclosporine on some gingival health parameters of renal transplant patients. *J Clin Periodontol* 1987; 14: 610.
10. Barclay S, Thomason JM, Idle JR, Seymour RA. Incidence and severity of nifedipine induced gingival overgrowth. *J Clin Periodontol* 1992; 19: 311.
11. Brown RS, Sein P, Corio R, Bottomley WK. Nitrendipine - induced gingival hyperplasia. *Oral Surg* 1990; 70: 593.
12. William G Shafer, Maynard K Hine, Bernat M Levy. Developmental disturbances of the perioral structures. In: *Text book of Oral Pathology*. 4<sup>th</sup> ed. A Prisma Indian, 1993. PP. 23-24.
13. Gupta N, Maheshwari S. Advanced gingival fibromatosis. *J Ind Dent Assoc* 1996;167:46-47.
14. Stewart RE. Periodontal diseases in children. In: *Pediatric Dentistry and Clinical Practice*. Mosby Company, 1982. pp. 623-639.
15. Mcdonald RE, Avery DR. Gingival and periodontal diseases. In *Dentistry for the Child and Adolescent*. 7<sup>th</sup> ed. Mosby Company; 2000. pp. 452-453.
16. Zachin SJ, Weisberger D. Hereditary gingival fibromatosis - report of a family. *Oral Surg Oral Med Oral Pathol* 1961; 14: 825-835.
17. Brightman VJ. Benign tumors of the oral cavity including gingival enlargement. In *Burkets of Oral Medicine, Diagnosis and Treatment Plan*. 8<sup>th</sup> ed. 1984. pp. 367-371.