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Gingival Fibromatosis: A Report of a Severe Case and Literature Review

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ABSTRACT

Hereditary Gingival fibromatosis (HGF) is a rare benign disease characterised by a progressive increase in gingival tissue that can occur as isolation or as part of a syndrome. As aesthetics may be affected in severe cases, the quality of life of patients, especially children, may be compromised. A 10-year-old girl presented with generalized diffuse gingival enlargement affecting the maxillary and mandibular arches, extending on the buccal and lingual/palatal surfaces and covering the incisal/occlusal third of the tooth. A scalpel and diode laser were used to perform a gingivectomy. The histological study and clinical symptoms all supported the diagnosis. Initially, during the mixed dentition stage, frequent periodontal scaling and oral hygiene reinforcement were carried out. The healing process was uneventful, the patient was pleased with her aesthetics and was able to resume her dental hygiene routines. Removable orthodontic appliance treatment was carried out three months following the procedure. To prevent gingival hyperplasia, oral hygiene was emphasized, and the teeth were polished once a month.

1. INTRODUCTION

Hereditary gingival fibromatosis (HGF) is a non-hemorrhagic, fibrous gingival overgrowth that can cover all or part of a tooth (Bhutada et al., 2017). Although there are few cases of this illness (1/175,000 inhalants), several cases may arise in the same household. HGF may manifest independently or in conjunction with other symptoms (Sangle et al., 2018). Despite having variable penetrance and expressivity, it has an autosomal dominant inheritance pattern. Notably, 20% of patients do not have any family history of the illness (Li et al., 2021).

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It is uncertain what mechanism causes gingival expansion in the aetiopathogenesis of HGF. According to researchers, there is a decrease in the matrix metalloproteinases (MMPs) responsible for collagen degradation and an increase in subepithelial fibroblast proliferation, collagen, and fibronectin synthesis (Fatih et al., 2024)(Li et al., 2021). It seems that the inflammatory stimulus of eruption causes levels of transforming growth factor ß1 (TGFß1) to increase (Fatih et al., 2024). An increase in TGFß1 causes increased fibroblast proliferation, and these cells continue to be phenotypically activated throughout the synthesis of collagen and fibronectin. This would lead to an imbalance in the formation and breakdown of the extracellular matrix (Li et al., 2021).

Clinically, HGF manifests as either a predominantly painless, fibrous, and exhibits noticeable stippling, non-hemorrhagic gingival hyperplasia in the absence of inflammatory symptoms, or as a locally restricted, pale pink connected gingiva with solid and lobular consistency (Almiñana-Pastor et al., 2017). Few cases describe an early diagnosis in the primary dentition, and it is uncommon to be detected at birth (Sangle et al., 2018). Most importantly, given the age at which it typically manifests, it can become a psychological burden and lower the patient's self-esteem in addition to interfering with speaking, lip closure, and chewing.

The differential diagnosis of HGF is drug-induced gingival hyperplasia, specifically linked to phenytoin, cyclosporine, and nifedipine consumption. According to the studies, the pathognomonic gingival tissue excess may create novel ecological niches for bacteria, influencing plaque buildup and pseudo pocket evaluation (Almiñana-Pastor et al., 2017). Diastema, retention of primary or impacted teeth, misaligned teeth, delayed eruption, pseudo-pockets without attachment loss, large lips, open and crossbite, and labial incompetence are the most frequent clinical sequelae in children. In addition, there is halitosis and early periodontal disease. In addition to the detrimental effects of poor aesthetics on their relationships, HGF patients may experience issues with speech, eating, and swallowing (Sangle et al., 2018).

The preferred course of treatment for HGF is surgical excision of extra gingival tissue because the condition does not heal on its own. The gingivectomy is a highly effective method that is typically utilized (Kumari et al., 2021). The surgical method enhances the patient's quality of life by minimizing negative psychological effects and functional and aesthetic impairment. However, follow-up care and practical dental hygiene guidelines are required to prevent relapses.

Treatment options for children with hyperplastic gingival tissue include electrocautery, carbon dioxide laser, mini-invasive periodontal surgery using apically relocated flaps, and/or numerous surgical gingivectomies in conjunction with gingivoplasty (Kumari et al., 2021). Treatment selection is based on HGF severity. Gingivectomy, which can be carried out with an internal or external bevel incision, is the preferred treatment for HGF since it does not go away on its own. The recurrence rate is decreased when surgery is performed following the eruption of permanent teeth (Huang et al., 2022). Nonetheless, the detrimental psychological impact of HGF on the patient and the potential functional impairments it may cause make it worthwhile to take the chance of a recurrence and undertake surgery at a younger age. Herein, we describe a rare non-syndromic case of generalized gingival fibromatosis in a child, including clinical and microscopical features, as well as highlighting the importance of the correct diagnosis of this condition.

2. CASE DESCRIPTION

A 10-year-old female presented with a painless gingival enlargement that hindered mastication and compromised aesthetics. During the anamnesis, the patient's mother denied any systemic changes or chronic drug use but did report gingival enlargement from age five. Gingival overgrowth was initially observed during primary dentition but intensified during permanent dentition. She also mentioned that the pregnancy and birth did not coincide. There are no congenital or acquired disorders or a family history of genetic defects. Past medical history was not contributory. Blood investigation revealed nothing abnormal, ruling out any systemic condition (Table 1). An extraoral clinical examination indicated skeletal pattern Class II with a convex profile, procumbent and incompetent lips, vertical growth pattern, gummy smile, Class II canine and molar relationship, increased overjet of 9 mm, and a very deep bite (90%). In the upper and lower mixed dentition, both free and attached gingiva exhibited uneven hyperplasia and hypertrophy. The swelling had a primarily fibrous nature, pink color, and firm in texture. It was also accompanied by patches of melanic pigmentation and a noticeable hyperemia of the gingival borders in the anterior vestibular region (Fig. 1A). It stretched toward the midline in the palatal region, partially obliterating the palate's depth (Fig. 1B). As seen in Figures 1A&B, the teeth were misaligned and the crowns were either fully or partially erupted in the fibrous tissue. She also presented with high gingival attachment and diastema between teeth 11 and 21. The dental panoramic tomograph (DPT) and cone beam tomography (CBCT) view showed normal development of dentition and generalised bone loss (Fig. 2). All permanent teeth were present and in their normal developmental stages. The diagnosis of inherited gingival fibromatosis was made based on the combined clinical and morphologic data.

Table 1. Blood investigation revealed nothing abnormal, ruling out any systemic condition.

Parameters	Normal values	Values of the child
White cell count	4-10 x 10^3/µL	7.51 x 10^3/µL
Red blood cell	3.80 -4.80 x 10^6/µL	3.9 x 10^3/µL
Haemoglobin	12-15 g/dL	13.2 g/dL
Mean corpuscular volume	83-101 fL	80.4 fL
Mean corpuscular hemoglobin	27-32 pg	27.3 pg
Mean corpuscular hemoglobin concentration	32.3-34.3 g/dL	32.5 g/dL
Hematocrit	36-46%	40.6%
Platelets	150-410 x 10^3/µL	295 x 10^3/µL
Neutrophils	2-8 x 10^3/µL	2.9 x 10^3/µL
Lymphocytes	27-47%	38%
Absolute Neutrophils	0.91-5.5 x 10^3/µL	3.2 x 10^3/µL
Absolute Lymphocytes	0.65-3.05 x 10^3/µL	1.2 x 10^3/µL
Absolute Monocytes	0.16 -0.72 x 10^3/µL	0.38 x 10^3/µL

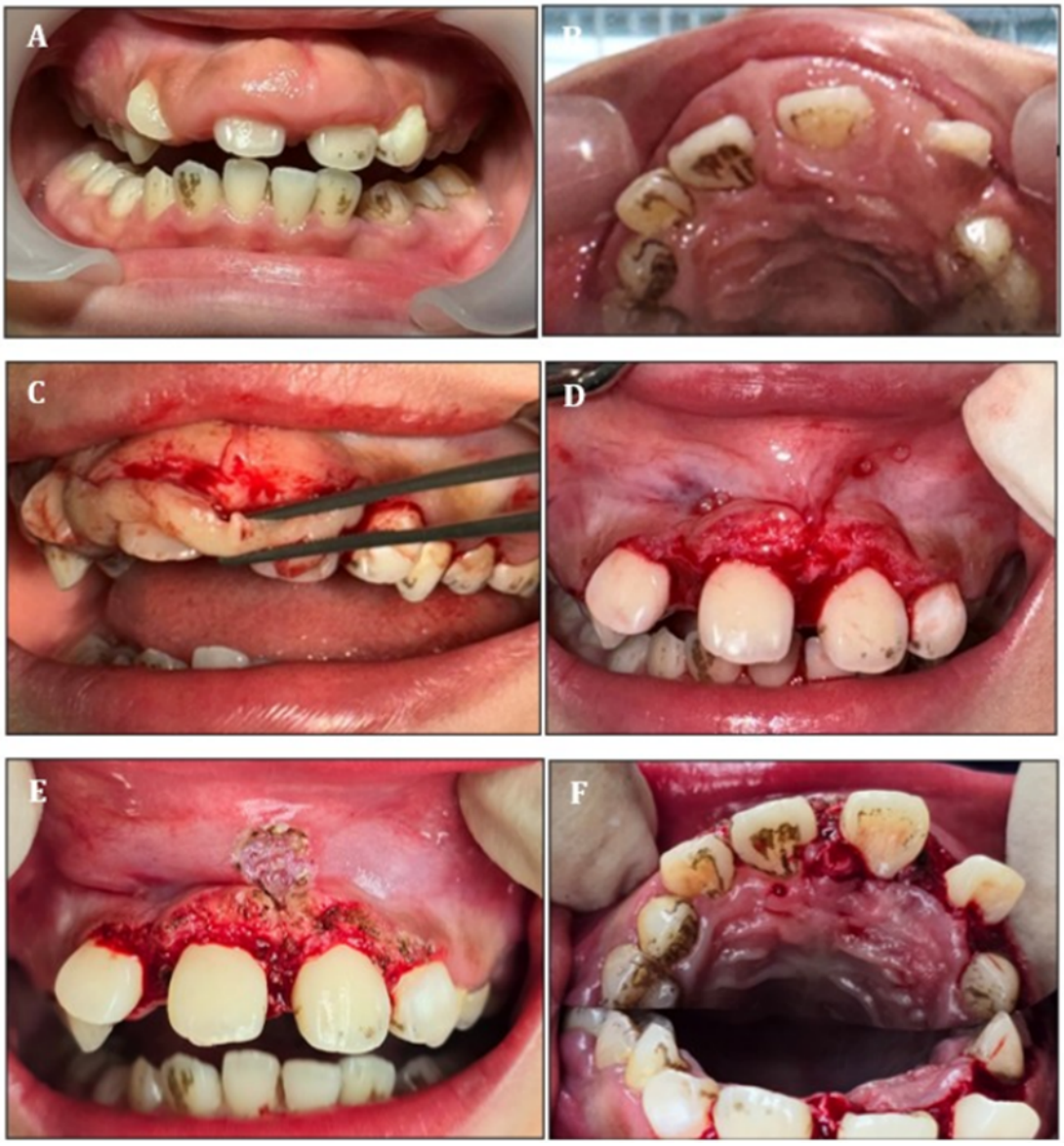


Fig. 1. (A) Pre-operative view of the maxilla and mandible; (B) Pre-operative view of the palate; (C) Intrasurgical view of external bevel incision gingivectomy showed thick attached gingiva; (D) Intrasurgical view of external bevel incision gingivectomy on upper anterior region; (E) Intrasurgical view of frenotomy of the high frenal attachment; (F) Palatal view post gingivectomy.

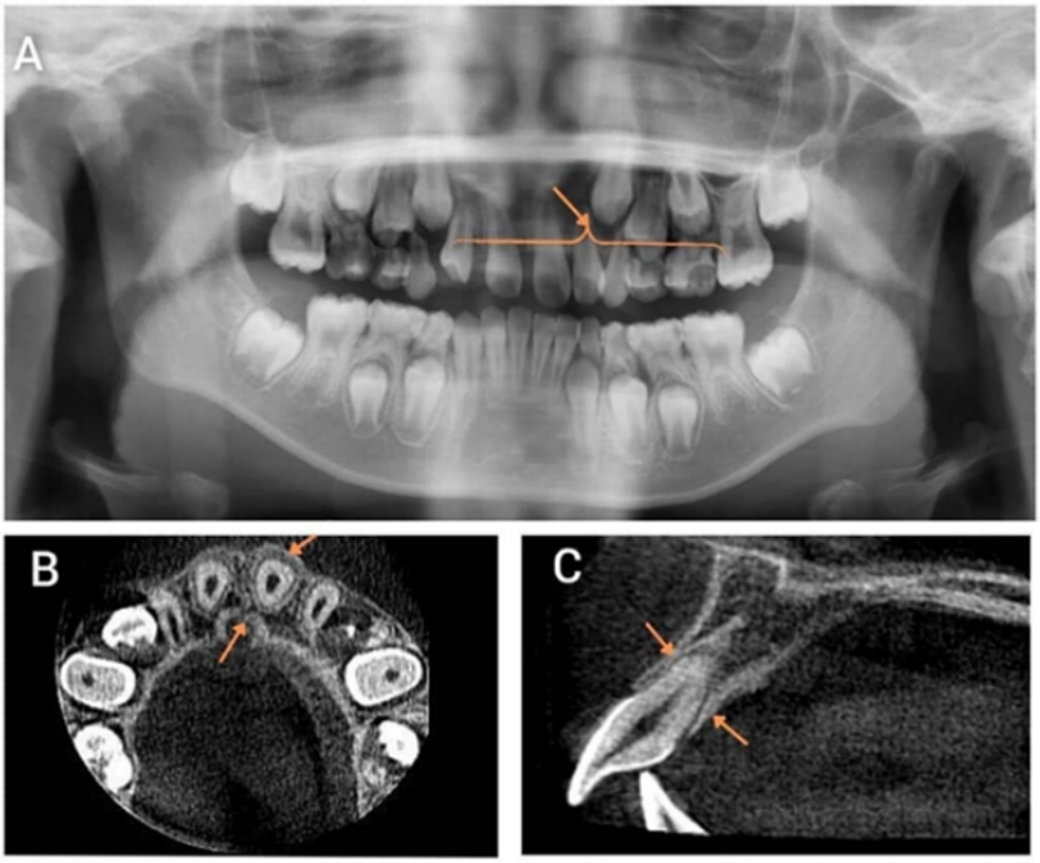


Fig. 2. Pre-operative dental panoramic tomograph view showing generalized bone loss. Figures 2B and 2C depict cone beam computed tomogram view showing adequate thickness of bone crest on a labial and palatal region on an axial and sagittal plane respectively.

2.1. Intervention

Under local anesthesia, the external bevel gingivectomy was used to surgically remove the enlarged gingival tissue from the buccal and palatal surfaces of the maxilla (Fig. 1D, E &F). The patient felt at ease throughout the surgery, and there was no bleeding. The specimen was sent for histopathological examination and revealed gingival fibromatosis. The hyperplastic gingival tissues showed a mature fibrous tissue composition with high levels of collagen fibres and focal inflammatory cell infiltration, consistent with the characteristics of gingival fibromatosis (Fig. 3). The patient was also instructed to use 0.2% chlorhexidine mouth rinse twice a day for three days straight. The patient continued practicing proper dental care following the gingivectomy, and the crowns were sufficiently exposed. The gingival morphology in the upper teeth dramatically improved after three months of review (Fig. 4). The patient was enrolled in the follow-up program at intervals of 1, 3, and 6 months and after 1 and 2 years. Orthodontic therapy was commenced 3 months after the gingivectomy was initiated and is currently ongoing for an upper removable appliance to correct increased overjet and overbite. The periodontist advised basic periodontal therapy, including teeth polishing, scaling, reinforcement of oral hygiene, and monitoring of tooth eruption, given the patient's mixed dentition. Periodontal maintenance therapy was carried out every six months throughout

this time. Patients with hereditary gingival fibromatosis (HGF) showed significant improvements in their functional, cosmetic, and psychological aspects following surgical therapy. Patient satisfaction rose, oral function improved, and aesthetic results improved as a result of these procedures. Patients showed a notable improvement in attitude during follow-up appointments, looking noticeably happier and more satisfied with their outcomes. For one year following the surgical procedure, the patient went back on occasion for follow-up appointments but showed no symptoms of recurrence.

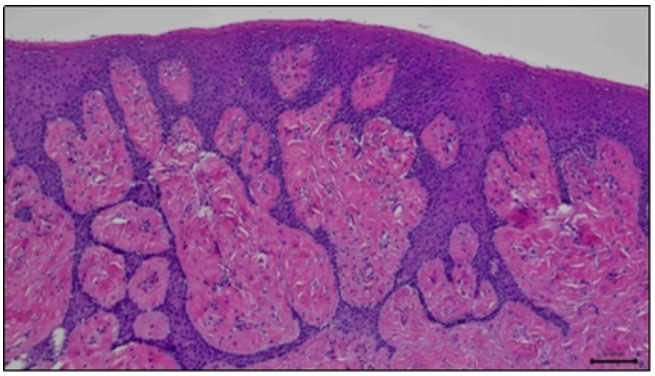


Fig. 3. Dense fibrocollagenous connective tissue surfaced by parakeratinised stratified squamous epithelium with thin rete ridges (H& E, original magnification 40x).

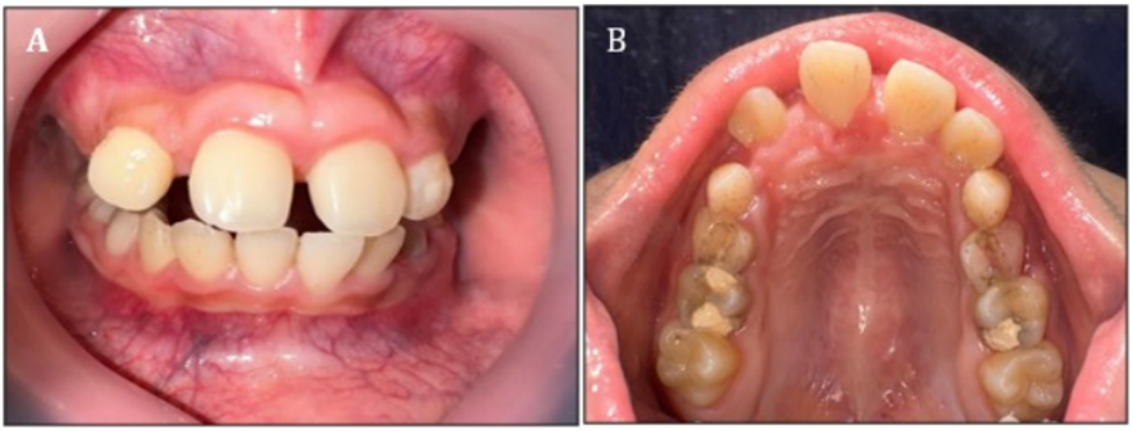


Fig. 4. Post-operative after 3 months (A) frontal view; (B) Palatal view.

3. DISCUSSION

HGF is an uncommon disorder of the gingival tissues marked by the hypertrophy of both free and connected gingiva (Kumari et al., 2021). HGF is classified into two types, nodular type and symmetric type. The nodular type is distinguished by the presence of numerous tumors in the dental papillae, whereas the symmetric type is marked by homogeneous growth of the gingiva (Huang et al., 2022).

The severity of gingival overgrowth can range from a mild enlargement of an isolated interdental papillae to a segmental or uniform and marked enlargement that affects one or both of the mandible (Kumari et al., 2021). In severe involvement, teeth are almost completely covered. The onset of gingival overgrowth

usually coincides with the eruption of the permanent dentition or with the eruption of the primary dentition (Bhutada et al., 2017). In this case report, we present a case of severe nonsyndromic idiopathic gingival fibromatosis and its management. We diagnosed idiopathic gingival fibromatosis due to the absence of any contributing family, medical, prenatal, or medication histories (Almiñana-Pastor et al., 2017).

Congenital or inherited factors can cause HGF. Literature indicates autosomal dominant inheritance with chromosomal anomaly in 2p21-p22 and 5q13-q22, and hormonal and dietary variables may be the cause of HGF (Almiñana-Pastor et al., 2017). One potential explanation has been proposed to be a mutation in the Son of Sevenless-1 (SOS-1) gene (Kumari et al., 2021). HGF may present as an isolated condition or as part of a syndrome, associated with various clinical manifestations such as intellectual disability, hypertrichosis, epilepsy, auditory impairment, growth retardation, and extremity deformities . In current cases, the patients did not disclose any systemic diseases or medication usage.

A slow-growing, asymptomatic gingival enlargement, either localized or widespread, is the clinical manifestation of HGF (Ko et al., 2016). A delayed eruption, an open bite, lip trap, mouth breathing, diastemas, altered phonation, swallowing and chewing, improper tooth alignment, halitosis, and other findings are among the other findings (Ko et al., 2016). Our case report corroborates these findings. Moreover, the proliferation of gingival tissue may intensify and increase the likelihood of bacterial plaque accumulation, ultimately resulting in periodontal disease. Diffuse gingival overgrowth has a broad clinical differential diagnosis that encompasses a range of inflammatory, reactive, and neoplastic diseases with unique biological characteristics (Patel et al., 2024). A definitive diagnosis of HGF was made on the basis of clinical presentation, family history, and histopathological features (Bhutada et al., 2017).

It is essential to differentiate HGF from other forms of gingival enlargement, such as inflammatory gingival enlargement, drug-induced gingival enlargement, enlargement due to hormonal or nutritional factors, pyogenic granuloma, neoplastic gingival enlargement, and enlargement linked to systemic diseases (Bhutada et al., 2017). When making a differential diagnosis including HGF, a number of disorders should be taken into account, including neurofibromatosis, leukaemia, hemifacial hypertrophy, local trauma, and the use of drugs such as phenytoin, cyclosporine, barbiturates, or calcium channel blockers (Patel et al., 2024).

First-line treatment for HGF is currently recommended to be long-term and progressive and includes surgery, periodontal care, orthodontic treatment, and extraction of teeth (Mariani et al., 2021). Depending on the severity of gingival fibromatosis, several treatments are used, such as electrocautery, carbon dioxide laser, and surgery. Consider the following factors before choosing the surgical technique: i) Patient age: The external bevel approach is quicker, requires no mucoperiosteal flap raise, and requires no sutures for younger patients or those who are less cooperative (Mariani et al., 2021); ii) Eruptive stage: where the eruption is complete, an internal bevel is the better option since it facilitates apical tissue placement following flap elevation and enables osteoplasty if required (Gao et al. 2019).

Regardless of the therapeutic method employed, dentists must examine the patient's age, medical history, severity of the condition, existence or absence of any diseases, as well as the patients' views and preferences. The therapy objectives should encompass steps to rectify mastication and phonation, facilitate healthy growth and development, and, if feasible, mitigate emotional repercussions stemming from aesthetic enhancement (Mariani et al., 2021).

Gingivoplasty and external bevel gingivectomy are two typical surgical procedures employed in the past. These techniques use a scalpel to remove or sculpt the extra gingival tissue. Electrosurgery and laser

surgery are two more effective ways to remove the enlarged tissue. Reduced bleeding, less discomfort during surgery, and improved restoration of the natural gingival margin and anatomy are all advantages of these procedures (Gao et al. 2019). In this case report, the maxillary labial and buccal gingiva underwent an external bevel gingivectomy, and extra tissue from the lingual and palatal sides was removed using a diode laser. The use of the diode laser technique in this case report resulted in reduced bleeding, minimized pain during and after the procedure, and a significant decrease in the amount of local anesthesia required. Additionally, the laser improved visibility, shortened chairside time, reduced operator fatigue, and enhanced patient acceptance (Almiñana-Pastor et al., 2017). The laser has several advantages over a scalpel when performing gingival lesion surgery. It may coagulate and close blood vessels, evaporate tissue, create precise incisions, and enhance the healing process because of its antibacterial qualities (Gawron et al., 2016). According to several studies, laser technique improved shape, visibility, and hemostasis during incision, especially in the posterior palatal and lingual regions (Almiñana-Pastor et al., 2017)(Gawron et al., 2016). This is consistent with previous research showing that hypertrophic soft tissues treated with laser had less tissue damage, edema, and postoperative pain than surgically treated areas (Gao et al. 2019). Furthermore, laser technique generates a superficial thermal incision, which lowers inflammatory reaction and post-operative discomfort when compared to other surgical procedures (Bhutada et al., 2017).

In this case report, the patient reported great levels of satisfaction with the aesthetic outcomes after a month. It is a painless, safe, and successful therapeutic option due to its low recurrence rate and simplicity of the procedure (Bhutada et al., 2017). After surgical therapy, the overall recurrence rate for HGF is 34.92%, indicating a high likelihood of recurrence (Gawron et al., 2016). Therefore, it is crucial to maintain excellent oral hygiene and schedule routine periodontal follow-ups during orthodontic treatment (Mariani et al., 2021). Furthermore, we performed orthodontic treatment after surgery to align the teeth and establish long-term periodontal stability.

Future studies should concentrate on assessing the recurrence rates and long-term efficacy of different surgical methods for HGF. To evaluate the results of conventional versus cutting-edge treatment techniques, like lasers and electrosurgery, comparative studies are required.

4. CONCLUSION

In order to determine whether HGF is present in gingival overgrowth, whether it is localized or diffuse, a complete medical and family history, as well as careful clinical and radiographic evaluations, are required. An accurate clinical diagnosis, multidisciplinary treatment comprising periodontal, orthodontic, and maintenance treatment of the HGF patient, and routine follow-up exams led to satisfactory long-term outcomes. To achieve sustained periodontal outcomes, long-term maintenance and follow-up are also necessary to reduce the risk of recurrence.

5. ACKNOWLEDGEMENTS

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6. CONFLICT OF INTEREST STATEMENT

The authors agree that this research was conducted in the absence of any self-benefits, commercial or financial conflicts and declare the absence of conflicting interests with the funders.

7. AUTHORS' CONTRIBUTIONS

Dayang Fadzlina Abang Ibrahim, the first author and a corresponding author, conceived, wrote, revised the article, and provided the data for the case report and submitted the revised manuscript. Soonia Laxmmi, the second author, conceptualised the central research idea and provided the theoretical framework. Aisah Ahmad and Lorend Telajan Achol, the third and fourth authors, provided logistic support and anchored the review and revisions. All authors have critically reviewed the case report and are responsible for the content and the manuscript.

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9. APPENDIX

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