Diagnosis and Treatment of a Hereditary Gingival Fibromatosis Case

Min ZHOU[1,2], Li XU[1], Huan Xin MENG[1]

Hereditary gingival fibromatosis (HGF) is a rare condition characterised by severe gingival hyperplasia, which could result in serious aesthetic and emotional problems and functional impairment. Here the present authors report a case of a 28-year-old female patient with generalised severe gingival enlargement covering almost all of the teeth and diagnosed as HGF. Her family history was of significance, since her father and 3-year-old daughter suffered from the same symptoms. Many studies have agreed that surgical removal should be used in the treatment of HGF, and gingivectomy is the most common method. This study tried both external and internal bevel incisions. The results suggest that the former is better for shaping gingival contour, if the attached gingiva is adequate. Correct physiological contour of the marginal gingiva, good oral hygiene and periodic recall can decrease recurrence risk. Post-surgical follow-up after 26 months demonstrated no recurrence and the patient was satisfied with her appearance.

Key words: *hereditary gingival fibromatosis, treatment*

Gingival enlargement may occur as a side effect of systemic medications, including phenytoin, cyclosporine and nifedipine, or because of leukemic infiltration. These lesions are to some extent plaque-dependent, or may be due to the presence of leukemic infiltration. However, gingival enlargement may also be of genetic origin; in such cases, lesions are known as hereditary gingival fibromatosis (HGF)[1]. HGF is a rare disease, with a phenotype frequency of only 1 in 750,000 people, with males and females equally affected[2]. The gingival tissue of HGF patients contains large amounts of interstitial collagen and other extracellular matrix molecules. The increased proliferation of HGF fibroblasts and their increased production of extracellular matrix molecules such as collagen and fibronectin may contribute to the clinical gingival enlargement characteristics of HGF[3]. HGF may be an isolated disease, or it may be part of a syndrome associated with other clinical manifestations, such as hypertrichosis, mental retardation, hearing loss and abnormalities of extremities. Isolated forms of HGF are compatible with a normal life span, but the aesthetic and dental-associated alterations can considerably reduce quality of life and may result in serious emotional and social problems and functional impairment[4]. This report presents one case of an HGF patient, with the clinical features, diagnosis and treatment. The periodontal condition and aesthetic appearance of the patient improved considerably after the treatment. Post-surgical follow-up after 26 months demonstrated no recurrence.

[1] Department of Peridontology, Peking University School and Hospital of Stomatology, Beijing, P.R. China.
[2] Tongji University School of Stomatology, Shanghai, P.R. China.

Corresponding author: Dr Li XU, Department of Peridontology, Peking University School and Hospital of Stomatology, #22 Zhongguancun Nandajie Haidian District, Beijing 100081, P.R. China. Tel: 86-10-62179977; E-mail: xulihome@263.net

Case report

A 28-year-old Chinese female patient was referred to Peking University Stomatology Hospital with a complaint of generalised gingival progressive overgrowth since childhood. The enlarged gingiva caused difficulties in speaking and eating and it had also compromised her aesthetic appearance remarkably. She did not have any

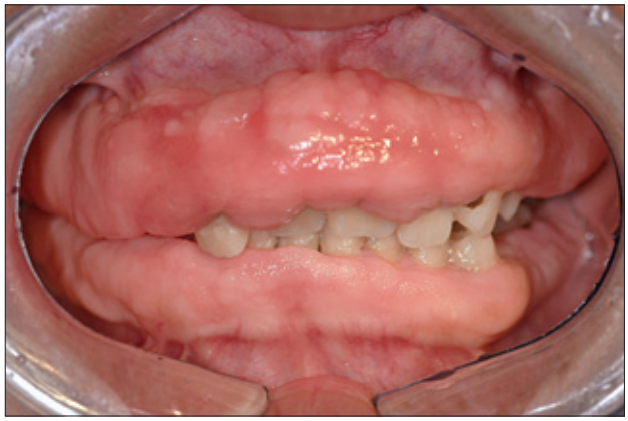

Fig 1 Intraoral view of the generalised severe gingival enlargement at the initial visit.

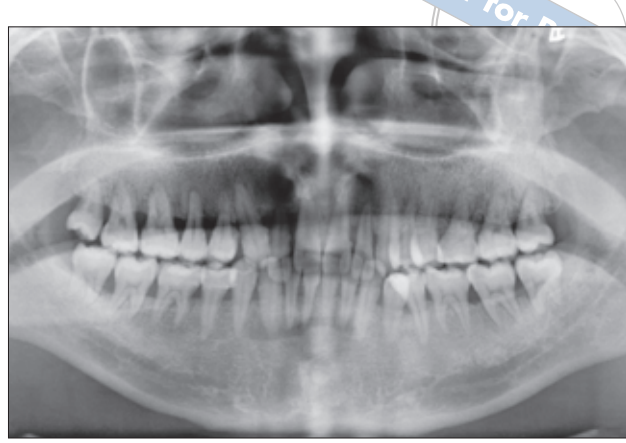

Fig 2 The panoramic radiograph showed mild horizontal bone loss.

other symptoms, such as pain, neuroparalysis or bleeding following brushing, and she denied mouth-breathing habits, systemic diseases and significant medical history. She had never visited the dentist before.

On examination, the patient did not reveal any abnormality except thick and protruding lips. She exhibited generally good health and no signs of hypertricosis or mental retardation. Intraoral examination revealed her oral hygiene was poor and plaque deposits were abundant. Large amounts of calculus were noted on the lingual surfaces of the mandibular incisors and subgingivally in all quadrants. Generalised severe gingival hyperplasia involved both the mandibular and maxillary arches, covering most of the tooth surfaces. The gingiva was pink in colour, with a firm and dense consistency (Fig 1). The false pocket depth was about 9 to 12 mm, with a slight tendency to bleed on probing. Apart from the right maxillary lateral incisor, which exhibited degree II mobility and severe migration, there were no obvious loose teeth. The panoramic radiograph showed generalised mild horizontal bone loss (Fig 2).

The patient's family history was of significance because her father suffered from the same symptoms and became edentulous at the age of 50 years. Her 3-year-old daughter exhibited slightly hyperplastic gingival tissue, with much wider and thicker keratinized gingiva than normal (Fig 3).

Based on her presenting a generalised severe gingival overgrowth without medication history and her especially significant family history, a diagnosis of HGF was made rather than drug-induced gingival enlargement or leukemic infiltration. It was obvious from the bone resorption that she also had chronic periodondtitis.

Histopathological examination of the tissue removed during gingivectomy showed the underlying stroma was almost full of hyperplastic dense collagen fibres, with a small amount of inflammatory cell infiltration. The epithelium was wider than normal and slightly hyperkeratotic, with elongated papillae (Fig 4). This confirmed the diagnosis.

The treatment goals were to solve the functional and aesthetic problems and prevent recurrence. Thus the treatment protocol comprised oral hygiene instructions (OHI); non-surgical periodontal treatment, including scaling and root planing; gingivectomy and gingivoplasty; and periodic recall.

The patient was given the periodontal initial (cause-related) therapy including OHI, scaling and root planing, after which the gingiva became pink, firm in consistency and without bleeding on probing.

She received gingivectomy with local anaesthesia quadrant by quadrant over four visits: the interval between each surgery was approximately 2 weeks. After a nerve block and local infiltration anaesthesia with articaine, a horizontal straight incision was made at the middle third using a scalpel (blade No. 15) to remove the bulk of the gingival tissue and expose the teeth below. Then a scalloped external bevel incision was made at the bottom of the pocket, shaping the gingival contour to give a thin and properly festooned margin to the remaining gingiva. The incised tissues were carefully removed by means of a curette or a scaler. Following meticulous debridement, with thorough removal of residual calculus and excessive granulation tissues, the gingival contour was checked and corrected by means of a scalpel or a pair of scissors to ensure there was a 'physiological' gingival contour from the alveolar ridge to the crown for optimal adaptation to the teeth. The wound surface was irrigated with normal saline and then covered by a periodontal dressing. The patient was instructed to use 0.12% chlorhexidine for oral rinsing twice a day for 1 week after each surgery. A

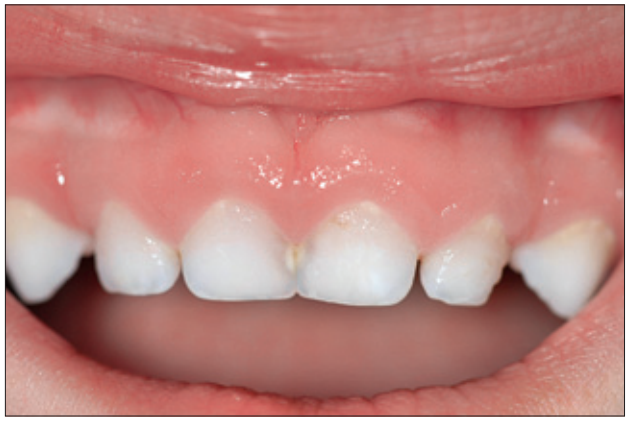

Fig 3 Mild gingival hyperplasia of the patient's 3-year-old daughter.

prophylaxis scaling was given monthly until 6 months after the surgery (Fig 5).

Discussion

Studies have agreed that treatment for HGF should comprise surgical removal of the hyperplastic ginviva, often in a series of gingivectomies, including external or internal bevel gingivectomy in association with gingivoplasty, an apically positioned flap, electrocautery and CO_2 laser[4]. However, mechanical treatment before surgery is still very important and necessary for such hyperplastic disease. During the operation, it was observed that where the soft tissue bled severely, there was always residual calculus below, and the serious bleeding made it difficult to visualise the surgical site. Good initial treatment could reduce bleeding and improve the operation field.

In previous reported cases, authors recommended an internal bevel incision or flap procedure[4-6]. Sengun[7] suggested that a periodontal flap procedure causes less postoperative discomfort as a result of the minimally cut tissue surface compared with a gingivectomy. However, the patient did not report pain or other discomfort after the gingivectomy without any analgesic, even after the removal of the periodontal dressing the next day. This may be attributed to the hyperplastic gingival tissue being almost entirely made up of dense collagen bundles[1] and lacking nerve fibres.

The present authors tried an internal bevel incision, but this proved not as satisfactory as an external bevel incision, as the internal bevel gingivectomy made it difficult to shape the interdental papillae and thin the tissue. Thus another surgery was performed using an external bevel incision in the same section. For those cases with pseudopockets and adequate attached gingiva, an external bevel gingivectomy might produce a better gingival contour.

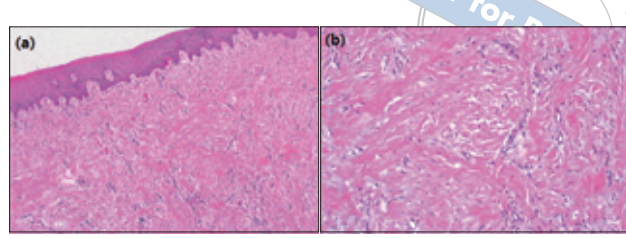

Fig 4 Histopathological examination showed the underlying stroma was almost full of hyperplastic dense collagen fibres: a) haematoxylin and eosin stain, original magnification ×40; b) haematoxylin and eosin stain, original magnification ×100.

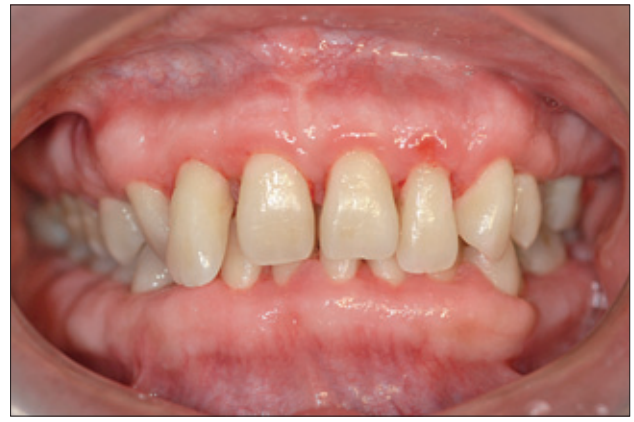

Fig 5 Intraoral appearance 6 months postoperatively.

A laser can be used for the excision of gingival growths. A CO_2 laser, the most commonly used type, permits adequate contouring of the tissue and controls haemorrhage, providing a dry operating field[8]. Roed-Petersen[9], treating mentally retarded patients with phenytoin-induced gingival hyperplasia, used CO_2 laser gingivectomy to prevent intra- or postoperative bleeding and to avoid the need for a surgical dressing (to protect against accidental swallowing). The majority of the patients also did not need any analgesics postoperatively. However, the accumulation of charred tissues, forming a carbonised layer on the wound surface, appeared to act as a protective covering, making further lasing ineffective. The cost of a laser for the dental practitioner, loss of tactile feedback, unpleasant odour and delay in healing impede the application of lasers in dentistry[10,11]. Furthermore, Er:YAG and Nd:YAG lasers are not recommended for major soft tissue surgery[1].

Despite recurrence being a common feature, there were cases reporting no relapse after a long follow-up period. There was no obvious recurrence during 3- and 5-year follow-up periods in two cases reported by Lin[13], and even during a 14-year follow-up reported by Guhan[14].

As most HGF patients also suffer from periodontitis, they should be given periodontal treatment and supportive therapy to facilitate the oral hygiene maintenance and decrease the periodontal bio-burden. Baptista[15] reported an HGF case with gingivectomy under general anaesthesia. After the surgery, the patient refused further periodontal treatment, and did not make a return visit. There was a mild recurrence 20 months after the surgical procedure. Sengun[7] also reported a recurrence case associated with insufficient oral hygiene during the uncontrolled period. It is believed that meticulous removal of calculus and granulation tissues, good oral hygiene maintenance and regular follow-ups help to prevent recurrence. However, Emerson[16] demonstrated that the degree of enlargement did not appear to be related to oral hygiene or to the amount of calculus, and that a correct physiological contour to the marginal gingival is more important in preventing recurrence. Although relapses are not uncommon, and Seymour et al[17] have suggested that the condition does not recur if the teeth are extracted, Ramer[18] believes that the psychological benefits resulting from cosmetic improvement, even temporary, far outweigh the risk of recurrence.

Many studies have demonstrated that elevated synthesis of transforming growth factor beta could promote cellular proliferation and abnormally high collagen and fibronectin production and reduce the synthesis and activity of extracellular matrix-degrading metalloproteinases[4]. Maybe in the future the treatment could be combined with cytokine modulatory therapy to achieve a better therapeutic efficacy and decrease the risk of recurrence.

Most cases of HGF are related to an autosomal-dominant inheritance, although autosomal-recessive inheritance has also been reported[1,19]. However, the penetrance seemed fairly low in the autosomal-dominant mode in a study by Martelli-Junior et al[20]. They studied a family spanning five generations with 117 family members and revealed autosomal-dominant inheritance with a sibling recurrence risk of 0.085 and an offspring recurrence risk of 0.078. The present case was also linked to an autosomal-dominant mode of inheritance. The patient and her father and daughter were all symptomatic for this disease. The daughter, aged 3 years, showed slightly hyperplastic gingival tissue, which should be kept under observation and interference of the eruption of permanent teeth should be avoided.

Conclusion

This case reports the treatment of an HGF case. External bevel gingivectomy is recommended as an easy and convenient surgical technique. Periodontal initial therapy comprising OHI and scaling and root planing is also very important for controlling inflammation and to reduce the probability of relapse.

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