A 15-year Follow-Up of a Gingivectomy Procedure for Idiopathic Gingival Fibromatosis: A Case Report and Literature Review

[*]Keisuke Seki / [**]Shuichi Sato

Few long-term reports exist concerning the treatment of idiopathic gingival fibromatosis, which is a rare autosomal dominant genetic disorder associated with non-inflammatory, benign, and chronic fibrous gingival proliferation and which causes serious esthetic problems.

The aim of this study was to report a case of idiopathic gingival fibromatosis treated with a gingivectomy using an inverse bevel flap method and comprehensively followed up for 15 years. A female patient visited a pediatric dentist at 7 years of age; however, a gingivectomy was not performed until the age of 20 years because of an uncertain prognosis. Now, more than 15 years after the gingivectomy, there has been no significant recurrence and the disease is well managed. Treatment by gingivectomy with an inverse bevel flap approach may provide long-term prevention of recurrence of gingival fibromatosis into adulthood. The aim of this study was to obtain new findings on the pathogenesis and prognosis of this rare disease and to review the case reports previously published.

Keywords: *idiopathic gingival fibromatosis, inverse bevel flap procedure, gingivectomy, long-term follow-up, recurrence*

INTRODUCTION

Gingival fibromatosis is a rare autosomal dominant genetic disorder associated with non-inflammatory[1,2], benign, and chronic fibrous gingival proliferation, with an estimated phenotypic frequency of 1:750, 000[3]. This condition exhibits non-plaque induced gingival overgrowth that develops at the time of eruption of the permanent teeth and often causes serious esthetic problems caused by proliferation of the gingiva, as well as functional problems such as delayed eruption of permanent teeth and odonto-parallaxis[4]. Gingival fibromatosis can be classified into a familial[5] form, with multiple cases occurring among related individuals, and a non-familial (idiopathic)[6] form, in which no other family members are affected. Genetic mapping has shown that there are two loci at 2p21 and 2p16-p13 on chromosome 2p that are reported to be associated with gingival fibromatosis[7,8]. Furthermore, a heterozygous frameshift mutation in the SOS1 (son of sevenless homolog 1) gene has been shown to be associated with 2p211. The modulation of gingival remodeling with changes in the proliferative potential of fibroblasts, collagen fibers, and extracellular matrix (ECM) may result in a variety of phenotypic symptoms. Gingival proliferation is caused by increased deposition of interstitial collagen type I, a major component of ECM in gingival connective tissue. It has been reported that TGF-β1 affects the metabolism of ECM and that TGF-β1 and TGF-β2 levels are increased in fibroblasts, especially in the tissue found in hereditary gingival hyperplasia[9]. Gingivectomy is performed as a symptomatic treatment, despite a high possibility of recurrence[10]. Because of the rarity of this disorder, many short-term reports have focused on the efficacy of surgery, but there is little information about the long-term prognosis into adulthood

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and the pathogenesis of the condition remains unclear. Gingival fibromatosis is often detected by pediatric dentists; however, there is little information available to guide the diagnosis. This article describes the long-term clinical course of a patient with gingival fibromatosis with successful maintenance after gingivectomy, with the aim of obtaining new findings on the pathogenesis and prognosis of this rare disease.

Case report

A 20-year-old woman presented with a chief complaint of poor gingival esthetics, incompetent lips, and gaps between the teeth. Although she was aware of gingival proliferation from the age of 7 years, the local dentist postponed performing a gingivectomy until she was an adult. The patient was referred by her pediatric dentist to our periodontology department in February 2003. The attached gingiva showed marked generalized chronic hyperplasia, with a smooth and hard surface and normal coloration (Figure 1). The lesion was present around all the erupted teeth and the buccolingual width of the alveolar ridge was markedly increased (8–15 mm). The proliferated gingiva covered half of the crown and a probing depth of 4–6 mm was detected around all teeth. The plaque control record was 34% and the patient was brushing twice a day. No inflammatory symptoms were observed. The upper and lower anterior teeth showed 1–2 mm of interdental separation and mild crowding, but no tooth mobility. The patient was a mouth breather, but she had no bruxism. Her family members were all healthy, and no similar gingival symptoms were reported in her parents, siblings, or other close relatives. An interview revealed no syndromes or systemic diseases. The patient's medical history showed no evidence of hypertrichosis, intellectual disability, or use of antihypertensive or antiepileptic medications.

Periapical radiographs showed no abnormalities in the number of teeth, delayed eruption of permanent teeth, or vertical bone resorption. All the roots were short and the anterior teeth of the mandible were characteristically conical (Figure 2a). The results of cephalometric analysis showed that the facial features were oval and symmetrical and the lateral features were convex, with a tendency for the maxillary anterior teeth and upper and lower lips to protrude, but no skeletal problems were observed. No tension of the mentalis muscle was observed during lip closure. U1 to FH and the SN plane were large, SNA and SNB were within the standard range, and there was no protrusion of the alveolar process (Figure 2b). No abnormal findings were seen in the peripheral blood results. Based on these findings, idiopathic gingival fibromatosis was diagnosed.

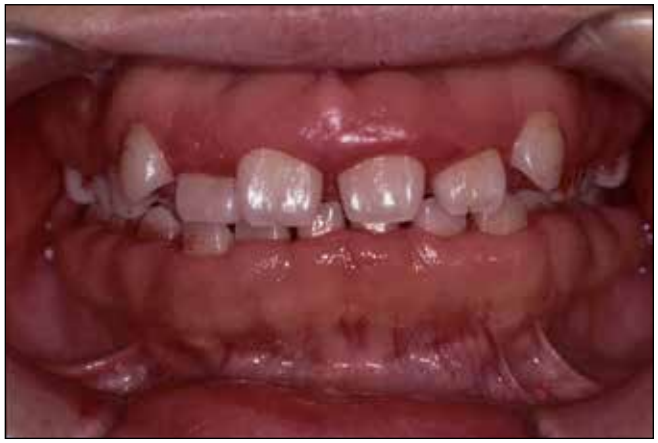

Figure 1. Intraoral presentation after dental plaque staining at the first visit (March 2003, plaque control record: 34%, bleeding on probing: 5.4%). Interdental separation and odontoparallaxis were observed, but there was no tooth mobility.

Gingivectomy with an inverse bevel flap

Immediately after the patient's oral hygiene had improved after initial periodontal therapy, a gingivectomy was performed. A gingivectomy using an inverse bevel flap procedure[11] was performed under intravenous sedation by dividing the mouth into four sections in the following order: maxillary anterior, mandibular anterior, left molar, and right molar. First, the positions of the cemento-enamel junction were marked from the gingival surface using a periodontal probe on a crown-exposed setup model, and the primary incisions were made continuously at these bleeding points using a scalpel (Feather® Surgical Blade #11, Feather Safety Razor Co. Ltd, Osaka, Japan) (Figure 3a,b). Secondary incisions directed apically were added into the connective tissue, leaving the epithelium intact (Figure 3c). Finally, the thick layer of connective tissue remaining on the bone surface was removed through a tertiary incision using a #15 scalpel (Figure 3d) and the flap was repositioned and sutured with 4-0 black silk. This procedure was repeated in the other areas. The maxillary anterior region is shown in Figure 4.

As postoperative medication, oral antibiotics (amoxicillin hydrate) thrice daily for 3 days were prescribed after surgery to prevent infection. Painkillers (loxoprofen sodium hydrate) were used according to the patient's symptoms. Following each surgery, all sutures were removed after 1 week and the patient was instructed to maintain her oral hygiene. After 21 days, epithelialization was almost complete and the patient was able to take regular food. Toothbrushes were changed from the soft type used immediately after surgery to a medium-hard type after healing.

Histological findings

The resected gingiva was sent for histological examination. Dense collagen fibers with few cellular components were observed and small circular areas of cellular infiltrate were found around the blood vessels. An irregularly elongated epithelial image was observed within the stratified squamous epithelium (Figure 5).

Follow-up

Postoperatively, the crown was exposed and the patient reported esthetic improvement. The patient was satisfied with her treatment. Two years after the gingivectomy, partial gingival regrowth was observed in the maxillary anterior section and interdental papillae. No plaque accumulation was observed, and the patient underwent only a minor gingivectomy under local anesthesia (Figure 6). Although migration of the teeth to their natural position was perceived postoperatively[12], orthodontic treatment has been performed since 2008 by a specialist and professional mechanical tooth cleaning has been performed every 6 months for 18 years including while the patient was pregnant (Figure 7). The child born in 2014 has not experienced any gingival proliferation associated with the eruption of permanent teeth.

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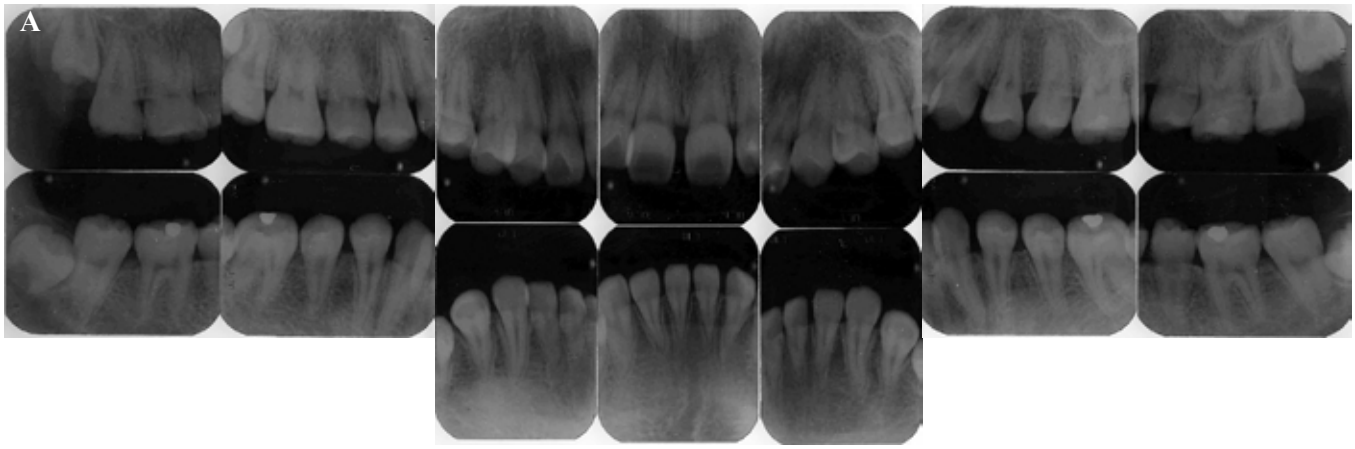


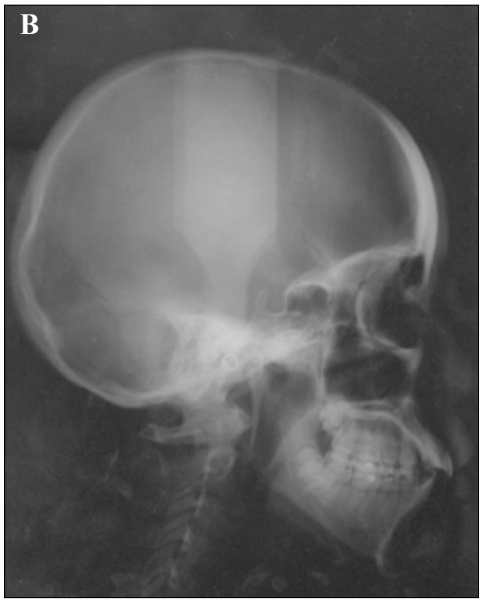
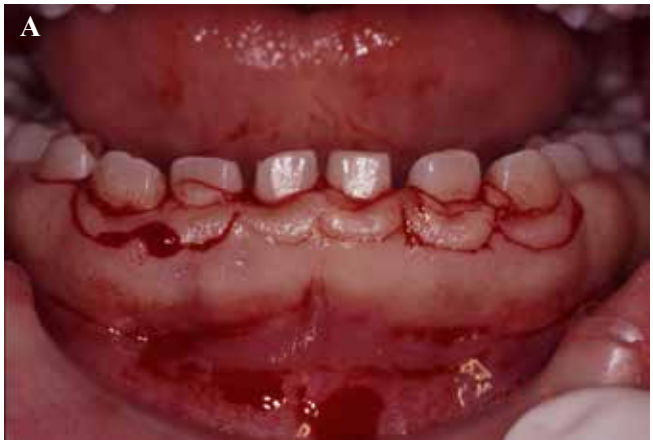
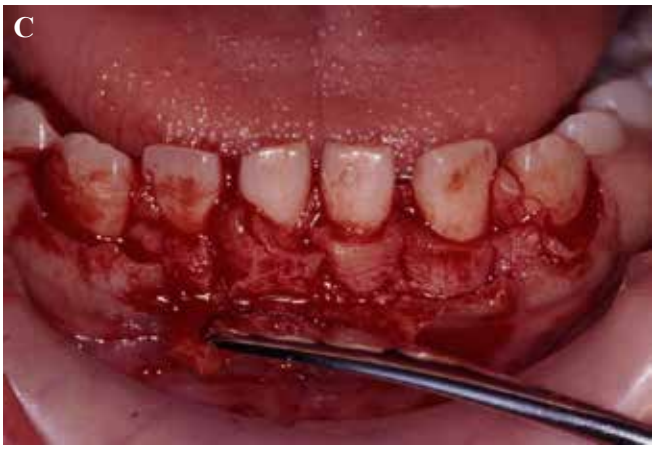


Figure 2. Periapical radiographic findings at the first visit. Long-term gingival overgrowth may have led to shortening of the roots (anterior, mandibular) (a). Lateral cephalometric radiographic findings revealed no protrusion of the alveolar process (b).





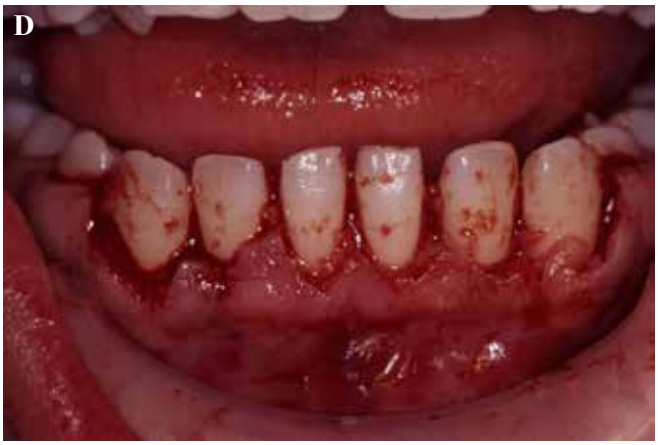


Figure 3. Inverse bevel flap procedure (#33–#43). A primary incision was made along the cemento-enamel junction (a). Excess gingiva was removed vertically (b). A tightly bound thick connective tissue layer remained (c). The flap was repositioned with the connective tissue removed (d).

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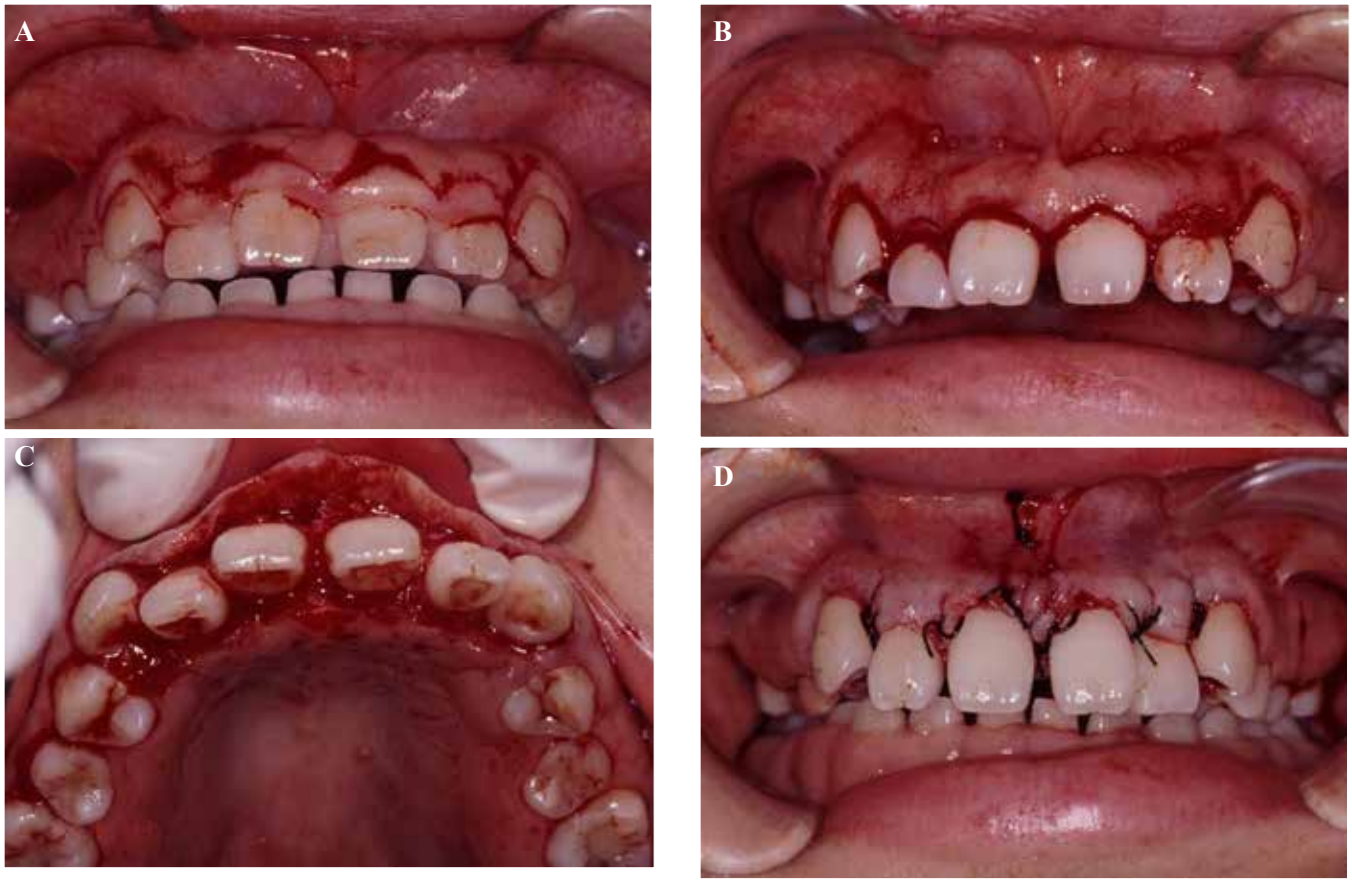

Figure 4. Inverse bevel flap procedure (#13–#23) showing the primary incision (a). After the removal of excess gingiva (b). This procedure aims to reduce the thickness of connective tissue (c). View after suturing (d).

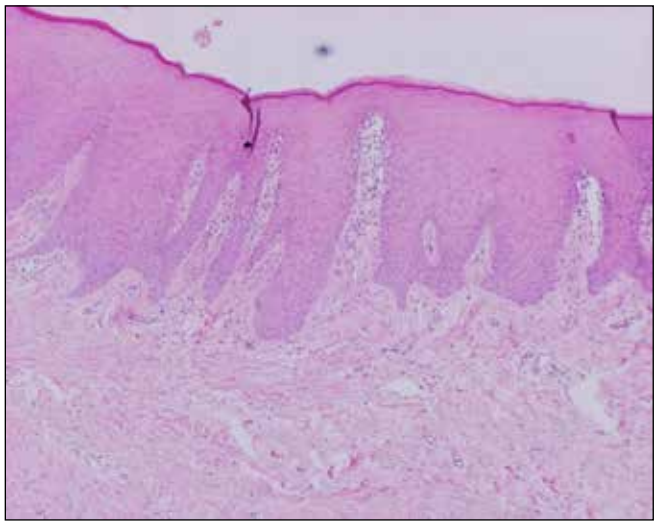

Figure 5. A gingival biopsy demonstrates the elongation of the rete ridges (HE low-power field).

Figure 6. Gingival regrowth after the complete gingivectomy. An additional gingivectomy was performed.

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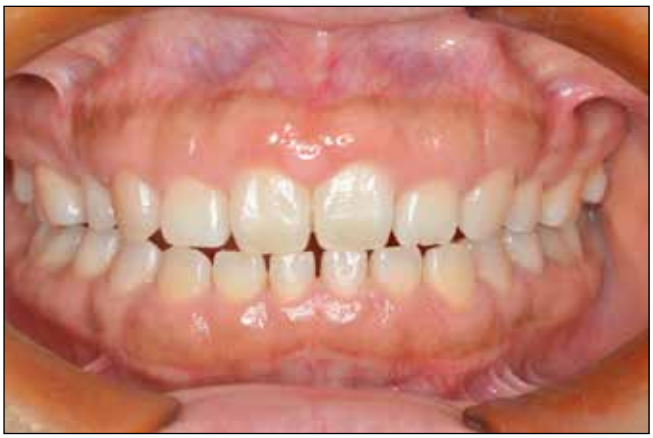

Figure 7. At 15 years postoperatively (aged 38 years). No gingival regrowth was observed and healthy periodontal tissue was maintained.

DISCUSSION

In the present case, the family history of gingival fibromatosis was negative and a diagnosis of idiopathic gingival fibromatosis was made. The thick connective tissue layer was reduced using an inverse bevel flap procedure. The good longitudinal results reported here may be a result of the removal of modulated tissue and a reduction in local inflammatory stimulation. This procedure may also be effective in cases where gingival proliferation is observed during orthodontic treatment and excessive gingival overgrowth remains after orthodontic bracket removal. Although a diode laser technique is often used for gingivectomies to decrease discomfort[13], this technique is mostly suitable for gingivectomies of small areas. It is unclear whether gingivectomy in childhood is effective in preventing pathological abnormalities of the periodontal tissue, but in this case, even though it was performed in adulthood, there was no recurrence, and a long-term stable course was achieved.

To review the relevant literature for this case, we searched for articles published up until 2021. The Medline via PubMed database was searched using the keywords "idiopathic gingival fibromatosis" and "management" and 19 case reports were found from the past 47 years. Hand-searching of the articles revealed that the postoperative follow-up period was 1 year in most reports. Because the reliability of achieving recurrence prevention was questionable in many cases, a follow-up period of 2 years or more was set as an inclusion criterion. Articles that did not state the length of the follow-up period and those not written in English were omitted with a manual search. Six papers fulfilled these criteria[14-19] (Table 1). Only one case reported a maintenance period exceeding 10 years[14], and the present case is the longest follow-up report without recurrence. Three cases reported recurrence, and one case required multiple gingivectomies[14,16,18]. The gingival regrowth in the present case was the least severe of any reported. Only two cases with concurrent disease were reported[14,18], while there were many reports of cases without systemic disease, including this case. Otherwise, a previous study reported a case in which orthodontic treatment was undertaken immediately after gingivectomy[20]; however, the prognosis remains unclear due to the short observation period. In the present case, orthodontic treatment was performed in adulthood and the favorable outcome suggests that gingival proliferation can be controlled and well managed.

Although gingival fibromatosis is not an inflammatory disease, the products of inflammatory cell migration may contribute to the activation of fibroblasts. It is undeniable that poor oral hygiene can trigger gingival regrowth. Hence, there is a close relationship between thorough improvement of the local oral hygiene environment and the prevention of overgrowth recurrence. In addition to the gingivectomy itself, orthodontic treatment also improves the plaque retention factors; therefore, prolonged professional mechanical tooth cleaning may have contributed to the good results.

In addition to stimulation at the time of tooth eruption, the presence of the permanent teeth may also induce gingival overgrowth[21].

Table 1: Case reports of Idiopathic gingival fibromatosis management

Authors (y)	Patient	Treatment	Follow-up	Recurrence	Concurrent disease
Kamolmatyakul et al 14 (2001)	14 F	Gingivectomy (External bevel) 3 times during the age of 5-6 years	13.5 years	Continuous	Mental retardation Chronic administration of phenobarbital
Kavvadia et al 15 (2005)	11 M	Gingivectomies at the age of 7 and 9 years Prosthetic rehabilitation with overdentures	2.5 years	None	Multiple impacted primary teeth Absence of eruption of permanent teeth
Shetty et al 16 (2010)	13 F	Gingivectomy (Loop electrode and a soft tissue diode laser usage)	4 years before	Treated 4 years ago, recovery was reported	
Saini et al 17 (2013)	13 M	Gingivectomy (External bevel)	2.5 years	None	
Gita et al 18 (2014)	14 M	Gingivectomy	2 years	Slight recurrence in the overgrowth	Jones syndrome Progressive deafness
Pol et al 19 (2016)	14 M	Gingivectomy (Electrosurgical)	2 years	None	
Seki et al (present report)	20 F	Gingivectomy (Inverse bevel) Orthodontic treatment	Over 15 years	None	

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However, age-related factors are currently unknown, and in this case a good long-term outcome was achieved in adulthood. This report may assist in bringing significant changes to therapeutic strategies and the prognosis of gingival fibromatosis in the future.

CONCLUSIONS

This long-term report provides useful information about the successful management of periodontal tissue that can be applied to the treatment planning of gingival fibromatosis. Maintenance of good oral hygiene may improve the prognosis and prevent recurrence of gingival fibromatosis for more than 15 years in adulthood.

List of abbreviations

SOS1: sons of sevenless homolog 1; ECM: extracellular matrix; TGF: transforming growth factor

Acknowledgments

None

Funding

None

Conflicts of interest

Keisuke Seki and Shuichi Sato declare that they have no competing interests.

Authors' contributions

K.S. conceived and designed the report; performed the surgical treatment and maintenance; acquired, analyzed, and interpreted the data for the work; and drafted and critically revised the article. S.S. conceived and designed the study and critically revised the article. Both authors approved the final version of the article to be published.

Ethics approval and consent to participate

Patient information and data collection for this study took place at Nihon University School of Dentistry Dental Hospital, Japan. All activities were conducted in full accordance with ethical principles, including the CARE guidelines. The patient gave her written consent for inclusion before she participated in the study. The patient gave her written informed consent for publication of this study.

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