



## CASE STUDY

### HEREDITARY, NON-SYNDROMIC, GINGIVAL FIBROMATOSIS, DIAGNOSIS AND MANAGEMENT: CASE REPORT

\*<sup>1</sup>Soukaïna Oujdad, <sup>2</sup>Noureddine Leshaf and <sup>3</sup>Ihsane Ben Yahya

<sup>1</sup>Faculty of Dental Medicine of Casablanca, Resident, Department of Oral Surgery and Pathology, Rue Abou Alâa Zahr, Casablanca 21100, Morocco

<sup>2</sup>Orthodontist, Private practitioner, Rue Mohamed Fidouzi Ex Jenner Quartier Des Hopitaux, Casablanca 20100, Morocco

<sup>3</sup>Faculty of Dental Medicine of Casablanca, Professor and Head of the Department of Oral Surgery and Pathology, Department of Oral Surgery and Pathology, Rue Abou Alâa Zahr, Casablanca 21100, Morocco

#### ARTICLE INFO

##### Article History:

Received 07<sup>th</sup> December, 2018

Received in revised form

28<sup>th</sup> January, 2019

Accepted 07<sup>th</sup> February, 2019

Published online 30<sup>th</sup> March, 2019

#### ABSTRACT

Hereditary gingival fibromatosis (HGF) is a benign gingival affection characterized by a pathological, diffuse or local, gingival overgrowth. This paper reports a case of familial HGF in a 21 years old female patient.

#### Keywords:

Fibromatosis, Gingival,  
Hereditary, Electrosurgery.

*Copyright © 2019, Soukaïna Oujdad et al. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution and reproduction in any medium, provided the original work is properly cited.*

## INTRODUCTION

### Case report

A 21 years old patient was referred to our department, of oral surgery and pathology, by her orthodontist regarding a gingival overgrowth causing in an unaesthetic smile and interfering with the final stages of the orthodontic treatment. The anamnesis showed no history of epilepsy or mental disorder, no intake of any drugs that may have induced a gingival enlargement. The patient reported that she became aware of her situation around puberty, when she noticed that her gums started covering the crowns of her permanent molars shortly after their eruption, but it was predominantly in the posterior region, and the patient was mostly concerned by her teeth malposition. The patient underwent an orthodontic treatment for 2 years, during which she noticed a slow and constant increase of gingival expansion. Upon examination, the extraoral appearance was normal, and no external swelling or asymmetry was noticed. The intraoral examination revealed a general gingival enlargement in both arches. The overall aspect of the covering tissue was pale pink, non-hemorrhagic,

and firm on palpation, covering approximatively the entire crowns of the maxillary and mandibular molars. The anterior maxillary incisors were half covered on the palatal side, and the anterior mandibular teeth were half covered on both buccal and lingual sides. Periodontal probing indicated generalized pseudo pockets with no sign of bleeding. No pathological tooth mobility was noticed. Dental examination concluded on the presence of temporary fillings on the inferior molars and the maxillary incisors: 36, 37, 46, 47, 12 and 21. A class. I stage 2 dental caries on the premolars: 34 and 44 Fig.1. The panoramic radiograph showed mild alveolar bone loss on the mesial side of the 38 and the 47. A radio-opacity consistent with an endodontic treatment and a well defined radiolucent small image attached to the apices of the 37. An extensive region of demineralization extended to the pulp, a periodontal enlargement and a periapical small radiolucent image on the 47 Fig. 2. The family evaluation revealed that the patient did not come from a consanguineous marriage. Her older brother was diagnosed with a hereditary gingival fibromatosis 13 years earlier and underwent a gingivectomy. Two of her two paternal aunts and her father have also the same condition Fig.3. In light of these informations, we have concluded that the diagnosis was a hereditary, non-syndromic, gingival fibromatosis. The treatment consisted of a quadrant by quadrant excision of the excess tissue using electrosurgery unit, with the blade electrode and under coagulation setting.

#### \*Corresponding author: Soukaïna Oujdad

Faculty of Dental Medicine of Casablanca, Resident, Department of Oral Surgery and Pathology, Rue Abou Alâa Zahr, Casablanca 21100, Morocco.



Figure 1. Endobuccal picture of the patient upon first examination

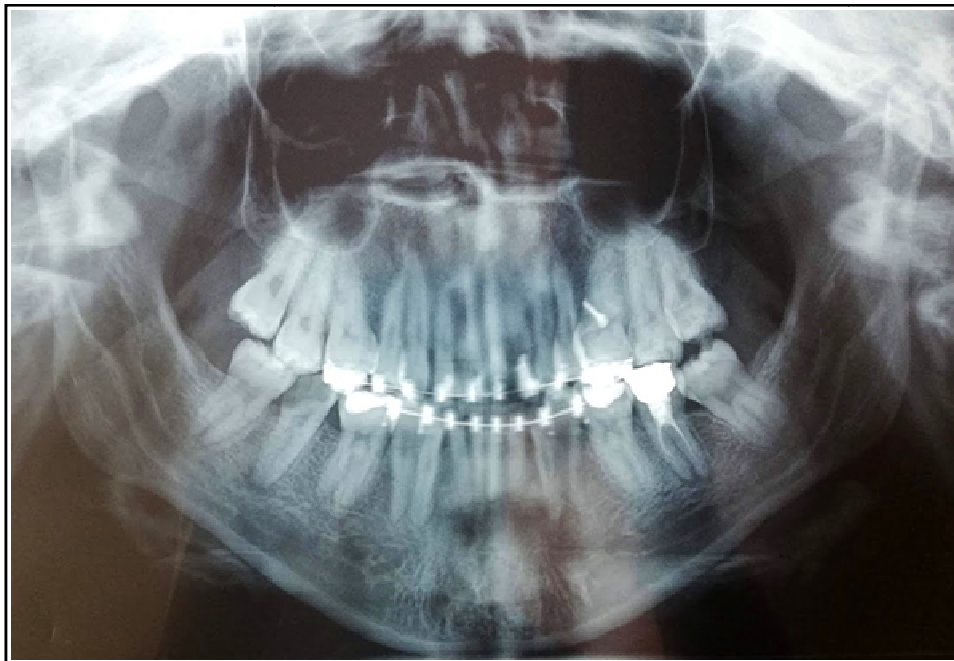


Figure 2. Panoramique Radiograph

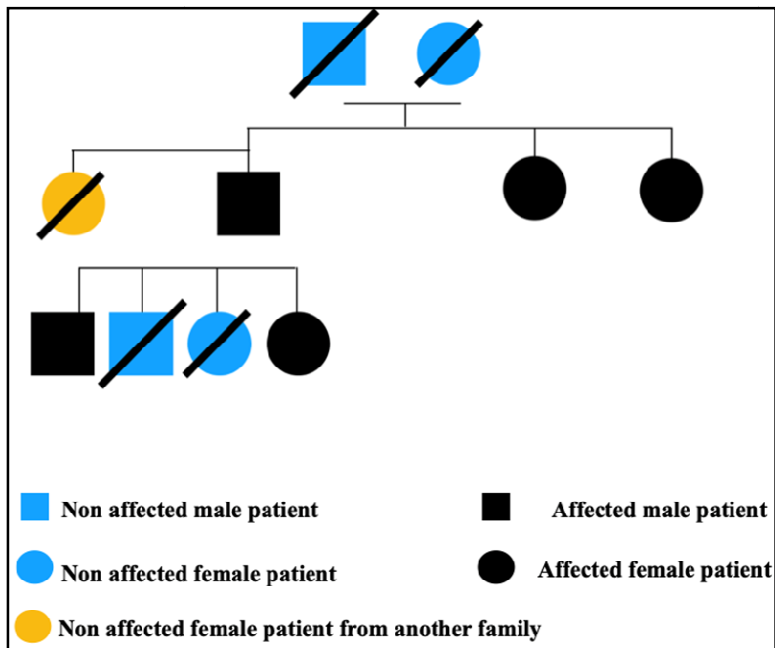


Figure 3. Family Pedigree

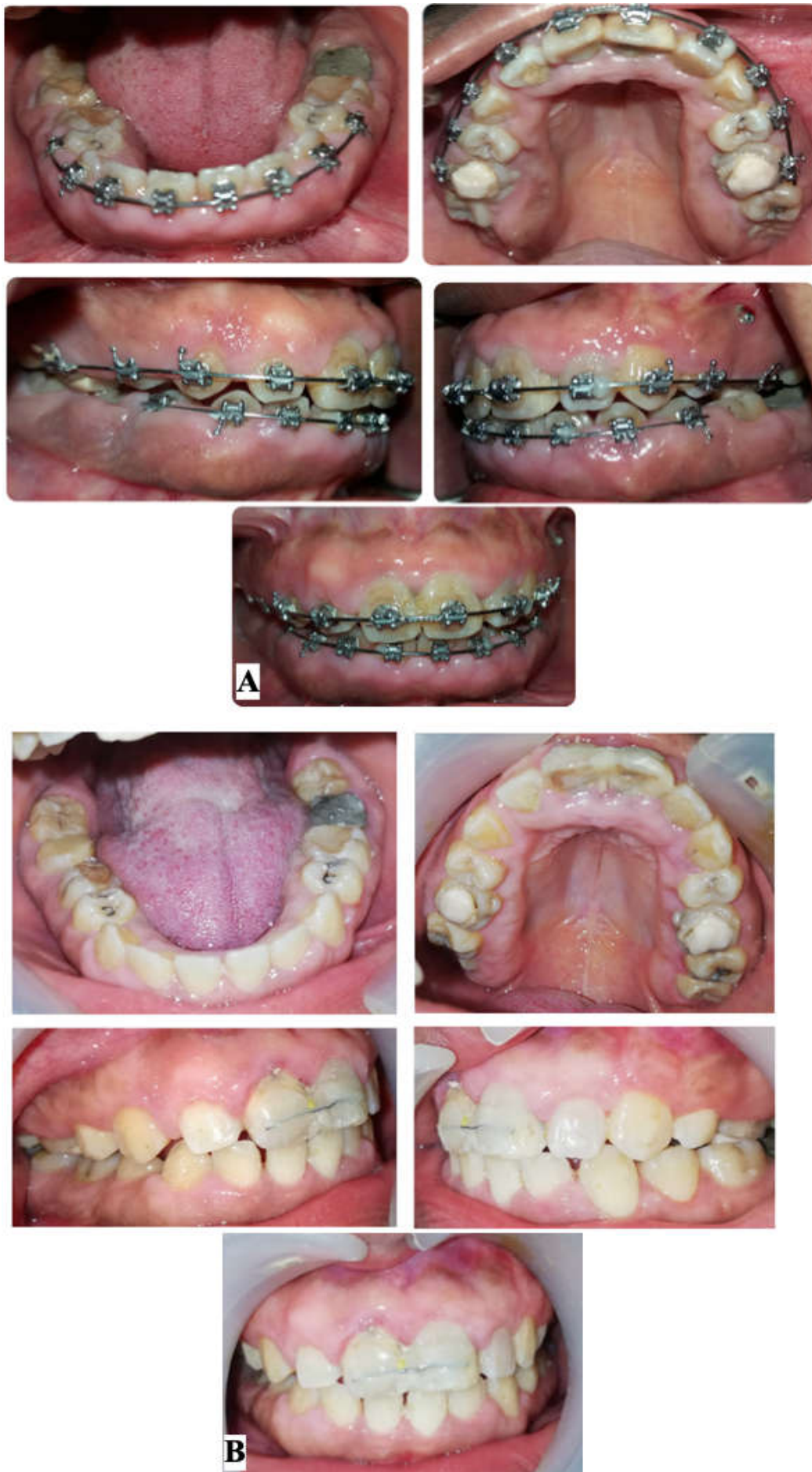


Figure 4. Before (A), and after (B) treatment photographs



Patient was prescribed an antibiotic (oral amoxicillin; 1g 2times/day for 7days), an analgesic (paracetamol 1g every 6hours for 2days) and a chlorhexidine mouthwash (0, 12%, 3 times a day for 7days) after every surgery Fig. 4.

## DISCUSSION

Hereditary Gingival fibromatosis (HGF) is a benign rare condition of genetic origin, identified by a slow overgrowth of the gingival tissue. In the 2018 classification of periodontal and peri-implant diseases and conditions, HGF is classified under non-plaque induced gingival diseases section as a genetic/developmental abnormality (Holmstrup *et al.*, 2018). The main differential diagnosis of HGF is with drug-induced gingival hyperplasia (DIGO), which is a side effect of medication intake. Three pharmacological classes have been implicated: phenytoin (an Antiseizure), cyclosporin (an Immunosuppressor), and nifedipine (a Calcium channel-blocker used as antihypertensive). There is also a form of gingival fibromatosis that cannot be linked to any precise cause (genetic or medication), called idiopathic gingival fibromatosis (IGF). Real numbers about HGF are not well known. Early reports have estimated its prevalence as 1:750,000 (Fletcher, 1966). Males and females are equally affected. This condition may occur in members of the same family, and sometimes with different intensity and expressivity. In general, HGF occurs as an isolated condition or as a part of genetic syndromes such as: Craniofacial dysmorphism, Progressive deafness, Murray-Puretic-Drescher (infantile systemic hyalinosis), Amelogenesis imperfecta, Oculodental syndrome.

It is known to be transmitted as an autosomal dominant trait or, less commonly, an autosomal recessive one. The autosomal dominant forms are usually isolated (non-syndromic). In our case, the family pedigree revealed that all children of the first generation were affected, and 2 of the children of the second generation were positive which confirm the dominant character of the disorder. Linkage analysis have identified a major gene locus for HGF on chromosome 2p21-p22 (Hart *et al.*, 1998; Hart *et al.*, 2002) and to chromosome 5q13-q22 (5). Extended genetic linkage identified a mutation in the Son of Sevenless 1(SOS-1) gene, as a possible cause of isolated (non-syndromic) gingival fibromatosis. Son of sevenless-1 (SOS-1) is a gene involved in the growth of normal healthy gums, but when mutated results in gingival fibromatosis (Hart *et al.*, 2002). Clinically, HGF is a slowly progressive overgrowth of gingival tissue, that is of normal color, non-hemorrhagic, and firm in consistency. The onset almost always occurs with the eruption of primary or permanent dentition. It has been noticed that the presence of teeth is necessary for HGF to occur because the condition disappears or recedes with the absence of the teeth. It affects the masticatory mucosa (the marginal and attached gingiva and the interdental papilla) and can cover parts of, or entire crowns resulting. The excess gingival tissue can cause diastemas, teeth displacement, or in severe cases teeth retention. In fact the unaesthetic prejudice caused by HGF is almost always the main reason for a consultation. In advanced cases, the overgrowth gingiva covers entire crowns, and may deform the palate, leading to functional problems as well (Coletta and Graner, 2006). In our case the patient reported noticing having her gums covering the crowns of her permanent molars shortly after their eruption. Fortunately the enlargement was mostly located in the posterior areas, so it did not have an important aesthetic prejudice, but it may have

enhanced the malposition of her teeth Medical history determine whether the condition is inherited or acquired, the presence of other diseases, and the prior therapies used. Family evaluation confirm the hereditary aspect of the condition. In our case the patient did not have any history of medication intake that may be responsible a drug induced gingival fibromatosis. The family pedigree was more than convincing of the diagnosis of the HGF with an autosomal dominant type of inheritance (Dhadse *et al.*, 2012). HGF does not resolve spontaneously, but can be controlled. The treatment varies according to the degree of severity. When hyperplasia is minimal, careful plaque removal and/or good scaling with adequate hygiene are usually effective. But in most cases where the aesthetic prejudice is the chief complaint, surgery is the treatment of choice, which consists of excess tissue resection by gingivectomy and gingivoplasty (Almiñana-Pastor *et al.*, 2017). In this case, we used electrosurgery unit to perform gingivectomy and gingivoplasty, for these major reasons: The thickness and fibrous consistency of the gingiva quality, which is more manageable and timesaving by the electrosurgery unit. Coagulation effect of the electrosurgery provides a bloodless field allowing a better vision.

**Faster healing process:** even if the healing results of electrosurgery are controversial, in our case it allowed us to make a gingivoplasty without raising a flap, and with no need to suture. The major inconvenient that seemed to be not in favor of the electrosurgery might be the risk of thermal overheating on the bone and roots (Samuel, 2016).

Recurrence is a common feature of HGF, and the patient should be noticed of the possibility to undergo repeated surgical procedures. The period between the surgery and the relapse varies between individuals. Some authors reported long periods of gingival stability: Günhan and al. reported a follow-up of 14 years with no recurrence. The surgical technique employed has not been shown to influence the risk of recurrence (Günhan *et al.*, 1995).

## Conclusion

This case report is a part of familial hereditary gingival fibromatosis with a dominant pattern of inheritance. While the use of medical laser knowing its climax in dentistry, electro surgery unit is almost abandoned. In this case the use of electro surgery was the best choice to reduce the excess tissue and with no per or post operative bleeding.

## REFERENCES

- Almiñana-Pastor, P.J., Buitrago-Vera, P.J., Alpiste-Illueca, F.M. and Catalá-Pizarro, M. 2017. Hereditary gingival fibromatosis: Characteristics and treatment approach. *J Clin Exp Dent.*, 9(4):e599-e602. Published 2017 Apr 1. doi:10.4317/jced.53644.
- Coletta, R. D. and Graner, E. 2006. Hereditary Gingival Fibromatosis: A Systematic Review. *Journal of Periodontology*, 77: 753-764.
- Dhadse, P.V., Yeltiwar, R.K., Pandilwar, P.K. and Gosavi, S.R. 2012. Hereditary gingival fibromatosis. *J Indian Soc Periodontol.*, 16(4):606-9.
- Fletcher, J. 1966. Gingival abnormalities of genetic origin: A preliminary communication with special reference to

- hereditary generalized gingival fibromatosis. *J Dent Res.*, 45:597-612.
- Günhan, O., Gardner, D. G., Bostanci, H. and Günhan, M. 1995. Familial gingival fibromatosis with unusual histologic findings. *Journal of Periodontology* 66, 1008–1011.
- Hart, TC., Pallos, D., Bowden, DW., Bolyard, J., Pettenati, MJ. And Cortelli, JR. 1998. Genetic linkage of hereditary gingival fibromatosis to chromosome 2p21. *Am J Hum Genet.*, 62:876–83. doi: 10.1086/301797.
- Hart, TC., Zhang, Y., Gorry, MC., *et al.*, 2002. A mutation in the SOS1 gene causes hereditary gingival fibromatosis type 1. *Am J Hum Genet.*, 70(4):943-54.
- Holmstrup, P., Plemons, J. and Meyle, J. 2018. Non-plaque-induced gingival diseases. *J Clin Periodontol.*, 45 (Suppl 20):S28–S43.
- James, PL. and Prasad, SV. 1971. Gingival fibromatosis: report of case. *J Oral Surg.*, 29(1):55-9.
- Samuel, B. 2016. Low, Lasers in Surgical Periodontics, Principles and Practice of Laser Dentistry (Second Edition), Mosby, 51-66, ISBN 9780323297622.
- Xiao, S., Bu, L., Zhu, L., Zheng, G., Yang, M., Qian, M., Hu, L., Liu, J., Zhao, G. and Kong, X. 2001. A new locus for hereditary gingival fibromatosis (GINGF2) maps to 5q13-q22. *Genomics*, Jun 1; 74(2):180-5.
- Xiao, S., Wang, X., Qu, B., Yang, M., Liu, G., Bu, L., Wang, Y., Zhu, L., Lei, H., Hu, L., Zhang, X., Liu, J., Zhao, G. and Kong, X. *Genomics*, 2000 Sep 15; 68(3):247-52.

\*\*\*\*\*