ORIGINAL ARTICLE

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Abstract:

The association of a pronounced gingival enlargement (GE) and severe aggressive periodontitis (AP) without underlying diseases or syndromes is rarely reported. We aimed to report a rare case of such association in a young female patient and to review the literature regarding the coexistence of these two diseases. A 20-year-old female patient was referred due to an extensive and generalized gingival enlargement along with teeth mobility with one-year length. Medical and family histories were non-contributory. Intraoral examination revealed a diffuse and smooth-surfaced overgrowth, normal in color with fibrous consistence covering nearly all molars crowns of both upper and lower arches. Histopathological examination revealed excessive amounts of collagen fibers and mild inflammation. The clinical and histopathological features did not fit in any known syndrome or disease. Thus, the diagnosis of generalized idiopathic gingival enlargement associated with severe aggressive periodontitis was concluded and the patient was referred to specialized periodontal treatment. The real meaning of the association between GE and AP remains unknown. Further investigation is desirable to understand the coexistence of these two diseases.

Keywords: Gingival Hyperplasia; Aggressive Periodontitis; Periodontal Diseases

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INTRODUCTION

Gingival enlargement (GE), overgrowth (GO) and fibromatosis (GF) are clinical terms used interchangeably to designate abnormal growth of gingiva¹. GE may be genetically inherited, either as an isolated feature or as part of a syndrome, medication-related or idiopathic². Despite the broad use of such terms, some authors may argue that gingival fibromatosis is a different condition, usually associated with the eruption of both deciduous or permanent teeth³. In either case, a diffuse or local overgrowth of gingiva that can be worsened due to inflammation can be observed.

The loss of periodontal support is commonly observed in older patients as a result of periodontal disease progression. Younger patients may present a severe form of periodontal disease known as aggressive periodontitis (AP). AP is genetically inherited, despite its pathogenesis is not clearly elucidated^{4,5}. The association of severe periodontal loss and a pronounced gingival enlargement without underlying syndromes or diseases is rarely reported, especially if no clear genetic inheritance is identified^{6,7}. We aim to report a new case and to review the literature regarding this rare association.

CASE PRESENTATION

A 20-year-old female patient was referred by her private dentist to the stomatology service of University of Sao Paulo, School of Dentistry, Sao Paulo, Brazil, due to an extensive and generalized gingival enlargement along with teeth mobility with one year of duration. The patient reported no previous episodes of GE and her medical and family history were non-contributory in relation to her clinical findings. The only medication she was taking was an oral contraceptive. Allergy to triethanolamine was also reported. Hematological exams matched normal parameters.

The intraoral examination revealed a diffuse and smooth-surfaced gingival overgrowth, normal in color, more exacerbated at the left side, covering nearly all molars crowns of both upper and lower arches (Figure 1 A-D). On palpation, the lesion presented fibrous consistence and was asymptomatic. A good oral hygiene was also observed. The patient presented moderate to deep periodontal pockets, moderate to severe periodontal attachment loss, teeth with mobility degree 1 to 3 mainly at incisors and molars. Further radiographic examination revealed advanced alveolar bone loss (Figure 2 A-B). She was using a chlorhexidine gluconate 0.12% mouthwash regularly without noticing any improvement.

An incisional biopsy was performed in the upper left first molar buccal gingiva region along with gingival crevicular fluid culture and an antibiogram. The histopathological examination revealed excessive amounts of collagen fibers and mild inflammation (Figure 3). The results from the crevicular fluid culture and antibiogram were negative for aerobic and anaerobic microbiota. A drug treatment including amoxicillin 500 mg and metronidazole 400 mg was prescribed for 14 days⁸. Despite noticing a gingival amelioration, the patient felt nausea and interrupted antibiotics in the 4th day. Therefore, the drug treatment was replaced to amoxicillin and potassium clavulanate without clinical improvement.

The clinical and histopathological features did not fit in any known syndrome or disease. Hence, the diagnosis of generalized idiopathic gingival enlargement associated with severe aggressive periodontitis was concluded. The diagnosis of aggressive periodontitis was based on the age of the patient, low plaque levels and severity of periodontal destruction⁹. The patient was referred to specialized care in periodontal therapy where she is still under treatment.

DISCUSSION

Gingival enlargement (GE) is not a disease itself but rather encompasses a lot of definitions describing an abnormal gingival swelling at any age. Idiopathic cases are less frequent reported than the ones associated with hereditary factors, including syndromes and non-syndromic conditions, inflammatory diseases of gingiva, non-genetic diseases and medications^{2,10}. On the other hand, aggressive periodontitis (AP) is a specific type of periodontal disease in which the amounts of biofilm are inconsistent with the extensive periodontal destruction. AP is genetically inherited and shows predilection for clinically healthy young patients^{4,11}.

GE and AP are clear distinct entities whose single association seems to be rarely reported. Table 1 discloses the previously reported cases of such association without underlying syndrome or diseases diagnosed. A total of 12 articles were found through Pubmed database but 3 were excluded due to the impossibility of accessing the full article version. The median age of the patients was 20 years old and women were much more affected than men.



Figura 1. Clinical aspects of the lesion. The gingival overgrowth was extensive, affecting both anterior (A-B) and posterior regions (C-D).



Figura 2. Radiographic aspects. The patient presented moderate to severe teeth mobility that matched on radiographic examination to severe periodontal loss mainly in incisors (A) and molar (B) regions.

Authors and year	Age/Gender	Clinical aspects	Etiology	Duration	Country
12(2004)	24y./F	Bimaxillary anterior and generalized AP	Hereditary	-	USA
13(2007)	15y./F	Generalized GE and AP	Idiopathic	Recurrent	India
7(2009)	23y./F	Generalized GE and AP	Idiopathic	6-7 years	India
14(2009)	16y./F	Bimaxillary posterior and localized AP	Idiopathic	2 years	India
15(2013)	15y./F	Bimaxillary unilateral GE and localized AP	Idiopathic	1 year	India
6(2013)	18y./F	Generalized GE and AP	Idiopathic	1 month	India
16(2014)	19y./F	Generalized GE and AP	Idiopathic	6-7 years	India
4(2014)	20y./M.	Bimaxillary posterior and localized AP	Idiopathic	3 months	India
17(2015)	30y./F	Generalized GE and AP	Idiopathic	Recurrent	India

Table 1. Reported cases showing the single association between GE and AP without any other signs or relation to other diseases.



Figura 3. The histopathological examination revealed excessive amounts of collagen fibers and mild inflammation.

The etiology of almost all cases was unknown, being the family and medical history non-contributory as it was observed in the present case. However, it is difficult to ascertain the real contribution of the family history once the patients' relatives are hardly examined by a dentist. There is also the possibility of these relatives to have discrete gingival alterations or incipient periodontal disease unnoticed by them¹². Therefore, a possible hereditary explanation for all cases, including this case report, can never be ruled out.

Besides the hereditary background, other possible explanations can be raised. One may argue that GE is a secondary event of the inflammatory periodontal disease but the remarkable growth, extension and duration of the enlargement in this case discredit this hypothesis. The biopsy specimen showed only a mild inflammatory infiltrate incompatible to what is usually observed in GE caused by inflammation¹⁰. Furthermore, an apparent endemic coexistence of these two diseases can be suspected since almost all case reports are from India^{16,17}. However this is debatable because there are few publications and the present case is just the second described in the western world. We may be facing a new syndrome or simply an occasional finding². The real meaning and importance of the association between GE and AP remains unknown. Therefore, further investigation is desirable to really understand the coexistence of these two diseases.

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