

Juvenile spongiotic gingival hyperplasia: Presentation of two clinical cases

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Abstract

Juvenile spongiotic gingival hyperplasia is a benign lesion, first described by Darling and cols. in 2007, there are several etiopathogenic factors proposed. It is characterized by being in the anterior attached gingiva of the maxilla, clinically it is observed as macular or elevated, with a papillary, granular, or smooth surface, bright red in color, and its presentation can be localized or generalized. It is an asymptomatic entity and is not associated with biofilm-induced inflammation. We present two female patients (17 and 10 years old) with relevant and unprecedented medical history respectively, who presented multifocal lesions of bright red color, painless of velvety texture of soft consistency in the maxilla and jaw in which a conservative therapeutic setting and basic periodontal therapy, observation and controls were performed, did not show regression of the lesions after 6 months of observation. It is important for the dentist to recognize this entity and to make the different differential diagnoses, as these could be clinically very coincidental, to carry out an optimal treatment strategy.

Keywords: Gingival hyperplasia; Gingivitis; Gingival disease; Gingival lesion

1. Introduction

Juvenile spongiotic gingival hyperplasia (JSGH) is a clinicopathological entity that was described by Darling and cols, based on immunohistochemical evidence, hematoxylin and eosin (H&E) sections of 24 cases [1] [2]. In its beginnings it was described that it affects young patients between the first and second decade of life, predilection for the female gender and affects the vestibular attached gingiva of the maxillary anterior zone, in most cases it is separated from the marginal gingiva by a band of healthy gingival tissue, appearing as a localized exophytic lesion, with defined borders, sessile base and bright red color [3]. This pathology was not included in the Armitage 1999 Classification System of Periodontal Diseases and Conditions or the 2018 updated classification. Currently, approximately 221 cases have been described in which it has been demonstrated that it is not a pathology totally of juvenile predilection because there are reports of cases of patients whose presentation was during the sixth decade of life, indeterminate male-female ratio, that is, without predilection for one or the other gender, likewise it has been located in multiple areas of the attached gingiva in both arches, it was evidenced that being exophytic, this entity should be called "hyperplasia" and there are reports of localized and generalized cases [4]. On the other hand, it is considered as the gingival counterpart of acute spongiotic dermatitis [5]. It is currently described as a patchy or raised lesion, with a papillary, granular, or smooth surface, bright red in color with a velvety appearance; patients do not report any symptoms and may bleed when brushing their teeth [6].

The etiopathogenesis is not fully clarified, but there are several mechanisms proposed as possible causes of the disease, among which are: viral infections such as HIV or HPV [7], high levels of estrogen or progesterone [8], allergic, bacterial,

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and fungal factors, external factors such as trauma or orthodontics [3] and local factors such as biofilm or mouth breathing [5]. There is a possibility that it is an entity of odontogenic origin, since it tends to be in the anterior sector of the maxilla [3], so there is a theory that due to the reduction of space when the primary teeth are exfoliated there is residual tissue of the junctional epithelium after the eruption of the permanent teeth [2]. This has been corroborated with immunohistochemical studies finding CK-19 positive [9]. On the other hand, it has also been thought that the epithelial tissue in this area is simply more susceptible to trauma or minor irritants, because it is less keratinized [10].

It has been determined that a biopsy is not necessary to reach a diagnosis since, through its characteristics such as the presence of lesions in the absence of plaque and absent response to periodontal treatment, we can corroborate its presence [2]. The differential diagnosis is a challenge for the professional because we must discern between pathologies such as: biofilm-induced gingivitis, pyogenic granuloma, pubertal gingivitis, irritative fibroma, peripheral giant cell granuloma, hemangioma, peripheral ossifying fibroma, squamous papilloma [3][1][7].

Histologically, three variants have been described: exophytic / papillary, flat and micropapillary [6]. In the epithelium, elongation of the rete pins, epithelial hyperplasia, intracellular edema and spongiosis of the spinous layer are observed. Exocytosis of pro-inflammatory cells, especially neutrophils, can also be found [7].

In the connective tissue the presence of localized inflammatory infiltrate with plasma cells, neutrophils and lymphocytes can be reported. It is important to recognize that lesions that are smaller and milder may not have epithelial hyperplasia, but spongiosis and inflammatory cells, it should be noted that this lesion does not cause bone resorption [1]. All these characteristics are like other pathologies such as fibroma or biofilm-induced gingivitis, so histology by itself is not a diagnostic reference, but it is necessary to correlate the clinical history and the evolution of the lesion with non-invasive procedures [2].

It is considered that the treatment of preference is conservative, waiting for its spontaneous resolution and assisting it with periodontal hygienic therapy so that the condition does not worsen, in extensive lesions or very close to the gingival margin it is better to avoid invasive treatment in order not to leave scars or affect the aesthetics of the gum by possible gingival recession [2]. In the context of other treatments, there is the cold cut excision, using a scalpel with the possibility of affecting esthetics [7], laser photo biomodulation, since an antimicrobial effect is obtained, reduction of inflammation cell migration [5], increasing the expression of the bFGF gene, an important factor in the regeneration of periodontal tissue, therefore, it will provide an optimal healing. The disadvantages of this technique are the high cost of the equipment, and the professional must be trained in the use of the laser to avoid adverse effects [8]. Cryotherapy also has advantages for this pathology, it can be used in patients whose behavior and tolerance to treatments is negative, and in these cases, topical steroids can be used as an adjuvant [4].

The evolution is slow, but with spontaneous resolution, after invasive treatments a recurrence of 6 to 16% has been observed [4], even a report indicated a recurrence of 25%, so it is a pathology to be considered by professionals. It has not been described that spongiotic gingivitis generates any type of lesion in the affected area after its presentation [1].

Therefore, the purpose of this report is to show preliminary findings in two cases of juvenile spongiotic gingival hyperplasia in a healthy patient and a systemically compromised patient. Informed consent was obtained from all individual participants included in the study.

2. Case reports

2.1. Patient 1

Female patient, 17 years old, institutionalized, who enters the consultation without self-help, in a wheelchair, with a history of spastic cerebral palsy diplegia type, total gastrostomy, severe intellectual disability, language and psychomotor developmental disturbances and focal non-motor epilepsy. Medicated with Valproic Acid 7cc every 8 hours, Fluoxetine 5cc every day, Trazodone 50mg one per day and Phenytoin 100mg every 12 hours.

The intraoral examination showed multiple ulcers on the inner side of the lower lip with a well-defined erythematous margin and a yellowish gray pseudomembranous center of approximately 2 to 4 mm, 5 days of evolution with a history of recurrence.

At the level of adherent, marginal and interproximal gingiva in the maxilla and jaw, multifocal lesions of bright red color, painless, velvety texture of soft consistency, multifocal. The lesions near teeth 2.1-2.2 are approximately 6mm in

diameter, tooth 1.1 of 3mm, teeth 1.2-1.3-2.3 of approximately 2mm, in anteroinferior teeth is approximately 8mm and generalized calcified biofilm is observed (Figure 1).



Figure 1 A and B Images of maxilla and jaw, indicating the location of the lesions, and ulcerations in the lower lip

Conservative therapeutic management, observation and periodic check-ups were performed. Additionally, a periodontal hygienic phase due to the generalized calculus. It should be emphasized that the etiology of these lesions is not biofilm, but they could aggravate the lesion.

After two months, a control appointment is made, and plaque type lesions are still observed in the attached, free, and interproximal gingiva. Periodic check-ups will continue until resolution is expected (Figure 2)



Figure 2 A and B maxilla and jaw respectively, where the presence of the lesions can still be seen

2.2. Patient 2

Female patient aged 10 years, who enters the appointment by her own means, with no medical history of relevance. On intraoral examination, the patient showed bright red, painless, multifocal lesions of velvety texture and soft consistency, multifocal, with respect to the adherent and marginal gingiva in the upper maxilla and jaw. The pathology near teeth 1.1-1.2 are approximately 5mm in diameter, tooth 4.3- 4.4 and 3.3 -3.4 are approximately 3mm (Figure 3).



Figure 3 A) Presence of lesion in maxilla at 1.1 level, presence of biofilm. B) Jaw, less bright lesions are observed

Conservative therapeutic management, observation and periodic controls were also carried out. Additionally, periodontal hygienic phase due to generalized calculus and presence of biofilm.

After two months, a control appointment was made, and plaque type lesions were still observed in the attached, free, and interproximal gingiva. Periodic controls will continue until resolution is expected. (Figure 4).



Figure 4 Control after two months, the pathology is still present

3. Discussion

Juvenile spongiotic gingival hyperplasia is a pathology that is little known by dentists in their clinical practice. It is currently considered an unusual finding, as the literature reports approximately 221 cases [8]. This number may be an overestimate or an underestimate, since this entity was recently described and the diagnostic criteria may be confused with other pathologies [8][11].

Its nomenclature has been a matter of discussion in different reports because some authors suggest the change to "spongiotic odontogenic gingivitis" or "spongiotic gingivitis with odontogenic metaplasia (SGOM)" because they consider more appropriate terms, highlighting the odontogenic origin of these lesions and the different histopathological characteristics reported [6].

Regarding the epidemiology there is some controversy, Vieira et al [8] in 2019 in their literature review did not find a clear gender preponderance, on the other hand DeSeta et al [2] in 2020 in their case series report found a predilection for presentation in the first two decades of life (mean age 12 years), with higher frequency in women (2:1) and a predominantly Caucasian trend was observed in a US group (82% white, 14% Hispanic and 4% Asian) coinciding with our report with two female patients aged 10 and 17 years respectively.

This condition was not included in the Armitage 1999 Classification System of Periodontal Diseases and Conditions or in the 2018 updated classification. Therefore, a lesion located at the gingival margin could be mistaken for another biofilm-induced pathology if the diagnosing dentist is not familiar with this condition.

Lafuente et al [3] in 2019 have described several differential diagnoses including: plaque-induced gingivitis, pyogenic granuloma, squamous papilloma, plasma cell gingivitis and fibroepithelial hyperplasia which differ by clinical presentation, influence of sex hormones, pathogenesis and response to periodontal setting procedures and oral hygiene instructions.

Some therapeutics have been proposed, universally a conservative and non-invasive approach is performed with periodic check-ups for resolution. On the other hand, it was feared that surgical treatment of lesions close to the gingival margins could lead to recession of the gingival tissues; therefore, this ran the risk of worsening esthetics [2]. DeSeta et al [2] in 2020 in their study indicated that scalpel excision or photodynamic therapy was previously considered the gold standard. However, a significant proportion of lesions have been noted to recur after surgical approaches and the literature reports recurrence rates ranging from 6% to 16% depending on dissection and laser interventions. Roberts et al [12] reported the use of 9300nm CO₂ laser during 3 sessions with a difference of one month between each session, with this, no significant side effects were observed, but only a superficial ablation by denaturation of proteins where they obtained a better healing, leaving an extremely small erythematous area, improving the aesthetics of that area. They also indicated the advantage of laser treatment, since it did not require anesthesia, painless postoperative period, and no need for medication. For their part, Nogueira et al [4] in 2017 propose cryotherapy as an alternative treatment modality through the use of the open spray technique with liquid nitrogen which was very well accepted by pediatric patients, the main advantages of this technique in cases of localized juvenile spongiotic gingival hyperplasia include

ease of application, relative absence of discomfort, absence of bleeding and satisfactory aesthetic results and the authors propose to perform more studies with a greater number of cases and follow-up to demonstrate the real effectiveness of cryotherapy.

On the other hand, Moine et al [13] in 2018 refer the use of a 70% aqueous solution of trichloroacetic acid (TA) to treat this pathology by inducing a denaturation and destruction of the lesions due to chemical coagulation of the affected tissue and in their case report they demonstrated that the technique offers many advantages in terms of effortless application and manipulation, good tolerance in pediatric patients, low cost and non-invasive, but more studies need to be done with this therapeutic alternative. Decani et al [15] in 2021 report a case of spontaneous resolution without surgical treatment after 19, 31 and 43 months of control.

Meanwhile, Mawardi et al [14] in 202 in their systematic review found that of the 97 cases that were treated by surgical excision, 12 had recurrence within a median follow-up of 29 months, two cases were treated with cryotherapy, one with photodynamic therapy and one case with superficial cauterization with topical clobetasol, all with no reported recurrence. One case was treated with desquamation and chlorhexidine application with no significant response but based on the available evidence data either method may have the most predictable outcome, however, further studies are needed to confirm these findings and explore alternative management options.

4. Conclusion

Juvenile spongiotic gingival hyperplasia is a pathology that can affect systemically healthy or compromised patients. It is opportune to report this entity so that the general dentist can become familiar with the clinicopathological characteristics, recognize it, make the pertinent consultations and differential diagnoses to carry out an optimal treatment and substantially improve the prognosis of this underdiagnosed pathology.

Compliance with ethical standards

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Disclosure of conflict of interest

The authors declare that they have no known competing financial interests or personal relationships that may have influenced the work reported in this document.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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