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LOCALIZED JUVENILE SPONGIOTIC GINGIVAL INFLAMMATION: A REPORT ON 3 CASES

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Abstract

Background and aims. *A new pathological entity with distinct clinico-pathological features has been recently described and termed as juvenile spongiotic gingivitis. Histopathological associated features are unique and characterized by prominent intercellular edema (spongiosis) and neutrophil infiltrate. The aims of this paper were to: introduce juvenile spongiotic gingivitis to the dental and pediatric communities, to report three cases based on clinical and histopathological findings, and to discuss the most common clinical differential diagnoses. The cases were documented at baseline and follow-ups. The clinical appearance of the lesions described in this paper correspond to the pattern described by the literature: 1) localized lesions as bright red slightly raised overgrowths, most often with a subtle papillary or finely granular surface; or 2) multifocal masses or raised papular lesions with a pebbly texture. The first intention treatment approach was personal and professional plaque control. Because of the lack of a good clinical response to conventional therapy, excisional biopsies were performed, which helped establish the diagnosis. The plaque control was reinforced and additional antiseptic local treatment was administered. A real improvement in the local gingival conditions was recorded for all the patients. However, because of the persistence of some bright reddish gingival masses in one of the patients these lesions were treated by surgical excision. The overall clinical outcome was good and stable after one year.*

Conclusions. *The presented cases might raise awareness of this condition among orthodontic specialists because orthodontic treatment could not be applied until the gingival gum disease was resolved.*

Keywords: gingival diseases, surgery, edema, histology.

Background and Aims

A new pathological entity with distinct clinico-pathological features has been recently described and termed as juvenile spongiotic gingivitis (JSG) [1]. The term includes small, bright red, easily bleeding, localized or generalized gingival overgrowth, with a papillary or velvety texture. Usually the lesions are painless, and around 20% are associated with bleeding when brushing teeth. The size of the lesions range from 2 mm to 10 mm in diameter (median 6 mm) [2]. One year later an alternative designation for this disease was suggested: localized juvenile spongiotic gingival hyperplasia [2]. This new denomination was proposed as almost all the monitored lesions (94%) displayed gingival overgrowth as determined by clinical and histopathological features rather than a pure inflammatory process with minimal to no tissue swelling [2]. Histopathological features are

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unique and characterized by prominent intercellular edema (spongiosis) and neutrophilic infiltrate. These lesions have a striking predilection for the gingiva and do not appear to be related to plaque [1]. The disease mostly affects the anterior facial gingiva, with 84% occurrence on the maxillary gingiva and 16% on the mandibular gingiva [1]. The condition occurs with a 2.3:1 female predominance in patients who are approximately 12 years old, on average [2].

This new pathological entity was retrospectively diagnosed in a series of patients from different archives as juvenile spongiotic gingivitis/hyperplasia [1,2]; this entity had been most frequently misdiagnosed as puberty plaque-induced gingivitis because of its predilection to occur in juveniles [3]. Several reports have indicated a significant increase in gingivitis as children enter puberty and during the pubertal period [4,5]. This increase is believed to be related to an alteration in the subgingival microflora [6] due to the exuberant hormonal supply. The severity of puberty plaque-induced gingivitis was related more closely to plaque buildup than to hormones [7]. However, even if there are common clinical signs that could drive to misdiagnosis, the resemblance between JSG and puberty gingivitis ends at this point. While the removal of local factors by oral hygiene techniques was the key to manage hormone-related gingivitis, a lack of response to conventional plaque control was remarked for JSG [1].

Moreover, microscopically, plaque induced gingivitis is associated with chronic inflammatory modifications: increase of the number of vessels, dilatations of the vessels, chronic inflammatory infiltrate [8]. Typically, this condition lacks significant epithelial spongiosis, so it may be ruled out from the differential diagnosis on a microscopical basis [1].

However, the limited age range of the patients under study by Darling et al.(2007) [1] and Chang et al.(2008) [2] suggests the possibility that puberty might play an etiologic role in this new diseased gingival entity. The hormonal change itself does not cause gingivitis, but exaggerates response of the gingiva to local irritation [2]. Other hormone-related gingival alterations with an associated histological inflammatory pattern have been described in association with menstrual cycles, pregnancy, and taking contraceptive medication [9].

The aims of this paper were to: introduce JSG to the dental and pediatric communities, to report three cases based on clinical and histopathological findings and to discuss the most common clinical differential diagnoses.

Patients and methods

Three patients with gingival lesions corresponding to the features described for JSG were addressed to our department in the last 3 years. The medical history of the patients was reviewed using the current questionnaire and verbal confirmation from the parents. Medically, the patients were healthy and reported no medication use. However, for the third patient presenting unusual gingival modifications, a hematological evaluation was recommended; normal values were recorded. The patients had never received periodontal therapy.

The cases were clinically documented at baseline: full mouth periodontal examination, oral hygiene evaluation [10], bleeding on probing [11], orthopantomography, and photographs. The examination of the patients followed a strict protocol [12]. Follow-up photographs were taken and periodontal monitoring of the clinical parameters was recorded. For each JSG case the clinical diagnosis was based on the following characteristics: 1) bright red, painless, singular or multifocal, thickened patches of the attached gingiva, marginal gingiva and/or interdental papillae; 2) the inconsistency between the quantity of plaque at the level of gingival margin and the development of the lesions; 3) the pubertal period of diagnosis [1]; 4) the lack of an appropriate response to conventional therapy [2]. The histological evaluation further confirmed the supposed diagnosis. The individual general and specific characteristics of the patients are available in table I.

For all the patients the lesions were localized only on the buccal aspect of the gingiva and interdental papillae

Tabel I. Demographic and clinical characteristics of the patients with JSG

No	Gender	Age	Affected areas	Oral hygiene (10)	Pocket depths in affected teeth
1	F	14	Anterior mandibular teeth	26%	2-3 mm
2	M	13	Right mandibular anterior teeth	32%	2-3 mm
3	F	15	Generalized, maxillary anterior teeth most affected	43%	3-4 mm

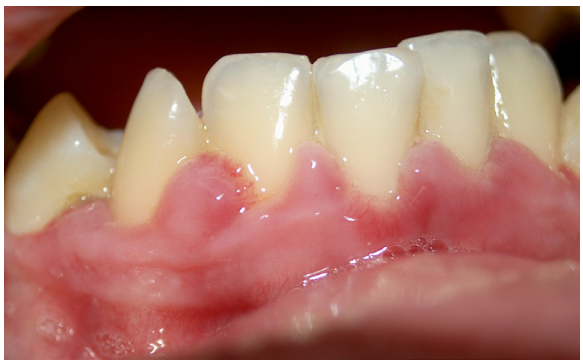

Figure 1. *Patient 1*: Elevated red, pebbly lesions at the level of anterior mandibular teeth

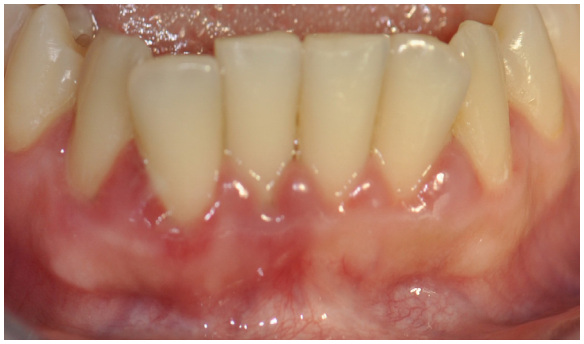

Figure 2. *Patient 2*: Slightly elevated reddish lesions on the level of right mandibular incisors

were touched (Figures 1-3).

Patient no 3 revealed multiple, reddish, highly prominent lesions at the level of the attached gingiva, with irregular outline and a feathery change of the gingiva; these particular lesions alternating with gingival modifications are characteristic for a common chronic gingival inflammation (Figure 3).

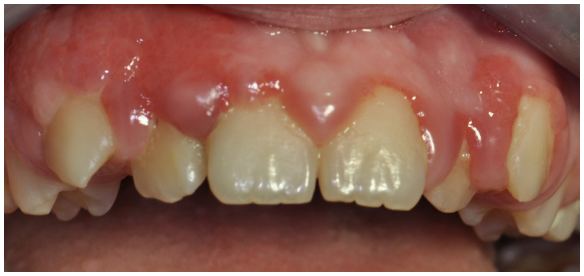

Figure 3. *Patient 3*: Raised, elevated, pebbled, red lesions mostly located at the level of maxillary canines

The first treatment approach was personal and professional plaque control. Because of the lack of a good clinical response to conventional therapy, excisional biopsies were performed and submitted for histopathological examination. Biopsy specimens were examined by light microscopy using hematoxylin and eosin–stained 5-mm tissue sections.

In all patients, the microscopic examination showed long epithelial rete pegs, marked edema in the epithelial layer and lamina propria (spongiosis), loss of keratinization, and intense intraepithelial hemorrhage. Only for two of the specimens was neutrophil infiltrate present. For patient no 3 the same histopathological picture was observed apart from the presence of the inflammatory cells (Figure 4).

Plaque control was reinforced including the professional mechanical debridement and additional antiseptic local treatment was administered to the patients (Corsodyl® Glaxo SmithKline, GB). A real improvement of the local gingival conditions was recorded in the patients. However, in patient no 3 some elevated, bright

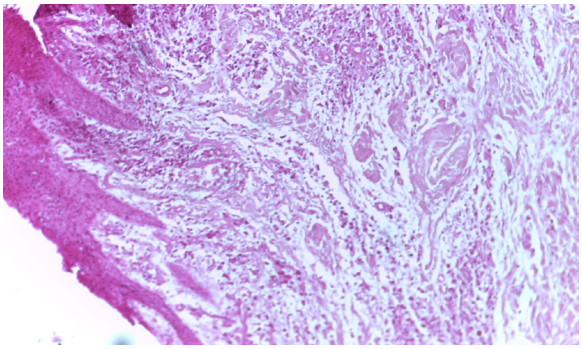

Figure 4. *Patient 3*: histological aspect of the gingival biopsy (HEx10)

reddish tissue masses persisted on the attached gingiva corresponding to the maxillary canines. The lesions were treated by surgical excision associated with a good clinical outcome (Figure 5). The patients were included in a very strict maintenance program. The clinical results were stable after one year.

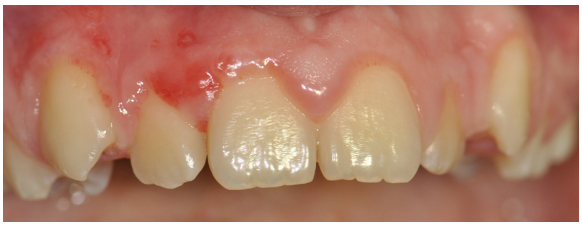

Figure 5. *Patient 3*: 14 days after the surgical excision of the lesions

Discussion

This paper presented three cases that could be described as gingival disease which was initially named JSG and after that reconsidered as a spongiotic gingival hyperplasia. The clinical appearance of the lesions described in this paper correspond to the pattern described in literature: 1) often localized lesions presented clinically as bright red raised overgrowths, most often with a subtle papillary or finely granular surface; or 2) sometimes multifocal masses or raised papular lesions with a pebbly texture [1,2,13].

The clinical localization of the lesions for the presented cases was slightly different from that described by Darling et al. (2007) [1] and Chan et al.(2008) [2] as they mentioned the localization of the gingival modifications mostly at the level of attached and marginal gingiva. In our patients the lesions also involved the interdental papillae. The gingival mucosa of the anterior maxilla is the most common location but, in some cases, the mandibular buccal gingiva was reported to be affected [13] as we recorded.

As two of our patients presented only slightly raised lesions we did not consider the diagnosed entities as hyperplasia. We remarked that even in the presence of minor gingival modifications, the lesions corresponding to JSG had characteristic clinical features: the vivid reddish color and the pebbly/granular aspect of the surface.

The incidence of JSG is unknown and cannot be estimated from the records of a biopsy service. JSG is a newly described pathological entity that is not included in the actual accepted classification of periodontal diseases [14] and probably it was one of the reasons of the misdiagnosis. JSG had been reported as misdiagnosed after a reevaluation of 52 patients' files was performed. Hematoxylin and eosin– stained slides were reviewed and the diagnosis of JSG was confirmed independently by 3 oral pathologists [2]. The first subject of confusion was puberty plaque-induced gingivitis, which is a chronic gingival inflammation induced by dental plaque and characterized clinically by the onset of an exuberant inflammation of the marginal and attached gingiva, with increased gingival bleeding [15]. The confusion is not surprising as pubertal plaque-induced gingivitis is frequently present in adolescents. Based on the new evaluation which corrected the former diagnosis, other pathological entities proved to be wrongly described: inflammatory papillary hyperplasia, chronic mucositis/ gingivitis, or inflammatory fibrous hyperplasia [2].

However, JSG could be clinically a subject of diagnostic confusion and thus clinical differential diagnosis must consider some similar diseased entities. The fact that the cause of JSG is not well-elucidated complicates furthermore the clinical identification of the disease. Pyogenic granuloma is another common clinical entity which could be taken into consideration for a differential diagnosis. Clinically, these lesions are most commonly described as a mass lesion rather than a pure inflammatory one. The raised lesions are frequently found in patients with orthodontic fixed appliances [2]. Sometimes, enlargement of the gingiva may occur as a hormone-related gingival disease, known also as a pregnancy tumor. Microscopically, the gingival enlargements show a mixture of variable amounts of vascular proliferation, inflammation, and fibrotic changes.

The localized and apparently inflammatory nature of JSG lesions leads to speculation about a hypersensitivity reaction to environmental agents or a foreign body reaction. Most of the JSG lesions are too localized to be diagnosed as hypersensitivity reactions; moreover, no foreign material was observed microscopically to sustain a foreign body reaction [2].

The possible etiology of JSG was investigated in the literature. Some authors ruled out the plaque-induced etiology as the JSG lesions on the attached gingiva were observed to be often separated from marginal gingiva and dental plaque by a strip of normal gingiva. Moreover, the lack of response to oral hygiene procedures in these patients was reported [1,2]. The same authors stated that the primary cause of JSG could be an alteration in the epithelium with a loss of keratinization and increased porosity, as seen in junctional epithelium, causing a decreased defense against oral antigens and a subsequently local inflammation. For the patients monitored by the present study, oral hygiene conventional procedures led to certain clinical improvements of the gingival appearance. On the other hand, the lack of the inflammatory infiltrate in the biopsy specimen of the third patient is somewhat surprising because of the inflammatory nature of the clinical aspect.

Inflammatory papillary hyperplasia could be easily microscopically diagnosed because of the histologic evidence of inflammation and papillary hyperplasia. However, the clinical features differentiate it from JSG (frequent palatal localization, older age of development).

The presence of strong staining for cytokeratin CK19 throughout all levels of the epithelium in JSG could reflect its odontogenic origin raising the possibility that the disease could represent an ectopic junctional epithelium at the level of the attached gingiva in association with a secondary inflammatory response [1].

The fact that JSG is benign and sometimes characterized by epithelial hyperplasia raises the possibility of an HPV etiology. The HPV-associated lesions, such as papilloma, verruca vulgaris, condyloma accuminatum, and focal epithelial hyperplasia, can become inflamed but most lesions are not inflamed and rarely show prominent spongiosis. Moreover, JSG does not show koilocytosis or cells in mitosis suggestive for an HPV etiology [2].

Conclusions

1. The gingival lesions described by the present study could be obviously reduced by the reinforcement of oral hygiene and short-term use of topical antimicrobial agents.

2. The diagnosis of JSG requires clinical pathological correlations; the biopsy is an appropriate approach to establish the diagnosis and to rule out other periodontal pathologies.

3. The presented cases might raise awareness of this condition among orthodontic specialists, as an orthodontic treatment could not be applied until the resolution of the gingival pathology.

Acknowledgments

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