Vol. 00 No. 00 ■■ 2025

Clinical and aetiological factors in spongiotic gingival hyperplasia

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Background. Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a rare gingival lesion characterized by exuberant epithelial proliferation. Its etiology remains unclear, with few studies exploring its clinical and histopathological features. This retrospective study aimed to analyze the demographic, clinical, and etiological characteristics of LJSGH across 13 diagnostic centers in Latin America.

Methods. Data were extracted from clinical records, including variables such as age, sex, clinical phenotype, anatomical site, comorbidities, potential triggering factors, and therapeutic approaches.

Results. Seventy cases were included. The mean patient age was 15.9 years, with a female-to-male ratio of 1.41:1. Most lesions were unifocal (72.9%), and the anterior maxillary gingiva was the predominant location (60%). The exophytic protruding pattern was the most common (45.7%). Systemic comorbidities were rare (4.3%), and 58.5% of cases showed no identifiable factors. Regarding potential local etiological factors, 27.14% of patients had teeth malposition in the affected region, and 14.28% were undergoing orthodontic appliances. The presence of biofilm was detected in 47.14% of the cases. Surgical excision was the most frequent treatment (57.1%).

Conclusion. This study supports SGH as a reactive, odontogenic lesion influenced by local and/or multifactorial triggers. Future studies should investigate its multifactorial etiology, focusing on periodontal, microbiological, and histopathological factors. (Oral Surg Oral Med Oral Pathol Oral Radiol 2025;000:1−10)

Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a recently characterized entity, originally described by Darling and subsequently by Chang.[1,2] LJSGH presents a distinctive clinicopathologic pattern, with no clear sex predominance and pathogenesis. Originally, females were thought to be affected more than twice as often as males, but recent studies have suggested a potential male predominance.[3,4] While LJSGH primarily affects children and young adults, cases have also been reported in elderly patients. Additionally, there have been clinical reports documenting LJSGH with a multifocal pattern, challenging the appropriateness of the terms *juvenile* and *localized* in all instances.[5]

The most frequent clinical presentation is a solitary red overgrowth with an irregular, granular, and velvety surface affecting the anterior area of the maxillary gingiva.[2,6] Histopathologically, Spongiotic Gingival Hyperplasia (SGH) is characterized by a nonkeratinized hyperplastic stratified squamous epithelium with a papillary architecture and shows

Statement of Clinical Relevance

This Latin American descriptive study provides deep insights into the clinical and etiological aspects of Localized Juvenile Spongiotic Gingival Hyperplasia, reinforcing previous studies on this lesion and its probable pathogenesis.

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Received for publication Mar 12, 2025; returned for revision Apr 27, 2025; accepted for publication May 8, 2025.

2212-4403/$-see front matter
https://doi.org/10.1016/j.oooo.2025.05.003

spongiosis with intercellular edema and neutrophilic infiltration. In the connective tissue, beneath the elongated papillae, there is evidence of vasodilatation and acute and chronic inflammation with inflammatory cell exocytosis.[4,6,7] SGH is considered a rare lesion and represented only 0.069% of 31469 biopsies submitted to an oral pathology laboratory during a 7-year period.[3] Nevertheless, the nonspecific histopathological features of SGH can be observed in other gingival disorders. Therefore, accurate clinicopathological correlation is essential for a definitive diagnosis. When such correlation is not properly assessed, misdiagnosis as other reactive conditions is possible, potentially explaining the seemingly low incidence of this specific entity.[8]

Over the past decade, the current literature has shown an increase in case reports and case series, enhancing the understanding of the disease and revealing diverse clinical patterns, such as multifocal lesions and cases in older individuals. This has sparked a debate regarding the nomenclature of SGH. For instance, Theofilou et al. suggested the term "spongiotic odontogenic gingivitis" or "spongiotic gingivitis with odontogenic metaplasia" to emphasize the odontogenic origin and the inflammatory nature of the disease.[6]

The real gap in the literature lies in the lack of comprehensive, data-driven descriptions of risk factors associated with SGH. While SGH lacks a response to periodontal treatment, suggesting a weak link to biofilm, some associations with calculus have been described.[4] Although the papillary architecture of the lesion might suggest an association with human papillomavirus (HPV) infection, SGH typically lacks the histopathological features needed to diagnose HPV-related lesions. Moreover, a recent study showed no evidence of HPV DNA in this entity.[3,4] Based on the histological similarity of the lesion epithelium and the junctional epithelium (JE) of the gingival sulcus, it has been proposed that SGH may develop from an *exteriorization* of the JE.[2,9] Lafuente Ibañez et al. hypothesized that the hyperplasic epithelium of SGH may originate from remnants of the JE of deciduous teeth that persist in the gingiva during the eruption of permanent teeth. External irritative factors such as trauma, biofilm, or tooth brushing might then trigger an inflammatory response, leading to the growth of the characteristic reddish tumor in SGH.[8] However, these clinical factors are not well documented in the available literature. This study aims to characterize a large case series of SGH, highlighting demographic, clinical and etiological factors through a multicentric Latin American collaboration.

MATERIALS AND METHODS

Study design

A cross-sectional study was conducted on cases with a clinical-histopathological diagnosis of SGH, based on a multicenter collaboration involving various Oral Medicine and Pathology Diagnostic Centers in Argentina, Brazil, Colombia, Costa Rica, Chile, Mexico, and Venezuela. The STROBE guidelines for reporting cross-sectional studies were followed.

Participants and recruitment

Cases with histopathological diagnosis of SGH according to the current criteria were included from all the participating centers. The histopathological study was carried out according to a specific protocol for this pathology.[3,5,8] Data were extracted from the clinical records and clinical and histopathological databases of each participating center, covering the period from 2016 to 2023. Cases that were ultimately diagnosed as reactive gingival lesions, excluding the diagnosis of SGH based on histopathological examination, were not included in the study. Moreover, cases lacking sufficient data—such as low-quality photographic documentation, incomplete clinical or medical records, and missing histopathological reports—were also excluded from the study.

Variables and measurement

The following variables were extracted from the available records, photographic documentation, and databases from all the participating centres: age, sex, and clinical phenotype. SGH was categorized into 3 novel categories proposed by the authors based on the clinical phenotype: a) Flat—micropapillary: a slightly elevated area of mucosa with a flat or micropapillary surface (a flat lesion or one with a slight increase in volume with an irregular surface), b) Exophytic with papillary surface: an elevated lesion with a visibly irregular surface of granular appearance, which can be manipulated with instruments, and c) Protruding: an elevated, exophytic lesion with a base that may be sessile or pedunculated, with a well-evidenced papillary surface appearance.

Additionally, the anatomical site(s) involved were recorded according to uni- or multifocality, the tooth or teeth involved by the lesion, and the existence of previous systemic comorbidities. Regarding local factors that could be associated with SGH development, it was also recorded whether the tooth associated with SGH was in occlusal trauma or malposition and the existence of orthodontic appliances (fixed or removable, or other orthodontic treatments where the tooth is subjected to external forces and tensions).

The association with biofilm or dental plaque was considered positive when the established criteria for the Simplified Oral Hygiene Index was greater than 1

in the tooth or teeth affected by the lesion. The criteria for classifying debris were as follows: 0: No debris or stains are present; 1: Soft debris covering not more than one-third of the tooth surface, or the presence of extrinsic stains without other debris regardless of the surface area covered; 2: Soft debris covering more than one third, but not more than two-thirds, of the exposed tooth surface; 3: Soft debris covering more than two-thirds of the exposed tooth surface.[10,11] Furthermore, bleeding during periodontal probing was recorded. The therapeutic approach for each case was also recorded.

Control of sources of bias

A virtual consensus meeting was held once all the clinical images from the collaborating centers had been collected. Moreover, the data recruitment method was described, and the process for each center to consistently record the included variables was calibrated, incorporating details from photographic documentation, clinical records, and databases at each participating site. The classification of clinical subtypes was explained before proceeding with the categorization of the included cases. Each center categorized the lesions independently. In case of discrepancies in category assignment, the problematic case was sent to 2 researchers from the leading center of this study (GG and RP) for final categorization and agreement.

Statistical analysis

A narrative descriptive synthesis was provided, using mean, standard deviation, and range for quantitative variables, and relative-absolute frequencies for qualitative variables. This analysis was performed using the software Infostat 2020 (Universidad Nacional de Córdoba, Argentina).

Missing data

The missing data were attributed to the absence of information in the original medical records. These were documented as "no data" (ND) during the descriptive analysis, and were excluded from the statistical analysis.

Ethical statement

The study was approved by the Ethics Committee and Advisory Committee on Teaching and Research at Hospital Señor del Milagro, Salta, Argentina—Protocol 05.08.24, and the Research Ethics Committee of Universidade Federal do Rio de Janeiro (Protocol 5.458.602) and the Scientific and Academic Committee on Research of the Facultad de Odontología, Universidad Nacional de Córdoba (CAIS-FO). This study was conducted following the Helsinki Declaration of 1975, as revised in 2000. All patients signed informed consent and identities were anonymized.

Revision of literature and search strategy

A comprehensive literature search was conducted using PubMed-MEDLINE to identify diagnosed cases of SGH, including case series or descriptive studies with at least 5 cases, published between 2014 and 2024 with the following keywords: ([Localized Juvenile Spongiotic Gingival Hyperplasia] OR [Spongiotic Gingivitis]) AND [clinical]) AND [etiological factors]) AND [risk factors] AND [treatment]. A full-article review was performed following an assessment of the article title and abstract, where possible. Exclusion criteria included no full-text access. Additionally, manual searching of the reference lists in the articles, as well as manual searches of tables of previous literature searches were performed. The literature review aimed to compare our findings with updated published evidence regarding clinicopathological and demographic factors, potential etiologies associated with the lesion, and commonly used treatments (Table II).

RESULTS

Seventy cases of SGH were included, based on a multicenter contribution from 13 diagnostic centers for Oral Medicine and Oral Pathology in Latin America—Figure 1A (21 cases from Argentina, 19 from Venezuela, 9 from Chile, 8 from Brazil, 7 from Costa Rica, 5 from Mexico, and 1 from Colombia), with a mean age of 15.9 years (range: 7-73 years) and a median age of 13 years (SD = 2.5). The sex distribution showed a slight female predominance (58.5%, n = 41) compared to male cases (41.5%, n = 29) with a female-to-male ratio of 1.41:1 (Table I). Four cases were excluded due to insufficient data, missing photographic documentation, and the lack of confirmatory histopathology.

Regarding clinical variables, the most common clinical phenotype was the protruding category (45.7%, n = 32), followed by flat red plaque with a micropapillary surface (30%, n = 21), and exophytic-papillary pattern (24.2%, n = 17). Most cases presented as unifocal (72.8%, n = 51). Figure 2 shows different clinical patterns of the included cases of SGH including cases of multifocal gingival involvement. Regarding histopathological characteristics, all cases showed the classical hyperplasic and spongiotic epithelium presented in SGH (hematoxylin-eosin staining). No additional diagnostic techniques or immunohistochemistry were required. The lesions showed fragments of mucosa covered by nonkeratinized stratified squamous epithelium, with a papillary surface, acanthosis, spongiosis, focally elongated and interconnecting rete pegs, as well as neutrophilic exocytosis. The underlying connective tissue was vascular, with dense inflammatory infiltrates composed mainly of neutrophils, lymphocytes, and plasma cells.

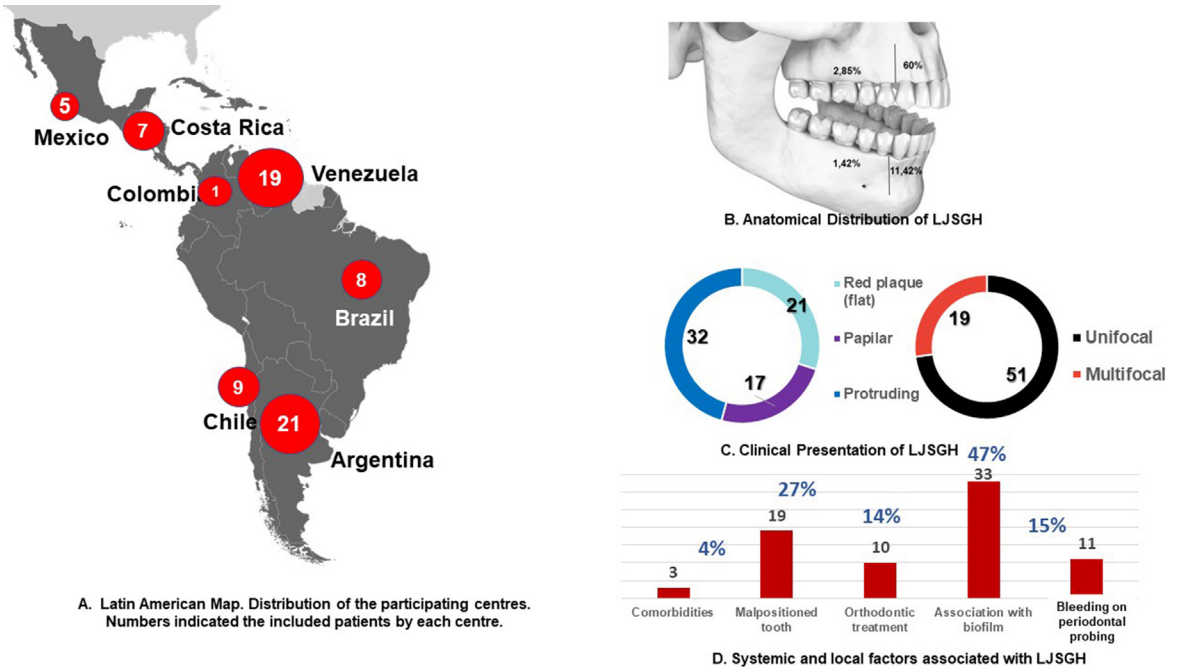

Fig. 1. (**A**) Latin American map. Distribution of the participating Oral Medicine Centres and the origin of the included patients. (**B**) The anatomical distribution of Spongiotic Gingival Hyperplasia. (**C**) Clinical presentation of Spongiotic Gingival Hyperplasia according to the clinical phenotype and uni or multifocal involvement. (**D**) Systemic comorbidities and potential triggering factors associated with Spongiotic Gingival Hyperplasia.

In terms of anatomical location, the anterior maxillary region was the most affected anatomical region (60%, n = 42), followed by the anterior mandibular area (11.4%, n = 8). In 12.8% of cases (n = 9), the lesions involved multiple areas of the jaws, including bimaxillary involvement (Figure 1B and C). All lesions were associated with permanent teeth, with 60% of the cases involving the upper incisors. Additionally, 52% of the patients had mixed dentition (permanent and deciduous teeth), while the remaining 48% had fully permanent dentition.

Systemic comorbidities were observed in only 3 patients (4.2%), including 2 pediatric patients, one with Down syndrome and one with vitiligo, and the oldest patient with SGH who reported hypertension. In 80% of the cases (n = 56), no associated medical conditions were recorded.

Regarding potential local etiological factors, 27.1% of patients (n = 19) had teeth malposition in the affected region, and 14.2% (n = 10) were undergoing orthodontic treatment at the time of the first consultation (Figure 2E and F). The presence of biofilm was detected in 47.1% of cases (n = 33) with a Simplified Oral Hygiene Index of more than 1, while 15.7% (n = 11) presented with bleeding on periodontal probing (Figure 1D). In 41 cases (58.5%) no potential etiological factors (neither local nor systemic) were recorded.

In terms of treatment, surgical excision was the most common therapeutic approach (57.1%, n = 40), followed by periodontal therapy in 21.4% of cases (n = 15). The use of trichloroacetic acid was employed in 8.6% (n = 6) of patients, and spontaneous resolution was observed in 10% of cases (n = 7). Table I shows the clinicopathological and demographic characteristics of the included cases.

Despite 40% missing data regarding lesion follow-up, multifocal SGH cases exhibited a higher recurrence rate compared to unifocal cases (6/10; 60% vs. 4/27; 14%). In some multifocal cases, classification as recurrence was challenging due to the extensive nature of the lesions, with certain areas remaining untreated.

DISCUSSION

To the best of our knowledge, this is the first multicenter study reporting the largest series of SGH cases in the literature, providing a comprehensive analysis of demographic, clinicopathological features, and potential etiological factors associated with its development. Table II summarizes case series published over the past ten years that have included more than 5 cases. While the mean age range (15-97 years) aligns with that reported in the majority of previous studies, the slight predominance of SGH in females observed in this Latin American cohort represents an interesting and

Table I. Demographic, clinical and diagnosis variables

	Total	
	N	%
	70	100
Age		
Mean (range)	15.97 (7-73)	
Median	13	
Sex		
Male	29	41.5
Female	41	58.5
Clinical phenotype		
Flat or micropapilar	21	30
Exophytic-papilar	17	24.28
Protruding	32	45.71
Clinical pattern		
Unifocal	51	72.86
Multifocal	19	27.14
Location		
Anterior maxilar area	42	60
Posterior maxilar area	2	2.85
Anterior mandibular area	8	11.42
Posterior mandibular area	1	1.42
No data	8	11.42
Multifocal with bimaxillar involvement	9	12.85
Comorbidities		
Yes	3	4.25
No	56	80
No Data	11	15.75
Malpositioned tooth		
Yes	19	27.14
No	47	67.15
No data	4	5.71
Orthodontic treatment		
Yes	10	14.28
No	56	80
No data	4	5.71
Association with biofilm		
Yes	33	47.14
No	32	45.71
No data	5	7.14
Bleeding on periodontal probing		
Yes	11	15.71
No	53	75.71
No data	6	8.57
Treatment		
Surgical excision	40	57.14
Trichloroacetic acid	6	8.6
Periodontal therapy	15	21.42
Others	2	2.85
Autorresolution/no treatment	7	10

noteworthy finding. This was also reported in the study by Theofilou et al., which included 18 SGH cases from Greek patients.[6] A relevant finding, which is consistent with previous authors is the existence of cases of SGH in adults, including elderly patients, which supports the hypothesis that it is not exclusively a condition of children or young individuals.[12,13]

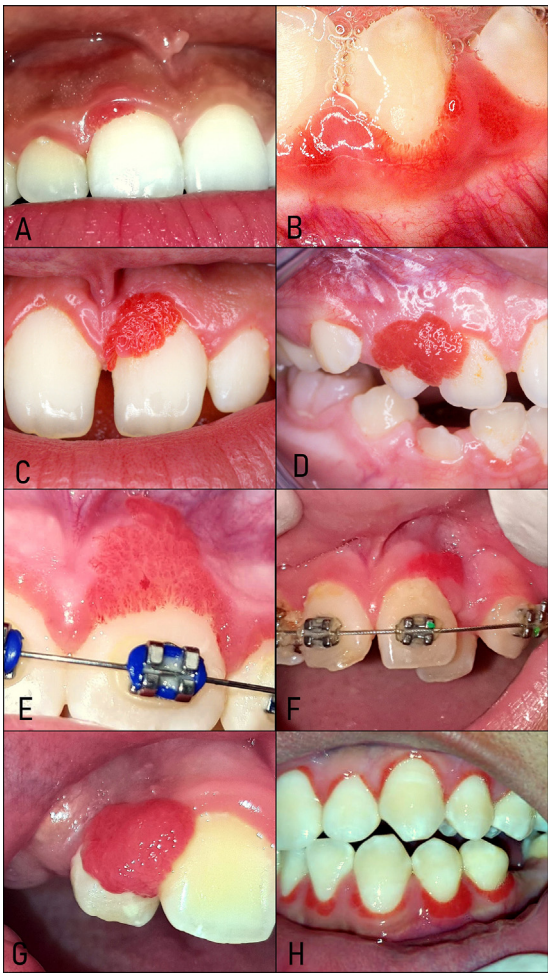

Fig. 2. Different clinical presentations of Spongiotic Gingival Hyperplasia. The clinical features are consistently bright red with a papillary or micropapillary surface. Variability in size and the elementary lesion of this entity could be associated with its stage or degree of evolution: (**A**) A flat, slightly elevated, well-demarcated lesion with a bright red micropapillary surface located on the maxillary central incisor. (**B**) A slightly elevated, flattened lesion emerges from the gingival margin with a micropapillary surface on a mandibular canine. It is noted the presence of biofilm. (**C**) A vestibulo-lingually flattened elevated lesion with an intense red colour and a papillary, lobulated surface on a maxillary central incisor. (**D**) A lesion involving 2 malpositioned maxillary teeth with dental trauma. The lesion is flattened vestibulo-lingually, with an intense red, multilobulated surface. (**E**) A red plaque with a distinctly granular and irregular red surface involving part of the crown and gingiva of a maxillary central incisor. The patient had fixed orthodontic appliances and the lesion was associated with dental biofilm. (**F**) A papillary lesion with gingival margin enlargement affecting the maxillary central incisor and extending to the lateral incisor, which is palatally displaced and in malocclusion with occlusal trauma; the patient is undergoing orthodontic treatment. This case was associated with a visible dental biofilm. (**G**) The protruding phenotype. An exophytic, pedunculated, granular, and papillary lesion involving the maxillary incisors. (**H**) A clinical

The clinical findings observed in SGH in this study are similar to the clinical descriptions published in the current literature.[3,4,6,8,12-14] According to our records, the clinical phenotype described at the time of diagnosis could correspond to the evolutionary stage in which the lesion is found. Theofileu et al. were the first to categorize SGH lesions, considering only the epithelial architecture observed in histopathological findings.[6] The present study aimed to categorize the lesions from a clinical perspective. While this is the first study to categorize the clinical appearance of this entity, this classification and nomenclature should be validated by other groups. We proposed 3 clinical patterns of SGH: a) Flat pattern: slightly elevated area of gingival mucosa with a flat or micropapillary surface, b) Exophytic-papillary pattern: an elevated lesion with a visibly irregular granular surface, which can be manipulated with instruments, and c) Protruding pattern: an exophytic lesion with a base that may be sessile or pedunculated, with a well-defined papillary surface appearance; the latter being the most frequent phenotype (32; 45.7%) (Figure 2). When these exophytic patterns of SGH are intensely red and have a tumoral appearance, differential diagnoses may include pyogenic granulomas and other reactive gingival lesions, as reported in previous studies.[3,8] Although Category III, or protruding may evoke an exophytic neoplastic lesion, we believe it is the most appropriate name. Other alternatives, such as "nodular," could refer strictly from a semiological or clinical perspective to a lesion that is more palpable than visible. We believe that the term “protruding” aligns with the semiological description of these large, papillary, and sometimes exuberant SGH cases. When the lesions are more flattened and multifocal, they become a diagnostic challenge, and the differential diagnosis should include other conditions such as plasma cell gingivitis, chronic desquamative gingivitis, or linear gingivitis.[15,16] In such cases, a biopsy is essential to establish a definitive diagnosis. Interestingly, in some instances where the lesion exhibited vestibulolingual flattening, muscular parafunctions of the lips were observed. The intense pressure exerted by the orbicularis oris muscle against the lesion may produce a flattening effect in this direction, thereby influencing the clinical phenotype. This phenomenon warrants further investigation in future studies.

The anatomical location most frequently observed in the anterior vestibular gingiva of the maxilla aligns with findings reported by other authors; the most commonly affected teeth were the maxillary central and lateral incisors.[3,6,8,12,13] It is noteworthy that this site is by far the most frequent location (42; 60%), followed by the same region in the mandibular gingiva. It is also interesting to highlight that the posterior region is rarely a site for the development of SGH. The reasons for the high frequency of this anatomical localization remain a subject of debate, although they could be directly associated with the origin and nature of this lesion. The preference of this lesion for the anterior region might be attributed to the fact that this area often exhibits a reduction in space for erupting teeth. This condition could favor the persistence of rests of the JE, both in patients with a single lesion and in cases where the involvement is multifocal.[8] Some authors have speculated that given this lesion’s origin is associated with a reactive odontogenic lesion arising from the JE, the teeth involved are typically among the first to erupt in the oral cavity. As a result, they remain in the mouth for a longer period, thus having more time to be exposed to various inflammatory stimuli that could potentially act as triggering factors for SGH.

In addition to the scientific evidence provided by immunohistochemical studies and the absence of reported SGH cases associated with implants, the hypothesis that this lesion originates from the JE is further supported. This is also based on the understanding that the JE formed around implant-supported restorations derives from the oral mucosal epithelium rather than the odontogenic epithelium.[1,4,17] Additionally, Mackenzie et al. demonstrated that epithelium with the phenotypic characteristics of JE could be developmentally exteriorized onto the surface of the marginal gingiva,[18] and this phenomenon has been proposed as another potential cause of SGH.[17]

Furthermore, the high prevalence of SGH in the anterior and maxillary regions suggests that specific anatomical and functional factors may predispose these areas to lesion development. One such factor is the increased length of the JE, which tends to be greater in these regions due to the conical morphology of the teeth and the need for a larger epithelial attachment surface to maintain periodontal health. This anatomical feature may result in a higher number of epithelial cells, providing a more susceptible substrate for inflammatory phenomena triggered by microbial stimuli, as well as for proliferation and growth disturbances. Additionally, these regions are more frequently exposed to functional forces, fixed orthodontic appliances (particularly in younger individuals), and oral

case demonstrating the bimaxillary multifocality of this entity. In the mandibular region, lesions involve the incisors, canines, and premolars, typically presenting as micropapillary lesions affecting the gingival margin, while in the maxilla, the lesions exhibit a more linear configuration, posing a challenge in both diagnosis and treatment.

Table II. Case series ofLocalized Juvenile Spongiotic Gingival Hyperplasia from the last 10 years

Authors, year of publication	*Origin of the included patients*	*N*	*Females*	*Males*	*Age Media and range*	*More frequent clinical description*	*More frequent anatomical location*	*Etiological aspects*
Argyris et al.[3]	USA	21	7	14	13 (8-36)	Well-demarcated, exophytic, erythematous, soft, and hemorrhagic on palpation lesions with granular or slightly papillary surface.	Maxillary gingiva was predominantly involved	One patient was under orthodontic treatment when the lesion appeared and another reported history of previous trauma in the area. One case with mouth breathing.
Allon et al.[4]	USA	10	5	5	11.9 (8-16)	Red velvety papillary-surfaced lesion.	None recorded	None recorded
Vargo et al.[12]	USA	28	13	15	14.5 (3-64)	Granular or pebbly red plaque.	Anterior facial gingiva	None recorded
Wang et al.[13]	USA	27	10	17	13 (7-72)	Solitary red, papillated lesion.	Anterior maxillary gingiva.	None recorded
Lafuente-Ibañez de Mendoza et al.[8]	Spain	10	5	5	13 (9-17)	None recorded	Attached buccal gingiva of the anterior sector (9 in the maxilla and one in the mandible)	None recorded
Innocentini et al.[14]	Brazil	8	4	4	11.7	Red lesions. All lesions were painless and not bleeding	None recorded	One patient associated the lesion with the onset of trauma and another with a previous history of an eruption cyst.
Theofilou et al.[6]	Greek	18	10	8	19.4 (8-57)	Solitary bright red erythematous plaques	Anterior labial maxillary gingiva	None recorded.
Gilligan et al., 2025 (present study)	Latin America	70	41	29	13 (15.97)	Well-demarcated, exophytic, erythematous, lesions with papillary surface. Novel descriptions of 3 clinical phenotypes: a-flat and micropapillary, b-exophytic-papillary c-protruding appearance	Maxillary gingiva. Anterior Vestibular area	27% of cases with malpositioned teeth 14.28% with orthodontic treatment 47.14% associated with dental biofilm. 4.25% with comorbidities 58.5% with no potentially etiological factors (neither local nor systemic)

habits such as mouth breathing. Moreover, the fact that maxillary incisors typically erupt with a slight labial inclination and are subjected to nonaxial masticatory forces may lead to increased exposure of the JE to direct mechanical stimuli.

In this context, some new prosthetic preparation techniques for gingival margin contouring involve intentional invasion of the JE, resulting in an intrasulcular wound, removal of the cemento-enamel junction, and the generation of a new periodontal ligament. Interestingly, there are reported cases where inflammation and revascularization in the gingival sulcus, caused by these prosthetic techniques, appear as a reddish micropapillary surface resembling the early stages of SGH.[19] Figure 2A and B show cases of early SGH with a slight reddish micropapillary surface arising from the gingival sulcus that is very similar to those gingival changes described by the aforementioned procedures. These concepts may support theories identifying the JE as the origin site of proliferative phenomena leading to SGH.

Other characteristics specific to these regions might involve mechanisms of inflammation in the underlying connective tissue due to more active vascularization, differences in the permeability of the JE to bacterial products, and the presence of immune cells such as Langerhans cells (which are present in the sulcular epithelium but not in the JE). One of the proposed mechanisms for SGH pathogenesis involves mechanical injury to the JE, leading to cell damage, inflammation, hyperplasia, and ultimately the clinical presentation of SGH.[3,17] Consistent with the findings of Lafuente et al., external irritative factors such as trauma, dental plaque, or tooth brushing may trigger an inflammatory response and the proliferation of the reddish mass over less resistant epithelial remnants.[8] All these hypotheses should also be explored in future studies.

In our study, more than 50% of the cases were not associated with any identifiable local or systemic factors (e.g., comorbidities, dental biofilm assessed through periodontal indices, orthodontic factors, malpositioned teeth, etc.). Conversely, in nearly 40% of the cases, the lesions were associated with local stimuli, such as orthodontic forces or occlusal stresses resulting from malpositioned teeth. These forces may stimulate the JE which under normal conditions is not adapted to withstand such mechanical stress. These findings suggest that although some cases of SGH may not be linked to identifiable local or systemic factors, a subset of cases supports a multifactorial etiology (e.g., the combined influence of proliferative stimuli such as biofilm accumulation in patients with orthodontic appliances). Moreover, the marked age predilection observed suggests a potential role of pubertal sex hormones in the etiopathogenesis of SGH. However, our study also identified a significant proportion of affected males and adult patients, chronologically distant from puberty, and without comorbidities or endocrine disorders. Previous authors have described no association with endocrine alterations, as evidenced by the absence of progesterone and estrogen receptors, which were confirmed through immunohistochemical assays.[1,3,17]

Regarding treatment, most cases were managed through surgical excision; however, in some instances, lesion reduction, partial improvement, or complete resolution was achieved following periodontal therapy. This outcome has also been reported in some cases published in the literature.[20] In cases of complete involution, the relationship with biofilm or gingival-periodontal alterations must be demonstrated to avoid inconsistencies in the literature regarding its etiology. It is maintained that SGH is not a lesion fully induced by biofilm, although, in this study, a significant percentage was associated with its presence. This supports the recommendation for basic periodontal treatment, focusing on oral hygiene measures, which may later be complemented by surgical removal if necessary. Interestingly, the mechanism under the spontaneous regression of SGH can be supposed as based on the pathogenetic hypothesis of odontogenic developmental lesion. SGH has similar histological features of ectopic JE, localised on the attached gingiva, and putatively exteriorised from the gingival sulcus. The lesion could regress with the passing of developmental age.[4,21] Treatment with trichloroacetic acid has been described in 6 cases with favorable outcomes, although scientific evidence supporting its use is limited.[22] Surgical removal using a scalpel, electrosurgery, or laser not only allows for the collection of a specimen for microscopic analysis but also ensures complete removal, although cases of recurrence have been reported.[23] The use of cryotherapy was described by Nogueira et al. in 2016 and has been proposed for pediatric cases, being much better tolerated than surgery, achieving good results.[24] On the contrary, corticosteroids have been used as an adjuvant treatment in some cases in our series, indicating partial resolution, in agreement with what has been described in the literature.[17] Our data suggest that multifocal SGH cases exhibited a higher propensity for recurrence compared to unifocal presentations. In some instances of multifocal SGH, distinguishing true recurrence from persistence of untreated lesions proved challenging due to the extensive nature of the disease, particularly when certain areas remained unaddressed. It is noteworthy that multifocal lesions often failed to resolve, even following periodontal therapy. The findings of Siamantas et al., which describe bimaxillary multifocal manifestations with variable progression patterns—including complete resolution, partial regression, and persistent areas despite 0.5% topical chlorhexidine application—further highlight

the limited potential for spontaneous resolution. Moreover, treatment decisions in multifocal cases are often influenced by patient (or parental) preferences to avoid simultaneous surgical interventions, favoring instead observation or more conservative management.[5] The study conducted by Nogueira et al. reported recurrence in 2 lesions that had been completely excised and subsequently treated with cryotherapy. However, the authors did not explore potential causes for the recurrence of these lesions.[24]

A limitation of this study is the retrospective data collection, which resulted in incomplete records for a subset of patients. This also precluded the assessment of other potential etiological factors, such as hormonal disorders or mouth breathing, which have been previously implicated in the literature. Moreover, the retrospective design necessitated the use of a periodontal index that may not fully capture the true periodontal condition of the patients at the time of presentation. Despite this, our findings regarding periodontal status are consistent with prior reports. Finally, the lack of comprehensive data on recurrence and long-term follow-up limits our ability to draw definitive conclusions about the comparative efficacy of different treatment modalities.

CONCLUSION

This large Latin American case series corroborates existing reports on SHGs, providing a substantial contribution to the literature owing to its considerable sample size. Our findings reinforce the hypothesis that SGH represents a reactive lesion of odontogenic origin, exclusive of periodontal tissues, and potentially elicited by diverse localized irritants.

Furthermore, the descriptors "localized" and "juvenile" appear to be misnomers and should be re-evaluated. Future research should focus on elucidating the multifactorial etiology of local irritants associated with SGH, as well as investigating the periodontal, microbiological, histological, and immunohistochemical factors implicated in the initial proliferative events of its pathogenesis.

DECLARATIONS OF INTEREST

None.

ACKNOWLEDGMENTS

The authors express their gratitude and acknowledge the kindness, collaborative efforts, scientific rigor, and encouragement for research fostered in recent years by the participating centers involved in this work and by the entire Latin American community within the field of Oral Medicine and Pathology.

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REFERENCES

1. Darling MR, Daley TD, Wilson A, Wysocki GP. Juvenile spongiotic gingivitis. *J Periodontol*. 2007;78:1235-1240. https://doi.org/10.1902/jop.2007.060502.
2. Chang JYF, Kessler HP, Wright JM. Localized juvenile spongiotic gingival hyperplasia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2008;106:411-418. https://doi.org/10.1016/j.tripleo.2008.04.024.
3. Argyris PP, Nelson AC, Papanakou S, Merkourea S, Tosios KI, Koutlas IG. Localized juvenile spongiotic gingival hyperplasia featuring unusual p16INK4A labeling and negative human papillomavirus status by polymerase chain reaction. J Oral *Pathol Med Off Publ Int Assoc Oral Pathol Am Acad Oral Pathol*. 2015;44:37-44. https://doi.org/10.1111/jop.12214.
4. Allon I, Lammert KM, Iwase R, Spears R, Wright JM, Naidu A. Localized juvenile spongiotic gingival hyperplasia possibly originates from the junctional gingival epithelium-an immunohistochemical study. *Histopathology*. 2016;68:549-555. https://doi.org/10.1111/his.12774.
5. Siamantas I, Kalogirou EM, Tosios KI, Fourmousis I, Sklavounou A. Spongiotic gingival hyperplasia synchronously involving multiple sites: case report and review of the literature. *Head Neck Pathol*. 2018;12:517-521. https://doi.org/10.1007/s12105-018-0903-9.
6. Theofilou VI, Pettas E, Georgaki M, Daskalopoulos A, Nikitakis NG. Localized juvenile spongiotic gingival hyperplasia: microscopic variations and proposed change to nomenclature. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2021;131:329-338. https://doi.org/10.1016/j.oooo.2020.10.024.
7. Kalogirou EM, Chatzidimitriou K, Tosios KI, Piperi EP, Sklavounou A. Localized juvenile spongiotic gingival hyperplasia: report of two cases. *J Clin Pediatr Dent*. 2017;41:228-231. https://doi.org/10.17796/1053-4628-41.3.228.
8. Lafuente-Ibáñez de Mendoza I, Alberdi-Navarro J, Marichalar-Mendia X, Mosqueda-Taylor A, Aguirre-Urizar JM. Characterization of juvenile spongiotic gingival hyperplasia as an entity of odontogenic origin. *J Periodontol*. 2019;90:1490-1495. https://doi.org/10.1002/JPER.19-0022.
9. Solomon LW, Trahan WR, Snow JE. Localized juvenile spongiotic gingival hyperplasia: a report of 3 cases. *Pediatr Dent*. 2013;35:360-363.
10. Greene JG, Vermillion JR. The simplified oral hygiene index. *J Am Dent Assoc*. 1964;68:7-13. https://doi.org/10.14219/jada.archive.1964.0034.
11. Methods and indices - Malmö University official Website-Oral Health Country/Area Profile Project. Accessed August 13, 2024. https://capp.mau.se/methods-and-indices/
12. Vargo RJ, Bilodeau EA. Reappraising localized juvenile spongiotic gingival hyperplasia. *J Am Dent Assoc 1939*. 2019;150:147-153.e2. https://doi.org/10.1016/j.adaj.2018.10.001.
13. Wang MZ, Jordan RC. Localized juvenile spongiotic gingival hyperplasia: a report of 27 cases. *J Cutan Pathol*. 2019;46:839-843. https://doi.org/10.1111/cup.13549.
14. Innocentini LMAR, Santos TT, Reis MBL, et al. Juvenile spongiotic gingival hyperplasia: clinicopathological features of eight cases and a literature review. *Oral Health Prev Dent*. 2020;18:953-958. https://doi.org/10.3290/j.ohpd.a45435.
15. Younis RH, Georgaki M, Nikitakis NG. Plasma cell gingivitis and its mimics. *Oral Maxillofac Surg Clin N Am*. 2023;35:261-270. https://doi.org/10.1016/j.coms.2022.10.003.
16. Mortazavi H, Amid R, Moscowchi A, Yousefi-Koma AA, Yousefi-Koma H. Differential diagnosis and treatment protocols for desquamative gingivitis: a systematic review. *Dent Med Probl*. 2024;61:747-758. https://doi.org/10.17219/dmp/156167.
17. Fernandes DT, Wright JM, Lopes SMP, Santos-Silva AR, Vargas PA, Lopes MA. Localized juvenile spongiotic gingival hyperplasia: a report of four cases and literature review. *Clin Adv Periodontics*. 2018;8:17-21. https://doi.org/10.1902/cap.2017.170018.
18. Mackenzie IC, Rittman G, Gao Z, Leigh I, Lane EB. Patterns of cytokeratin expression in human gingival epithelia. *J Periodontal Res*. 1991;26:468-478. https://doi.org/10.1111/j.1600-0765.1991.tb01797.x.
19. Rinaldi T, Santamaría-Laorden A, Orejas Pérez J, Godoy Ruíz L, Serrano Granger C, Gómez Cogolludo P. Periodontal healing with fixed restorations using the biologically oriented preparation technique combined with a full digital workflow: a clinical case report. *Healthc Basel Switz*. 2023;11:1144. https://doi.org/10.3390/healthcare11081144.
20. Caciva R, Belardinelli P, Grenón M. Gingivitis espongiótica. Manejo clínico no quirúrgico. Reporte de un caso. *Rev Asoc Odontológica Argent*. 2024;112:e1120833. https://doi.org/10.52979/raoa.1120833.1246.
21. Decani S, Lodi G, Sardella A, Varoni EM. Localised juvenile spongiotic gingival hyperplasia: a case of spontaneous resolution and a literature review. *Eur J Paediatr Dent*. 2021;22:159-162. https://doi.org/10.23804/ejpd.2021.22.02.14.
22. Moine L, Gilligan G. Trichloroacetic acid for localized juvenile spongiotic gingival hyperplasia: a case report with a novel treatment. *Pediatr Dent J*. 2018;28:47-51. https://doi.org/10.1016/j.pdj.2018.05.003.
23. Goldberg AS, Fatahzadeh M, Bortnik V, Drew H. Novel treatment approach of localized juvenile spongiotic gingival hyperplasia with use of Nd:YAG laser. *Clin Adv Periodontics*. 2023;13:253-257. https://doi.org/10.1002/cap.10241.
24. Nogueira VKC, Fernandes D, Navarro CM, et al. Cryotherapy for localized juvenile spongiotic gingival hyperplasia: preliminary findings on two cases. *Int J Paediatr Dent*. 2017;27:231-235. https://doi.org/10.1111/ipd.12278.