Localised juvenile spongiotic gingival hyperplasia: A case of spontaneous resolution and a literature review



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Abstract

Aim Localised juvenile spongiotic gingival hyperplasia (LJSGH) is a benign lesion occurring in young patients as gingival erythema and overgrowth, typically localised on gingiva of maxillary incisors. The aim of this work is to report a case of LJSGH where complete spontaneous regression was achieved together with a review of the literature on the topic.

Case report An 8-year-old girl was referred for a gingival painless lesion, which had appeared spontaneously one year before and was refractory to periodontal treatment. Intraoral examination showed a well-defined, red gingival overgrowth involving the left maxillary central incisor, without involving the marginal gingiva. The clinical diagnosis of LJSGH was made, due to the pathognomonic aspect. The patient was periodically recalled for 43 months; at the last visit, the lesion was spontaneously resolved.

Results LJSGH is not plaque-related and not responsive to periodontal treatment. Surgical removal of the lesions correlates with high recurrence, while spontaneous resolution over time has been hardly demonstrated.

Conclusion Follow-up of LJSGH may be an option of care, alternative to surgery, in selected cases.

KEYWORDS Oral medicine; Gingival lesion; Childhood.

Introduction

Juvenile spongiotic gingivitis is a benign lesion, described for the first time by Darling et al. [2007]. Since almost all lesions were associated with localised gingival erythema and overgrowth rather than a pure inflammatory lesion, in 2008, Chang et al. recommended to use the term "localised juvenile spongiotic gingival hyperplasia" (LJSGH) to define more accurately this condition [Chang et al., 2008]. The exact incidence is still unknown, and available reports suggest the occurrence mainly in Caucasians during puberty [Allon et al., 2016; Argyris et al., 2015; Chang et al., 2008; Darling et al., 2007; Decani et al., 2013; Flaitz and Longoria, 2010; Grossmann et al., 2014; Nogueira et al., 2017; Petruţiu et al., 2014; Solomon et al., 2013]. LJSGH very rarely has been reported in patients older than 20 years [Argyris et al., 2015; Chang et al., 2008; Darling et al., 2007; Moran et al., 2012]. The female predilection has not been confirmed by all reports [Chang et al., 2008; Darling et al., 2007].

After the original description in 2007, just few cases of LJSGH have been reported and, to the best of our knowledge, the spontaneous healing of LJSGH has never been described in details to date.

Clinically, LJSGH appears as a well-confined erythema on the attached gingiva, typically associated with a gingival overgrowth showing a granular, velvety or pebbly surface [Darling et al., 2007]. LJSGH can have a multiple or single gingival localisation, involving typically the facial gingiva of anterior tooth, mostly of the maxilla, without including the marginal gingiva, although exceptions to this rule are possible [Darling et al., 2007]. Usually asymptomatic, LJSGH is not directly related to dental plaque or calculus deposition, and it can be seldom associated with bleeding during tooth brushing or trauma.

The aetiology of LJSGH is unknown and the pathogenesis is still controversial. The negligible association with dental plaque and calculus does not support the role of bacteria. The age of the patient may suggest an odontogenic developmental aetiology, and Darling et al. [2007] speculated that these lesions could represent an ectopic junctional epithelium (JE) because of histological and immunophenotypic similarities with this tissue, in turn more prone to be in inflamed by negligible irritant factors. Recent immunohistochemical findings further support this theory, since the pattern of cytokeratin (CK) expression in LJSGH (CK1/10, CK4, CK8/18, and CK19) is evocative of JE profile [Allon et al., 2016; Darling et al., 2007]. The role of puberty, instead, is controversial. It is mainly based on its infrequent finding in adulthood, but the absence of estrogen and progesterone receptors in the lesion, its localized feature, the possibility of affecting children in prepubescent age in a consistent number of cases do not agree with a potential effect of sex hormones [Chang et al., 2008].

LJSGH does not resolve after conventional periodontal treatment, and the surgical treatment has a recurrence rate up to 25% within one year [Darling et al., 2007; Solomon et





FIG. 1 Attached gingiva (facial aspect) of the left maxillary central incisor with a well-defined flat erythema, without the involvement of marginal gingiva. FIG. 2The same lesion at 5-month follow-up visit, one month later an accidental trauma occurred.

al., 2013]. Spontaneous resolution has been supposed after an unpredictable time span [Darling et al., 2007], but it has been reported in just one study [Siamantas et al., 2018], specifically the case of multifocal LJSGH treated with surgical therapy, who experienced the recurrence of the lesions after 4 months from intervention; after 15 months, the spontaneous resolution of the lesions (except one) occurred [Siamantas et al., 2018].

Our report aims at describing in details the spontaneous resolution of a case of LJSGH, who did not receive any surgical treatment.

Case report

An 8-year-old female patient came to our clinic, referred by her paediatrician for a painless gingival lesion, which had appeared spontaneously one year before. During the collection of clinical history, the patient reported previous atopic dermatitis and kiwifruit allergy, and the recurrent and mild bleeding of a gingival red lesion, during oral hygiene procedures or accidental traumas. The lesion persisted even after the scaling performed by her dentist. Intraoral examination showed a well-defined, red gingival overgrowth involving the left maxillary central incisor, not including the marginal gingiva (Fig. 1). No plaque or calculus were detected, the periodontal probing depth was normal. On palpation, the lesion was soft and painless. The clinical diagnosis, based on the pathognomonic aspect, was of LJSGH. In agreement with the patient's parents, due to the benign nature of the lesion, we decided to follow-up the lesion and we further instructed the patient on the proper oral hygiene procedures at home, with the recommendation of further dental scaling at her own dentist.

Four months later, an accidental trauma, as reported by patient, produced the partial detachment of the lesion (Fig. 2), which reappeared after further six months (Fig. 3), this time involving also the marginal gingiva. The lesion was again followed-up at 19-month, 31-month, and 43-month visits when the complete spontaneous resolution occurred (Fig. 4).

Discussion and conclusion

So far, only few reports have described cases of LJSGH (Table 1), probably because of its recent identification as clinical self-standing entity and the potential misdiagnosis with plaque-related gingivitis [Darling et al., 2007]. The final diagnosis can be posed, as in our report, taking into account the pathognomonic signs, i.e. red gingival overgrowth not including the marginal gingiva at the anterior maxillary incisive, and the lack of improvement after periodontal treatment [Darling et al., 2007]. In those cases showing the involvement of the marginal gingiva, further clinical signs are important for the final diagnosis, such as the aspect of bright red patch with papillary, granular, pebbly, or velvety surfaces; biopsy will be useful to discriminate doubtful cases. Differential diagnosis includes puberty gingivitis [Murakami et al., 2018], which is the most common provisional hypothesis made by the clinician because of the gingival erythema suggestive of a local inflammatory process. However, LJSGH differs considerably from that condition because of the unresponsiveness to oral hygiene procedures, and the absence of a plaque-related pathogenesis. LJSGH can be also misdiagnosed, particularly in presence of gingival overgrowth, with pyogenic granuloma, peripheral giant cell granuloma, human papilloma virus (HPV)-related lesions, foreign body granuloma, small superficial lymphangioma. In presence of these clinical suspicions, biopsy is recommended. Histopathological features of LJSGH include loss of keratinisation of the stratified squamous epithelium,





FIG. 3 At 10-month follow-up visit, the lesion reappeared, this time involving marginal gingiva.

FIG. 4 The condition resolved spontaneously at 43-month follow-up visit, without any treatment.

References	Cases of LJSH	Age (years)	Treatment	Resolution	Recurrence
Darling et al., 2007	24 (12 m, 12 f)	5-28 (mean: 12)	- Surgical (n=23) - Follow-up (n=1)	- Yes (n=23) - No (n=1) (recorded as"recent case")	- Yes, after 2 months n=4 (in one case, recurred lesion was stable at 2 yrs) - No n=10 (follow-up 6 months-18 yrs) - Recent cases n=2 - NR n=7 - Recorded as "recent case" n=1
Chang et al., 2008	52 (36 m, 16 f)	7-39 (1 older than 20)	NR	NR	3 cases recorded as "recurrent"
Flaitz and Longoria, 2010	1 (m)	11	Chlorhexidine	Partial (after 14 days of chlorhexidine treatment)	NR
MacNeill et al., 2011	1 (m)	14	Cauterization (CO_2 laser) + clobetasol	Yes	No (follow-up: 6 months)
Moran et al., 2012	81	adults	NR	NR	NR
Solomon et al., 2013	3 (2 m; 1 f)	15, 11, 9	- Surgical (n=2) - NR (n=1)	- Yes (n=2) - NR (n=1)	- Yes, 4-6 months (n=2) - NR (n=1)
Decani et al., 2013	1 (f)	11	Follow-up	NR	NR
Grossmann et al., 2014	1 (femae)	6	Chlorhexidine	Partial (after 14 days of treatment)	NR
Petruţiu et al., 2014	3 (1 m, 2 f)	13, 14, 15	Surgical (n=3)	NR (n=3)	NR (n=3)
Argyris et al., 2015	21 (14 m, 7 f)	8-36 (mean: 13)	- Surgical, scalpel (n=18) - Surgical, laser (n=3)	Yes (n=18) Yes (n=3)	- Yes, after 20-21 months (n=2) - No (n=16) (follow-up: 2-55 months) - No (n=3) (follow-up: 8-18 months)
Allon et al., 2016	10 (5 m, 5 f)	9-16	NR	NR	NR
Kamperos et al., 2016	1 (f)	22	Surgical	Yes	No (follow-up: 3 months)
Nogueira et al., 2017	2 (f)	11	- Surgical (n=2) - Cryotherapy (n=2) (after recurrence)	- Yes (n=2) - Yes (n=2)	- Yes, after 20-30 days (n=2) - No (n=2) (follow-up: 5-6 months)
Kalogirou et al., 2017	2 (m)	12	Surgical (n=2)	Yes (n=2)	No (follow-up: 17 months)
Siamantas et al., 2018	1 (m) with multiple localization	19	Surgical	Yes	Partial to complete recurrence after 4 months Spontaneous regression of all but one lesion after 15 months
Vieira et al., 2019	1 (m)	9	Photodynamic therapy	Partial	NR
Wang 2019	27 (17 m, 10 f)	7-72 (mean: 13)	NR	NR	NR
Vargo 2019	28 (15m, 13f)	3-64 (mean 14.5)	Surgical	Yes	n=3: after 17 months, after 45 months, 10.5 years. No: n=25
Innocentini 2020	8 (4m, 4f)	6-24 (mean 11.6)	Treatment: surgical (n=3)/follow-up+biofilm control (n=5)	Unclear	

TABLE 1 Cases of localized juvenile spongiotic gingival hyperplasia (LJSGH) and related treatment, resolution and recurrence (NR – not reported).

which shows epithelial hyperplasia with a papillary architecture and spongiosis, prominent intercellular oedema and neutrophil exocytosis [Allon et al., 2016]. The connective tissue underneath the elongated papillae has signs of acute and chronic inflammation [Allon et al., 2016]. The papillary architecture, in particular, may be suggestive of the role of HPV, but LJSGH fails to show the typical histopathological features observable in HPV-related lesions [Argyris et al., 2015].

No consensus on LJSGH management is available, to date. Periodontal therapy has been advocated as first-line treatment, useful for excluding the role of plaque and calculus deposition and the diagnosis of puberty gingivitis. In two cases, the local application of chlorhexidine 0.12% three times a day produced, after 14 days, the partial clinical regression of multifocal lesions [Flaitz and Longoria, 2010; Grossmann et al., 2014]. Surgical treatment using a scalpel was the most commonly proposed approach, since it was often associated with excisional biopsy for histopathological confirmation of the lesion.

Recurrence rate, however, is high and should be interpreted carefully, since studies often considered a too short follow-up or did not explicit this outcome. Overall, the literature shows a rate of recurrence ranging from 6% to 25% [Argyris et al., 2015; Chang et al., 2008; Darling et al., 2007]. Solomon and co-workers [2013] described the LJSGH relapse within few months from surgical removal of the treated cases, similarly to Nogueira et al. who found recurring lesions after surgery, then successfully treated using cryotherapy with a still complete resolution at the 6-month follow-up [Nogueira et al., 2017]. Laser surgery has been applied in three cases, showing successful lesion removal, which was stable up to 8–18 months after treatment [Argyris et al., 2015]. A further report described the conservative treatment of this condition via laser CO_2 cauterisation in combination with the application of 0.05% clobetasol ointment for 4 weeks; the lesion entirely healed and no evidence of recurrence was recorded at 6 months-follow up [MacNeill et al., 2011]. Recently, photodynamic therapy has been also proposed, but evidence is limited to one case report showing

the partial resolution of the lesion [Vieira et al., 2018].

Although no prospective study has ever been carried out to confirm the hypothesis, it has been suggested that many cases of LJSGH undergo spontaneous resolution [Darling et al., 2007], as observed in our report about three years from the first visit. Darling et al. [2007] stated in their paper: "Discussion of this condition with a broad range of dental colleagues, including general practitioners and specialists, suggests that the condition is more common than our files would indicate and that most cases resolve spontaneously after a variable and unpredictable time period. This would be consistent with the rare occurrence in adults compared to the more common occurrence in juveniles". The literature, however, described spontaneous resolution of LJSGH in just one report, describing the surgical therapy of multifocal LJSGH lesions in a patient, who recurred after 4 months and spontaneously healed after 15 months [Siamantas et al., 2018]. Three studies reported a follow-up of the lesions after periodontal care and oral hygiene instruction, without surgical intervention [Darling et al., 2007; Decani et al., 2013; Grossmann et al., 2014]: in two cases, the patient follow-up was not clearly specified and resolution not stated [Darling et al., 2007; Decani et al., 2013], while in one case chlorhexidine was also prescribed for 14 days resulting in a partial clinical response of multifocal lesions [Grossmann et al., 2014].

The mechanism under the spontaneous regression of LJSGH can be supposed as based on the pathogenetic hypothesis of odontogenic developmental lesion. LJSGH has similar histological features of ectopic JE, localised on the attached gingiva, putatively exteriorised from the gingival sulcus [Allon et al., 2016; Darling et al., 2007]. The lesion could regress with the passing of the developmental age.

This report demonstrates the complete spontaneous resolution of a case of painless LJSGH and suggests the follow-up of those asymptomatic lesions, having the pathognomonic features of LJSGH, and for which the biopsy is not required for diagnostic confirmation, as an option of care alternative to surgical treatment, which is often prone to relapse. In case of doubts, the clinician should ask oral medicine specialist for consultation and, in presence of not-pathognomonic clinical aspect, perform the diagnostic assessment by incisional biopsy. Careful explanations of LJSGH benignity, pathogenesis and possible alternatives of care are always recommended for an informed consent. In case of the "wait-and-see" approach, the recall visit should always carefully evaluate the periodontal health and encourage a high level of oral hygiene at home, since bleeding during tooth brushing might hinder the correct cleaning of the area. The patient and his parents should be aware that complete regression may require a long time, and, in those cases where pain, bleeding and aesthetics are a concern, the "wait-and-see" approach is not advisable. They should be also informed that, even with significant spontaneous resolution of the lesion, recurrence may occur and that it is still unknown wether the recurrence rate is greater or lesser than with surgical intervention. Larger clinical studies are needed to determine how frequently are and how long the lesions take to resolve, and how frequently they relapse.

This non-invasive approach might result more acceptable for paediatric patients and their parents than surgery, unless the latteris specifically required for symptoms, anxiety or aesthetic concerns.

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Conflicts of Interest

The authors have no conflicts of interest relevant to this article to disclose.

Author contributions

SD examined and treated the patient, SD and EV conceived the idea for the case report. EV led the case report writing, with editing and contributions by SD, AS and GL.

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