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Periodontology

Part 2: Localised Juvenile Spongiotic Gingival Hyperplasia: histopathological and clinical features

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Abstract

Localised juvenile spongiotic gingival hyperplasia (LJSGH) is a poorly understood but distinctive inflammatory hyperplasia. It is an exophytic lesion lined with non keratinised hyperplastic stratified squamous epithelium characterised by acanthosis, papillomatosis and spongiosis. Despite the name, it is not limited to juveniles but it can affect also adults. Its aetiology is unknown and the pathogenesis is controversial. Histological and immunophenotypic similarities suggest that LJSGH may result from a less keratinised junctional or sulcular epithelium. The excisional biopsy was considered the treatment of choice for a very long time. However, new evidence has suggested the efficacy of a conservative approach as it was seen that spontaneous remission of the lesion was possible. Even if LJSGH is not a plaque-induced lesion, the wait-and-see approach involves recall visits to evaluate the periodontal health and to reinforce oral hygiene instructions.

KEYWORDS Gingival diseases; Oral pathology; Juvenile hyperplasia; Spongiotic; Exophytic lesion.

Introduction

The term Localised Juvenile Spongiotic Gingival Hyperplasia (LJSGH) was coined in 2008 by Chang et al. [2008] to review the clinical and histological features of "Juvenile Spongiotic Gingivitis" originally described by Darling et al., in 2007 [2007].

Clinically, LJSGH is a well-circumscribed exophytic painless lesion with micropapillary or granular surface of a bright red colour (Fig. 1). LJSGH seems to be a non-plaque related lesion: in some cases it develops on the attached gingiva and is separated from the marginal gingiva by healthy tissue, where biofilm deposits are absent. In addition, most lesions do not respond favourably to periodontal treatment and improvement in oral hygiene [Chang, Kessler et al., 2008; MacNeill, Rokos et al., 2011; Kalogirou, Chatzidimitriou et al., 2017].

In most cases LJSGH appears as a solitary gingival mass (0.2-1 cm in diameter) located on the labial maxillary marginal gingiva. Posterior maxillary, palatal and mandibular localisations are less frequent [Silveira, Toral-Rizo et al., 2021]. A multifocal presence is rare but possible (Fig. 2), therefore the term "localised" is not always justified [Solomon et al., 2013; de Freitas Silva et al., 2015; Nogueira et al., 2017]. The lesion is usually non haemorrhagic upon palpation and probing (Fig. 3), but may bleed during teeth brushing [Kalogirou et al., 2017]. The mean age of the reported cases is 12 years [Deseta et al.,2020] with a wide age range from 5 to 78 years [Darling et al., 2007; Siamantas et al., 2018; Silveira et al., 2021], usually affecting patients during the first two decades of life [Chang et al., 2008; Siamantas et al., 2018; DeSeta et al., 2020]. Due to case reports describing multiple LJSGH-like lesions and to the wide age range of presentation, Solomon et al. [2013] suggested an alternative definition for such lesions, i.e. "spongiotic gingival hyperplasia".

The aim of this work is to summarise the histopathological and clinical features of LJSGH for improving its diagnostic and treatment approach to this condition.

Analysis of histopathological and clinical features of LJSGH

A 7-year observational study conducted in an oral pathology division showed that among all the biopsies accessed 0.069% were diagnosed as LJSGH [Argyris et al., 2015]. LJSGH occurs more frequently in Caucasians, and to date gender predominance is unclear [Chang et al., 2008; Argyris et al., 2015].

Histopathologically, LJSGH is an exophytic lesion that is lined with non-keratinised hyperplastic stratified squamous epithelium showing papillomatosis. It is characterised by acanthosis, interconnecting rete pegs and spongiosis of the spinus layer with prominent intercellular oedema and neutrophilic infiltration [Allon et al., 2016]. In the underlying connective tissue, beneath the elongated papillae, there is evidence of acute and chronic inflammation with inflammatory cells exocytosis, vasodilatation and congestion of the superficial capillary network (Fig. 4C). This lesion is not accompanied by underlying bone resorption [Kalogirou et al., 2017].

The aetiology of LJSGH is unknown and its pathogenesis is controversial [Decani et al., 2021]: some evidence suggest that it may result from a less keratinised junctional or sulcular epithelium which is more susceptible to local irritants and minor local trauma [McNamara and Kalmar 2019]. Trauma, orthodontic treatment and mouth breathing have been described as coincidental events [Chang et al., 2008; Kalogirou et al., 2017].

FIG. 1 Bright red, exophytic, granular surface in the area of tooth 1.2 in a 9-year-old male. This lesion was diagnosed as LJSGH.

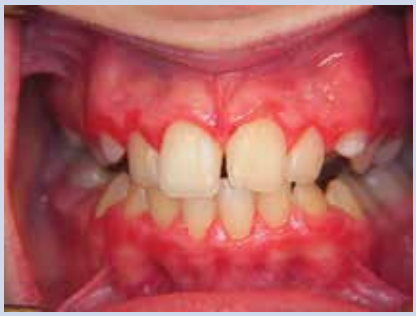

FIG. 2 Multiple lesions diagnosed as LJSGH in a 10-year-old male.

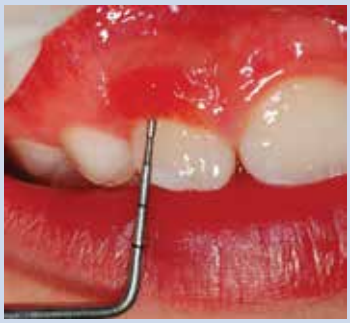

FIG. 3 LJSGH around tooth 1.2 in a 9-year-old male. No bleeding on probing.

Histological and immunophenotypic similarities between LJSGH and the junctional epithelium (JE) have been documented. The pattern of cytokeratin expression in LJSGH (CK1/10, CK4, CK8/18, and CK19) is evocative of a JE profile [Darling et al., 2007; Allon et al., 2016; Decani et al., 2021]. Immunohistochemistry for citokeratins CK19 and CK18 was performed in some studies showing an intense CK19 cytoplasmic expression and a mild CK18 expression in all cell layers whereas the adjacent normal gingival epithelium was CK19+ uniquely in the basal cell layer [Kalogirou et al., 2017]. Moreover, JE specifically expresses CK19 and CK14 proteins in all epithelial layers, in absence of CK7, as occurs in the LJSGH hyperplasic epithelium [Lafuente-Ibanez de Mendoza et al., 2019]. Based on these results, LJSGH may originate from strands of the primary teeth's JE that remain in the gingiva during the eruption of the permanent successor; external irritative factors would then trigger the inflammatory response and provoke the growth of LJSGH lesions. LJSGH frequent localisation in the anterior maxilla might be due to the common lack of space for the permanent teeth that could favour the maintenance of JE strands [Lafuente-Ibanez de Mendoza et al., 2019]. Moreover, it is well known that the anterior part of the maxilla is the most subject to local trauma.

An infective aetiology suggested by epithelial hyperplasia was not confirmed by virus testing for HPV (Human Papillomavirus), HHV (Human Herpesvirus) and Polyomavirus [Mora-Gonzalez, 2014; Argyris, et al., 2015].

The differential diagnosis is limited due to the distinctive features of LJSGH [Burlini et al., 2013; Tadini et al., 2015]. Clinicians may misdiagnose this lesion as puberty gingivitis, foreign body gingivitis, pyogenic granuloma or squamous papilloma [Chang et al., 2008; Kalogirou et al., 2017; Cargini P. et al., 2012]. However, histopathological findings of LJSGH are distinctive and should exclude other clinical diagnosis. The absence of estrogenic and progesterone receptors in the LJSGH lesions, its recurrent localised feature and the frequent diagnosis during childhood do not support a potential effect of sexual hormones [Chang, Kessler et al., 2008; Garracho-Rangel JA. et al., 2018]. Pyogenic granuloma is distinguished from LJSGH by a highly vascular proliferation and haemorrhagic nature, epithelial atrophy and surface ulceration. The absence of parakeratosis in LJSGH excludes the possible diagnosis of papilloma.

Excisional biopsy of the lesions is the current treatment of choice, as LJSGH does not respond to periodontal treatments and oral hygiene improvement [Siamantas et al., 2018], and is justified by the need of a histopathological confirmation of the lesion (Fig. 4). Cryotherapy was described as an efficient alternative treatment for multiple lesions [Nogueira et al., 2017]. The efficacy of topical steroid therapy is not supported by analytic studies: it was ineffective in one case [de Freitas Silva et al., 2015], in another case associated to a transient clinical improvement [Fernandes et al., 2017]. The recurrence of multifocal lesions was greater than for isolated LJSGH (38.5% VS 6%–28.6%) [Darling et al., 2007; Chang et al., 2008; Argyris et al., 2015; Siamantas et al., 2018] and may occur from 2 months [Darling et al., 2007] to 5 years after initial excision [Chang et al., 2008]. Risk factors for recurrence have not been identified [Solomon et al., 2013].

However, a surgical therapy is not always indicated, especially

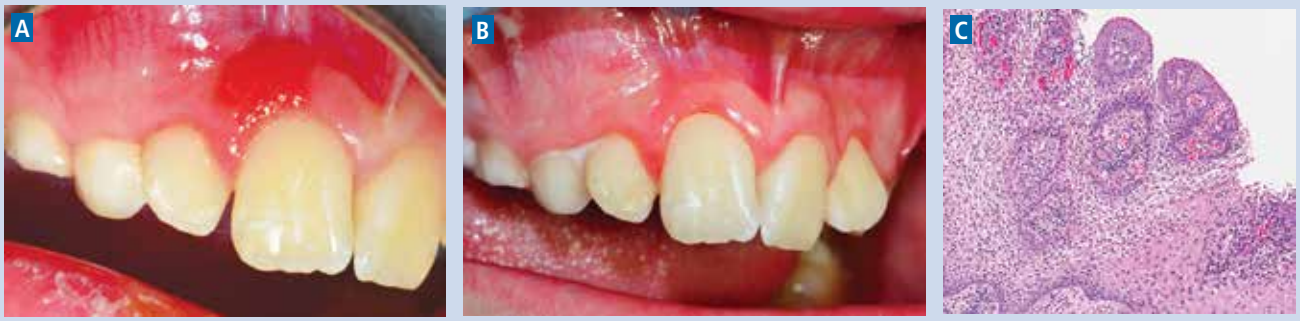

FIG. 4 LJSGH lesion around tooth 1.1 in a 9-year-old male
A: Pre-surgical photograph showing the buccal attached gingiva of the right maxillary central incisor with a well-defined flat LJSGH lesion.
B: Four-weeks post-surgical healing of the attached gingiva of the right maxillary central incisor after excisional treatment of LJSGH lesion. The whole lesion was excised; the gingiva appears slightly inflamed at the interdental papilla between teeth 1.1 and 1.2.
C: Photomicrograph of the LJSGH lesion showing a papillary non-keratinised surface, spongiosis, epithelial hyperplasia, interconnecting rete pegs and dilated and congested vessels, as well as chronic inflammatory infiltrate.

if performed in aesthetic areas, due to the risk of gingival recessions and when dealing with young children [Arcuri et al., 2020]. In fact, spontaneous resolution has been reported [Nasim et al., 2014; Ozkan et al., 2014]. The duration of the lesions reported show a very wide resolution time, ranging from three weeks to a few years [Vargo and Bilodeau 2019]. In 2007 Darling et al. [2007] hypothesised a possible spontaneous resolution of LJSGH yet in the following years few cases have been documented. Decani et al., [2021] reported a case of spontaneous resolution in an 8-year-old female after 43 months, and Siamantas et al., [2018] after 15 months in a 19 year-old woman. Deseta et al., [2020] documented 10 cases that were successfully managed with hygiene recalls and intensive oral hygiene instructions. Use of sodium-lauryl-sulphate-free and age-appropriate fluoridated toothpaste were recommended to all patients; the percentage of spontaneous resolution was 60% over a follow-up period ranging from 13 months to 5 years and 11 months. Similarly, a case series [Innocentini et al., 2020] documented a mild improvement of 5 cases following clinical observation and plaque control after a mean follow-up period of 11 months [Mummolo et al., 2019].

Spontaneous resolution of LJSGH could be attributed to the elimination of a possible, yet unknown, causative factor [Siamantas et al., 2018] and this could justify the wait-and-see approach consisting of recall visits to evaluate periodontal health and to reinforce appropriate oral hygiene habits.

Conclusion

Larger clinical studies are needed to determine frequency, mean time of spontaneous resolution and recurrence frequency. The wait-and-see approach might result more acceptable for paediatric patients and their parents than surgical excision considering the benign nature of LJSGH and the possibility of a spontaneous resolution.

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