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Original Article

Localized Juvenile Spongiotic Gingival Hyperplasia: A Case Report

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Abstract

Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a recently discovered oral lesion in children and young adults, which can be misdiagnosed with gingivitis associated with dental plaque and other well recognized reactive lesions. Clinically, JSGH presents as bright red gingival overgrowth with papillary surface, most commonly at attached and marginal gingivae of the upper anterior region. Differential diagnoses may include pyogenic granuloma, peripheral giant cell granuloma, peripheral ossifying fibroma and hyperplastic gingival inflammation. Histologically distinctive, JSGH is characterized by non-keratinizing stratified squamous epithelium with papillary architecture, intense neutrophilic exocytosis and spongiosis. The objectives of the present case report were to report a localized juvenile spongiotic gingival hyperplasia case, describe its clinical manifestations, histological features and treatment protocol, and emphasize clinical awareness of the occurrence of this uncommon oral mucosal lesion among dental general practitioners.

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Key words: *(in alphabetical order): Benign tumor; Gingival overgrowth; Gingival swelling; Gingivitis; Localized juvenile spongiotic gingival hyperplasia; Pyogenic granuloma; Reactive lesion*

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Introduction

Localized juvenile spongiotic gingival hyperplasia (LJSGH) is recently discovered as an oral lesion in young adult, which can be misdiagnosed with a simple reactive gingivitis. Following its first clinical recognition as 'juvenile spongiotic gingivitis' in 2007 by Darling et al (Darling et al., 2007), this uncommon oral lesion, however, remains poorly recognized in clinical observation among clinicians. In 2008, Chang et al proposed an alternative term for this condition namely 'localized juvenile spongiotic gingival hyperplasia' (LJSGH), following its commonly observed characteristics of gingival overgrowth, rather than a pure inflammatory feature (Chang et al., 2008). Chang et al also reported based on a total number of 52 cases, that epidemiological aspects of LJSGH reveal a male-to-female ratio of 1:2.3 and age range between 7 and 39 years, with the majority of cases being in the second decade (mean 11.8 years, median 12 years). The race distribution appeared to be 82% Caucasian, 14% Hispanic and 4% Asian (Chang et al., 2008).

Clinical features of JSGH can be found as bright red gingival hyperplasia, easily bleeding with papillary surface texture. Both localized and generalized can be seen. Most common location occurred locally at attached gingiva of upper anterior region, and some presented with gingival margin involvement. Occasionally, labial marginal gingiva of the anterior lower arch and attached gingiva adjacent to root apex were also reported (Chang et al., 2008). Histologically, JSGH is characterized by non-keratinizing stratified squamous epithelium with papillary architecture, intense neutrophilic exocytosis and spongiosis (Darling et al., 2007).

The objectives of the present case report were to report a localized juvenile spongiotic gingival hyperplasia case, describe its clinical manifestations, histological features and treatment protocol, and discuss the most common clinical differential diagnoses. This helps emphasize the clinical awareness of the occurrence of such uncommon intra-oral mucosal lesion among dental general practitioners.

Case Report

A 12 years old Thai boy showed the symptom of gingival swelling. From interviewing, the exact duration of that swelling could not be recorded, but it appeared to be recently increased in size. The lesion was once appeared at this area about a year ago. It had been complete excised without histopathologic examination. The lesion seemed to be asymptomatic. The clinical presentation was easily bleeding gingival overgrowth. Past medical history showed no history of systemic diseases and drug therapy. The clinical examination of head and neck soft tissues indicated that it was within normal limits. Intraoral clinical examination, as shown in Fig. 1A and 1B, showed a solitary bright red exophytic and papillary lesion with granular surface and soft consistency. The lesion was located on both labial (approximately 15-mm diameter) and lingual (8 mm-diameter) interdental papilla regions between teeth 42 and 43. Calculus occurs on the buccal and lingual surface of the 6 lower anterior teeth. The teeth were negative to percussion test with no tenderness of surrounding labial and lingual vestibular area.

In Fig. 1C, a periapical radiograph of teeth 42-44 revealed intact tooth structure, no bony destruction with intact lamina dura, normal trabeculae pattern and normal PDL space. Clinical diagnoses included pyogenic granuloma, peripheral giant cell granuloma, peripheral ossifying fibroma and hyperplastic gingival inflammation. According to its non-aggressive behavior, pedunculated characteristic, location, accessibility of the lesion and possible curability by local excision, excisional biopsy was recommended (Marx and Stern, 2012) and therefore performed under local anesthesia in this patient. Surgical excision of the lesion was accomplished with scalpel excision. The teeth were root planed to remove calculus. Immediate postoperative active bleeding could successfully be controlled by direct pressure application.

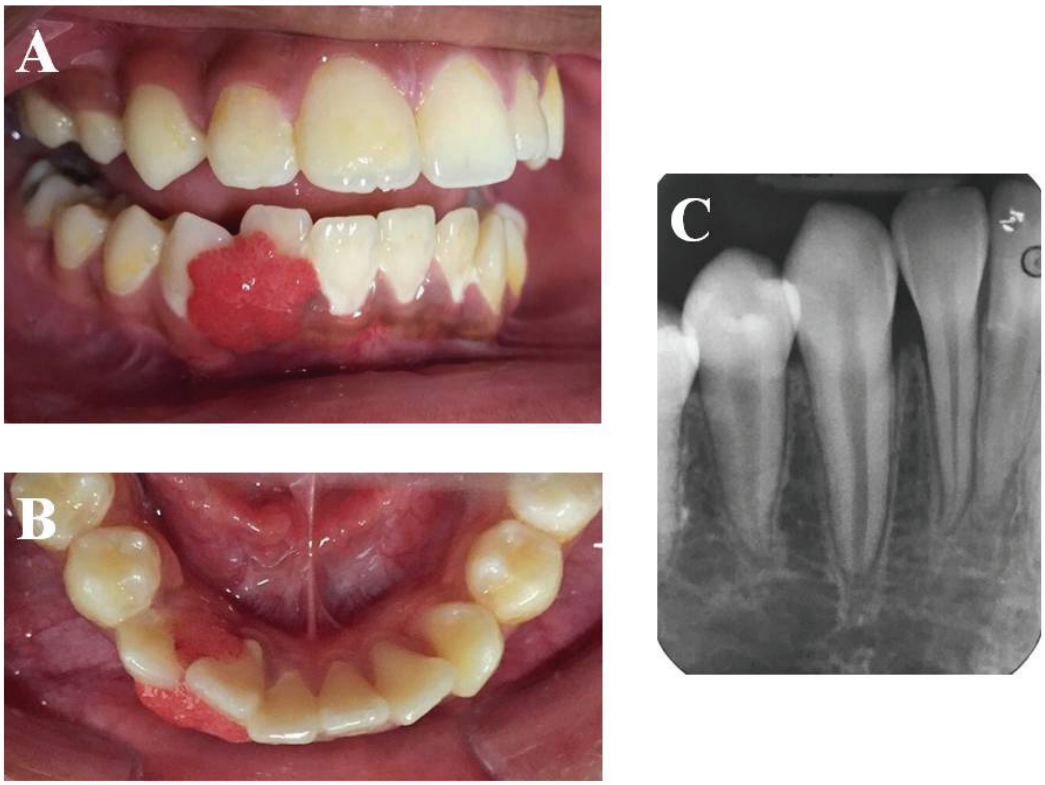

Fig. 1 Pre-operative findings. A: Intra-oral feature (frontal view), B: Intra-oral feature (occlusal view), C: Periapical radiograph

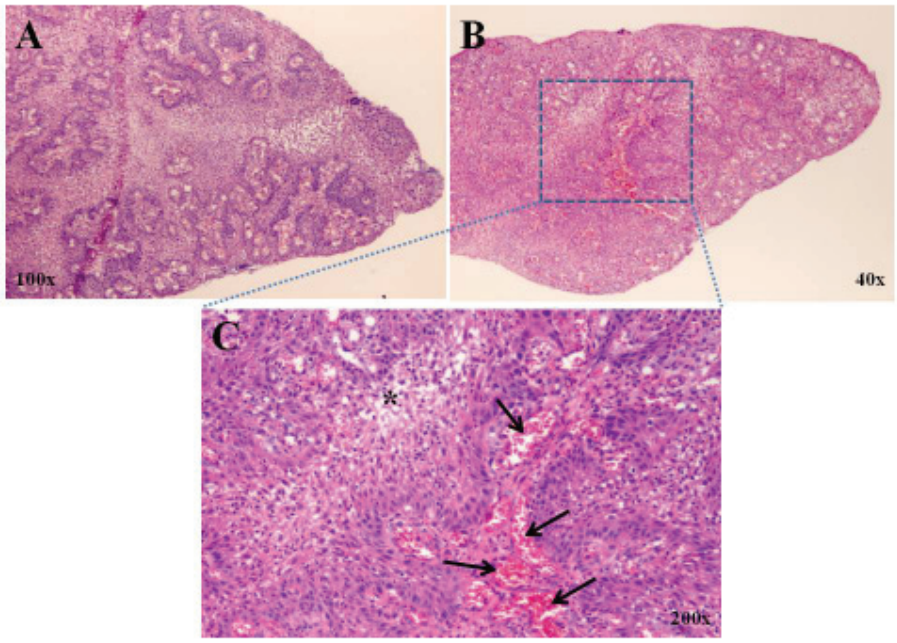

Fig. 2 Histopathology photographs

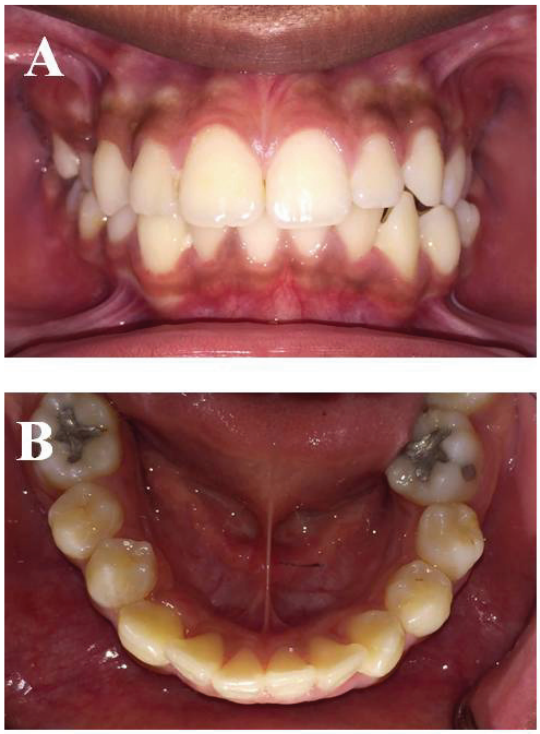

Fig. 3 Eight month post-operative findings. A: Intra-oral feature (frontal view), B: Intra-oral feature (occlusal view)

In Fig. 2, histopathologic examination of the biopsy specimen revealed an exophytic soft tissue mass with lobulated architecture (Fig. 2A and 2B). It was surfaced by non-keratinized stratified squamous epithelium. The surface epithelium showed severe intercellular edema (asterisk in Fig. 2C) with hyperplastic and interconnected rete ridges proliferating into underlying connective tissue stroma (Fig. 2A and 2B). Exocytosis of neutrophils was present. Intervening stroma contained multiple congested blood vessels (arrows in Fig. 2C) and scattered acute and chronic inflammatory cells. The histopathological diagnosis was localized juvenile spongiotic gingival hyperplasia.

After surgical excision of the lesion, the lesion had resolved. At the 8 months post-treatment appointment, there was total resolution of the lesion appearance with only a mild inflammation of the gingival margins that appeared to be associated with dental plaque (Fig. 3). No calculus deposition was observed on all of lower anterior teeth. Periodical follow up was advised due to its previously reported recurrence (Chang et al., 2008; Darling et al., 2007).

Discussion

Firstly reported in 2007 by Darling et al., 24 patients with uncommon histopathological characteristics of an oral mucosal lesion, namely juvenile spongiotic gingivitis, were introduced to the clinical world. To the best of our knowledge, this is the first published case report of LJSGH in Thailand. The present reported case possessed some features which are different from those previously published (Chang et al., 2008; Darling et al., 2007; Decani et al., 2013; Noonan et al., 2013; Solomon et al., 2013). We reported here that the lesion involved interdental papillae, unlike the previously reported location on the attached gingiva and marginal gingiva. Moreover, the gingival mucosa of the anterior maxilla has been reported to be the most common clinical finding location whereas the affected area of the patient

presented in this report involved the mandibular buccal gingiva, which is less common compared with the previously reported LJSGH. While the size of such lesion can vary between 2 mm and 10 mm with the average of 6 mm in diameter (Chang et al., 2008), the patient in this case report possessed a LJSGH sized approximately 15 mm in diameter. To the best of our knowledge, this is thus far the largest size of LJSGH reported in the literature. However, it is not yet known whether the size of the lesion could be of clinical importance. Understanding the pathogenesis and progression of the lesion may be beneficial for preventing and managing this condition.

Regarding its clinical characteristics, clinicians might be perplexed by other commonly observed reactive gingival conditions such as pyogenic granuloma and drug-induced gingival overgrowth. In order to successfully provide proper differential diagnosis, clinicians must also consider other related lesion existences. Intrinsic and extrinsic factors, such as systemic diseases, current use of medications and localized dental plaque, also result in overgrowth of gingival tissue (Agrawal, 2015; Doufexi et al., 2005) The patient described in this case report has been ruled out for systemic diseases and medication taking. Moreover, the patient's intra-oral presentation did not correspond to clinical behaviors of both benign and malignant tumors, rather more consistent with a reactive condition. It is thus categorized this discovered lesion in a reactive lesion group due to its slow and painless growth of gingiva with adequate plaque control (Rossmann, 2011). Therefore, possible differential diagnoses may include pyogenic granuloma, peripheral giant cell granuloma, peripheral ossifying fibroma, plaque-associated inflammatory gingival enlargement, LJSGH. The definitive diagnosis must be based on the clinical and pathological correlations of the lesion.

In the current patient, the lesion was a recurrent lesion of a lesion previously excised by a previous dentist without tissue biopsy. Biopsy is important to establish a final diagnosis, as well as to determine the nature and characteristics of the lesion (Jain, 2011). Therefore, all specimens obtained from biopsy should be sent for a histopathological examination.

Recurrence of LJSGH has previously been reported (16.7% in Darling et al.'s study and 6% in Chang et. al.'s study) (Chang et al., 2008; Darling et al., 2007). Limited responsiveness to conventional periodontal treatment and routine oral hygiene maintenance was also reported (Chang et al., 2008; Darling et al., 2007). Therefore, periodical recall is recommended. It is not yet known regarding the etiology and pathogenesis of LJSGH. However, it has been suggested that LJSGH may originate from the junctional epithelium of gingiva (Allon et al., 2016). It is also possible that LJSGH may be associated with human papillomavirus (HPV) infection since the low levels of human papillomavirus (HPV) DNA could be detected in certain LJSGH samples (Argyris et al., 2015). Further studies are undoubtedly required to investigate its precise etiology and pathogenesis.

Although clinical characteristics of LJSGH and pyogenic granuloma seem similar, LJSGH is histopathologically distinguishable from pyogenic granuloma. LJSGH is commonly an epithelial hyperplasia, while pyogenic granuloma is rather a fibrous and vascular hyperplasia. Unlike pyogenic granuloma, unique characteristics of LJSGH show subtle papillary architecture with exophytic epithelial hyperplasia with spongiosis and inflammatory exocytosis, which may suggest differences in etiology, pathogenesis and development of these lesions.

Treatment options for LJSGH include surgical excision and regularly scheduled follow-up for spontaneous remission. In the present case, the lesion was treated by surgical excision, which may produce gingival margin defect. However, no gingival defect was observed in the patient in this case report. Simon et al (2011) demonstrated that LJSGH can also be

successfully managed by conservative therapy, a mild surface cauterization followed by topical application of a 0.05% clobetasol ointment four times per day for a period of 4 weeks, thereby avoiding potential gingival margin defects that may result from a surgical ablation of the lesion.

Conclusion

The recently described uncommon gingival hyperplasia lesion, namely LJSGH, can be distinguished from other reactive gingival conditions by its histopathological characteristics while clinical features alone can bring clinicians to confuse in making differential diagnosis. Excisional biopsy is recommended prior to any further management, if required. Regarding its recurrence, routine follow up should be addressed and explained to patients.

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