Localized Juvenile Spongiotic Gingival Hyperplasia: Report of Two Cases

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Objective: Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a painless gingival swelling that histologically exhibits hyperplasia of the non-keratinized stratified squamous epithelium, intercellular edema and spongiosis of the spinus layer, and exocytosis of inflammatory cells. LJSGH pathogenesis remains to be elucidated, while a possible origin from the gingival sulcus epithelium is nowadays proposed. Study design: We report two cases of LJSGH with immunohistochemical evaluation of cytokeratins (CKs) 18 and 19. Results: Both cases concerned 12-year-old boys, who presented with a well-circumscribed bright red pedunculated papillary swelling on the marginal gingiva of the left maxillary lateral incisor. With the provisional diagnosis of LJSGH, the lesions were excised under local anesthesia and histological examination supported the final diagnosis of LJSGH. In both cases, the lesional epithelium showed intense and mild positivity for CK19 and CK18, respectively, while the adjacent normal gingival epithelium expressed CK19, but not CK18, only in the basal cell layer. The postoperative course was uneventful in both patients and no recurrence has been reported. Conclusion: LJSGH is a recently introduced entity that is worth attention in the clinical pediatric dentistry. Clinical and histological examination is required for the final diagnosis, while immunohistochemistry has shed light to LJSGH pathogenesis.

Key words: gingiva, hyperplasia, juvenile, spongiotic, immunohistochemistry, cytokeratin 19; cytokeratin 18.

INTRODUCTION

Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a gingival hyperplasia with unique clinicopathologic presentation1 that was initially described as juvenile spongiotic gingivitis (JSG) and considered to be the gingival counterpart of acute spongiotic dermatitis.2 The term LJSGH is preferred by most authors as it better delineates its features and emphasizes its hyperplastic nature. However, multifocal gingival involvement1-4 and cases in adults5 have been described; therefore the lesion may be neither localized nor juvenile.

LJSGH seems to be a rare lesion. In one study it represented 0.069% of 31,469 specimens accessioned in an oral pathology laboratory,1 and in the English language literature there are approximately 10 well documented publications.1-4,6-11 However, cases of LJSGH may have been misdiagnosed as pyogenic granuloma or gingivitis, in particular puberty gingivitis.1,2

LJSGH typically presents on the facial maxillary gingiva of young patients as a localized and solitary slightly mass up to 1cm in diameter, with bright red color, and papillary, granular, pebbly or velvety surface that is painless, but may bleed on teeth brushing.1,3 Its etiology remains obscure and an origin from the junctional epithelium of the gingiva is suggested.1,2

Only a few case reports of this unusual lesion have been published in the pediatric dentistry literature, although pediatric dentists are the health providers more likely to first examine a patient in this age group. Therefore, the clinical, microscopic and immunohistochemical features of two new cases of LJSGH are described, and the pertinent literature is reviewed.

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Case reports

Case 1
A 12-year-old boy was referred by his dentist for evaluation and management of a painless erythematous swelling on the left side of the facial maxillary gingiva, noticed approximately 6 months before presentation. His parents reported that it “slightly shrank” following plaque debridement 5 months ago, while rinses with Povidone-iodine oral antiseptic solution twice daily for 2 weeks were ineffective. The patient’s medical and family history was noncontributory.

Intraoral examination revealed a well-circumscribed, bright red, pedunculated swelling with granular surface, approximately 1 cm in greatest dimension, on the marginal and attached gingiva of the left maxillary lateral incisor that seemed to grow from the dentogingival junction (Fig. 1). The lesion was non-hemorrhagic on palpation and examination with a periodontal probe. The surface of the corresponding tooth was covered by dental plaque. No other intraoral or extraoral abnormalities were observed. A dental X-ray did not show bone involvement.

The clinical diagnosis was LJSGH and the lesion was totally excised under local anesthesia.

Case 2
A 12-year-old boy was referred by his pediatric dentist for diagnosis and management of an asymptomatic red mass on the facial anterior maxillary gingiva, of approximately 2 months duration. The patient had been diagnosed with “ulcerative colitis” a year before presentation.

Clinical examination revealed a well-defined bright red, pedunculated mass, involving the facial marginal gingiva of the left maxillary lateral incisor that extended to the interproximal area between the two maxillary left incisors. It measured approximately 1 cm in greatest dimension, had a granular surface and was soft, non-hemorrhagic on palpation. The patient oral hygiene was poor, as indicated by the presence of calculus deposits in most of his teeth. The rest of oral mucosa was within normal limits. A dental X-ray did not reveal bone resorption.

The lesion was consistent with LJSGH and excised under local anesthesia.

Microscopic findings
Grossly, both lesions had solid, gray cut-surface and measured 1 cm in greatest dimension. Microscopic examination of 5 μm thick formalin-fixed and paraffin-embedded tissue sections stained with hematoxylin and eosin showed a mucosal fragment covered by spongiotic and acanthotic non-keratinized stratified squamous epithelium, displaying papillomatosis, intercellular edema and neutrophilic exocytosis (Fig. 2A, 2B, 2C). The underlying connective tissue was edematous and vascular, with many dilated and congested vessels, and was diffusely infiltrated by lymphocytes, plasma cells, and neutrophils. Examination under polarized light did not show birefringent exogenous material and Periodic acid-Schiff staining did not reveal fungal hyphae. The final diagnosis was LJSGH.
The cases presented herein showed the diagnostic microscopic features of LJSGH, i.e. papillary, acanthotic, non-keratinized stratified squamous epithelium, intercellular edema and spongiosis of the spinus layer, and exocytosis of acute or chronic inflammatory cells.1-3 Both our patients were 12-year old boys. Most patients with LJSGH are 10-15-year old, the age range is 5-39 years and the average age is 11.8-13 years.1,2 In three large series only 3 cases were found in patients older than 20 years,1-3 but in a recent study,5 19 cases consistent with LJSGH were found in patients aged 21-84 years, after retrospectively reviewing a sample of pyogenic granulomas. A 2.3:1 female predominance1,2 or equal sex distribution,2 as well as a racial predilection for Caucasians1,3 is reported. Time to diagnosis is 1-36 months,1,2 and in our patients the localization of the lesion on the facial maxillary gingiva helped the prompt recognition of the lesion by the parents. As in our cases, the facial maxillary gingiva are more commonly involved,1-3 while few lesions are described on the posterior maxilla2,3 and rare cases on the mandibular1,3 or lingual gingiva.2,4 The marginal gingiva is mainly affected, but the attached or interproximal gingiva may be also involved, as was evident in case 2.1,2 Multifocal lesions seem to be rare,1,4 but this may be an underestimation, as it is possible that such cases may be diagnosed as puberty gingivitis and not subjected to biopsy. The clinical and histological differential diagnosis of LJSGH has been thoroughly discussed in previous studies.2,4 The most common clinical diagnosis is pyogenic granuloma, and the differential diagnosis may include peripheral giant cell granuloma, benign vascular lesions, such as hemangioma and arteriovenous malformation, vascular lesions associated with immunosuppression, such as Kaposi sarcoma and bacillary angiomatosis, and Wegener granulomatosis. Clinical diagnosis in multifocal cases of LJSGH includes puberty gingivitis, plaque-associated gingivitis, allergic reaction to drugs or dental materials, plasma cell gingivitis, and “desquamative” gingivitis in an autoimmune disease, while linear gingival erythema should be considered if there is indication of immunodeficiency. Microscopically, neutrophilic exocytosis may be seen in erythema migrans, oral psoriasis, Reiter’s syndrome, and chronic hyperplastic candidiasis, lesions that do not bear clinical similarity with LJSGH, while spongiosis may be seen in secondary syphilis and inflamed lesions of odontogenic origin.
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CONCLUSION

Familiarity with the clinicopathologic features of LJSGH by pediatric dentists should allow the recognition of more cases of this under-diagnosed rather than rare lesion. Interestingly, although LJSGH usually affects children and adolescents, only scarce case reports exist in the pediatric literature. Description of more cases can help in elucidating its pathogenesis and improving its management.

REFERENCES