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Juvenile Spongiotic Epithelial Hyperplasia: Case Report

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Abstract **Case Report**

Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a rare gingival lesion that has a distinctive form of inflammatory hyperplasia. The lesion has specific features both clinically and pathologically and it may or may not involve multiple sites. In this paper we present a case with lesion that clinically in harmony with LJSGH in upper central incisor area, confirmed by biopsy. A 13-year-old boy was referred to our Assalam Dental Center in Zliten, Libya, presented with circumscribed, erythematous overgrowth on the right maxillary incisor gingiva. With the provisional diagnosis of LJSGH, total excision of the lesion was performed. Clinical, microscopic and histopathogical examination confirmed the diagnosis of LJSGH in maxillary incisor site. The excised lesion showed no recurrence after 18 months of follow up appointments.

Keywords: ocalized juvenile spongiotic gingival hyperplasia, inflammatory hyperplasia, diagnosis.

INTRODUCTION

Juvenile spongiotic gingivitis is a rare benign lesion with unique clinic-pathological features. These lesions have a prominent predilection for the gingiva and do not appear to be related to plaque (Darling *et al*., 2007). The disease mostly affects the anterior facial gingiva, with 84% occurrence on the maxillary gingiva and 16% on the mandibular gingiva (Darling *et al*., 2007). The condition mostly affects young patients who are under 20 years and rarely affecting older patients (Chang *et al*., 2008, Darling *et al*., 2007). There is no confirmed gender predominance and young Caucasians seems to be the preferred patient group (Darling *et al*., 2007, Chang *et al*., 2008, Argyris *et al*., 2015). Chang *et al*., (2008) stated that the lesion was localized gingival overgrowth rather than pure inflammation hence he gave the term localized juvenile spongiotic hyperplasia. Clinically, the condition includes a bright red, easily bleeding, small localized or generalized gingival overgrowth, with a velvety or papillary texture. It is usually painless, and around 20% of the cases are associated with bleeding when brushing teeth (Petruţiu *et al*., 2014). Even though LJSGH is by definition a localized lesion, there are patients presenting with more than one lesion clinically consistent with LJSGH have been seen (Chang *et al*., 2008, Solomon *et al*., 2013) The size of these lesions ranges from 2 mm to 10 mm in diameter (Chang *et al*., 2008). Histopathological investigation of the epithelial tissue reveals papillomatosis, acanthosis, spongiosis, interstitial edema and inflammatory cell exocytosis. The underlying connective tissue is edematous and vascularized with diffuse inflammatory infiltration by lymphocytes, neutrophils and plasma cells (Allon *et al*., 2016, Darling *et al*., 2007, de Freitas Silva *et al*., 2015).

We present the case of a 13 year old boy with LJSGH on the facial margin of his maxillary left central incisor with confirmation of the diagnosis by biopsy. The excised lesion shows no recurrence 18 months later.

CASE REPORT

A 13-year-old boy presented for diagnosis and management of painless bright red elevated gum on the facial margin of his maxillary left central incisor. Medical and dental history is normal and patient had previous extraction with no complications. According to the patient gingival erythema was first noticed more than a year ago associated with gingival bleeding when brushing. Gingival overgrowths developed since and following dentist advice, dental plaque removal and

tooth scaling has been carried out. Although close follow up and tooth and gum cleaning program was followed, the lesion persisted after one year of scaling was performed.

Intraoral examination revealed a well-circumscribed, bright red, pedunculated overgrowth with papillary surface measuring about 0.8 cm on the facial margin and attached gingiva of the maxillary left central incisor (Figure 1). Taking into account the treatment history of the case and the clinical manifestation, a provisional diagnosis of LJSGH was given and following discussion and agreement of the parents and the patient a total excision of the lesion was performed under local anesthesia (Figure 2) and the tissue was fixed in buffered formalin.

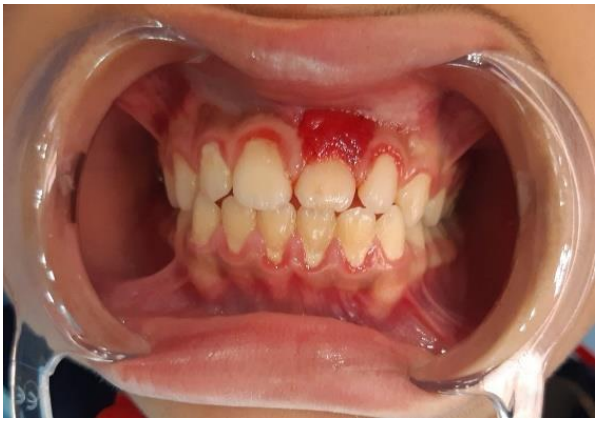

Figure 1: Pre-operative photo shows the lesion above the left central incisor

The Patient was advised to use mouth wash and follow strict mouth hygiene including regular brushing of his teeth. Laboratory microscopic examination showed several sections of mucosal lesion covered by non-keratinized hyperplastic squamous epithelium with edema of the stratum spinosum and neutrophils exocytosis, numerous small dilated blood vessels and mixed inflammatory cells infiltrate.

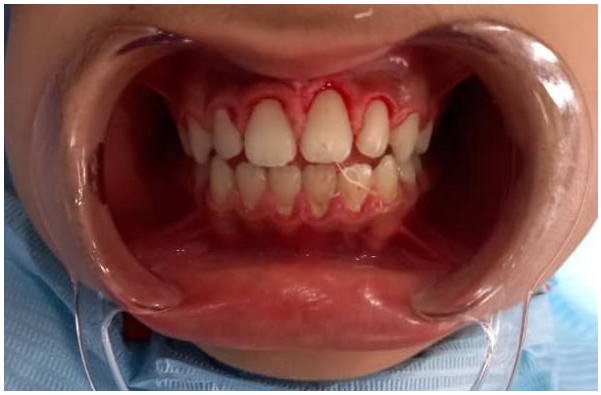

Figure 2: Half an Hour Post-Operative

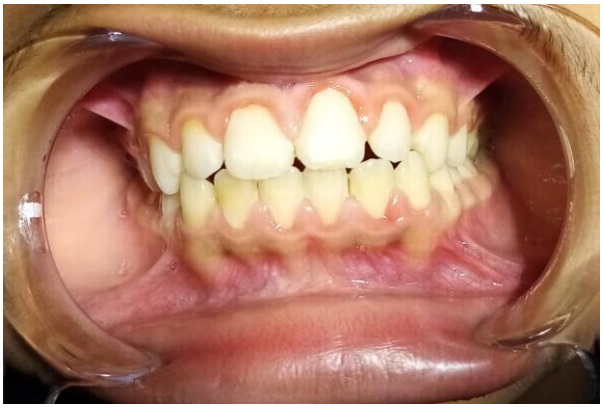

Figure 3: 3 Months following surgery

DISCUSSION

There are relatively few reports that have described LJSGH cases and this may be due to the relatively recent identification of the case as clinical stand-alone entity or may also be due to misdiagnosis with plaque related gingivitis (Darling *et al*., 2007). The clinical feature of red gingival overgrowth not including the marginal gingiva at the anterior maxillary incisive, and the lack of improvement after periodontal treatment can lead to the diagnosis (Darling *et al*., 2007). In the cases that showed the involvement of the marginal gingiva, additional clinical signs are vital for the final diagnosis, such as the characteristic bright red patch with papillary, granular, pebbly, or velvety surfaces; however, biopsy will be valuable to discriminate doubtful cases. Although a lot of cases may be misdiagnosed with puberty gingivitis due to the presence of the gingival erythema that suggest a local inflammatory process, LJSGH differs considerably because of the unresponsiveness to oral hygiene procedures, and the absence of a plaque-related pathogenesis. The condition can also be misdiagnosed with peripheral giant cell granuloma, pyogenic granuloma, human papilloma virus (HPV)-related lesions, foreign body granuloma, small superficial lymphangioma especially in presence of gingival overgrowth (Decani *et al.*, 2021). Loss of keratinization in the stratified squamous epithelium which shows epithelial hyperplasia with a papillary architecture and spongiosis, prominent intercellular oedema are leading features of LJSGH (Allon *et al*., 2016). The connective tissue underneath the elongated papillae may indicate acute and chronic inflammation (Allon *et al*., 2016). The papillary architecture 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). Many treatment plans of LJSGH have been suggested and there is no agreement on specific a plan as yet. Periodontal therapy has been promoted as first-line treatment which will be helpful in excluding the role of plaque induced and puberty gingivitis. Some also suggested that local application of chlorhexidine 0.12% three times for 14 days lead to partial clinical regression of multifocal lesions (Flaitz and Longoria, 2010, Grossmann *et al*., 2014).

Cryotherapy has also been suggested to be the most suitable treatment in areas cosmetically unseen (Nogueira *et al*., 2017). Others reported topical steroids therapy although it is transitory (Fernandes *et al.*, 2018). Laser surgery has also been used successfully in removal of the lesion with no recurrence after 8-18 months. Photodynamic therapy has been suggested recently but the evidence is limited (Vieira *et al.*, 2019, Mawardi *et al*., 2021). The need of excisional biopsy for histopathological confirmation of the lesion made the surgical treatment using the scalpel the most common used approach, however, the reported recurrence is high (Decani *et al*., 2021).

We are reporting one case of LJSGH which was reviewed and treated at Assalam Dental Centre, Zliten, Libya in 2018 and to our knowledge this is the first case report in our city.

The literature reported that the case usually affects youths; Darling *et al*., (2007) reported that 71% of his patients were between 10 and 14 years old, while most (55%) of Chang *et al*., (2008) and Allon *et al*., (2016) (70%) patients were between 11 and 15 years old. Vargo and Bilodeau (2019) reported a wide range of age in his study, between 3 and 64 years old, with median of 14.5 years old. As there are few studies reported LJSGH in adults (Chang *et al.*, 2008, Argyris *et al*., 2015, Siamantas *et al*., 2018) so it seems that LJSGH is most common in young age groups but not limited to. Our patient was 13 years old which falls in the same age range mentioned in the literature. As the lesion occurs in young people, it is usually misdiagnosed as plaque induced puberty gingivitis, however, unlike both of these conditions, LJSGH does not respond to conventional measures of oral hygiene such as brushing and flossing (Darling *et al*., 2007, Chang *et al*., 2008) as what happened in our case.

There is potential for LJSGH cases to recur after biopsy procedure, however in our case we surgically removed the lesion in the process of taking the biopsy and we followed up the case for 18 months so far with no recurrence (Figure 3).

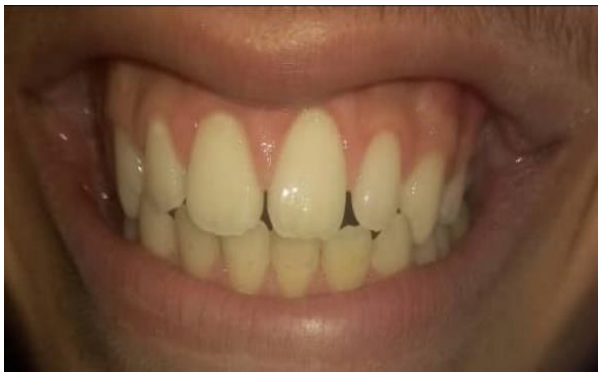

Figure 3: Post-operative follow up (18 months)

CONCLUSION

Localized Juvenile Spongiotic Gingival Hyperplasia is an infrequent lesion that clinicians may come across in the clinic. This lesion usually affects the maxillary gingiva as a red, papillated lesion. Although it can affect both adults and children, it has predilection towards younger age groups. We surgically excised the lesion and followed it for 18 months with no recurrence despite other studies reported high recurrence rate. Larger clinical studies are needed to determine how long the lesions take to reoccur.

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