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CASE REPORT

Aggressive localized juvenile spongiotic gingival hyperplasia (LJSGS) in a 33-year-old patient: a case report

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Abstract:

The aim of this study is to report a case of an extensive and aggressive gingival lesion, compatible with localized juvenile spongiotic gingival hyperplasia (LJSGH), demonstrating the difficulties of diagnosis and management. A 33 years-old male patient was referred to Department of Oral and Maxillofacial Surgery at the Erasto Gaertner Hospital with a lesion on anterior maxillary gingiva, with 3-months evolution period. The patient had complete permanent dentition and good oral hygiene. The gingival hyperplasia involved the gums of teeth 11 and 12, and was solitary, asymptomatic, red, flaccid and bleeding on palpation. No palpable lymph nodes were found. An incisional biopsy was performed, with anatomopathological result of "squamous mucosa with pseudoepitheliomatous hyperplasia and lymphoplasmocytic infiltrate with neutrophils". The lesion was fully excised and the anatomopathological report confirmed the biopsy showing no malignancy in the sample and negative for the investigation of fungus. Biofilm control, mouth rinses with hydrogen peroxide, Chlorhexidine Digluconate 0.12% and Amoxicilin 500mg+Potassium Clavulanate 125mg have not demonstrated efficacy, and the patient kept showing recurrence of the lesion in short time even after the complete excision. Against this situation, the patient was referred to an infectologist, by the suspicion of a resistant microorganism. He started antibacterial therapy with Sulfamethoxazole 800mg+Trimethoprim 160mg that showed good results after 3 months. Currently, the patient remains in follow-up without signs of recurrence. LJSGH is a clinicopathological benign entity that appears in young patients as a soft, bleeding and reddish mass, mainly in the buccal gingiva of the upper anterior maxilla and lonely in most cases. The management of this condition is very difficult because of the few cases described in literature, and establishing the correct diagnosis is essential to ensure that the appropriate treatment is followed.

Keywords: Gingiva; Gingival Diseases; Gingival Hyperplasia; Pathology; Oral.

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INTRODUCTION

Localized juvenile spongiotic gingival hyperplasia (LJSGH) is a rare and poorly understood condition that affects most commonly the anterior maxillary gingiva of juvenile patients[1,2,3]. It was originally described by Darling et. al. (2007)[4] as juvenile spongiotic gingivitis. Later, in 2008, Chang et. al.[5] proposed its current nomenclature, keeping the terms "spongiotic" and "juvenile", as 96% of their patients were between 5 and 15 years old and spongiosis was the most found histological characteristic. However, multifocal gingival involvement and cases in adults have been described; therefore, the lesion may be neither *localized* nor *juvenile*[3]. Theterm "hyperplasia" was given, as 94% of patients had gingival growth, by clinical or histopathological descriptions[3].

Many etiopathogenic mechanisms have been proposed in order to explain and understand the possibledevelopment of JSGH, such as irritation due to plaque build-up, stimulation of gingival tissues by hormonal factors, and infection of human papilloma virus; nevertheless, none of these explain completely the appearance of this specific lesion[6].

The classic presentation of LJSGH consists of an asymptomatic, small, red and single lesion, which may be easily bleeding or ulcerated[1]. From the clinical features, the most common differential diagnoses are pyogenic granuloma, peripheral giant cell lesion, granulation tissue, hemangioma, and peripheral ossifying fibroma[1].

The microscopic findings may include elevated areas of spongiotic and variably acanthotic, nonkeratinized stratified squamous epithelium, with elongated rete ridges and atrophy of the epithelium overlying long connective tissue papillae[1,2]. The immunohistochemical studies of JSGH noticed the similarity between the hyperplasic epithelium of JSGH and the junctional epithelium (JE)[6].

The treatment is still undefined, and a lot of new conducts has been described, although the surgical approach is still considered the gold standard, but some authors affirm that the technique is invasive with controversial results[2]. Photodynamic therapy (PDT), criotherapy and antibiotics treatments has been shown in literature as alternatives.

As a rare disease, the demographic factors, related to LJSGH, such as gender and age remain undeterminated[2], and there are still few reports in the literature and no treatment guidelines available.

Therefore, since LJSGH is a rare entity described in the literature, the aim of this study is to report a case of an extensive and aggressive gingival lesion, which the primary hypothesis is LJSGH, based on clinical and histopathological descriptions. Our focus is demonstrating the difficulties of diagnosis and management of the condition.

CASE REPORT

A 33-year-old male patient was referred to the Department of Oral and Maxillofacial Surgery at the Erasto Gaertner Hospital with a lesion on the anterior maxillary gingiva, with a 3-month evolution time. The patient had complete permanent dentition and good oral hygiene.

He presented a gingival hyperplasia involving the free and inserted gums of teeth 11 and 12. The lesion was solitary, asymptomatic, red, flaccid and bleeding on palpation (figure 1). No palpable lymph nodes were found.

Figure 1. Initial aspect of the lesion.

The patient denied any type of comorbidity and reported only being a social drinker and presenting with gastritis. A complementary CT scan of the face showed intact maxillary bone structures.

An incisional biopsy was performed, with anatomopathological report of pseudoepitheliomatous hyperplasia and lymphoplasmatic inflammation. The lesion was then fully excised under local anesthesia and patient was prescribed Amoxicillin 500mg, Potassium Clavulanate 125 mg for 14 days, mouth rinses with hydrogen peroxide 10 volumes, chlorhexidine digluconate 0.12% and analgesia with Paracetamol 750mg. The anatomopathological report confirmed the biopsy showing no malignancy in the sample and negative in the investigation of fungus.

Postoperatively, the patient presented significant bone loss with root exposure of teeth 11 and 12 and recurrence of the lesion extending between teeth 13 and 21. At this time, the patient maintained good oral hygiene, with no accumulation of dental biofilm and no mobility of the teeth involved.

The lesion continued to show significant increase in its size during the next visits (figure 2). A new incisional biopsy was performed, showing again unspecific inflammation and immunohistochemical analysis was consistent with granulation tissue with inflammatory infiltrate predominantly plasmocytic, dense, and with absence of malignant neoplasia in the sample (figure 3).

Figure 2. Aspect of the lesion after 3 months of evolution, even after total excision.

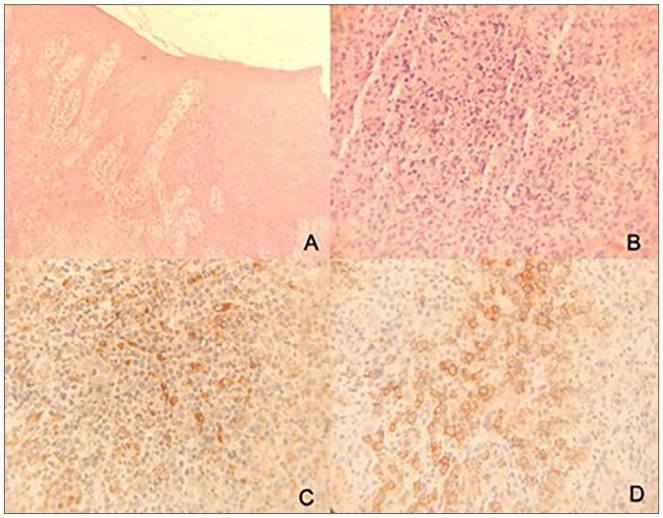

Figure 3. Histopathological aspect: A - Mucosa exhibiting pseudoepitheliomatous hyperplasia; B - Dense lymphoplasmacytic infiltrate; C - Positive staining for CD68 and D - for CD138, characterizing the inflammatory infiltrate.

In order to deepen the diagnostic investigation, the patient was referred to an infectologist, to evaluate a possible infectious process associated with the lesion. He started antibacterial therapy with Sulfamethoxazole 800mg + Trimethoprim 160mg, and 3 months after starting treatment the patient showed a progressive decrease in the size of the lesion.

Currently, the patient remains in follow-up without signs of recurrence (figure 4), but a reconstruction procedure with free gingival graft is being planned by the surgeons to fix the gingival defect caused by the lesion. Therefore, the first and only diagnostic hypothesis, based on clinical and histopathological characteristics and the case outcome, is LJSGH.

Figure 4. Aspect of the patient's gingiva today, after almost three years.

DISCUSSION

The classic presentation of LJSGH consists of a small, solitary, localized and red lesion, that may be easily bleeding or ulcerated[1] and according to the literature, the most common site is the anterior maxillary gingiva[3]. As presented in our case, the patient presented a solitary, red and bleeding lesion affecting the maxillary gingiva without a clear etiology. Furthermore, the cases are predominantly of single lesions[6] that do not reach large proportions, with a median size of 6 mm[4], while in our patient, the lesion reached an average size of 2 cm. Regarding symptoms, our patient had no complains associated with the lesion, which corroborates the literature[4]. In addition, he denied having comorbidities or allergies, and tests for viral and fungal infection were negative, and laboratory exams confirmed no other systemic condition.

Although its etiology is still not well clarified, its characteristics do not demonstrate a relationship with biofilm-induced inflammation, HIV, HPV, estrogen and

progesterone levels, allergic factors, linear gingival erythema, or fungal infections[2, 8]. Allon et al.[8] suggested that JSGH occurs because of an exteriorization of the junctional epithelium during the eruption of permanent tooth. Diverse irritative factors would then act over this epithelium, triggering the inflammatory proliferation and tumoral appearance of this lesion.

The predilection for younger people is indicative of hormonal influence, but no estrogen and progesterone receptors are found, while LJSGH may appear in prepubertal children and adults.[3] Other factors considered are trauma that was documented in just one patient; orthodontic appliances that were present in only 8 in 52 and 1 in 21 patients with LJSGH; and mouth breathing that is not consistent with the mostly localized distribution of the lesion[3].

As a rare disease, the demographic factors related to LJSGH, such as gender and age remain indeterminate[2]. Variations among the age range of patients are observed, with many publications addressing a mean age of 11 to 13 years[4, 5, 9,10], while other more recent studies describe cases with a mean age of 18 years or older[1, 6, 8].

The treatment of LJSGH is difficult and there are still no guidelines available, although some authors have shown improvement with dental biofilm control[7]. In cases of multifocal lesions, surgical treatment is not indicated due to aesthetic dissatisfactions[11]. To Inoccentini et al. (2020)[9], the surgical excision should be chosen when no recession is expected or when gingival plastic is possible. However, in our case, the biofilm control does not demonstrated great impacton the treatment, so the staff opted for surgical excision due to the aggressiveness demonstrated by the lesion and also because of the recurrences.

Some authors reported non-invasive treatments such as cryotherapy, especially in pediatric patients[12]. The use of photodynamic therapy has shown to be effective in treating similar inflammatory conditions, in addition to its selective toxicity, low invasiveness, and rare side effects, just like Vieira et. al.[2] have described. Others authors affirm that LJSGH generally does not regress when only periodontal treatment is performed, as it is not related to dental biofilm, and its recurrence after surgical excision is about 6-16%[13]. Topical steroid therapy was ineffective in one case[14], whereas in another case resulted in transient clinical improvement of the lesion.[15] As the lesion may spread and often recur, just like in this case report, the patients should be regularly re-examined.[16]

In our experience, the indication for surgery versus conservative treatment depends on the extension and number of areas involved by the lesion, as the tissue growth is not always reversible. In our case, surgical excision did not demonstrate to be effective, with recurrence of the lesion less than one month after the procedure.

This case was considered so challenging because even after performing numerous biopsies, the anatomopathological and immunohistochemical reports remained unspecific and the surgical and periodontal treatments were not effective. Therefore, as the team was in a hospital service with a vast multidisciplinary team, we chose to refer the patient to the infectious disease medical team, who started the antimicrobial treatment (Sulfamethoxazole 800mg + Trimethoprim 160mg) due to the clinical characteristics of an infectious and aggressive lesion. The proposed treatment proved to be effective, showing that not all lesions have a definitive diagnosis, but the clinical knowledge and experience of the professional can define effective treatments.

CONCLUSION

Despite being a small and non-aggressive lesion in the vast majority of cases, LJSGH may present in a more aggressive and extensive form, challenging and resistant to treatment, that could cause aesthetic losses to the patient. The management of this condition is very difficult because of the few cases described in literature, and establishing the correct diagnosis is essential to ensure that the appropriate treatment is followed. For this reason, we consider the report of this case important, to stimulate the development of studies that guide the treatment of this type of lesion.

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