

# Overcoming the Recurrence Dilemma of Oral Lobular Capillary Hemangioma (Pyogenic Granuloma): Case Series of an Alternative Therapeutic Approach

**\*Morolayo T. OGUNWEMIMO, \*Ibukun O.OJO, \*Olusoji ONABANJO, \*Olubunmi O. AKINYEMI, \*Babatope A. OGUNLEYE, \*Opeyemi M. ADEWOLE, \*Tolulope OGUNDIRAN, \*Solomon O. NWHATOR**

*\*Department of Preventive and Community Dentistry, Obafemi Awolowo University Teaching Hospital Complex,*

## Correspondence

Dr. S. O. Nwhator  
Department of Preventive and Community Dentistry  
Obafemi Awolowo University Teaching Hospital Complex  
Email: [periodontologist2010@gmail.com](mailto:periodontologist2010@gmail.com)

## ABSTRACT

**Background:** Lobular capillary hemangioma (LCH) is a reactive inflammatory lesion that has a high recurrence rate. The aetiology is unknown but risk factors such as trauma, chronic irritation in form of plaque and calculus, hormonal imbalance etc have been implicated. Male: female ratio is 1:2. Excision is the mainstay management with sclerosant therapy been advocated as an alternative. Intralesional bleomycin was previously used in this center as an alternative to reduce recurrence.

**Objective:** To report the resolution of two cases of recurrent lobular capillary hemangioma using intralesional bleomycin solution injection combined with surgical excision.

**Results:** Case reports of 2 male patients managed by the Periodontology Unit, Obafemi Awolowo University Teaching Hospital Complex, Ile-Ife.

## Keywords:

## INTRODUCTION

Pyogenic granuloma is a common benign lesion of vascular origin in the head and neck region, occurring in the skin and mucous membrane (Wollina et al., 2017). The name “pyogenic granuloma” is a misnomer as there is neither pus associated with the lesion (Vaidya et al., 2018) nor is histologically a granuloma (Vilman et al., 1986). The current preferred name-lobular capillary hemangioma (LCH) (Wollina et al., 2017) is considered more appropriate because histologically its histology is characterized by distinct proliferation and lobular arrangement of capillaries (Patrice et al., 1991).

LCH with its atypical presentation and behaviour may clinically mimic malignant tumours and the lesion has a predilection for females in their reproductive years. The objective was to report the resolution of two cases of recurrent lobular capillary hemangioma using

intralesional bleomycin solution injection combined with surgical excision.

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## CASE REPORT

Patient A was referred to our Periodontology clinic with a history of swelling in the labial and palatal aspect of the gingiva of teeth 21 to 23. Tooth 21 was extruded and with a diastema between 11 and 21. The patient claimed the current swelling started as a tiny palatal nodule 6-8 months prior to presentation, but increased rapidly in size within the last month to presentation with aesthetic challenge and some speech impairment.

The oral hygiene was poor with sub-gingival accumulation of plaque and calculus. A periapical radiograph showed no bony involvement (Figures 1a-c) and a clinical diagnosis of lobular capillary hemangioma was made. Professional scale and polish preceded surgical excisional biopsy performed (the sixth excision) and a non-eugenol periodontal dressing applied. The histopathologic examination returned a diagnosis of lobular capillary hemangioma. Within two weeks of the excision, the lesion grew rapidly, became more hemorrhagic, extended from tooth 11 to 23 and measured about 2.5cm by 2.0cm labially and palatally (Figures 2 a-b). Since this was the first excision at our center and the sixth overall excision, a decision was reached by our periodontology team to adopt an alternative approach by the injection of a sclerosant. This was performed four weeks post operatively using an intralesional injection of 5 units of Bleomycin (Pingyangmycin) dissolved 1.8mls of Lignocaine and mixed with 2mls of 0.9% of normal saline. 1.8mls of

the resultant solution was injected into the labial component and the remaining into the palatal component of the lesion.

Similar, patient B, a 68yr old retiree who presented with a history of a palatal swelling of 9 months duration, in relation to tooth 11, 12, 13 and 14. An impression of LCH was made and surgical excision performed. Histology confirmed the diagnosis but upon recurrence within a week of excision, a re-excision with intralesional injection of given. Similarly, the third patient, a 50yr old male who presented with a history of a recurrence of LCH within 2 months, palatal to tooth 11. Our clinical impression of LCH was made and histology confirmed the diagnosis of LCH. The lesion recurred again within a week of re-excision at our centre. Due to the large size and aesthetic concerns, another re-excision (the third) was performed but this time with intralesional solution comprising 5mg bleomycin+ 1.8mls lignocaine+ 1ml 0.9% normal saline.



Figures 1a ,1b and 1c showing radiograph at presentation palatal and labial view at presentation



Figures 2a and 2b showing a recurrence extending from tooth 11 to 23 within 2 weeks of re-excision



Figure 3a-b showing resolution of palatal swelling and markedly improved labial swelling



Figure 4a-b showing maintenance of gingival health without recurrence at one year post-operative review

## RESULTS AND REVIEW OF THE LITERATURE

Case 1 --within four weeks, both the labial and the palatal components of the lesion completely regressed and one year review showed stable results with maintenance of gingival health and no recurrence (Figures 4a-b). Similarly, in case 2 and 3, 18 month review and 6 week review showed

maintenance of gingival health with no recurrence respectively. (Figures 5a-c and 6a-c)

In all three cases presented above, the lesions have completely resolved with no recurrence till date.

Lobular capillary hemangioma has been known by many names since Hartzell first discovered it in 1904.



Figure 5a-c showing patient B at presentation, recurrence at 2 weeks & 6 week review after re-excision/sclerosant injection

Figure 6a-c Patient C at presentation, 2weeks recurrence & 18 month post re-excision/sclerosant injection



The commonest being “Pyogenic granuloma” which is a misnomer as explained above. It occurs on the skin and in the head and neck region, most commonly in the gastro-intestinal tract, especially in the oral cavity. Its aetiology is unknown (Koo et al., 2007) but postulated risk factors include chronic low -grade irritation (in form of calculus and plaque, restoration etc. (Cai, et al., 2017) and trauma. (Patrice et al., 1991; Rachappa et al., 2010) Others include hormonal influences, medications (Rosa et al., 2017) and iatrogenic stimulations including guided tissue regeneration and bone marrow transplant.

Though the pathophysiology of this lesion is not well understood, two theories have been proposed viz the intrinsic and extrinsic theories. The intrinsic theory points to mutation of a critical gene in the precursor stem cell with clonal expansion of this single cell hemangioma formation (Régnier et al., 2007). The extrinsic theory says abnormal hormonal milieu induces tissue hypoxia with angiogenesis stimulation through vascular endothelial growth factor (VEGF) (Vara et al., 2017) leading to hemangioma formation (Mulliken & Glowacki, 1982). Other theories focus on an imbalance between angiogenic promoters and inhibitor leading to capillary overgrowth (Gowda & Owens, 2017). It was also said that the low apoptotic rate in pyogenic granuloma is responsible for its rapid growth due to the bcl-2 proteins (Nakamura, 2000). The management of LCH includes the removal of the causative factors (scaling and polishing), and if the patient is pregnant, encouraging her to keep the mouth as healthy as possible and to wait few months after delivery before any definitive management is done.

The management of lobular capillary hemangioma varies from excision to the use of laser, pulsed dye to sclerosant for recalcitrant cases and recurrences. Mainstay treatment consists of excision down to the periosteum, with no recurrence in the oral ones reviewed<sup>14</sup> but with the lowest rate of recurrence in other sites studied. (Giblin and Clover, 2007) Other forms of treatment are cryosurgery, laser surgery in addition to electrodesiccation and chemical cauterization depending on the size, site and patients' wishes.<sup>1</sup> All the above treatments mentioned above tend to heal by secondary intention; thereby limiting its use in the aesthetic zones of the mouth and also tend to be messy with recurrence rate of up to 50%. But it has been found out that pulsed dye laser treatment in pregnant women most especially leads to recurrences.

The use of sclerosant has been in existence for long with different materials ranging from corticosteroid to alcohol but the use of Bleomycin was first documented in literature in 1997 for a paediatric case

of capillary hemangioma, followed by its use in ocular cases of non-proven but clinically appearing cases of pyogenic granuloma. Sclerotherapy has also been attempted in the management of LCH to reduce its recurrence rate. Sclerosants that had been used include sodium tetradecyl sulfate, monoethanolamine oleate, steroid (Kiran et al., 2018). Another sclerosant that has been used is Pingyangmycin (Bleomycin) especially in recurrent cases with good results obtained like in the case discussed below. Most cases of Lobular capillary hemangioma tend to occur commonly in the female, (Lawoyin et al., 1997; Koo et al., 2007; Cai et al., 2017) although this is not so in every case. The commonest site in the head and neck region is the anterior maxilla, most especially on the gingiva (Lawoyin et al., 1997; Rachappa et al., 2010) which is regarded as an aesthetic zone in contrast to what was observed in the study done by Lawoyin et al. in Ibadan; where the site commonly affected was the lower anterior labial region (Cai, et al., 2017). Any treatment done must be as conservative as possible and also done to avoid recurrences. Excision offers a good choice especially when carried out to the periosteal layer with removal of the causative agent (Wollina et al., 2017) but due to the paucity of the resources in this part of the world does not always offer freedom from scarification and morbidity (extraction of affected tooth) (Cai et al., 2017), and possible recurrences as high as 16% (Kiran et al., 2018) although some studies recorded high success and low relapse percentages (Giblin & Clover, 2007).

Alternative medical therapy has been sought for over the years to prevent the recurrence of LCH and many have shown promising result starting from steroid use, interferon therapy, vincristine, propranolol and thalidomide and sclerosants such as bleomycin, sodium tetradecyl sulphate (STS). All have been found to be effective but some with serious side effects.

Sclerotherapy is targeted at eliminating small vessels, varicose veins and vascular anomalies by injection of sclerosants (Sacchidanand & Purohit, 2013). Sodium tetradecyl sulphate administration gave various results from complete resolution with no recurrence to partial resolution, using it alone or in combination with other agents (Kiran et al., 2018). Other sclerosants include absolute alcohol and corticosteroid. Complications associated with their use include non-specific necrotic changes and local nerve injury (STS) and allergy (absolute alcohol) with resultant soft tissue edema (Cai et al., 2017).

Bleomycin is an antineoplastic antibiotic, particularly effective against some neoplasms in the human beings, especially squamous carcinoma and lymphomas. It was found to be effective against

inoperable hemangioma and in low doses to be a good sclerosant in treatment of vascular anomalies. When co-administered with dexamethasone, it was found to achieve a 90% reduction of 80% of the patients. The mechanisms of action of bleomycin was attributed to its inhibition of growth factor induced endothelial cell invasion, endothelial cell growth and migration, and also, induces endothelial cell apoptosis (Cai et al., 2017) and in vitro was said to be due its inhibition of neovascularization. (Mabeta & Pepper, 2011) No systemic complication has been published except for localized reactions at the site such as ulceration, pain, local swelling and temporary paraesthesia. In another study, the side effect was hyperpigmentation. (Pienaar et al., 2006) In this case, only localized swelling was reported for about 5days. For a recurrent LCH (pyogenic granuloma), intralesional injection of Bleomycin (Pingyangmycin) was found to be effective and no tissue destruction and also preservation of associated teeth and soft tissues.

## CONCLUSION

Intralesional bleomycin injection can be considered an alternative treatment approach in cases of confirmed recurrent LCH.

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