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## Research Article

# PYOGENIC GRANULOMA IN ASSOCIATION WITH PORT WINE STAINS – COINCIDENCE OR CORRELATION

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### ABSTRACT

Pyogenic Granuloma [PG] is considered to be a non neoplastic inflammatory lesion in the oral cavity. It is not a true granuloma despite its name. The etiology of pyogenic granuloma includes exogenous factors such as calculus, foreign materials in the gingival sulcus, and trauma. It most commonly involves 75% of the gingiva. Many recent reports have showed a rare association between the Pyogenic granuloma [PG] and port-wine stain [PWS]. Sturge Weber syndrome is a nonhereditary developmental condition characterized by a facial capillary malformation, ipsilateral leptomeningeal angioma, and vascular eye abnormalities. The facial capillary malformations are of characteristic dark red to brown black in color thus referred as facial Port Wine Stains. There are a wide range of treatment options available for treating PGs with PWS which includes excision, curettage, cryotherapy and lasers. A new treatment modality is now followed for recurrent PG which is the intralesional injection of liquid sclerosing agents referred as sclerotherapy. Thus in this case report, the sclerosant Sodium Tetradecyl Sulfate is employed for the treatment of recurrent pyogenic granuloma in a woman with concurrent presentation of PWS and it has proved to be a successful treatment modality for the resolution of the lesion. Thus sclerotherapy with Sodium Tetradecyl Sulfate can be used as an effective alternative for the treatment of recurrent PG associated with PWS.

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### INTRODUCTION

Incidences of localized growth are seen frequently in oral mucosae which are either reactive or neoplastic in nature. Pyogenic granuloma [PG] is one of the most common inflammatory tumor like growth of the oral cavity that is nonneoplastic in nature<sup>[1,2]</sup>. Despite its name, it is not a true granuloma. The etiology of pyogenic granuloma includes exogenous factors such as calculus, foreign materials in the gingival sulcus, and trauma<sup>[3]</sup>. These are usually painless lesions which are pink to red to purple in colour depending upon the age of the patient. The young pyogenic granuloma appears to be more vascular and the old ones appear to be more collagenized and pink. They present as a smooth or lobulated mass that is usually pedunculated but some lesions are sessile with ulcerated surfaces. They are soft on palpation and compressible and bleed easily on touch<sup>[4]</sup>. The size of the lesion varies from few millimeters to few centimeters in diameter. Oral pyogenic granuloma shows a striking predilection of 75% for the gingiva. It has been reported that the highest incidence occurred in the second and fifth decades and increasingly seen in women (2:1)<sup>[5]</sup>.

Microscopically pyogenic granuloma shows high vascular proliferation resembling granulation tissue and contains endothelium-lined vascular spaces. The epithelium is thin and atrophic and there is proliferation of fibroblasts and endothelial cell budding. In connective tissue there is infiltration of polymorphonuclear leukocytes, lymphocytes, and plasma cells<sup>[6]</sup>. CD31 is the immunohistochemical marker which is used to confirm the lesion's vascular nature<sup>[7,8]</sup>. Many recent reports have showed a rare association between the pyogenic granuloma and port-wine stain [PWS]

Sturge Weber or encephalotrigeminal angiomatosis is a nonhereditary developmental condition characterized by a facial capillary malformation, ipsilateral leptomeningeal angioma, and vascular eye abnormalities. The facial capillary malformations are of characteristic dark red color similar to the portuguese liquor and thus referred as facial PWS<sup>[9,10]</sup>. The pathogenesis of PWS remains debatable till now<sup>[11,12]</sup>. In this syndrome the oral changes occur in 40 % of cases ranging from small lesions to massive growth of the gingiva. When PG is associated with PWS it is usually seen in women of young age after trauma<sup>[13,14]</sup>. There are a wide range of treatment options available for treating PGs with PWS which includes

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cautery, cryotherapy, chemical cauterizations and lasers. Among this the most commonest is excision. Now for treating recurrent PG a new treatment modality is being followed which is the intralesional injection of liquid sclerosing agents referred a sclerotherapy<sup>[15,16]</sup>.

Sclerotherapy is defined as the targeted elimination of small vessels and vascular malformations by the injection of a sclerosant<sup>[3]</sup> These sclerosing agents cause vascular thrombosis and permanent damage to the endothelial vessels and vascular obliteration when injected into the blood vessels.<sup>[5]</sup> In this case report, Sodium tetradecyl sulfate (STS) is used as an intralesional sclerosing agent for a case of recurrent oral PG. The various other applications of STS are varicose veins, endoscopic sclerosis of gastroesophageal varices.

### CASE REPORT

A 22-year-old woman reported to the Department of Periodontology with a complaint of localized gingival mass in the maxillary left posterior tooth region for the past 3 months. The patient noticed that it was gradually increasing in size over the period of time. On clinical examination, it was noticed that the exophytic pedunculated mass originated from interdental papilla between 26 and 27, and the mass consisted of 2 parts with labial and lingual extension. The dimensions of the labial part was 1.5 x1.5 cm and the lingual part of the lesion was about 8 x8 mm [Fig 1]. The mass was pinkish red in color with soft in consistency. It exhibited smooth, lobulated, shiny surface with a tendency to bleed profusely on provocation. Bleeding time and clotting time were found to be within normal limits.

Extra oral examination of the skin showed a well-circumscribed reddish plaque covering the lower part of the face, circumscribing the perioral area which was present since birth [Fig.2]. No significant family history was noted. Patient gives history of two episodes of recurrence. The first time, gingival mass recurred after 1 year following excisional biopsy then it was diagnosed as PG histopathologically. The patient reported back with the recurring lesion after 2 months and the treatment plan was to perform the phase I therapy which includes scaling and root planning to reduce the inflammatory component of the lesion and surgical excision [Fig.3] following histopathological evaluation it was diagnosed as recurrent pyogenic granuloma. However the lesion recurred in three months and hence the treatment plan was modified to include thorough scaling and root planning followed by sclerotherapy with sodium tetradecyl sulphate. So to ensure safety of the drug, a prick test was performed by injecting 0.1 mL of the solution subcutaneously into the left forearm where changes were observed for 1 hr 15 min and no hypersensitivity reaction was reported. Local anesthesia was administered to the patient to anesthetize the area, followed by intralesional injection of STS 30 mg/mL [Setrol] at 5 sites (3 labial and 2 lingual) with 0.1 mL at each site [Fig.4]. Follow-up was carried out immediately, at 1 hr, 24 hrs, and on seventh day of injection. After 10 min of injection, a bluish tinge was noticed at the periphery of the lesion [Fig.5A] at the end of 1 hr, the lesion was completely blue in color [Fig.5B]. After 24 hr, the color of the lesion had changed to reddish black with a slight reduction in size [Fig.5C]. Subsequently repeat injection of sclerosing agent was done at weekly intervals for 3 weeks. The entire lesion was evaluated after sclerosant injections and excised at fifth week [Fig. 6]. Then the tissue was sent for histopathologic

assessment [Fig 7]. Based on these clinical and histopathologic findings, the lesion was diagnosed as PG [Fig.8 A&B]. Presently no recurrence has been noted during the follow up period of 6 months [Fig. 9]



Figure 1 Pre-operative Intra Oral clinical picture of the lesion



Figure 2 Extraoral: presence of PWS demarcated by lines



Figure 3 Clinical picture of the lesion 4 weeks after phase I therapy



Figure 4 Intra lesional injection of sclerosant [Setrol]

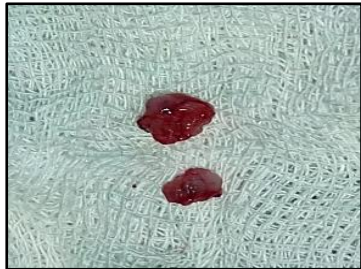




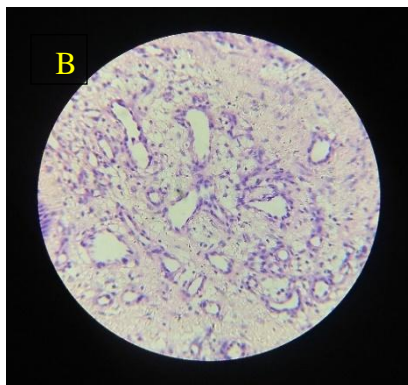
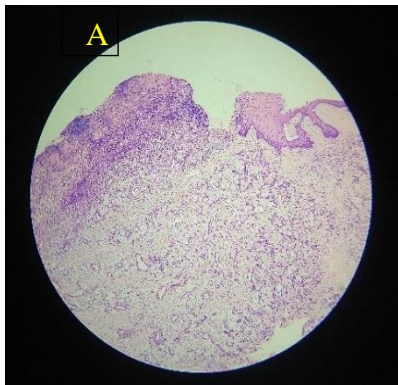
**Figure 5** Changes in lesion [A] Immediately [B] at 1 hour [C] at 24 hours



**Figure 6** Clinical picture 4 weeks after sclerosant therapy just before excision



**Figure 7** Excised tissues



**Figure 8** A&B Photomicrograph of lesion showing typical features of PG



**Figure 9** 6 months post-treatment photograph without any recurrence of lesion

## DISCUSSION

PG is a benign inflammatory lesion which mainly affects the skin and oral cavity. In the literature it has been documented that PG can recur over a period of time after initial treatment with the same features as that of the original one when it is associated with vascular abnormality like sturge weber syndrome. In this present case, the PG lesion was present in a 22-year-old woman involving the gingiva of the maxillary posterior teeth. The histopathologic features were found to be similar to those mentioned in the studies. Recurrent PGs grow rapidly and appear as smooth or lobulated exophytic lesions with senile or pedunculated bases. They are painless and usually hemorrhagic. Same characteristics are seen in the present case, in which PG has recurred for the third time. The association between PG and PWS is that this lesion tends to develop in PWS is associated with microscopic arteriovenous anastomoses, leading to the development of PG lesions in highly vascularized areas such as hands, face, tongue<sup>[17]</sup>. In the present case, the histopathology revealed hyperplastic stratified squamous parakeratinised epithelium replaced by pyogenic membrane .

The underlying connective tissue shows proliferating blood vessels, endothelial cells and aggregates of chronic inflammatory cells consistent with PG diagnosis. Anastomosis between thick- and thin-walled vessels was observed in the deeper tissues, suggesting the association of PG and PWS according to literature. The recommended treatment protocol for PG associated with PWS is complete surgical excision of the lesion. But owing to the recurrence of the lesion at the same site here in the above mentioned case report, sclerotherapy as an adjunct to scaling and root planing and excision was carried out.

Sclerosing agent produces damage to the endothelium of the vessels, and the entire vessel wall and transforms it into a fibrous cord. Literature shows the effective use of sclerotherapy in the form of intralesional injections for the treatment of AIDS-associated Kaposi sarcoma.<sup>[18,19]</sup> Ramirez-Amador et al has conducted a study and proved that intralesional STS is more effective in the treatment of AIDS related Kaposi sarcoma compared to vinblastine .Moon et al. have described that the treatment of extraoral PG is successful with the use of STS<sup>[20,21]</sup>. Thus the dentists while treatment planning must be aware of the possible association between recurrent PG and PWS. Because PG is vascular in nature, sclerosing solution may be appropriately primary or an adjunctive treatment modality. which is found to be effective with no adverse effect.

The port wine stains have usually unilateral distribution along with the course of trigeminal nerve. These port wine stains can

be a cosmetic problem and are treated by high dose of hydrocortisone, dermabrasion or lash lamp pulse tunable dye laser therapy which results in partial or complete clearing of the Port wine stain. In this case the patient was referred to Dermatologist for the cosmetic treatment of port wine stains, but due to the financial reasons the patient was not able to undergo the treatment. Dental management of the patient should be stressed on behavior management and preventive oral hygiene measures. Therefore patient and parent education along with plaque control measures should be strictly followed to prevent further complications.

## CONCLUSION

In Sturge Weber syndrome there is an increased risk of hemorrhage, thus the management of these patients is a challenging task. Precautionary measures to manage the risk of hemorrhage such as use of haemostatic agent, provision for blood transfusion should be carried out. Sclerotherapy is a simple and minimally invasive procedure, with negligible blood loss and no postoperative complications. It is appropriate to suggest that more cases are necessary to establish a standard treatment protocol for this association and larger samples are required for further confirmation of the findings

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