



Pregnancy tumor and facial port-wine stain: A case report

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ABSTRACT

Pyogenic granuloma is a common tumor-like growth of the oral cavity that traditionally has been considered to be non-neoplastic in nature. The pyogenic granuloma is a smooth or lobulated mass that is usually pedunculated, although some lesions are sessile. Pyogenic granuloma of the gingival is frequently developed in pregnant women, hence the terms pregnancy tumor and/or granuloma gravidarum are often used. The pyogenic granuloma has been reported to be associated with port-wine stain particularly during pregnancy, and after trauma, or laser treatment. The pathogenesis of pyogenic granuloma is unclear. This report describes the presentation of a large pregnancy tumor in a patient with an isolated facial port-wine stain.

Keywords: Port-wine stain, Pregnancy, Pyogenic granuloma.

Introduction

Pyogenic granuloma is a common tumor-like growth of the oral cavity that traditionally has been considered to be nonneoplastic in nature. Although it was originally thought to be caused by pyogenic organisms, it is now believed to be unrelated to infection [1]. The pyogenic granuloma is a smooth or lobulated mass that is usually pedunculated, although some lesions are sessile. The surface is characteristically ulcerated and ranges from pink to red to purple, depending on the age of the lesion [1]. Pyogenic granuloma is commonly seen on the gingiva, where it is presumably caused by calculus or foreign material within the gingival crevice. Hormonal changes of

puberty and pregnancy may modify the gingival reparative response to injury, producing what was once called a "pregnancy tumor". Under these circumstances, multiple gingival lesions or generalized gingival hyperplasia may be seen. Pyogenic granuloma is uncommonly seen elsewhere in the mouth but may appear in areas of frequent trauma, such as the lower lip, buccal mucosa, and tongue [2]. Although the pyogenic granuloma can develop at any age, it is most common in children and young adults. Most studies also have demonstrated a definite female predilection, possibly because of the vascular effects of female hormones [1].

Pyogenic granuloma of the gingiva is frequently developed in pregnant women; hence the terms pregnancy tumor and/or granuloma gravidarum often are used. Such lesions may begin to develop during the first trimester, and their prevalence increases up through the seventh month of pregnancy. The gradual rise in development of these lesions throughout pregnancy may be related to the increasing levels of estrogen and progesterone as the pregnancy progresses. After pregnancy and the return of normal hormone levels, some of these pyogenic granuloma resolve without treatment or undergo fibrous maturation and resemble a fibroma [1]. Microscopically, pyogenic granuloma are composed of lobular masses of hyperplastic granulation tissue. Some scarring may be noted in some of these lesions, suggesting that occasionally there may be maturation of the connective tissue repair process. Variable numbers of chronic, inflammatory cells may be seen. Neutrophils are present in the superficial zone of ulcerated pyogenic granuloma [2].

The treatment of patients with pyogenic granuloma consists of conservative surgical excision, which is usually curative. The specimen should be submitted for microscopic examination to rule out other more serious diagnoses. For gingival lesions, the excision should extend down to periosteum and the adjacent teeth should be thoroughly scaled to remove any source of continuing irritation. A recurrence rate of 3% to 15% has been reported in most studies [1].

Yuan et al. described the relationship between pyogenic granuloma and angiogenic factors in pregnancy. Women with pyogenic granuloma during pregnancy demonstrated significantly more basic fibroblast growth factor (FGF), more vascular endothelial growth factor (VEGF), and less tumor necrosis factor-alpha (TNF-alpha) [3]. The pyogenic granuloma has been reported to be associated with port-wine stain particularly during pregnancy, and after trauma, or laser treatment. The pathogenesis of this pyogenic granuloma is unclear [4].

Port-wine stain is a congenital capillary malformation with a prevalence of 0.3–0.5% and manifests on the mucosa or skin as pink or red, erythematous patches that can become darker with age. The head and neck region is the most common location, especially in the V1 and V2 dermatomes [3]. The following is a case report of a 26-year-old female with a right facial port-wine stain who developed a large pregnancy tumor of her right maxillary gingiva in a similar distribution to her port-wine stain.

Case Report

A 26-year-old female, at 24 weeks gestation, presented to the Oral and Maxillofacial Surgery Department at Yazd/Iran. She presented with a 9 week history of a lesion on her right maxillary gingiva that was rapidly enlarging, painful and bled easily with minor irritation. On physical exam, she had a large red-purple lesion of the right maxillary buccal and palatal gingiva. The lesion was soft and spongy in texture and easily bled upon manipulation. The patient also had a right facial port-wine stain, adjacent to the oral lesion, which had been present since birth. The port-wine stain involved the facial skin of her right cheek (Figure 1A, B).

Given the clinical appearance of the oral lesion and that it presented during pregnancy, a clinical diagnosis of pregnancy tumor was rendered. Sturge-Weber syndrome, a disorder often associated with facial port-wine stains, had been ruled out due to a lack of other clinical symptoms characteristic of the syndrome, such as neurologic and ocular abnormalities. A panoramic radiograph depicts interproximal bone loss between teeth numbers 13,14 and 15 (international nomenclature) and teeth numbers 14 and 15 were less mobile and therefore were maintained.

Local anesthesia was administered to the patient to anesthetize the area. The most prominent sublesion of the right posterior maxillary buccal and palatal gingiva was excised in full thickness fashion along with a cuff of healthy tissue. The flap was closed primarily without tension. The excised specimens were sent for histopathologic review (Figure 1C). Histopathologic analysis revealed hyperplastic squamous epithelium with foci of ulceration overlying granulation and edematous fibrous tissues with severe acute and chronic inflammatory cells infiltrate (Figure 1D).

A





Figure 1. (A) extraoral photography and presence of port-wine stain on the right side of the patient, (B) intraoral photography at initial presentation with patient in maximum open, (C) intraoral occlusal view of all right side maxillary teeth after excision of lesion and (D) histologic picture of lesion.

Discussion

Pyogenic granuloma is a nonneoplastic, benign inflammatory lesion involving the skin and oral cavity. Skin lesions are more common than those of oral mucosa. When present in the oral cavity, it most commonly involves the gingiva with its prevalence in women being twice than that of men, and usually occurring during the second decade of life [5]. Estrogen and progesterone increase expression of angiogenic factors and decrease granuloma cell apoptosis. Increased blood flow to the

region of the pyogenic granuloma through the enlarged capillaries of the port-wine stain may have predisposed the patient to the development of the pregnancy tumor as imaging showed a prominent facial artery [3,6].

Management guidelines for granuloma of pregnancy have been reported [7]. If possible, it is often favored to delay intervention until postpartum period since lesions can resolve when hormones stabilize. However, for large lesions that bleed and interfere with function, treatment should be rendered.

Various suggested treatment options include curettage, cryotherapy, laser ablation, sclerotherapy, corticosteroid injection, and surgical excision. Pyogenic granuloma associated with port-wine stain often recur and, therefore, complete surgical excision is generally favored [3]. Complete surgical excision of the lesion is commonly preferred for the treatment of recurrent pyogenic granuloma associated with port-wine stain, which is generally associated with recurrence. Curettage, cryotherapy, sclerotherapy, and lasers can also be used as alternate treatment modalities [8]. Ethanolamine (sclerosing agent) has been successfully used for the treatment of recurrent pyogenic granuloma associated with port-wine stain [4]. In this case we chose surgical excision and curettage for the treatment and all of the teeth were maintained and the patient was followed.

Conclusion

This patient's pregnancy tumor, located in the region of a right facial port-wine stain, was determined to be the result of hormonal influences from her pregnancy and vascular anastomoses to the region from the port-wine stain. Pyogenic granuloma in association with port-wine stain can be more resistant to standard treatment. In conclusion, clinicians and dentists should be aware of this possible association (pyogenic granuloma, recurrent pyogenic granuloma and port-wine stain to plan the patient's treatment.

Conflict of Interest

There is no conflict of Interest to declare.

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