SUMMARY

Nevus flammeus, which is also known as port-wine stain (PWS), is one of the vascular birthmarks. PWS occurs in 0.3% of the newborns in both genders. It is a capillary vascular malformation, characterized by a pink or red stain and may involve skin, soft tissue and/or bone. There are a very limited number of reports regarding intraoral involvement of PWS. We report 2 female patients with PWS from date of birth. The first patient was an 11-year-old female who applied to our clinics for the treatment of her non-aesthetic and deviated intraoral view and discoloration of her gingiva, and the second patient was a 56-year-old female who applied for the extraction of her wisdom tooth. Extraoral examination in both patients revealed a diffuse PWS on the right side of their face over the cheek, extending from the midline. While the first patient had reddish skin, gingiva on right site her both jaws and lips, the second patient had only her upper jaw and lip. Because of the first patient’s age, the treatment postponed to her 20’s, and the second patient did not accept any treatment. PWS is a rare and non-fatal condition; however, the unique appearance of these patients can lead to psychological problems especially in early ages.

Keywords: Port-Wine Stain; Gingiva, pigmentation

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CASE REPORT (CR)

Gingival Tissue Alterations in 2 Patients with Port Wine Stain

Introduction

Port-wine stains (PWS) are one of the vascular birthmarks and also known as nevus flammeus¹. PWS occur in 0.3 percent of the live newborns, affecting females and males equally in different racial ethnicities¹-⁶. PWS are congenital and superficial vascular and slow-flow capillary malformations. Adults may have raised papule or nodular components that are associated with soft tissue overgrowth⁶. They may also develop varicosities or granulomas. PWS are often located on the face and follows the trigeminal nerve. It is appearing at birth as a diffuse light pink macula and tends to become darker and thicker with age⁷. Unlike hemangiomas, PWS generally do not fade over time, and do not disappear; on the contrary, PWS expands while child is growing. PWS do not need treatment, but pulsed dye laser therapy is thought to be gold standard therapy for lightening lesions¹.

Histologically, PWS are clusters of superficial capillaries that are abnormally increased in size and number in the papillary layer of dermis. The pathogenesis of PWS is controversial. There is a progressive increase with age in both size and number of the capillaries that can result in disfiguring lesions. There are many negative psychosocial implications for a child growing up with such a facial birthmarks⁶.

Pigmentation is discoloration of the oral mucosa or gingiva due to wide variety of lesions and conditions. Oral pigmentation occurs in all races and ethnic groups and gingiva is the most frequently pigmented intraoral tissue. The most of the complaints and requests for treatment of gingival pigmentation arise from aesthetic reasons⁷. The prevalence of gingival pigmentation was higher on the labial part of the gingiva than on the buccal and palatal/lingual parts of the jaws. The shade of pigment was classified as very dark brown to black, brown, light brown-yellow⁸.

We report the cases of 2 female Turkish patients (11 and 56 years of age, respectively) with PWS.
Case Reports

Case 1
An 11-year-old Turkish girl presented to Kocaeli University, Faculty of Dentistry for the reconstruction of her unsymmetrical facial and intraoral view. During the extraoral examination, a diffuse port-wine staining on the right side of her face over the cheek, which was extending from the midline and the forehead to the upper and the lower lips, was noted (Fig. 1). Her parents reported that the stain appeared at birth and became darker.

Intra-oral examination showed that the gingival tissues of the right upper and lower jaws were reddish and darker than the left side (Fig. 2, a and b). Asymmetrical intraoral view was noticed. The upper right teeth and gingiva were positioned more coronal compared to the left side. Oral hygiene was good. Radiological examination revealed normal bone and jaw structures.

Due to her younger age, no treatment was planned for pigmentation and asymmetrical intraoral view.

Case 2
A 56-year-old Turkish woman applied to Kocaeli University, Faculty of Dentistry for the extraction of her wisdom tooth due to severe pain. She had a diffuse PWS on the right side of her face over the cheek, which was extending from the midline and from the forehead to the upper lip (Fig. 3). She stated that she already had PWS when she was born.

In the intraoral examination, it was seen that the gingiva of right side her upper jaw was more reddish than the left side (Fig. 4a). It was also seen that the right side of the palate was pigmented (Fig. 4b). This pigmentation has a perfect line through the central incisors. Radiological examination revealed normal bone and jaw structures.

She did not have any complain about her non-aesthetic facial view, and did not accept the treatment of her gingival pigmentation.
Discussion

PWS presents hypertrophy and progressive vascular dilation with age if it is not treated. PWS can be treated by curettage, cryotherapy, dermabrasion, tattooing, chemical cauterization, electric cauterization, photodynamic therapy, intense pulsed light, spectrophotometric devices, which are specially, design for PWS, cosmetic makeup; however, current standard treatment is laser. Early intervention during infancy is crucial for better results. Treatment with laser therapy also decreases the development risk of pyogenic granuloma. While the pulse-dye laser is gold standard therapy for PWS, argon lasers are one of the first lasers, which are used for treatment of PWS. Treatment is mostly well tolerated and is successful in approximately 70% of individuals. Laser treatment of haemangioma can be lighted the PWS since PWS therapy is resulted with reduction in the number of vessels; however, complete eradication of the PWS lesion is not warranted. Radiation therapy, smoking, UV exposure would be the reason for the development of the skin cancer in PWS.

A simple and effective technique for removal of gingival pigmentation is surgical excision of the epithelium and pigmented gingiva; there are many other therapeutic approaches for treatment of gingival pigmentation such as, cryosurgery, chemical abrasion, free gingival grafts, gingivectomy and laser. PWS involving gingiva does not require treatment, except for the aesthetics demands.

References

Conflict of Interest: The authors declare that they have no conflict of interest. No external funding, apart from authors’ institution, was available for this study.

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