Case Report

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Cryotherapy as a conservative treatment modality for gingival enlargement in a patient with Sturge-Weber Syndrome

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Summary **This case report describes a case of Sturge-Weber syndrome reported for unilateral gingival enlargement and bleeding from gingiva in maxillary left region. Initial treatment in the form of scaling and root planing was done but recurrence was observed after one year of follow up. Instead of performing conventional surgery, an alternative conservative treatment was planned in the form of cryotherapy with the help of closed nitrous oxide probe. Seeing the satisfactory results obtained, cryotherapy can be suggested as an atraumatic, bloodless and effective chair side procedure for treating vascular gingival enlargement.**

Keywords: Cryotherapy, hemorrhage, nitrous oxide, gingival enlargement, Sturge-Weber syndrome

1. Introduction

Sturge-Weber syndrome, a rare neurocutaneous disorder belongs to the group of neuroectodermal development anomalies called "phakomatosis" and is characterized by venous angiomas of leptomeninges over the cerebral cortex with ipsilateral angiomatous lesions of face, skull, jaws, and oral soft tissues (*1*). Facial lesions known as port wine stains include rosy-purple nevus flammeus lesions that are sharply demarcated, usually flat and occur unilaterally. Intraorally, angiomatosis on buccal mucosa and lips may present as purplish-red discoloration, which may involve soft palate, tongue, floor of mouth, and gingiva. Gingival lesions range from slight vascular enlargement to large growths.

Treatment modalities for vascular gingival enlargements include conscientious observation, radiation therapy, steroids, antimetabolites, sclerosing solutions and surgical removal of gingival overgrowth with electro-surgery or laser (*2*). Cryotherapy has been used in treatment of keratotic, hyperplastic, granulomatous, vascular, pigmented and salivary gland lesions (*3*). However, an established protocol and description of the procedure for diffuse vascular gingival enlargement is lacking in dental literature.

This case report describes the treatment of vascular gingival enlargement with cryotherapy and discusses the intricacies involved in management of oral manifestations of Struge-Weber syndrome.

2. Case Report

A 12-year-old female patient was referred to Department of Periodontics with swelling in left side posterior region and spontaneous bleeding from gums for last three years. Medical history revealed that she was suffering from Sturge-Weber syndrome and was on antiepileptic drug (phenytoin sodium) from age of six months. This drug was changed to sodium valproate since last two years. Extraoral examination revealed presence of port wine stains, facial asymmetry due to hemihypertrophy of left side of face and increase in nasal bridge width (Figure 1A). Intraorally bright red discoloration of alveolar mucosa and enlargement of gingiva was observed in maxillary left region from

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Figure 1. Clinical photographs. (A) Extraoral photograph showing facial asymmetry, portwine stains on the left side of face and increase in nasal bridge width; **(B)** Preoperative intraoral view showing gingival enlargement in maxillary left region and bright red discoloration of alveolar mucosa; **(C)** Normal contralateral side.

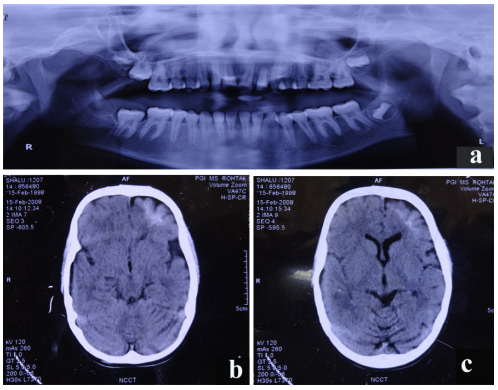

Figure 2. Panoramic radiographic and computed tomographic images. (A) Panormic radiograph showing no alveolar bone loss with respect to involved segment; **(B)** Computed tomographic scan showing hemiatropy of left cerebral hemisphere along with calcifications; **(C)** Normal tomographic findings on right side.

central incisor to second molar (Figure 1B). Enlarged gingiva covering more than one third of tooth structure extending from first premolar to second molar teeth was reddish pink, soft in consistency, non-tender and blanched on pressure. Gingiva and alveolar mucosa was normal on contralateral side (Figure 1C).

Radiographs showed no alveolar bone loss with respect to involved segment (Figure 2A). The complete hemogram was within normal limits. Computed tomography scan done in early childhood revealed subcortical gyriform calcification in left frontal lobe with prominent adjacent sulci representing mild cortical atrophy (Figures 2B and 2C).

A thorough plaque control regimen (scaling, root planing, oral hygiene instructions) was initiated to minimize gingival inflammation. After one month, gingival enlargement had decreased marginally and no bleeding was reported. After one year recurrence of gingival enlargement was observed in maxillary left region (Figure 3A). Cryotherapy, a non-invasive treatment option was planned as the conventional surgical intervention could not be carried out because

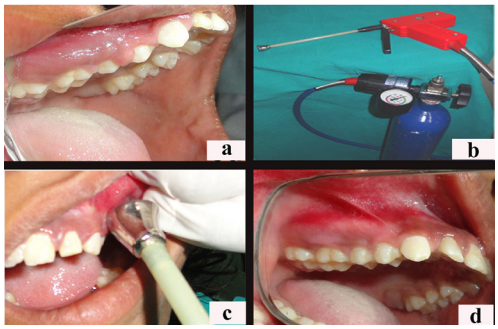

Figure 3: Clinical photographs and Cryotherapy apparatus. (A) Recurrence of gingival enlargement after one year of oral prophylaxis; **(B)** Cryotherapy apparatus showing nitrous oxide cryogun and cylinder; **(C)** Use of nitrous oxide cryoprobe to treat the gingival enlargement; **(D)** Complete resolution of gingival enlargement after one year of cryotherapy.

of risk of hemorrhage and patient's reluctance. The procedure was carried out, under topical anesthesia (2% lignocaine hydrochloride gel), using a closed nitrous oxide probe (Figure 3B) with a freezing cycle of 30 seconds and then thawing for 1 minute. This process was repeated twice with overlapping freezes (Figure 3C).

No postoperative pain and complications were seen on second day. Re-epithelialization of the lesion occurred at seventh day of follow up with remarkable regression of gingival enlargement. Complete regeneration and regression of enlargement was observed after one month. Resolution of pseudo-pockets was noticed and no recurrence of gingival enlargement was found after one year of follow up (Figure 3D).

3. Discussion

Dental rehabilitation of patients with Sturge-Weber syndrome is a complex process requiring initial conservative management and later surgical intervention. However, this has to be exercised with caution because the tissues may bleed profusely intraoperatively and postoperatively.

Cryotherapy is a method of lesion destruction through ischemic necrosis of the target tissue induced by rapid freezing in situ. This type of tissue destruction helps in bloodless field of surgery and is useful in treating vascular gingival enlargement. The basic technique involves rapid cooling, slow thawing and repetition of the freezing process to maximize tissue destruction (*4*). Most tissues freeze at -2.2°C, and tissue death occurs at a temperature of -20°C (*5*). Liquid nitrogen (-191°C), nitrous oxide (-80°C), and carbon dioxide (-79°C) are commonly used cryogens. The available cryotherapy apparatus is classified into open and closed systems. Open systems involve direct application of cryogenic fluid (usually liquid nitrogen) to the lesion with a cotton swab or spray. In closed systems the tissue is frozen by a cryoprobe

(*6-10*). Closed probe technique is useful as direct contact between cryogen and the tissue allows a more controlled and profound depth of freezing.

These probes follow the principles of Joule-Thompson expansion which enable substances to undergo a drop in temperature when moved from high pressure area to low pressure area. When nitrous oxide is released from high pressure inside the cryoprobe to the lower pressure cryotip, the drop in temperature allows freezing of the tissues to occur (*6,10*). Current protocols suggest for most benign mucosal lesions a 1-2 minute freeze/thaw cycle, for premalignant/malignant lesions three 2 minute freeze/thaw cycles and for smaller lesions, shorter freeze cycles (20-30 seconds) using a cryoprobe are adequate (*6,10*).

Ichingolo *et al*. compared traditional surgery, electro surgery, CO_2 and neodymium-doped yttrium aluminium garnet (Nd: YAG) laser in treating gingival hyperplasia in Sturge-Weber syndrome and found most encouraging results with Nd:Yag laser (*11*). In present case, cryotherapy was chosen because of its advantage of providing better hemostatic control which considerably reduced the surgical time along with faster and easier tissue healing as opposed to slow and complex healing in case of traditional scalpel surgery. After therapy, no pain was reported because of blockage of neural transmission to the area and there was absence of secondary infection. Through our observations, we have found cryotherapy as an effective conservative method of management of vascular malformations. In conclusion seeing the satisfactory results obtained, cryotherapy can be suggested as an atraumatic, bloodless, economical and effective chair side procedure for treating gingival enlargement without resorting to knife surgery in patients suffering from Sturge-Weber syndrome.

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