ORIGINAL RESEARCH PAPER
Volume - 9 | Issue - 12 | December - 2020 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

CRYO-SURGICAL MANAGEMENT OF ANGIOMATOUS GINGIVAL OVERGROWTH ASSOCIATED WITH STURGE WEBER SYNDROME (SWS) - A CASE REPORT.

Dental Science

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ABSTRACT

A 22 year old male patient reported with extra oral unilateral port wine stain on the right side of the face with intraoral diffused angiomatous gingival overgrowth in the maxillary posterior region and palate on the right side. It is identifiedon Roach scale as type I Sturge Weber Syndrome (SWS).Due to the typical involvement of multiple feeder vessels in SWS, tendency towards excessive bleeding intrasurgically and higher recurrence rates reported, cryotherapy is opted as a novel treatment option. The treatment outcome achieved suggests that cryotherapy can be an effective treatment option in treating intraoral hemangiomas.

KEYWORDS

Sturge weber syndrome, Angiomas, Gingival Overgrowth, Cryotherapy

INTRODUCTION

Sturge-Weber syndrome (SWS), also called encephalotri geminalangiomatosis, is a neurocutaneous disorder with angiomas that involve the leptomeninges (leptomeningeal angiomas) and the skin of the face, typically in the ophthalmic (V1) and maxillary (V2) distributions of the trigeminal nerve.[1] The cutaneous angiomas which are called the port wine stains, usually occur unilaterally along the dermatomes supplied by the ophthalmic and maxillary division of trigeminal nerve[2] which may be bilateral or totally absent or may extend to neck, limbs and other parts of the body.[3] The stain, varying from light pink to deep purple, is due to an overabundance of capillaries just beneath the surface of the involved skin. Involvement of the area supplied by ophthalmic division is pathognomic and can result in glaucoma and choroidal hemangioma.[4] Approximately 5% of patients have associated ocular involvement, mental retardation, and seizures due to the involvement of the vasculature of eye and central nervous system.

On the other handintra-orallyangiomatosis may involve lips, buccal mucosa, palate, gingiva, and floor of mouth.[5,6] These lesions are highly vascular with rich capillarity under the epithelial bed resulting in greater bleeding tendencies. Thereforein most of the cases reported till date non-surgical management of the intra-oral lesions is considered the preferred treatment option. Diffused angiomatous gingival overgrowths are reported in most of the cases of SWS and are managed non surgically[7]i.e, thorough and repeated scaling and root planning and reinforcement of oral hygiene instructions to prevent the recurrence of inflammation and excessive bleeding. However, in few cases with unestheticexcessive gingival overgrowths and interference with masticatory function surgical excisions were performed.

Case report

A 22 year old male patient reported to the department with a chief complaint of gingival swelling and bleeding gums on the right upper arch. History revealed that the reddish discoloration (port wine stain) was present on the face since birth and was gradually darkening with age. Family history was non contributory.(Figure 1) There was no visible sign of mental retardation and patient had history of occasional convulsions. Extra oral examination revealed that the facial discoloration had unilateral distribution involving the right side of face extending from the middle of forehead and involved the eye, half of nose, cheek, philtrum, and right side of upper lip. The lower lip and lower jaw were not involved. There were whitish patches on right side of the face with scaly appearance.Examination of the eyes revealed that the blood vessels in the right eye were dilated, patient lost his eye sight at the age of 15. Left eye appeared normal.Intra oral examination showed a sharply delineated extension of port wine stain on the right side showing diffused involvement of gingiva on the buccal side, and extending from the gingival margin to the soft palate on the palatal side(Figure 2). The overgrowth was extending from mesial aspect of right second premolar to mesial aspect of third molar (23mm) and vertically extending beyond mucogingival line(10mm), while the left side appeared normal. Patients oral hygiene maintenance was fair with minimal deposits and moderate bleeding on probing was present on the involved side. Gingival overgrowth was bright red in color and showed blanching on applying pressure suggesting angiomatous enlargement. On application of pressure on the external carotid artery blanching of the lesion was observed suggestive of direct innervation of the major vasculature. History and clinical findings are suggestive of SWS, and later the same was confirmed on consultation with neurophysician. According to the Roach scale, it is identified as type I SWS as the patient had a port wine stain and glaucoma on the right side with occasional epileptic convulsions and angiomas on face and intraorally[8].

Figure 1: Extraoral photographs showing Port wine stain on the right side of face

Figure 2: Intra-oral photographs showing angiomatous gingival overgrowth on right maxillary buccal and palatal sides

MANAGEMENT

A thorough plaque control regimen including scaling, root planing, oral hygiene instructions were given to the patient to minimize gingival inflammation. On re-evaluation after one month period, gingival inflammation reduced marginally with slight reduction in bleeding. However, the lesion still persisted with no change in extent and appearance. The management of intra-oral hemangiomas usually involve the ligation of feeder vessels after detection with contrast radiography followed by subsequent excision either surgically or by laser therapy. However in the current case due to involvement of multiple feeder vessels, the inability to detect the vessels and due to the probable difficulty in controlling hemorrhage during surgery, cryosurgical management of the lesion was considered as the treatment of choice.

The procedure was carried out, under topical anesthesia with 2% lignocaine hydrochloride gel. A closed nitrous oxide probe was used with a freezing cycle of 20 seconds over the entire area of the lesion and then thawing for 1 minute.(Figure 3) This process was repeated twice with overlapping freezes. Two weeks after therapy size of the lesion was considerably reduced. One month after complete healing the same protocol was repeated. On re-evaluation after 1 month complete remission of the lesion was clearly evident. Post operative evaluation at 1 month showed no signs of recurrence of the lesion.(Figure 4)

Figure 3: Cryosurgical management of gingival overgrowth

Figure 4: Re-evaluation at 6 months

DISCUSSION

SWS is caused by residual embryonal blood vessels and their secondary effects on surrounding brain tissues and the vascular plexus supplying the various parts on one side. Angiomas of SWS usually result from failure of regression of a vascular plexus, thus resulting in formationof angiomas of leptomeninges, face, ipsilateral eye and intraoral tissues. Although the leptomeningeal angioma in SWS is typically a static lesion, in few cases it is progressive in nature.[9]

Oral manifestations reported in 38% of cases includehemangiomatous lesions in the lip, oral mucosa, gum, tongue and palatine region.[10]These manifestations are generally unilateral, diffused and localized to few teeth or terminate abruptly in the midline. The greater frequency of intra-oral manifestations of SWS demands a need for knowledge of the clinical presentation of the disease and various treatment modalities. Diagnosis and dental management should be done after consulting with the specialists. The conventional management of angiomatous intra-oral lesions associated with SWS is usually non-surgical, while surgical management resulted in excessive bleedingin few cases.[11,12]

Cryotherapy, also known as cryosurgery, is a commonly used in-office procedure for the treatment of a variety of benign and malignant lesions. It is the second most common in-office procedure after skin excision for management of various extra-oral lesions in cosmetic dentistry. The principle in cryotherapy is necrosis of the tissue, which results from the freezing and thawing of cells. Adverse effects of cryotherapy are usually minor and short-lived.The current treatment modality resulted in uneventful healing with no intrasurgical and post surgical complications with good patient acceptance.

CONCLUSION

Based on the clinical outcome, Cryotherapy could be an effective and alternative conservative management of intra-oral angiomatous lesions associated with SWS.

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