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RESEARCH ARTICLE

LOCALISED JUVENILE SPONGIOTIC GINGIVAL HYPERPLASIA: A RARE CASE REPORT

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Abstract

Juvenile spongiotic gingival hyperplasia is a benign lesion was first described by "Darling" and "Cols" in 2007.Various etiopathogenic factors have been proposed for its presentation. It is characterized by being in the anterior attached gingiva of the maxilla, clinically can be appreciated as macular or elevated, with a papillary, granular, or smooth surface, bright red in color, and its presentation can be localized or generalized. It is an asymptomatic entity and is not associated with biofilm-induced inflammation. It is important for the dentist to recognize this entity and to make the different differential diagnosis and propose proper diagnosis and treatment planning.

Introduction:-

Localised juvenile spongiotic gingival hyperplasia (LJSGH) is an unusual gingival lesion with unique predilection for anterior gingiva in children and adolescents. It affects second decade of life with females getting more affected than males in ratio of 2:1. Its incidence has been very rare and represented only 0.069% in about 32,000 biopsies.[1,2]

Earlier called as Juvenile spongiotic gingivitis was describedfor the first time by Darling et al. in 2007 as benign lesion.Later on, with further research all these lesions were seen associated with localised gingival erythema andovergrowth rather than a pure inflammatory lesion Chang et al recommended to use the term "localised juvenilespongiotic gingival hyperplasia" (LJSGH) to define moreaccurately this condition in 2008.

Clinically it appears as a solitary gingival mass with granular surface and bright red colour. It is usually painless but has increased propensity for bleeding. Most common site of gingiva getting affected by LJSGH is the attached gingiva but variable marginal involvement can also be appreciated.[3]

Histopathologically, it is characterised by epithelial hyperplasia, spongiosis of spinous layer acanthosis, increased exocytosis of neutrophils, lymphocytes, plasma cells with interstitial edema.[4] The pathogenesis remains far from understanding however its derivation from exteriorized JE due to lack of space after primary teeth exfoliation is

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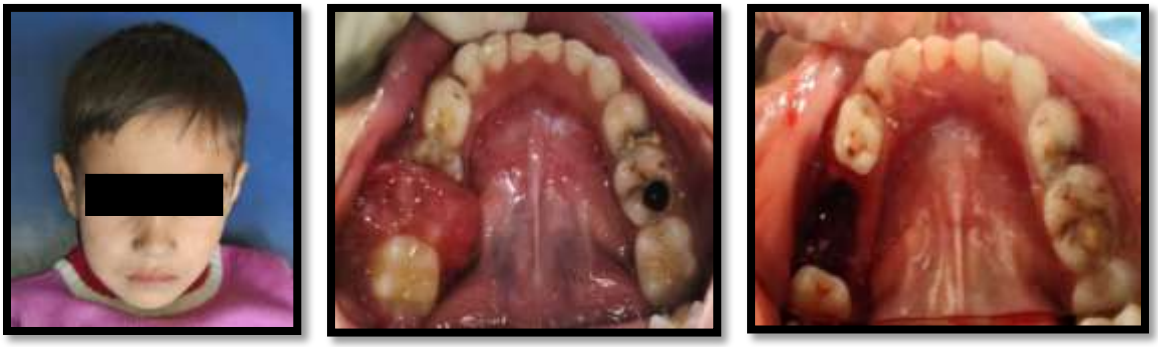
favoured as the two have many morphological and immunophenotypic similarities.[5]Recentimmunohistochemical findings further support this theory, since the pattern of cytokeratin (CK) expression in LJSGH

(CK1/10, CK4, CK8/18, and CK19) is evocative of JE profile. Plaque induced inflammation is not considered as the etiology for this lesion as there is lack of response to improved oral hygiene and does not regress with subsequent periodontal treatment.[2]

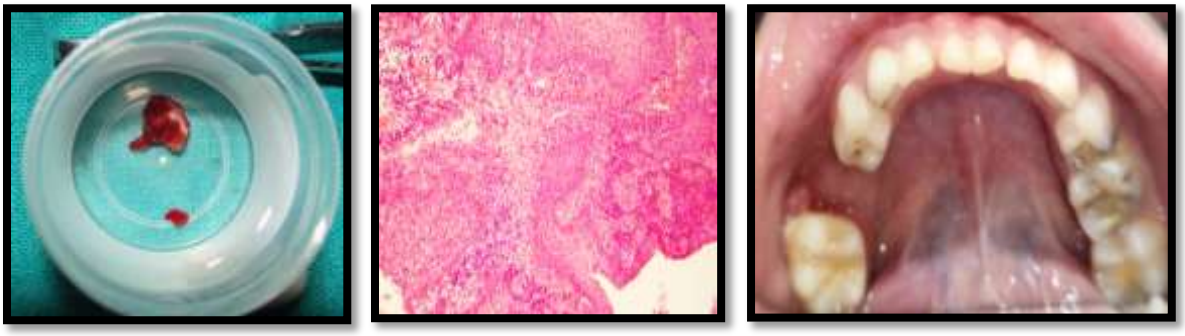
Case report

Aseven-year-old female patient reported to Pediatric Dentistry departmentinGovernmentDental College Srinagar for evaluation of erythematous overgrowth on right mandibular posterior region of 85. The lesion was initially of small size and increased gradually with extraoral swelling andhigh grade fever for about one month. On clinical examination, the lesion was painless, pedunculated involving marginal as well as attached gingiva on both sides of 85, bled profusely on manipulation, granular on appearance and oedematous in consistency. The oral hygiene of patient seemed satisfactory with non-contributory family or past medical or dental history.

Patient was advised for an IOPAR and OPG that revealed chronic pulpal and periapical involvement of 85 with extensive resorption seen. Based on clinical and radiographical examination, it was provisionally diagnosed as pyogenic granuloma. Differential diagnosis for the lesion can be puberty gingivitis, pyogenic granuloma, peripheral giant cell granuloma, human papilloma virus (HPV)-related lesions, foreign body granuloma, small superficiallymphangioma.



An incisional biopsy was carried out to confirm the diagnosis and perform appropriate treatment and so the specimen was sent to histopathological evaluation. However, it was confirmed to be a localised juvenile spongiotic gingival hyperplasia despite its unique presentation in the posterior area as an extensive lesion. Then, an excisional biopsy was planned and conducted. The entire lesion was excised completely and the area was debrided copiously with saline. Antibiotics and analgesics were prescribed and patient was recalled after one week for follow up.



Discussion:-

LJSGH was originally described by Darling et al. as juvenile spongiotic gingivitis as it was commonly found affecting young patients. However later many studies emphasised upon being this lesion as LJSGH due to overgrowth of gingiva in localised areas especially attached gingiva generalised involvement has not been documented with this lesion ever.[3,6]

It is a very rare condition not known by many dentists and literature reports presence of about 221 cases globally as of now. Regarding epidemiology there are contrasting views, Vieira et al reported no gender preponderance however DeSetaet al found female predilection of 2:1 along with presentation in first two decades of life.[7,8,9]

The pathogenesis is not clearly understood but various mechanisms have been proposed like viral etiology, increased estrogen or progesterone, allergic, bacterial, trauma etc.[10]The role of puberty, instead, is controversial. It is mainly based on its infrequentfinding in adulthood, but the absence of estrogen andprogesterone receptors in the lesion, its localized feature, thepossibility of affecting children in prepubescent age in aconsistent number of cases do not agree with a potentialeffect of sex hormones.[10,11]

Histopathological features found in this case included loss of keratinisation of the stratified squamous epithelium,epithelial hyperplasia with a papillary architectureand spongiosis, prominent intercellular oedema and neutrophilexocytosis which is consistent with that reported by Allon et al in his case of LJSGH.

Since the lesion is most often smallin size thus conservative approach with spontaneous resolution is considered however literature has shown increased chances of recurrence in such cases. Apart from this, large extensive lesionsusually require invasive treatment but with that also recurrence rate of about 6-16% has been reported. Other treatment modalities that can be executed are laser therapy, biomodulation, cryotherapy which can effectively be used in pediatric population due to ease of application, absence of discomfort and minimal bleeding.[11,12]

Mawardi et al published a systematic review in which it was found that 97 cases were treated by surgical excision, 2 with cryotherapy, one with photodynamic therapy and one with superficial cauterization with clobetasol. 12 cases showed recurrences after treating by surgical excision however none reported to recur after using other modalities. In this case report under study the proposed etiology could be chronic source of infection and sharp edge of 85 which could have stimulated the gingival hyperplasia. It was managed using surgical excision due to many published reports showing success using this modality and lack of enough evidence regarding other approaches. There was no evidence of recurrence after following patient for about a year which can be attributed to pathogenetic hypothesis ofodontogenic developmental lesion that regress withthe passing of the developmental age.

Conclusion:-

It can be concluded that this lesion is a rare lesion requiring further investigations to confirm clinical and histopathological findings and exploring various alternative approaches for managing such lesions, minimizing recurrence and improving prognosis.

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