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RESEARCH ARTICLE

UNILATERAL EXTENSION OF CAPILLARY HEMANGIOMA INVOLVING ORAL CAVITY – A RARE VARIANT

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ABSTRACT

Capillary haemangiomas are benign vascular tumours uncommonly encountered by the dentists. Capillary haemangiomas are developmental hamartomatous lesions of vascular tissue. Although most commonly haemangiomas may appear on the head and neck region, they very rarely manifest in the oral cavity. This article presents a rare case of capillary haemangioma showing unilateral involvement of oral cavity.

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INTRODUCTION

Haemangiomas are one of the rare benign oral tumours occurring due to dilatation of blood vessels.[1,2,3] They may be present at birth or may manifest during early years of childhood.[3,4]Approximately 80% of haemangiomas appear as a solitary lesion, but the rest 20% of may appear as multiple tumours.[5] The incidence of capillary haemangiomas varies from 0.5 to 1% of all the oral tumours.[6,7]Haemangiomas are most commonly seen in the head and neck region but they rarely occur in the oral cavity making it a rare variant.[1,8] The origin and pathogenesis of haemangioma is still controversial but it is said that haemangiomas contain numerous blood vessels with hyperplastic fast growing endothelial cells with a female predilection and is most commonly seen in Caucasians and individuals with low birth weights.[9] A haemangioma is a smooth, soft, lobulated growth or mass which may be either pedunculated or sessile.

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It can be of varied sizes ranging from a few millimetres to a few centimetres. It may vary in colour depending on its depth in the tissue from being pink to red or violet and appearing blanched when pressed.[10,11] Haemangiomas arising in the oral cavity usually appear on the gingiva.[10] No special treatment is needed for most patients with haemangiomas as they will heal and disappear spontaneously but a proper follow up does play a major role.[12] Haemangiomas which occur on the surface can easily be removed but deep lesions need a proper intervention [13]In this case report, we present a case of a 19 year old male patient who presented with enlargement of lower lip since birth.

CASE REPORT

A 19 years old male patient reported to the Department of Oral Medicine & Radiology with a chief complaint of enlargement of lower lip since birth. Patient's parents noticed an enlargement of his lip soon after his birth. Hence they took him to a private physician where he was advised some medications but he got no relief. Patient noticed that the enlargement of his lower lip was increasing with the increase in his age.

Hence, patient went to a private physician 2-3 yrs back, where debulking of the enlargement of his lower lip was done. Further,1 year post surgery, there was similar enlargement involving his lower lip, which was slowly increasing in size since then. There was no history of lip bitting. There was no relevant history of any other systemic diseases and his past dental history was not contributory. On extra oral examination, port wine stain was present on the skin of left side of lower side of face extending posterior to the ear & on the neck involving the upper part of left side of chest. (Figure 1) Patients face was bilaterally symmetrical and no abnormality was present in the temporomandibular joint. His lower lips were everted and incompetent due to diffuse swelling on lower lip. (Figure 2) On inspection, the swelling was diffused and extended anteroposteriorly from right corner of mouth to left corner of mouth & superoinferiorly from vermillion border of lip to the depth of labial vestibule. Swelling was reddish pink in colour with well-defined edges, surface was lobulated and indentations of upper lip were present on the swelling. On palpation, the swelling was non-tender, compressible, soft with well-defined borders, pulsations were felt on swelling and surface texture was rough. No discharge was present from the swelling.

Figure 1. Showing port wine stain on the skin of left side of lower side of face extending posterior to the ear & on the neck involving the upper part of left side of chest

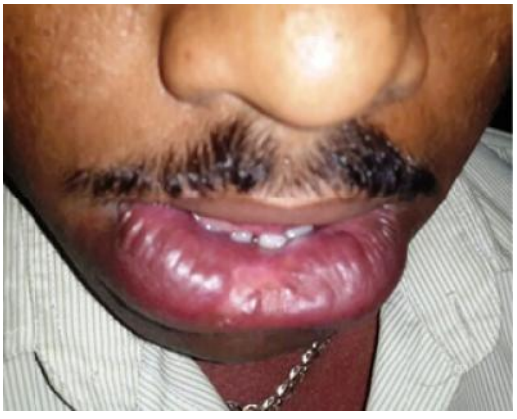

Figure 2. Everted and incompetent lower lip due to diffuse swelling

Figure 3. Showing spacing in the maxillary and mandibular anterior teeth

On intraoral examination, the hard tissue examination revealed presence of all permanent teeth. Spacing was seen between maxillary and mandibular anteriors (Figure 3). The gingivaappeared pale pink in color, with a scalloping contour, consistency was firm and resilient and there was generalized bleeding on probing of gingiva. Gingival recession was seen with maxillary anteriors. A provisional diagnosis of capillary haemangioma was made due to its clinical appearance and its growth involving the left side of face, back of the ear, neck, upper part of chest, lower lip, labial mucosa, floor of mouth, left side of soft palate, pharyngeal mucosa, ventral surface of tongue, buccal mucosa, alveolar mucosa on right & left side of mouth.

Figure 4. (A) papules on left buccal mucosa, (B)reddish papules on the lower labial mucosa, (C)reddish papules on left side of soft palate & pharyngeal mucosa, (D) papules on upper labial mucosa, (E) papules on right buccal mucosa

Patient was advised further investigations including PA Skull and MRI of head & neck region.PA skull did not show any significant findings. (Figure 7A) Post contrast MRI scan revealed a significant swelling of lower lip showing enhanced soft tissue (Figure 7B)The lower lip showed significant enhancement as compared to the rest of the soft tissues of face with vascular supply appearing to come from the facial artery. This may represent a slow flow vascular malformation.

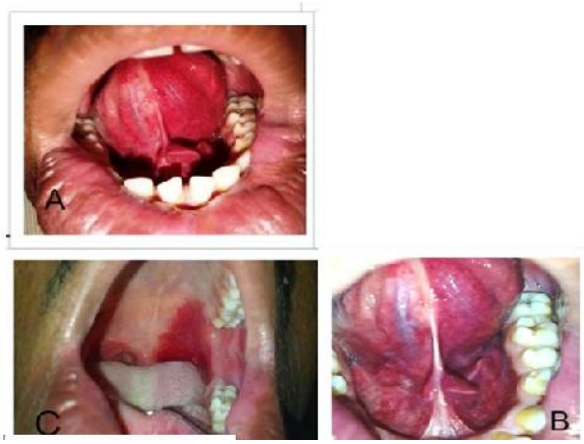

Figure 5. (A) showing diffuse swelling on left side of ventral surface of the tongue, (B) Reddish discoloration on floor of mouth, (C) Melanotic pigmentation seen over palate.

Figure 6. Reddish discolouration on (A) left lower buccal vestibule. (B) lower labial vestibule, (C) right lower buccal vestibule

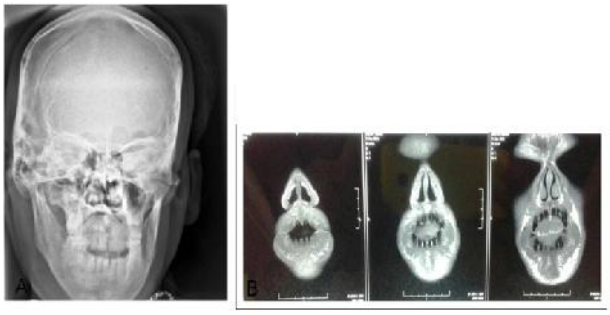

Figure 7. (A) PA skull showing no significant radiographic findings. (B) MRI scan showing significant swelling of lower lip with enhanced soft tissue within on post contrast examination

Hence a final diagnosis of Capillary Haemangioma was made on the basis of clinical and radiographic findings. Therefore, for esthetic correction a surgical treatment plan was planned for the patient which involved surgical debulking of lower lip. The patient was followed up regularly. On soft tissue examination, reddish papules were seen on the lower labial mucosa, left buccal mucosa, soft palate & pharyngeal mucosa on the left side of mouth (Figure 4A,4B,4C,4D,4E) The tongue appeared to be deviated on the right side with a normal dorsal surface.

However, the ventral surface of the tongue showed diffuse swelling on left side which was reddish in color, soft in consistency & non tender on palpation. Reddish discoloration was seen on the floor of mouth (Figure 5A, 5B, 5C, 5D). Reddishdiscoloration was also seen on alveolar mucosa of the lower labial vestibule and over right & left lower buccal vestibule (Figure 6A, 6B).

DISCUSSION

The word haemangioma is derived from a Greek word termed, haem- 'blood', angio –'vessel', oma-'tumour'.[3] One of the most common congenital abnormalities are vascular lesions.[14,15]Haemangiomas occur as solitary lesions in 80% of cases. The female to male ratio of capillary hemangioma is 3:1 and is commonly seen in Caucasians individuals. [16] Though the exact etiology of hemangioma is not known, it is believed by some of the authors and researchers that it is not a true lesion, but rather a hamartoma or some kind of a developmental anomaly. There are two types of vascular lesions namely haemangiomas and vascular malformations.[3]Haemangioma is a term which may be used to describe large number of vasoformative tumours. These are benign proliferation of blood vessels. Oral capillary haemangiomas lack a definitive criteria for the diagnosis and treatment.[18]Though benign these vascular tumours can cause disfigurement and life threatening issues. Haemangiomas are considered to be benign tumours of infancy (7%), affecting approximately 12% of whites, but rarely affecting individuals with dark skin. The incidence of intraoral capillary haemangiomas is extremely rare and may vary from 0.5-1.0% of all intraoral tumours.[22, 23, 24]

Nayouki Matsumoto et al., when studied 31 cases of intra oral capillary haemangioma found that most lesions of haemangioma were located on the buccal mucosa (45.2%), the tongue (35.5%), lip (9.7%), gingiva (6.5%), and palate (3.2%)[25.] Haemangiomas can be classified into three types: capillary haemangioma, cavernous haemangioma and mixed haemangioma. Out of the three variants of haemangioma, capillary haemangioma is commonly seen in the head and neck region, however, its occurrence in the oral cavity is very rare. Capillary haemangiomas contain small capillaries lined by endothelial cells drowned in a connective tissue stroma showing a rapid phase of growth following gradual involution.[19 , 20 , 21] Clinician may face a diagnostic dilemma while diagnosing a haemangioma as it may mimic other conditions clinically, radiologically as well as histopathologically. Since many years,clinicians have remained confused due to the vascular anomalies of head and neck region as a result of which they have faced difficulty in doing studies and have failed to derive a proper diagnosis and proper treatment plan. The differential diagnosis of haemangiomas includes pyogenic granuloma, peripheral giant cell granuloma, chronic inflammatory hyperplasia, epulis granulomatosa and squamous cell carcinoma.[26] Pyogenic granuloma most commonly affects the gingiva and is of two types when classified histologically as Lobular Capillary Haemangioma (LCH) type and the non- LCH type. The LCH type is characterised by proliferation of blood vessels organized in aggregates of lobules while the non-LCH type of pyogenic granuloma is characterised by vascular proliferation that may resemble a granulation tissue. Since Capillary Haemangioma and Pyogenic Granuloma possess similar clinical characteristics, it's very important to take a biopsy of

the lesion in order to come to a proper definitive diagnosis. The management of oral haemangiomas is dependent on various factors like the age of the patient, the area of occurrence and involvement of the lesion, size of the lesion and clinical aura of haemangioma.[27] Surgical excision with or without ligation of blood vessels along with embolization remains the treatment of choice for treating oral haemangiomas.[28] Though some recent treatment modalities have also been developed which includes use of Nd:YAG laser, steroid therapy , electrosurgery, CO_2 laser, cryosurgery and use of sclerosing agents. The risk of profuse intraoperative and postoperative bleeding must be taken into consideration while treating surgically. The case discussed in this case report is of significance as there was unilateral involvement of the head and neck region along with involvement of the oral cavity making it a rare entity. In the present case, there was a gradual increase in the size of the lesion with increase in the age of the patient. There was diffuse non tender swelling on the lower lip.The lesions of haemangioma were manifested in the lower labial mucosa, left buccal mucosa, soft palate and the pharyngeal mucosa on the left side in this case. There was diffuse swelling on the ventral surface of the tongue on the left side along with reddish colour of the mucosa. The floor of the mouth and the palate were also affected. Port wine stain were seen over the skin on the left side of the head and neck region making it unilateral in appearance. The patient was advised to undergo some radiologic investigations which included PA skull and MRI scan. On MRI findings, swelling was noted over the lower lip area and enhancement as compared to rest of the soft tissues of the face with vascular supply appearing to come from the facial artery. In this case debulking of the lower lip along with aesthetic correction was advised to the patient along with a proper follow up. No recurrence is reported till date.

Conclusion

Oral capillary haemangiomas are quite rare and hence the dental surgeons must be aware about the risks associated while diagnosing and treating a patient of haemangiomas. There may be profuse bleeding intraoperatively and postoperatively and hence it is of utmost importance to keep in mind the complications and to treat haemangiomas accordingly.

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