A case report on capillary hemangioma and Leukoplakia on Tongue

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Citation

ABSTRACT
The following case report is about a female patient of age 50-years, who came to OPD with the chief complaint of nodular swelling on tongue and white patch on left lateral border of tongue of 1 year duration. She gave history of tobacco lime quid keeping habit along with betelnut chewing habit for 5-6 times/day since last 20 years. On the basis of clinical examination provisional diagnosis of hemangioma over tongue dorsum was given and pyogenic granuloma was kept in differential diagnosis. For the white patch on left lateral border of tongue and buccal mucosa a clinical diagnosis of leukoplakia was given. The case was treated by surgery of leukoplakic patch and nodule over tongue dorsum was managed by excision and placement of collagen sheath over the area was done. Oral leukoplakia is not commonly seen with hemangioma; however, it might be present quite before the development of leukoplakia.

Keywords: leukoplakia, hemangioma, vascular malformation
1. INTRODUCTION

The vascular anomalies are usually characterized by a proliferative phase and by involutive phase in haemangioma. Haemangioma accounts for 60% to 70% of the conditions in the head and neck area (Okaji et al., 2011). Recently, guidelines by ISSVA (International Society for the Study of Vascular Anomalies) are provided to differentiate vascular anomalies. According to the new classification given by Mulliken et al., in 1982, vasoformative tumours are classified into two groups into haemangioma and vascular malformation. Histologically, there are two types of haemangioma they are capillary and cavernous forms. Capillary haemangioma made up of numerous minor capillaries which are lined by an endothelial cells coating. Cavernous haemangiomas are consisted of vessels which are thin walled, large, or sinusoids that have epithelial cells lining. These are further separated the outside layer of connective tissue septa (Neville et al., 2005).

Oral leukoplakia has been defined as “a predominantly the whitish non-scrappable lesion of the oral mucosa that cannot be characterized as any other lesion.” The First International Conference held on oral leukoplakia (1984) Malmo, Sweden: defined leukoplakia as “A white non scrapeable patch or plaque that cannot be similar pathologically or clinically as any other disease and is not related with any physical or chemical contributing agent excluding the use of tobacco” (Warnakulasuriya et al., 2007); however, the leukoplakic lesion itself is a potentially malignant lesion hence to be observed carefully” (George et al., 2011; Masthan et al., 2015). Various treatment modalities are used for oral leukoplakia which includes surgery and systemic treatment like antioxidants, carotenoids, and antifungal therapies (Kayalvizhi et al., 2016). Lodi and Porter suggested that carbon dioxide (CO$_2$) LASER vaporization leads to maximum recurrence while use of CO$_2$ LASER evaporation leads to minimum recurrence of leukoplakia. The cryosurgery showed up to 22% and conventional blade surgery showed up 13% recurrence rates (Lodi & Porter, 2008).

2. CASE REPORT

A 50-year-old female patient farm worker by occupation, reported to OPD with a complaint nodular swelling on dorsal surface of tongue on left side and an elevated white patch since 1year. She was having habit of tobacco lime quid keeping habit along with betelnut chewing habit since 20 yrs for 5-6 times/day. On clinical examination, a single nodular sessile swelling approximately 1x1.5 cm in size was present on dorsal surface of left side of tongue, roughly oval in shape, which was interspersed with whitish patch the colour of the nodule was pale pink (Figure 1).

> Figure 1 Haemangioma on dorsal surface of tongue

The nodule was firm in consistency. It was fluctuant to in some area and non slippery. On asking leading questions she said that the nodular swelling was present since almost childhood. She gave no history of bleeding from the nodular lesion. There was no similar type of swelling present throughout the oral cavity. Taking into consideration of childhood history, the provisional diagnosis of haemangioma was given and pyogenic granuloma was kept in differential diagnosis. Diascopy was done which revealed negative results. There was a large, irregular white patch involving tongue dorsum and lateral border of tongue. The patch over the left lateral border of tongue was thick, elevated and showed cracked mud appearance and was non scrapable (Figure 2).

The elevated white patch initially was small in size and gradually increased to cover almost the entire half of the dorsum of tongue. Diffused whitish non-scrappable patches were also present on right and left buccal mucosa. The clinical provisional diagnosis of homogenous leukoplakic lesions on tongue, buccal mucosa and haemangioma on dorsal surface of tongue was given. After doing required investigations, the surgical excision of leukoplakic patch and nodule over left dorsal surface of tongue was carried...
out and placement of collagen sheath was done. The process of electro-fulguration of white lesions over bilateral buccal mucosa was also carried out.

**Figure 2** Leukoplakia on left lateral border of tongue

After completion of surgical treatment, the excised specimen was sent for histopathological examination. The histopathological report showed parakeratinized stratified squamous epithelium and underlying connective tissue stroma. The underlying lesional connective tissue stroma comprises of densely packed bundles of collagen fibers. Numerous endothelial lined blood capillaries of varying shape and sizes are seen with intravasated and extravasated RBC’s. Proliferations of endothelial cells are seen in connective tissue stroma. Mild chronic inflammatory cell infiltrate is seen (Figure 3).

**Figure 3** Showing Capillary hemangioma

All these features suggested capillary hemangioma for the nodule over the tongue dorsum on left side and white patch on tongue revealed hyper-ortho-keratosis whereas the white patches on buccal mucosa showed hyperkeratosis with chronic inflammation. The specimen of white patch on tongue: H and E stained tissue section showed hyperparakeratinized stratified squamous epithelium and underlying connective tissue stroma. The basement membrane was undamaged. The features of dysplasia were however confined to the basal one third of the epithelium. The underlying connective tissue stroma showed presence of subepithelial band of inflammatory cell infiltrate; haphazardly arranged bundles of collagen fibers and fibroblasts. Endothelial lined blood vessels were seen. Deeper part of connective tissue showed muscle tissue (Figure 4).

All these features suggested of Hyperkeratosis with mild dysplasia. Then patient was under observation for 7 days. The medicinal treatment in the form of tab augmentin 625 mg BD, tab Chymorol forte BD, tab PAN-D 40 mg BD for 7 days was given. Patient was
recalled after 7 days for follow up. The surgical site was healthy. The post operative photos of dorsal surface and lateral border of tongue (figure 5 & 6).

![Image](image1)

**Figure 4** Showing hyperkeratosis with mild dysplasia

![Image](image2)

**Figure 5** Post operative photo of tongue dorsum

![Image](image3)

**Figure 6** Post operative photo of lateral border of tongue

3. DISCUSSION

Hemangioma affecting the head and neck region is a true neoplasm. It usually appears a few weeks following birth and then grows rapidly (Kamala et al., 2011). They are characterized by endothelial cell hyperplasia (Glick & Feagans, 2015). Hemangiomas show
higher occurrence in females. The head and neck area is more commonly affected. The face, oral mucosa, lips followed by tongue and trunk (Slaba et al., 2010). In the present case, the nodular lesion present on tongue was sessile and was about 1x1.5 cm in size approximately. Clinically, hemangioma is characterized by sessile or pedunculated soft, smooth or lobulated lesion, and can vary in any size from a small lesion as few mm to a large one up to several cm. The color of this lesion can be pink to red purple which blanches on applying pressure. The hemorrhage can occur either spontaneously or sometimes after minor trauma and is usually painless (Fisher et al., 2005).

The leukoplakia is the most frequently occurring precancerous lesion. The rate of malignant transformation vary among various studies, probably because suspicion about the differential diagnosis and may be that different criteria utilized for the diagnosis, the follow up periods, the morbidity and mortality linked to oral cancer might make leukoplakia an important health problem (Chandak et al., 2017; Fisher et al., 2005; Scheifele & Reichart, 2003; Hamadah & Thomson, 2009; Patton et al., 2008). An augmented clinical thought may help accomplish earlier diagnosis by minimizing the causes for delay (Lohe et al., 2017). The female reported in this case was a farm worker with tobacco habit. Taking into consideration the widespread nature of the leukoplakic patch and poor socioeconomic status it was necessary to treat the lesion early with long term follow-up to avoid malignant transformation. The tobacco related habits as recognized risk factor, socioeconomic status can be considered as probable risk factor for oral squamous cell carcinoma (Lohe et al., 2017).

4. CONCLUSION
The soft tissue hemangiomas can occupy almost any location in the oral cavity. They can be seen either single or multiple. The differential diagnosis can range from lingual thyroid, fibroma, pyogenic granuloma to minor salivary gland tumours depending on its location. The deep seated lesions may not give rise to change in coloration hence leads to confusion. Oral leukoplakia is not commonly seen with hemangioma; however it might be present quite before the development of leukoplakia. In the case presented here, the presentation of leukoplakic patch on tongue dorsum was unusual and may show malignant transformation in future. Therefore a close post- treatment follow- up is mandatory.

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Conflict of Interest
The authors declare that they have no conflict of interest.

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Consent
Appropriate signed consent was taken from the patient before writing this case report (Identity of the patient was not revealed in this case report).

REFERENCES AND NOTES


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