Original Research Paper

Volume-9 | Issue-11 | November - 2019 | PRINT ISSN No. 2249 - 555X | DOI : 10.36106/ijar

Clinical Science

MASSON'S TUMOUR OF ORAL CAVITY: REPORT OF TWO CASES WITH REVIEW OF LITERATURE

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ABSTRACT

BACKGROUND: Intravascular papillary endothelial hyperplasia (Masson's tumor) is a rare, benign lesion of the skin and subcutaneous tissue with reactive papillary proliferation of vascular endothelial cells. It was first described by Masson in 1923.

CASE REPORT: Two cases with asymptomatic pigmented lesion at the right buccal mucosa, present since 20 years, reported to the OPD without any relevant history Inspectory findings revealed a bluish red, 2 x 2 cm sized lesion with no secondary changes of overlying mucosa. On palpation, the lesion was nontender, nonpulsating, noncompressible, nonreducible, rubbery in consistency and no blanching on pressure. Auscultation didn't reveal any thrill or bruit. Based on the clinical findings, a provisional diagnosis of 'pigmented lesion of right buccal mucosa' was made. Excisional biopsy of the lesion was carried out under local anaesthesia. Histopathological report described the lesion as hemangiomatous growth with intravascular papillomatous endothelial hyperplasia which is evocative of Masson's tumour.

CONCLUSION: Intra oral Masson's tumor is very rare and less than a hundred cases have been reported in the literature. Clinically and histopathologically these pigmented lesions may behave deceptively mimicking angiosarcoma.

KEYWORDS : Vascular Lesion, Hemangioma, Pigmented Lesion, Buccal Mucosa, Masson's Tumor

INTRODUCTION

Masson's tumour is a rare, benign, low flow vascular lesion encompassing approximately 2% of the vascular tumors of the skin and subcutaneous tissue [(1)]. It was first described by Masson in 1923 as '**hemangioendotheliome vegetant intravasculaire'**.[(2)] Subsequently, the lesion has been described by a variety of nomenclature but the most suitable and least confusing nomenclature for the lesion is **intravascular papillary endothelial hyperplasia (IPEH)**. This vascular lesion can develop anywhere in the body, but more common in head & neck region, trunk and extremities. Intra oral lesions are very rare and less than a hundred cases have been reported in the literature till date.[(3)]

Clinically IPEH manifests as a slow growing, firm nodule or mass which is slightly raised from the base, sharply demarcated and overlying skin is red or blue in color. Histopathologically it is characterized by endothelial papillary tissue growth within a luminal structure often coupled with a thrombus in various stages of maturation[(1)]. Though the lesion is clinically and histopathologically benign, proper diagnosis and understanding of the disease is imperative because, it is manifested as a pigmented mass which can be misinterpreted and wrongly treated as hemangioma or angiosarcoma and it tends to recur if incompletely resected[(4)].

In this present paper, two cases of intra oral Masson's tumor located at buccal mucosa are presented with their clinical and histopathological characteristics (**Table 1**).

Case report

CASE 1

A 48 years old lady, reported for evaluation of apparently asymptomatic, pigmented lesion at right cheek mucosa which was present for last 20 years. There was no history of trauma or bleeding from the lesion and there was no family history of similar lesion. On inspection, the lesion was approximately 2 x 2 cm in size, bluish red in colour, sharply demarcated from adjacent tissue and overlying mucosa was normal in appearance (**Fig 1A**). On palpation it was non tender, non compressible, non reducible, firm and rubbery in consistency and there was no blanching on pressure or pulsation. Auscultation of the lesion revealed no thrill or bruit. The lesion was mildly elevated from the surface**.** Diascopy test was negative.

A provisional diagnosis of 'a pigmented lesion of buccal mucosa' was made with differential diagnosis of 'capillary haemangioma', 'Kaposi's sarcoma' and 'blue nevus'**.** An excisional biopsy of the lesion was carried out under local anaesthesia (**Fig 1B and C**). Histopath ologically it turned out to be a hemangiomatous lesion mimicking an organized thrombus occupying a luminal structure with copious papillary tissues lined by endothelial cells without any frank dysplasia, mitosis or necrosis. The description was evocative of intravascular papillary endothelial hyperplasia or Masson's tumour (**Fig 3A, B and C**).

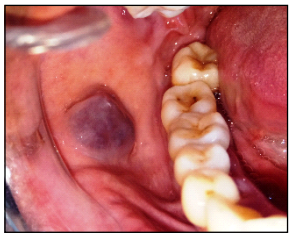
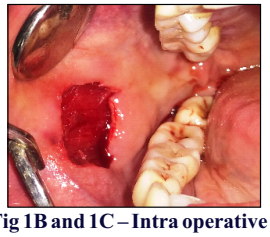


Fig 1A– Photograph of the lesion at (R) buccal mucosa



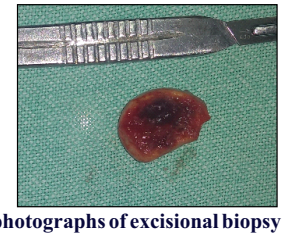


Fig 1B and 1C – Intra operative photographs of excisional biopsy

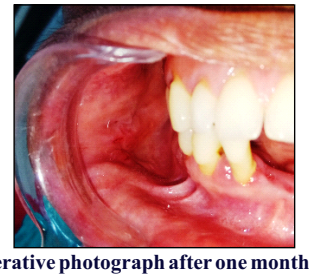


Fig 1D – Post operative photograph after one month

CASE 2

A 42 year old lady reported with a pigmented lesion at right buccal mucosa with similar clinical and subsequent histopathological presentation conceding successful excisional management (**Fig 2A to 2C**).

Both intra operative and post operative events were uneventful. Presently both the cases are under follow up review since one year

without any sign of recurrence.

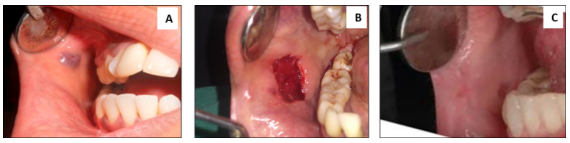


Fig 2: A – photograph of the lesion at (R) buccal mucosa; 2B – intra operative photograph of excisional biopsy; 2C – post operative photograph after one month.

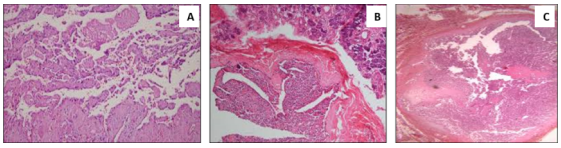


Fig 3: A, B and C – organized thrombus occupying a luminal structure with copious papillary tissues lined by endothelial cells.

Table 1: details of the cases

Ser No	Age/Sex	Site	Clinical presentation	Treatment done
1.	48/F	Right buccal mucosa	Asymptomatic, pigmented lesion of 2 x 2 cm size, bluish red in colour; non tender, firm and rubbery in consistency; negative diascopy test	Excisional biopsy
2.	42	Right buccal mucosa	Clinically asymptomatic bluish red, non tender lesion of 1x1 cm size; Negative diascopy test	Excisional biopsy

DISCUSSION

The etiopathogenesis of IPEH is speculative. Various hypotheses have been proposed and discussed in the literature. Some authors described it as a benign neoplastic process with proliferation of endothelial cells and papillary growth in the vascular lumen which undergoes degeneration and necrosis[3]. Another theory illustrated it as a reactive process of vascular endothelial cells induced by perivascular inflammation and reduced perfusion and subsequent angiolymphoid hyperplasia crop up from a thrombus with eosinophilia[5]. Few other investigators highlighted it as a pseudotumoral lesion originated from a thrombotic material succeeded by endothelial proliferation with papillary growth[6]. A possible hormonal role has also been put forward coupled with local angiogenic growth factors which contribute to proliferation of the endothelial cells[7]. Minor trauma and chronic irritation have been suggested as predisposing factors too[8].

IPEH can be found in the lumen of enlarged veins, hematomas and rarely in lymphagiomas also. Till date less than a hundred of cases of IPEH in the oral mucosa and lips have been reported in the literature. It is more frequent in 4th and 5th decade of life with a female predilection. Within the intra oral sites, lip mucosa is the commonest followed by tongue, buccal mucosa and mandibular vestibule[5].

Various clinical differential diagnoses can be acknowledged with which the lesion is often mistaken are hemangioma, hematoma, mucocele, thrombosed vein, phlebectasia, lymphagioma, traumatic fibroma, granuloma, minor salivary gland tumor and nevus[4]. The lesion can be differentiated from other vascular lesion by a negative diascopy test where IPEH doesn't blanch out.

Histologically IPEH is often erroneous to angiosarcoma. Differential characteristics for IPEH are well circumscribed lesion, intravascular location, thrombotic papillary growth, fibrohyalinized wedge of the papillae and hyperchromatic endothelial layers without any piling up[9]. Features like cellular pleomorphism, mitotic activity or foci of necrosis in inosculated capillary vessels are infrequent. It is important to differentiate this lesion clinically and histopathologically from angiosarcoma to prevent any misinterpretation and subsequent execution of overly aggressive treatment. Other histological differe ntiation includes hemangioma, mucocele, intravenous pyogenic granuloma, Kaposi's sarcoma, spindle cell hemangio endothelioma, malignant endovascular papillary angioendothelioma etc[3].

The cells lining the papillae can be confirmed as endothelial cells by immunohistomical staining demonstrating positive reactions with andibodies for ferritin in the early stages and then for vimentin and the factor VIII related antigen in mature stages. Immature cells show a negative reaction for CD34, a hematopoietic progenitor cell antigen[7].

An excisional biopsy with a healthy margin of 4-5 mm using conventional knife, electrocautery, soft tissue LASER or cryotherapy yields the best result. Recurrence after excision has been reported in a single case, probably due to inadequate removal of the primary lesion[1]. No malignant transformation has been reported till date.

CONCLUSION

This lesion is clinically important because it presents as a pigmented nodule or mass that can be mistaken clinically and histologically as angiosarcoma and treated as such. Due to its rare occurrence in oral cavity, a diagnostic dilemma exists amongst the clinician and this lesion is not usually considered in the differential diagnosis of oral pigmented lesions. We suggest that this entity should routinely be considered as a differential diagnosis and awareness in dentists and dermatologists of this lesion will prevent misinterpretation and overly aggressive treatment.

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