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Case Report

Extremely rare isolated and acquired oral presentation – Verrucous hemangioma

Tejraj P. Kale^{*,1}, S.M. Kotrashetti², Arjun Singh^{*}

Department of Oral and Maxillofacial Surgery, V.K. Institute of Dental Sciences, KLE University, Belgaum, Karnataka, India

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ABSTRACT

Imperial and Helwig originally described verrucous hemangioma as a vascular malformation that is present at birth, anatomic predilection for lower limbs, proportionate growth and equal sex distribution. Mucosal involvement, including the oral cavity, is seen occasionally as integration of a systemic variety or with multiple cutaneous lesions in other locations. Isolated oral involvement is extremely rare while acquired involvement of tongue without cutaneous or metabolic disease has not been reported in the literature. We present a case of verrucous hemangioma involving the lateral surface of the tongue in a 60-year-old male. Due to the unusual presentation we need to reconsider our current knowledge on the etio-pathogenesis of verrucous hemangioma and its clinical inclusion in the differential diagnosis of various verrucous lesions.

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1. Introduction

Vascular lesions and malformations, comprising of a broad category of lesions often referred to as vascular anomalies, are a heterogeneous group of clinicopathologically distinct entities. Clinicians traditionally have grouped these lesions under the generic term, hemangioma, sometimes classified as cavernous (large vessels) or capillary (small vessels) [1]. Most are congenital or occur at a young age and may show limited growth potential. Some lesions undergo spontaneous regression, which has led some to believe it to be a hamartoma rather than a tumour, while others require active intervention [2]. Imperial and Helwig [1] first introduced the verrucous hemangioma, usually found as a congenital solitary or multiple cutaneous lesions of lower limbs, and distinguished it from the angiokeratoma. Although it is a clinically specific lesion with characteristic histological features, its precise nature remains

unknown. The original description was that of a vascular malformation, that is, present at birth, anatomic predilection for lower limbs, proportionate growth and equal sex distribution [1]. Involvement of the mucosa, including the oral cavity, is seen occasionally as integration of a systemic variety or with multiple cutaneous lesions in other locations. Isolated oral involvement is extremely rare while acquired involvement of tongue without cutaneous or metabolic disease has not been reported in the literature.

2. Case report

A 60-year-old Asian Indian male, presented to our Oral and Maxillofacial unit with a complaint of a growth over the left half of the tongue. He was having trouble eating and had developed a psychological component of fear of it being a cancerous growth. He gave a history of extraction done of a lower left third molar at a local dentist 3 months prior to his presentation to us, which he considered traumatic, after which a small ulcerative lesion 1 cm × 1 cm appeared over the left anterior half of the tongue in the left second molar region. Over about a month after the extraction, the ulcer healed and appeared then as a small 1 cm × 1 cm growth which eventually progressed to its current size. There was no history of any associated fever during these events. The patient had no deleterious habits such as tobacco chewing or alcohol consumption. The family history was inconclusive for any hereditary or genetic correlation.

General examination did not reveal any significant findings such as lesions on the extremities or the chest and abdomen. On fur-

Abbreviations: RBC, red blood cells; CD, cluster of differentiation; GLUT, glucose transporter; MAP3K3, mitogen-activated protein kinase kinase 3.

^{*} Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

^{*} Corresponding author. Tel.: +91 9743764783.

E-mail addresses: tejraj kale@yahoo.com (T.P. Kale), kotra27@gmail.com (S.M. Kotrashetti), arjun193@gmail.com (A. Singh).

¹ Tel.: +91 9448472891.

² Tel.: +91 8722323369.

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Fig. 1. Lesion in situ on deviation of tongue to opposite side.

ther intraoral examination, a 3 cm in length and 2 cm in width irregular, mixed white and red, verrucous growth was seen over the left anterior 2/3rd of the tongue which was firm, sessile and nontender (Fig. 1). In the mandibular arch, an edentulous span was present posterior to the mandibular left canine, whose tooth preparation was done to receive a crown prosthesis, and there were porcelain fused to metal crowns over the lower anterior teeth. There were also fractured restorations and crowns of the right mandibular second molar, first and second premolar. All maxillary teeth were present with moderate amount of attrition and abrasion. No sharp cusps or points were present. No regional lymph nodes were palpable. Routine blood and urine investigations were ordered which were within the normal limits. Based on the clinical features, a provisional diagnosis of verrucous hyperplasia involving the left anterior lateral half of tongue was given. The treatment plan included a wide excision biopsy for the lesion. The patient was explained the risks associated and after obtaining informed consent the patient was taken up under general anaesthesia. After placing an anterior traction suture and retracting the tongue to the opposite side, an elliptical incision was given using an electrocautery around the periphery of the lesion maintaining a 5 mm margin of clinically healthy tissue (Fig. 2a). The lesion was excised (Fig. 2b) and the wound sutured using Polyglactin 910 sutures and sent for histopathology examination.

The histopathology of the specimen under low magnification of one area revealed papillary projections with connective tissue core,

surface keratinisation, keratin plugging, acanthosis and individual cell keratinisation. Other side of the tissue shows loss of epithelium, fibrinous exudates and necrotic debris. Numerous endothelial lined blood vessels engorged with RBCs were also evident (Fig. 3). Under higher magnification, the epithelium confirmed the findings as low magnification. The underlying connective tissue stroma was loose with numerous branching endothelial lined spaces with engorged RBCs. The stromal tissue in few areas showed myxoid degeneration. The deeper connective tissues stroma showed muscle tissue inter-spread with small to large capillary spaces with RBCs. A few chronic inflammatory cells such as lymphocytes, plasma cells and mast cells were also seen (Fig. 4). Based on these findings a histopathological diagnosis of verrucous hemangioma was given. Due to the rarity of the presentation in the oral cavity the margins of the lesion were further examined which were found to be close or clear margins.



Fig. 3. Photomicrograph (4×) of specimen showing papillary projections with surface keratinisation, individual cell keratinisation and plugging, acanthosis, fibrinous exudates and underlying loose connective tissue stroma with numerous endothelial lined spaces.

Postoperatively, the wound healed uneventfully and the patient has been followed up regularly for 9 months now without any signs of recurrence or new lesions (Fig. 5).

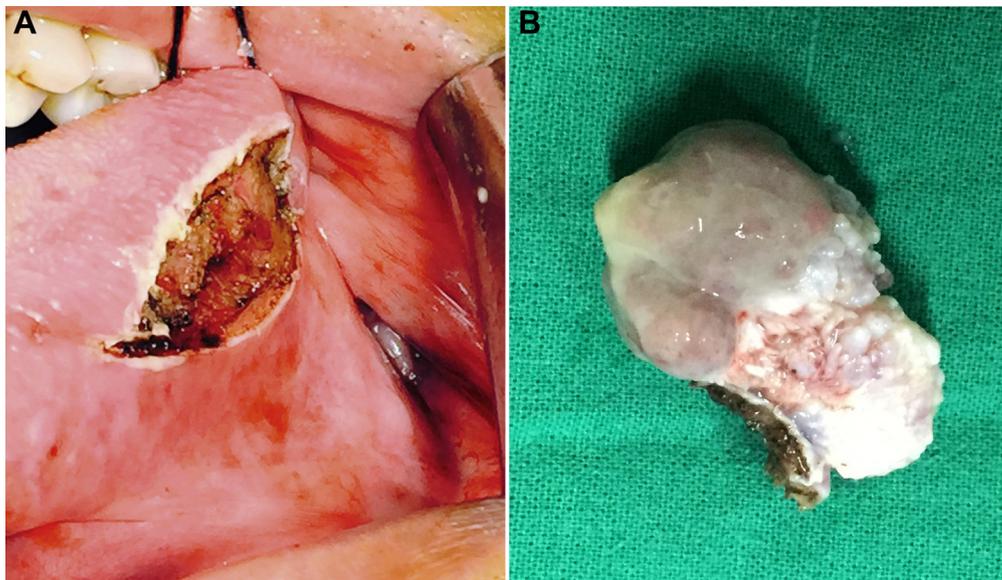


Fig. 2. (A) Site after excision of lesion. (B) Excised lesion.

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