

# Intramuscular Hemangioma of Upper Lip Mucosa in a Fifty-Four-Year-Old Man

Shokoofeh Jamshidi<sup>1</sup>; Shahrbanou Raedi<sup>2,\*</sup>; Mahsa Mahdian<sup>2</sup>; Lida Sami<sup>2</sup>

<sup>1</sup>Dental Research Center, Department of Oral and Maxillofacial Pathology, Dental School, Hamadan University of Medical Sciences, Hamadan, IR Iran

<sup>2</sup>Dental Research Center, Department of Oral Medicine, Dental School, Hamadan University of Medical Sciences, Hamadan, IR Iran

\*Corresponding author: Shahrbanou Raedi, Dental Research Center, Department of Oral Medicine, Dental School, Hamadan University of Medical Sciences, Hamadan, IR Iran. Tel: +98-9181079620, Fax: +98-8138354220, E-mail: dr.radi80@yahoo.com

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**Introduction:** Hemangiomas are the most common benign tumor of infancy that can occur anywhere in the body. Intramuscular hemangiomas (IMH) are accounting for approximately 1% of all cases and the muscles of extremities are the most common sites. Because of scarcity and variable clinical features of these tumors, we decided to report a case of IMH in the upper lip mucosa.

**Case Report:** The patient was a 54-year-old male that was referred to our clinic for swelling of upper lip mucosa and face asymmetry. The lesion was excised. The histological study revealed an IMH.

**Discussion:** Surgical excision might be an effective approach in IMH of upper lip. Complete resection minimizes the relapse rate of the tumor and results in favorable cosmetic outcomes.

**Keywords:** Hemangioma; Tumor; Muscle; Lip

## 1. Introduction

Hemangiomas are the most common benign tumor of infancy that are characterized by numerous big and fat endothelial cells and indistinct vascular lumen at primary stage. As the lesion mature, blood vessels become more evident (1-4). Intramuscular Hemangioma (IMH) is an uncommon tumor that was first described by Liston in 1843. It is characterized by proliferation of blood vessels between skeletal muscle fibers. IMH accounts for less than 1% of all hemangiomas. About 10% to 20% of cases occur in head and neck (1, 5). In these regions, masseter and trapezius are the most commonly involved muscles (6). Due to their deep location, scarcity, and variable clinical features of these tumors (3), we decided to report a case of IMH in the upper lip mucosa.

## 2. Case Report

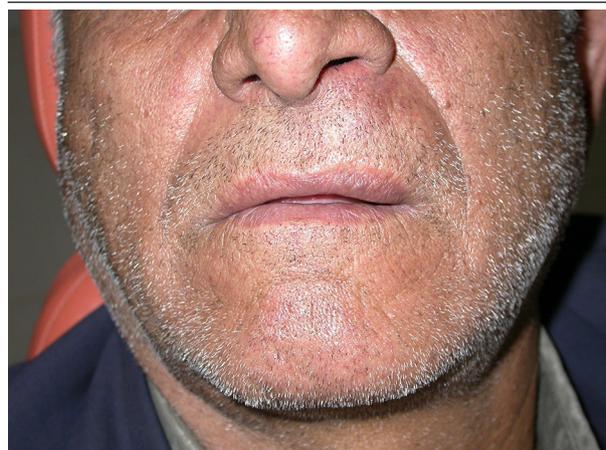
A 54-year-old male was referred to the Oral Medicine Department, Dentistry School of Hamadan University of Medical Sciences. His chief complaint was swelling of upper lip mucosa and asymmetry of his face (Figure 1).

There was no history of trauma or lip chewing prior to appearance of the swelling. The patient had no significant medical history. The intraoral examination showed a painless submucosal mass with firm consistency and tenderness to the touch. The lesion was well defined and completely differentiable from surrounding tissue.

Superficial mucosa was normal and intact. Swelling was measured as 2 × 3 × 5 cm (Figure 2).

Panoramic radiography did not show any changes. The initial differential diagnoses were fibroma, pleomorphic adenoma, and neurilemmoma (Figure 3).

Excisional biopsy was performed in the Oral and Maxillofacial Surgery Department. Histopathologic examination showed the proliferation of capillaries in skeletal muscle. Hence, the diagnosis of IMH was confirmed (Figure 4).



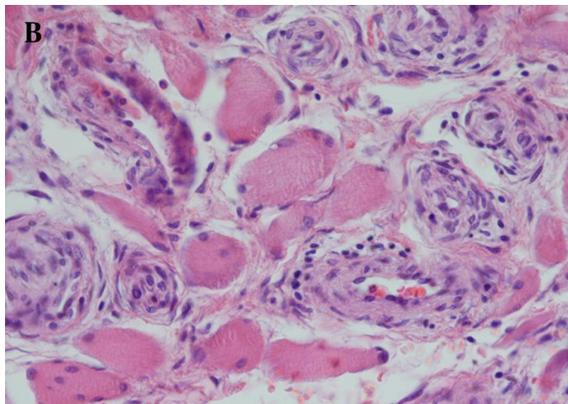
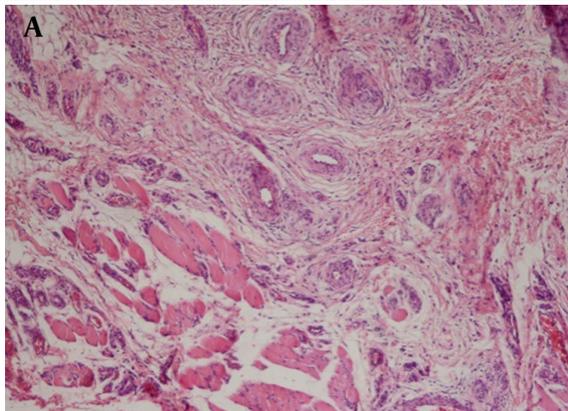
**Figure 1.** A 54-Year-Old Male With Swelling of Upper Lip Mucosa and Face Asymmetry



**Figure 2.** A Submucosal Mass With Firm Consistency and Tenderness to Touch in Left Upper Lip



**Figure 3.** Panoramic Radiography of the Patient Before Surgery



**Figure 4.** Histopathologic Examination Showing the Intramuscular Hemangioma Multiple Capillaries Interspersed With the Muscle Fiber (H & E stain; Magnification: a,  $\times 100$ ; and b,  $\times 400$ ).

### 3. Discussion

Hemangiomas are the most common benign tumors of infancy that can occur anywhere in the body (7). First described by Liston (8), IMHs are accounting for approximately 1% of all cases of hemangioma and the muscles of extremities are the most common sites for these benign tumors (7). About 94% of hemangiomas occur before the age of 30 with approximately 50% representing in the first decade (9). There is no predilection for either sex (3). In the study of Conley et al. on 393 patients with IMH, 28 and 15 cases were presented in the head and neck regions, respectively (10). The most common involved site in the orofacial region is the masseter (10-12). IMHs of the mentalis (13), digastric, mylohyoid (14), orbicularis oris (15-17), and sternocleidomastoid (18) muscles have been reported too.

Clinically, hemangiomas present as localized, rubbery swelling (18). The most frequent reason for seeking treatment is the cosmetic concern because of the facial asymmetry (16). Pain was reported in about 58% of lesions in one study (19). With a pressure, the size of hemangioma reduces and becomes pale, which is due of emptying blood vessels (20-22). Allen and Enzinger introduced a classification of IMHs according to the diameter of the vessels: (a) small vessel or capillary type ( $< 140 \mu\text{m}$ ); (b) large vessel or cavernous type ( $> 140 \mu\text{m}$ ); and (c) mixed type, consisting of both aforementioned sizes (23). In the head and neck, small vessel, large vessel, and mixed types appear in 30%, 19%, and 5% of cases, respectively. The highest recurrence rate (28%) is seen with mixed type (23). Capillary types have a rapid growth and the recurrence rate is 20% (5, 18). The present case was capillary type. The best treatment modality is complete resection of tumor with a safe margin of surrounding muscle due to the infiltrative characteristic of the tumor (18). Fortunately, the mass was well defined and was easily separated from adjacent normal tissue. Massive bleeding during resection has been reported during removing the orbicularis oris IMHs (15, 16); nevertheless, hemorrhage was not seen in our case because most of the vessels were ligated during removal of the mass. Surgery minimizes the risk of damage to adjacent structures and is an effective approach to cure the patients with low relapse rate (5). The patient was visited three, six, and twelve months after surgery and there was no clinical signs of recurrence or any other tumor-associated complaint.

Nonsurgical modalities such as cryosurgery, radiation, sclerotherapy, embolization, and electrocoagulation have been used in the treatment of hemangiomas (12, 13, 24, 25). Because of the destructive effect on normal tissues and lower rate of complete regression of the tumor, these modalities are not as effective as the surgical resection (5). In conclusion, surgical excision of IMH could be an effective approach in upper lip. Complete resection minimizes the relapse rate of the tumor and results in favorable cosmetic outcomes.

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