A case report of an oral hemangioma with unusual features

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Key Clinical Message

Hemangioma is a tumor that occurs due to the proliferation of endothelial cells in 4–5% of infants and affects the head and neck region in almost 70% of cases (1). The tumor in the mouth can appear as a soft, smooth or lobulated mass with or without a base and varying in size from a few millimeters to a few centimeters. This article reports a case of hemangioma with unusual clinical features in a 26-year-old male patient.

Keywords: Capillary hemangioma, Vascular Malformation, Hemangioma, Oral cavity.

1 | INTRODUCTION

Hemangiomas are benign soft tissue vascular tumors and true neoplasms of endothelial cells that are often present at birth or appear several weeks after birth and grow and spread rapidly as the child grows. However, many vascular lesions resolve spontaneously before or during puberty (2, 3). Head and neck hemangiomas are histologically divided into three types, namely capillary, cavernous and mixed. Capillary hemangiomas are more common than other types (4). This benign tumor is significantly more common in women than in men (3:1) and occurs in 80% of cases (5). Intraoral capillary hemangiomas are very rare and account for 0.5–1.0% of all intraoral neoplasms (6, 7). The most common site of occurrence of these benign tumors is the oral cavity, lips, tongue, and buccal mucosa (4). The clinical appearance of lesions in the oral cavity is diverse and different layers including skin and even muscles are involved, although the adjacent bone is rarely affected (8). The lesion does not pose any risk to the patient and surgical treatment is only necessary if the aesthetic appearance is affected or chewing is impaired or bleeding occurs.

2 | CASE PRESENTATION

2.1 | Case history/Examination

A 26-year-old male patient with complaints of swelling in the lingual part of the lower anterior teeth referred to the Faculty of Dentistry, Department of Oral and Maxillofacial Diseases, Mashhad University of Medical Sciences, Iran. No history of systemic disease or drug use was mentioned. In addition, no lymphadenopathy or facial asymmetry was observed on extra-oral examination. On intraoral examination, a prominent pinkish nodular lesion with a rubbery consistency and telangiectasia on the surface of the lesion were observed. The lesion was approximately 2.5 cm in size in the lingual area of teeth 24 and 25. The patient had had the course of the lesion for approximately 10 months, sometimes with trauma-related bleeding. Approximately one month previously, teeth 24 and 25 were extracted due to looseness and the area was curetted. However, the lesion had recurred in the same location several days after the extraction.

In the patient's dental records, the previous radiographs (OPG) were reviewed and translucency with clear boundaries was observed in the area of teeth 24 and 25. Based on the clinical appearance, location and history

of tooth loosening, and the translucency observed on the OPG, additional CBCT radiography was requested to rule out central lesions, including residual cysts. However, no pathological findings were reported in the area of the mandibular anterior teeth (Figure 1).

3 | METHODS

To complete the clinical examination, aspiration of the lesion was performed and blood discharge was observed. Based on the results of clinical examinations and the absence of intraosseous lesions in the area of the mandibular anterior teeth, the lesion was clinically diagnosed as a vascular malformation or a hemangioma. To confirm this, the non-invasive ultrasound technique was also used and the results showed that the lesion was a mass with unclear boundaries and heterogeneous echo containing three calcified areas with dimensions of 24 mm, 13 mm and 16 mm in the lingual area of the soft tissue of the gums. The front of the mandible was also observed to have significant blood supply on Doppler scan, and on ultrasound, the lesion was diagnosed as potentially calcified hemangioma and oral choristoma.

According to the clinical diagnosis of vascular lesions, a sample was collected from the lesion. During histopathological examination, the proliferation of endothelial spindle cells was observed with the formation of numerous blood vessels, often with a capillary view, divided into parts by the fibrous connective tissue. Some bone trabeculae were also observed in the lesion and the diagnosis of calcified capillary hemangioma and choristoma was made(Figure 2). According to spindle cells, histiocytes as well as bone cells in histopathology, immunohistochemical examination was recommended to rule out relevant lesions.

1. Immunohistochemical examination of vimentin was performed on connective tissue lesions observed in the background and reported as positive (Figure 2).

2. S100, Sox10, inhibin and calretinin were performed to detect and rule out negative nerve lesions, paragangliomas and neural crest lesions.

3. CD68 was performed to detect lesions originating from histiocytes, such as histiocytomas, which was negative and ruled out.

4. SMA was performed to examine the smooth muscle of the vessel wall with the origin of pericytes such as hemangiomas and pericytomas, and it was negative.

5. CD31 was performed to examine the lesions originating from endothelial cells. This was positive and suggested the vascular origin of the lesion. Therefore, due to the negativity of other markers, the diagnosis of capillary hemangioma was confirmed (Figure 3). On the patient's desire for the complete removal of the lesion, the patient was referred to a maxillofacial surgeon and after CT angiography of the lesion and a complete operation of the lesion was performed.

4 | CONCLUSION and RESULTS

Capillary hemangioma can be diagnosed differentially from other lesions such as vascular malformations or inflammatory hyperplasia, and histopathological findings are very helpful in the final diagnosis. Because hemangioma lesions are asymptomatic, the location and course of the lesion must be considered in the diagnosis. Vascular hemangioma lesions with unusual appearance and location are often regarded as a diagnostic problem for general dentists. When removing the hemangioma through a surgical incision, severe bleeding may occur, although in our case this did not occur during tooth extraction and surgery. Due to the small size of the lesion, easy access, and low risk of bleeding, surgical treatment was chosen in our case.However, due to the unavailability of the patient, further follow-up was not possible.

5 | CASE DISCUSSION

Hemangiomas are benign tumors of the capillary endothelium that occur in newborns between two and eight weeks of age, grow rapidly in the first year and resolve in 50% of cases by the age of five years (9). Hemangioma is one of the most common soft tissue tumors in the head and neck region. However, it rarely occurs in the oral cavity and is congenital in most cases, while its occurrence in adults is very rare (9). On

the other hand, hemangioma is the most common tumor in infants, occurring in 5–10% in the first year of life. It occurs as a single lesion in 80% of cases and as a multiple lesion in the rest. The clinical appearance can vary depending on the surface or depth of single or multiple lesions (10). It can bleed spontaneously or due to trauma and is often painless. In our case, it was a pink, baseless mass with a smooth surface and with no symptoms.

Hemangiomas affect various areas of the oral cavity, including the tongue, lips, and buccal mucosa, as well as the mandible. When the lesions occur in the gums, the interdental gum area is the most common site of infection (11). Matsumoto *et al.* studies 31 patients with intraoral hemangioma and found that most lesions were located on the oral mucosa (45.2%), followed by tongue (35.5%), lip (9.7%), gum (6.5%), and palate (3.2%) (6). In the case of our patient, the lesion occurred in the lingual part of the anterior gingiva of the mandible, far from the marginal gingiva. After initially ruling out bone involvement, the lesion was differentially diagnosed as amatous hyperplasia. However, taking into account the distance from the marginal gingiva, the observed appearance and the blood discharge during aspiration, amatous hyperplasia could also be excluded. Furthermore, the history of recurrence after removal of the lesion was misleading. In any case, and according to the clinical presentation and the result of aspiration, vascular lesions were ultimately suggested as the most likely diagnosis, although the location was unusual for intraoral hemangiomas.

The differential diagnoses of intraoral capillary hemangioma are vascular malformations and pyogenic granuloma, which are histologically similar and can be differentiated by biopsy and immunohistochemical examination (12). Since the history and course of the lesion are very helpful in diagnosing vascular anomalies, the unclear history of these lesions may lead to incorrect diagnoses and inappropriate treatments. In our case, such a misdiagnosis was made by a general dentist. To confirm the diagnosis of vascular lesions, histopathological examination is considered the diagnostic standard. Radiographic examination is necessary to differentiate such lesions from other intraosseous lesions or central hemangiomas (13).

On the initial panoramic radiograph, translucency was observed in the anterior region; therefore, and due to the inaccuracy of the OPG radiograph in the anterior maxilla, an additional CBCT radiograph was required for a detailed examination of the area. According to the CBCT radiograph, lesions of bony origin and intraosseous hemangiomas could be excluded. In addition, the patient had a history of loose teeth that appeared unrelated to the lesion and was likely caused by occlusal trauma.

Calcified spots were unexpectedly observed on ultrasound of the lesion, which were differentially diagnosed as choristoma (3). Choristomas are rare benign lesions that result from to the growth of normal tissue in an abnormal location and their etiology is unknown (14). Considering that choristoma in the lingual gingiva is very rare and that the clinical, radiographical, and histopathological evidences confirmed the presence of a vascular lesion, the calcifications were probably due to the slowing of peripheral blood flow and the consequent thrombosis and mineralization within the vessels (15). The rare thrombosis and phlebitis in hemangioma in our case was confirmed by the ultrasound and histopathological examination of the lesion.

Hemangioma treatment is only necessary when there is a functional disorder such as malaise and recurrent bleeding or for aesthetic reasons (16). Treatment includes surgery, laser, sclerosing agents, embolization, and corticosteroid therapy (17). Treatment of oral hemangioma depends on various factors such as age, size and extent of the lesion, site of involvement, and clinical features (18). Given the unusual location of the lesion on the gum, this case is of particular importance as dentists may mistakenly consider it a periapical lesion or a hyperplasia lesion.

FIGURE panoramic radiograph(A). intraoral view(B). CBCT(C)

FIGURE 2 Pathology with H&E staining, overview of the proliferation of endothelial cells, blood vessels and bone tissue (magnification 200x and 400x)(A,B,C) .Immunohistochemical staining (cd68) of background histiocyte cells was positive (D) Immunohistochemical staining (vimentin) was positive in connective tissue cells(E)

FIGURE 3 Immunohistochemical staining (CD31) related to positive endothelial cells (magnification x100

and x200)(A,B).

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