CASE REPORT

Angiolymphoid hyperplasia with eosinophilia in the angle region of the mandible

Birkan Taha Ozkan, Cennet Neslihan Eroglu, Levent Cigerim, Omer Gunhan[1]
Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, YüzüncüYıl University, Van, [1]Department of Pathology, Gulhane Military Medical Academy, Ankara, Turkey

Address for correspondence:
Dr. Cennet Neslihan Eroglu,
Yuzuncu Yil Universitesi, Dis Hekimligi Fakultesi, Kampus, Van - 65080, Turkey.
E-mail: neslihanakca2003@yahoo.com

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ABSTRACT

Angiolymphoid hyperplasia with eosinophilia (ALHE), also called epithelioid hemangioma, is a rare benign vascular lesion usually affecting the muscular arteries of the head and neck in female patients. Here, we report a 30-year-old male patient who presented with painless swelling in the angle region of the mandible. The diagnosis of the specimen, which was surgically removed under local anesthesia, was made as ALHE. The patient has remained uneventful for 3 years.

Key words: Angiolymphoid hyperplasia, angle of mandible, eosinophilia, epithelioid hemangioma

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a benign vascular lesion.[1] The most frequently involved regions are the head and neck, hands, penis and arms.[2-4]

ALHE typically manifests with single or multiple, red/brown dome-shaped papule or subcutaneous nodules. In some cases, nodules extend into the dermis or muscular layer. Peripheral eosinophilia and lymphadenopathy are observed in 20% of patients.[5-7] The etiology of ALHE remains indefinite, but it may be vascular.[6,8]

ALHE is more prevalent in Caucasian women aged 30–40 years.[9] Histologically, it appears as reactive proliferation of a small blood vessel that surrounds a muscle artery together with inflammatory infiltrates.[6]

CASE REPORT

A 30-year-old male patient presented to the Department of Maxillofacial Surgery with swelling in the right facial region. His anamnesis revealed that the patient had his right first molar tooth extracted owing to infection a year ago and that painless swelling in that region had been present ever since. Clinical examination of the swelling demonstrated a painless, hard, mobile, localized elastic lesion on palpation in the subcutaneous tissues extending in line with mandibular border. The lesion was located approximately between the angle of the mandible and the anterior aspect of the sternocleidomastoid muscle (2–3 cm below the inferior border of the mandible) [Figure 1a].

There was no lymphadenopathy in the region. No specific finding was detected on radiological examination. The lesion was explored at the level of the subplatismal plane via extraoral approach under local anesthesia through an incision made parallel to the mandibular border. The lesion, which appeared of vascular origin because of red-brown color, was capsulated and associated with a small artery on both the anterior and posterior borders [Figure 1b].

The borders of the lesion were ligated with polyglycolic acid suture to control bleeding . After total enucleation, subcutaneous tissues were sutured with 3/0 polyglycolic acid and the skin was sutured with 4/0 polyprolene. The excised specimen measuring 1.5 cm was sent to the laboratory for histopathological examination [Figure 1c].

Histopathological examination revealed a restricted tumoral lesion consisting of vascular configurations of various diameters. Vascular configurations were rich with capillary vessels arranged in a lobular pattern. There was a large blood vessel with patent lumen and thick wall in the middle of the lesion [Figure 2a].

Vascular configurations were lined by epithelioid endothelial cells. Endothelial cells showed no atypia. Edematous connective tissue with lymphocyte and eosinophilic

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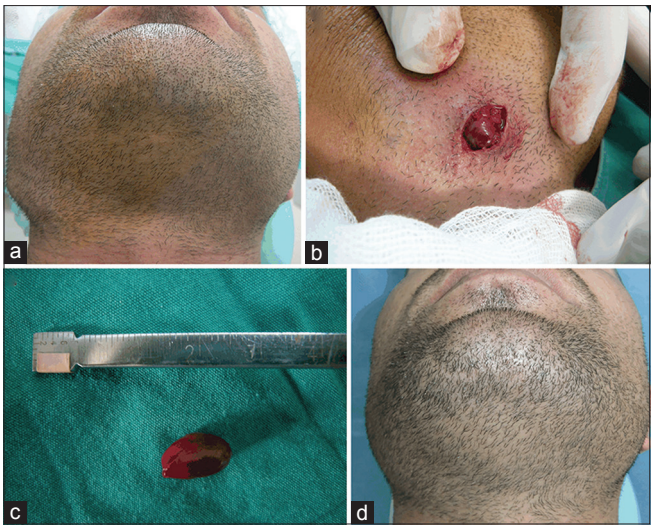

Figure 1: (a) Preoperative patient's extraoral view. (b) Intraoperative view of the lesion. (c) Resected surgical specimen. (d) 12th month Postoperative extraoral view

leukocyte infiltration were detected among vascular configurations [Figure 2b].

Under the light of these findings, the patient was diagnosed with epithelioid hemangioma. No esthetic problem or complaint or recurrence was encountered over the course of the 3-year follow-up period [Figure 1d].

DISCUSSION

Here, we report a case of ALHE in the angle region of the mandible in a 30-year-old male patient. The etiology of ALHE remains unknown, because it is not clear if it is primarily a vascular neoplasm, a lymphoproliferative process or a heterogeneous group of entities. Trauma, infections and renin or hyperestrogenic conditions (pregnancy or oral contraceptive agents) are considered to be the likely causes.[10,11] Infection was considered as the etiological factor in the present case report, because it was learned from his anamnesis that swelling caused by an infected first molar tooth a year ago induced the pathology in that region.

ALHE usually appears in head and neck region, frequently in the auricular area and usually measures about 2–3 cm in size. However, other authors have reported that lips, oral mucosa and tongue may also be involved.[3,12] In the present case, epithelioid hemangioma involved the angle region of the mandible. In this respect, the present case report might contribute to the literature of other cases of epithelioid hemangioma that might be presented in the future.

In the literature, it is usually mentioned that ALHE is an asymptomatic lesion, more prevalent among females and that lymphadenopathy is present in about 5–20% of patients.[12] Asymptomatic disease course may sometimes

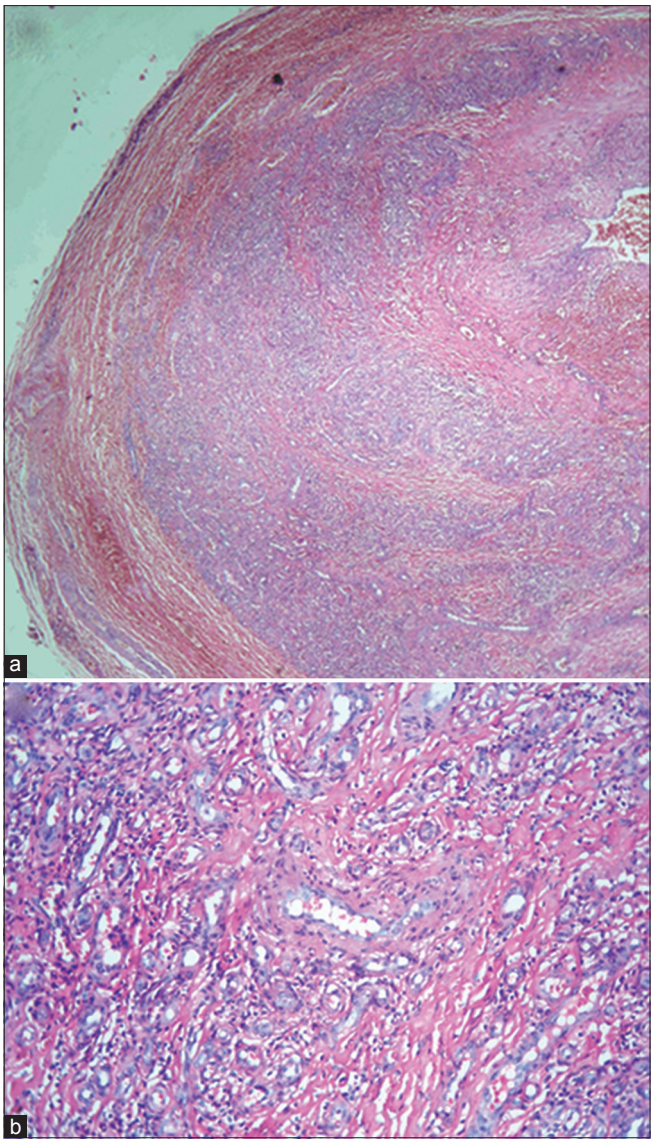

Figure 2: (a) Histopathological image of the large blood vessel (H&E stain, x40). (b) Photomicrograph showing lymphocyte and eosinophilic leukocyte infiltration along with blood capillaries (H&E stain, x200)

become symptomatic because of occasional recurrence of inflammation. We believe that lymphadenopathy might be detected in that period. Particularly, considering that ALHE is a reactive lesion, frequent palpation of the lesion by the patients may be triggering the inflammatory process.

ALHE must be histologically and clinically differentiated from Kimura disease, which is a chronic inflammatory condition characterized by large subcutaneous nodules in the head and neck region. It should be kept in mind that Kimura disease is more prevalent among young males; it is associated with increased immunoglobulin E levels and skeletal involvement may be encountered. Peripheral eosinophilia and lymphadenopathy may be encountered in both diseases. The most significant distinctive criterion is the fact that whilst

ALHE is pathology of vascular origin, Kimura disease is a chronic inflammatory process.[10,12]

In addition to Kimura disease, differential diagnosis of ALHE includes cutaneous lymphoma, cavernous hemangioma, pyogenic granuloma, Kaposi sarcoma and bacillary angiomatosis.[13]

Vascular proliferation in subcutaneous tissue, the presence of cobblestone-like endothelial cells lining the vessels, lymphocyte infiltration together with eosinophilia are important histopathological criteria for diagnosis.[14] These features were key in the diagnosis of the present case. Eosinophilic chemotactic factor released from mast cells is suggested as an extra agent contributing to eosinophilia, which adds to the inflammatory characteristics of such cases. Eosinophil counts typically return to normal after complete remission of the lesion.[15] Surgical excision is the preferred method for the treatment of ALHE. Alternative therapies include electrodessication, curettage, radiotherapy, cryotherapy, chemotherapy, corticosteroids, laser surgery and various agents, for example, interferon alpha 2b.[7] Spontaneous remission in such cases is possible within months, even years, but recurrences are frequent. Treatment is necessary in symptomatic cases and in situations that alter the patient's appearance.[10] In the present case, we preferred surgical excision for esthetic concern. We believe that complete surgical resection is a successful therapeutic option for such cases, because of its feasibility and practicality.

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