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Case Report

A rare case of angiofibroma of the mandible: A case report

Mohammed Israr Ul Khaliq^{a,*}, Ajaz A. Shah^b, Nahida Dar^c^a Post Graduate Scholar, Department of Oral and Maxillofacial Surgery, Govt. Dental College & Hospital, Srinagar, India^b Professor and Head, Department of Oral and Maxillofacial Surgery, Govt. Dental College & Hospital, Srinagar, India^c Post Graduate Scholar, Department of Oral and Maxillofacial Surgery, SGT Dental College, Gurgaon, India

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ABSTRACT

Angiofibroma is a rare, highly vascular nonencapsulated tumor, which is locally invasive. A rare case of angiofibroma of the mandible in a 16-year-old female patient is reported here. The lesion was excised along with surgical removal of right mandibular third molar tooth. Patient was followed up for 1 year without any recurrence.

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1. Introduction

Angiofibroma is a rare, highly vascular nonencapsulated tumor, which is locally invasive. Hippocrates described this tumor in the 5th century BC, but Friedberg first used the term angiofibroma in 1940.¹ Vascular and fibrous lesions are common in the head and neck area, including both benign and malignant subtypes. Angiofibromas are typically encountered in the nasopharynx among teenage males, accounting for the appellation “juvenile nasopharyngeal angiofibroma.” Cellular angiofibroma is a benign vascular neoplasm that typically arises in the vulva, perineal, and paratesticular region. Microscopically, the lesions exhibit multiple small, nondilated capillary channels, many of which contain erythrocytes. The lesions usually do not recur following simple excision. Recent evidence

indicates that cellular angiofibromas may be cytogenetically related to spindle cell lipoma.

2. Case report

A 16-year-old female patient reported to us with the chief complaint of swelling on the right side of the face since 1 year that had rapidly increased in size since the past 4–5 months. It was associated with occasional intermittent, spontaneous pain. The medical history was not significant. Extraoral examination (Fig. 1a) was suggestive of a swelling on the right side of the face. The overlying skin was normal in color and texture. Intraoral examination (Fig. 1b) revealed inflamed and tender mucosa in the right lower mandibular third molar region.

* Corresponding author.

E-mail address: drisrarch@gmail.com (M.I. Ul Khaliq).<http://dx.doi.org/10.1016/j.jobcr.2016.01.005>

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Fig. 1 – (a) Extraoral photograph of the patient showing swelling on right side of face. (b) Intraoral photograph of the patient showing inflamed right buccal mucosa in retromolar region.

Routine hematological investigations were found to be within the normal limits. Fine-needle aspiration of the swelling and the lymph node was performed, which revealed only blood on examination. Orthopantomogram (Fig. 2)

Fig. 2 – Orthopantomogram showing multilocular radiolucent lesion extending from right mandibular first premolar to mandibular third molar region on the right side without root resorption.

showed multilocular radiolucent lesion extending from right mandibular first premolar to right mandibular third molar region with no root resorption.

An incisional biopsy was done, which revealed numerous vascular channels and were bordered by single layer of endothelium consisting of dense fibrous tissue with smooth muscles. The vascular component was more in the periphery with collagen fibers arranged parallel to each other. Fibroblasts were interposed within collagen fibrils. These findings were suggestive of Angiofibroma. The lesion was excised along with surgical removal of right mandibular third molar tooth (Fig. 3a and b). Patient was followed up for 1 year without any recurrence (Fig. 4).

3. Discussion

Angiofibroma is a relatively rare tumor. Only a few cases have been reported in the maxillofacial complex in females.² The microscopic differential diagnosis for angiofibromas includes angiomylipoma, angiomyo fibroblastoma, juvenile nasopharyngeal angiofibroma, solitary fibrous tumor, spindle cell

Fig. 3 – (a, b) The lesion was excised along with surgical removal of right mandibular third molar tooth.

Fig. 4 – Patient was followed up for 1 year without any recurrence.

lipoma, and superficial angiomyxoma. All of these lesions are vessel rich with a fibroblastic or myofibroblastic element, variable sclerosis, and harbor focal aggregates of adipocytes. Angiomyolipoma is a fatty tumor with prominent muscular arterioles while angiomyofibroblastoma is a fibrovascular neoplasm with a loose, somewhat myxoid fibroblastic element. Juvenile nasopharyngeal angiofibroma is uniquely confined to the nasopharynx and has a very distinct histology with a homogeneous fibroplasia and delicate immature collagen with interposed sinusoidal-like vascular channels that lack prominent endothelium.³ The origin of angiofibroma is still disputed.⁴ Various etiological factors have been suggested. Schiff in 1959⁵ postulated that the tumor arises from an ectopic nidus of hormone sensitive fibrovascular tissue that is entrapped in periosteum. Recently, another fibrovascular lesion has been described in the genital/perineal region among females that is distinctly different from juvenile nasopharyngeal angiofibroma, both histologically and behaviorally.⁶ Despite being classified as benign, they may grow considerably and cause significant structural and functional damage. There are no clear explanations as to why a histologically benign tumor should erode bone and invade regional structures.⁷ Batsakis (1979)⁸ reported that the incidence of nasopharyngeal angiofibroma is 1.6 per 16,000 otolaryngology cases with a higher incidence in India and Egypt than in United States and Europe. Handousa et al. (1961)⁹ gave an incidence of 1 in 50,000.

4. Management

Angiofibroma can be diagnosed using CT, MRI, and magnetic resonance angiography.¹ The preferred management includes surgery with preoperative embolization.⁴ Other methods of

treatment that have been employed are irradiation, hormone therapy, cryotherapy, arterial ligation, and use of sclerosing agents.^{2,4} Future therapy may include intraarterial immunotherapy and the use of certain agents that inhibit angiogenesis.² The most widely preferred treatment includes surgical resection with often preoperative embolization.

Other treatment modalities, according to Jafek (1973), include¹⁰:

- a) Sclerosing agents
- b) Radiation therapy
- c) Preoperative hormone therapy
- d) Chemotherapy
- e) Arterial ligation

The future therapy should include intraarterial immunotherapy and use of certain drugs that inhibit angiogenesis.

5. Conclusion

Though angiofibroma occurs exclusively in nasopharynx, the significance of reporting this case lies in its involvement into the oral cavity including mandible, which is rare, and considering it among the differentials for swelling in the oral cavity.

Conflicts of interest

The authors have none to declare.

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