Case Reports in Odontology
ISSN:2410-0412
Journal homepage:
www.casereportsinodontology.org

Primary Intranodal Hemangioma in Submental Region

[PP: **15-18**]

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Abstract:

Hemangioma is a lymph node is an extremely uncommon finding. To the best of our knowledge, only three cases have been reported in the submental region. We present a case of slow growing primary nodal hemangioma of the capillary cavernous type, in a 37-year-old asymptomatic patient. It is important that this benign condition is recognised and not mistaken for malignant tumours or other vasoproliferative lesions.

Keywords:	*Hemangioma, Nodal Hemangioma, Submental, Lymphadenopathy*
ARTICLE INFO	The paper received on: **20/12/2018** Accepted after review on: **04/01/2019** Published on: **04/02/2019**

Cite this article as:
Ramalingam, K., Chinnasamy, A., Verma, D. & Gopinath, V. (2018). Primary Intranodal Hemangioma in Submental Region. *Case Reports in Odontology*. *5(2), 15-18. Retrieved from www.casereportsinodontology.org*

1. Introduction

Benign vascular tumours in the lymph node is an extremely uncommon finding [1]. To the best of our knowledge, only 36 cases of intranodal hemangioma have been reported in the medical literature [1-16] of them only three have been reported in the submental region [13, 17] . We present a case of primary intranodal hemangioma of the lobular type in the submental region, where the entire nodal parenchyma is replaced by the capillary tissue.

2. Case Presentation

A 37-year-old fit and healthy, female patient from a low-income group presented to the department of oral and maxillofacial surgery with a swelling on the lower third of the face on the right side in the submental region. The asymptomatic mass appeared to be of no significant concern, when the patient noticed it for the first time, two years ago, since then the mass had gradually enlarged. At the time of presentation, no significant extraoral gross asymmetry was noticed and the skin overlying the lesion appeared normal with no sign of tenderness, fever or suppuration. On palpation, the mass appeared soft, single, well defined, lobulated, oval in shape, measuring 1 to 1.5 cm mesiodistally, 1mm below the lower border of the mandible with no sign of fixation to underlying structures. An intra and extra oral examination of the neck was

performed. Further, a complete blood count was advised to rule out any neglected or abnormal finding. All the investigation results yielded no abnormality.

The clinician suspected it to be chronic submental lymphadenopathy that may or may not have malignant potential. It was decided to excise the lesion with no definitive diagnosis.

2.1. Gross Finding

The department of oral pathology received the excised mass and on examination it appeared as a soft single somewhat ovoid mass, with an irregular surface, measuring 21x24x13mm in size, dark bluish black in colour with areas of yellow less frequent on the surface suggesting nodal remnants (Figure 1). It was dissected into two halves and the inner aspect revealed dark brownish red vascular elements with occasional yellow fibro-fatty tissue. During the hemi section there was haemorrhage from the vascular remnants in the capsule leaving multiple empty locules. Almost the entire medullary portion of the node is replaced by vascular elements and the cortex showed occasional remnants of the residual node dispersed irregularly.

2.2 Histopathology

Hematoxylin and Eosin (H & E) stained tissue section showed the nodal parenchyma affected by a vascular proliferative lesion with remnants of the fibrous capsule (figure 2). The lesion was composed of endothelial cell proliferation into small sized vessels of varying calibre along with large cavernous spaces engorged with erythrocytes. Foci of chronic inflammatory cell infiltrate is also seen. Adipose tissue and few areas of fibrous connective tissue are also evident.

Correlating the clinical findings, gross pathology and histopathology, features was highly suggestive of a nodal hemangioma of the capillary/cavernous type in the sub-mental region

3. Discussion

Hemangiomas are not uncommon and can occur less frequently in almost every organ in the body. In the head and neck region they are noticed in the skin, mucosa and the soft tissue. The most comprehensive study [18] that involve a large number of cases of primary vascular tumours was conducted in 1992 [17]. Since then there has been occasional isolated case reports on primary nodal hemangiomas. To date, 19 authors have reported a total of 37 cases of primary nodal hemangiomas in medical literature of them only three have been reported by two authors in the submental region [13, 17]. Of the three reported in the submental region [13, 17] two were present in 45 and 21-year-old female patient and the one was in a 16-year-old patient. We present one additional case reported in a 37-year-old female patient.

Hemangioma in the soft tissue are rare and can be classified as intra and extra muscular [18, 19]. Intramuscular is more common within this rare entity and nodal hemangioma are considered extremely uncommon [20]. They are often infiltrative and less well demarcated. However, the nodal hemangioma that we present here is unusually well circumscribed and was surgically removed at ease. The vascular lesion developing within the lymph node was thought to be the most plausible cause because there was endothelial cell proliferation which rules out a vascular malformation. Further, vascular malformations show either arterial or venous morphology, which is not identified in our case.

Since it is not uncommon to see lymph node swelling with high vascularity being associated in a variety of infections, we thought it was a reactivation of nonspecific

inflammatory lymphadenopathy. However, the macroscopic appearance of irregular surface with multiple micro lobules is suggestive of burst in activity with a potential for proliferation. This gave us the suggestion that the node is being replaced with vascular proliferative element.

In the past, pathologist doubted if hemangioma of the lymph node ever exists [5, 21]. The findings of a nodal hemangioma by Gupta in 1964 [2] cast some doubts if they were primary nodal haemangioma or nodal angiomatosis [22]. Since then several isolated findings of nodal hemangioma have been reported in the medical literature. The number cases identified as nodal hemangioma is growing. It is important that this rare entity is recognised and included in the differential diagnosis and not to be mistaken for Kaposi sarcoma, angiosarcoma, metastatic cancer and other benign vasoproliferative lesions.

Conflict of Interest

The authors have no conflict of interests

Funding

This study is not supported by any funding organization

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Figures & Legends:

Figure 1: Upon removal of the submental lymph node

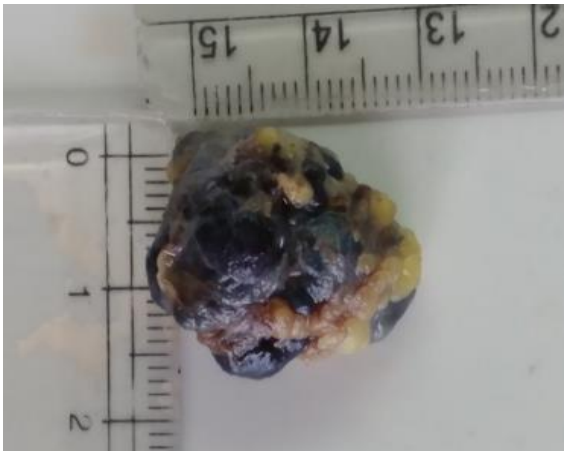

Figure 2: Gross appearance

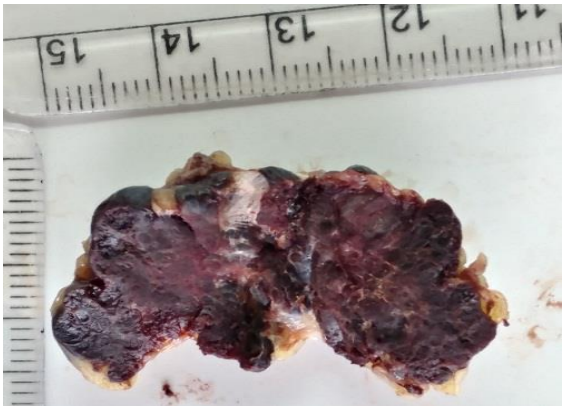

Figure 3: Vascular elements on Hemi Section

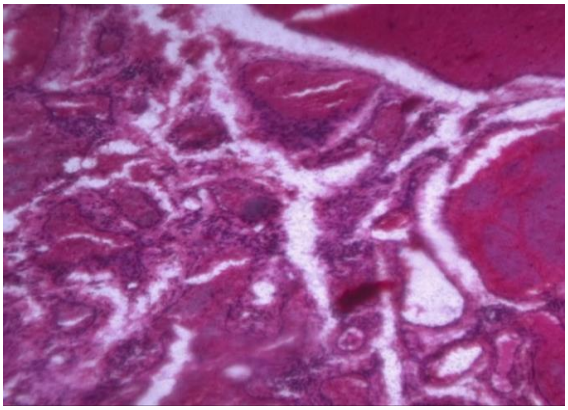

Figure 4: Vascular channels replacing the nodal parenchyma (H & E stain, 20x)