CASE REPORT

Jurnal Radiologi Dentomaksilofasial Indonesia December 2025, Vol. 9, No. 3: 183-187
P-ISSN.2685-0249 | E-ISSN.2686-1321

http://jurnal.pdgi.or.id/index.php/jrdi/index

A case presentation and management of a hamartomatous vascular lesion of a patient with kaposi sarcoma

Rana Erbalta[1*], Beyza Nur Cengiz[1], Melisa Öçbe[1], Fatih Mehmet Coşkunses[2], Vakur Olgaç[3]

ABSTRACT

Objectives: Intraosseous vascular lesions of the mandible are uncommon and often difficult to diagnose. This report presents the clinical, radiographic, and histopathological findings of such a lesion in an elderly patient with a history of Kaposi sarcoma, emphasizing diagnostic challenges and management considerations.

Case Report: A 73-year-old male with a medical history of Kaposi sarcoma was referred for evaluation of an edentulous mandible. Panoramic radiography and cone-beam computed tomography (CBCT) revealed a well-defined hypodense lesion adjacent to the mandibular canal, measuring 41 × 19 × 11 mm. The lesion demonstrated cortical thinning without expansion or perforation. An incisional biopsy revealed vascular lumens, and the final diagnosis was a hamartomatous vascular lesion. Considering the patient's advanced age and medical condition, conservative management with regular clinical and radiographic follow-up was chosen.

Conclusion: Although intraosseous vascular lesions of the mandible are rare, they should be included in the differential diagnosis of radiolucent jaw lesions. Radiographic and histopathological evaluations play a crucial role in diagnosis. In medically compromised or elderly patients, conservative management with close monitoring can be a suitable treatment option.

Keywords: *Hamartomatous lesions, vascular lesions, kaposi sarcoma*
Cite this article: Erbalta R, Cengiz BN, Öçbe M, Coşkunses FM, Olgaç V. *A case presentation and management of a hamartomatous vascular lesion of a patient with kaposi sarcoma*. Jurnal Radiologi Dentomaksilofasial Indonesia 2025;9(3)183-187. https://doi.org/10.32793/jrdi.v9i3.1436

[1]Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Kocaeli Health and Technology University, Kocaeli, Turkiye, 41275

[2]Kocaeli, Türkiye

[3]Department of Oral Pathology, Faculty of Dentistry, Istanbul University, Istanbul, Turkiye, 34104

*Correspondence to:
Rana Erbalta
rana.erbalta@kocaelisaglik.edu.tr

Received on: September 2025
Revised on: November 2025
Accepted on: December 2025

INTRODUCTION

Intraosseous vascular lesions are a rare, benign, and slowly progressive vascular tumor, accounting for approximately 0.5% to 1% of all benign bone neoplasms. While hemangiomas are more commonly found in soft tissues, their occurrence within bone is uncommon. Involvement of the facial skeleton is particularly rare, and when present, the mandible is affected in nearly two-thirds of cases.[1] They predominantly develop during the second decade of life, with a higher prevalence observed in female patients.[1,2] There is a lack of consensus among authors regarding the origin of the lesions. Some researchers consider it a true neoplasm, while others believe it represents a hamartoma resulting from the proliferation of intraosseous mesodermal cells undergoing endothelial differentiation.[1,2] They are considered to be benign tumors developing in the spongiosum with outward expansion, destroying the bony cortex.[3]

Hemangiomas are classified into three types based on the size of their vascular spaces: cavernous (the most common), capillary, and mixed.[4,5] Cavernous hemangiomas consist of large, endothelium-lined vascular spaces with minimal supporting stroma. Capillary hemangiomas are composed of smaller, densely cellular vessels with more prominent stroma. Mixed hemangiomas exhibit features of both cavernous and capillary types. All hemangiomas typically undergo an initial proliferative phase during early childhood, marked by rapid growth, increased vascularization, and heightened activity of endothelial and mast cells. This is followed by a regressive phase, characterized by increased fibroblastic matrix production and a reduction in the vascular component.[5] Their clinical presentation, diagnosis, and treatment approaches can vary significantly depending on the subtype, location, and patient-related factors. Most patients present with a painless, firm swelling of the jaw, sometimes accompanied by facial asymmetry, discomfort, pulsatile bleeding, bluish discoloration, mobile teeth, or changes in dental arch form.[1-4]

Orthopantomogram (OPG), Computed Tomography (CT), and Magnetic resonance imaging (MRI) are most frequently used diagnostic tools, and CT angiography is also useful for diagnostic

purposes.[1,2,4] In addition to these methods, cone-beam computed tomography (CBCT) has been widely used for radiologic evaluation in cases with maxillofacial involvement.[6,7] Radiographic features of vascular lesions are often nonspecific, making diagnosis challenging. The lesion typically presents as a radiolucent area, which may appear unilocular or multilocular and can exhibit patterns such as reticulated, honeycomb, or sunburst—particularly on tangential views. Due to the overlap in imaging characteristics, it can be difficult to distinguish vascular lesions from other lesions like ameloblastoma, odontogenic myxoma, fibrous dysplasia, and aneurysmal bone cyst.[1,2,6] In this case report, we aimed to present the preliminary evaluation with CBCT, histopathological assessment, and management of a vascular hamartomatous lesion.

CASE REPORT

A 73-year-old male patient was referred to the outpatient clinic of XXX University, Faculty of Dentistry, Department of Oral and Maxillofacial Radiology, for the evaluation of partially edentulous areas in the mandible. His chief complaint was related to missing teeth in the posterior region of the mandible. The patient's medical history revealed systemic hypertension managed with calcium channel blockers. Additionally, the patient had a documented history of Kaposi's sarcoma involving the soft tissue of the hand and leg.

As part of the routine diagnostic workup, a panoramic radiograph was obtained. Radiographic examination revealed a well-demarcated unilocular radiolucent area in the left mandibular premolar and molar region. Given the radiographic findings, further imaging with cone beam computed tomography (CBCT) was recommended to better characterize the lesion and its anatomical relations.

CBCT imaging demonstrated a hypodense, partially well-defined lesion extending from the mandibular canine to the second molar region. The lesion was in close proximity to the mandibular canal, with its peripheral borders adjacent to the neurovascular bundle. The dimensions of the lesion were measured as 41x19x11 mm. While cortical bone destruction was evident, there was no observable expansion or perforation of the buccal or lingual cortical plates (Figure 1).

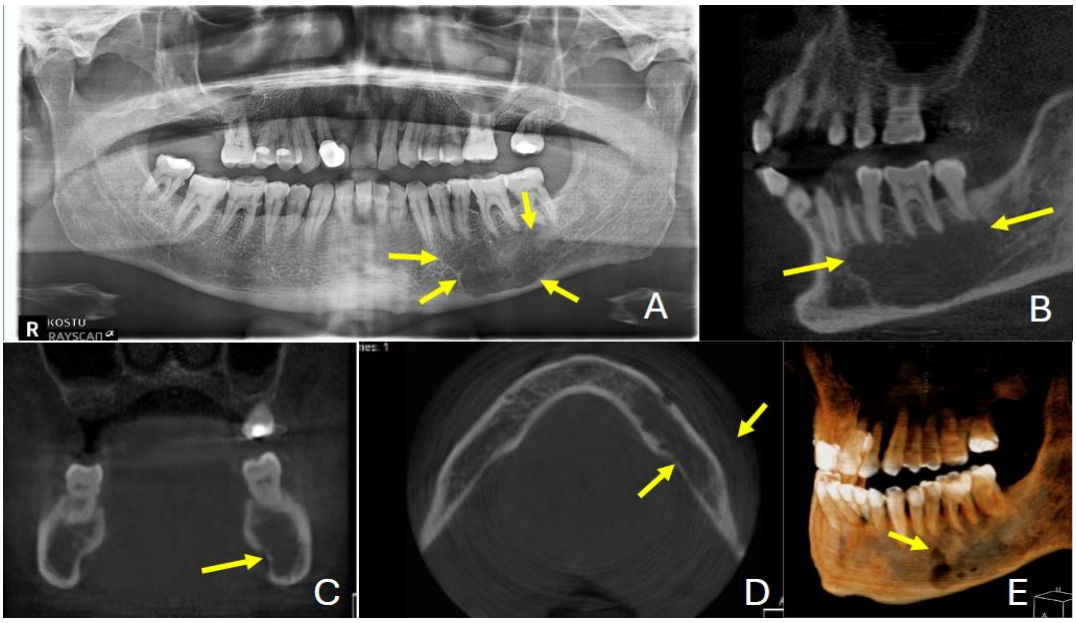

Figure 1. (A). Panoramic radiograph showing a hypodense lesion in the left mandible, extending from the canine to the second molar region. **(B)**. Oblique sagittal cone-beam computed tomography (CBCT) section demonstrating a partially well-defined lesion in close proximity to the mandibular premolars and molars, with evidence of destruction in the inferior mandibular cortex. **(C)**. Coronal CBCT section revealing cortical thinning of the mandibular cortex. **(D)**. Axial CBCT section illustrating cortical thinning and destruction in both the buccal and lingual aspects of the mandibular cortex. **(E)**. Three-dimensional reconstruction image showing trabecular bone loss within the lesion

Based on the radiographic appearance and anatomical extent, the differential diagnosis included odontogenic keratocyst, glandular odontogenic cyst, and aneurysmal bone cyst. Pulp vitality testing was conducted for the teeth within the affected area, and all teeth from the canine to the second molar responded positively to vitality tests, indicating vital pulp tissue.

To establish a definitive diagnosis, an incisional biopsy was planned. Under local anesthesia, a cortical window was created through the buccal bone to access the lesion. Aspiration was performed prior to biopsy, and the result was negative for fluid or pus. Upon entry into the lesion, a partially hemorrhagic soft tissue was encountered, with areas suggestive of vascularity and a friable consistency (Figure 2). A representative tissue sample was obtained, and the surgical site was closed primarily using resorbable sutures. The patient was prescribed a course of antibiotics (amoxicillin 1 g, twice a day for 7 days) and analgesics (paracetamol 500 mg, as needed for pain). Postoperative follow-up was scheduled for one week later.

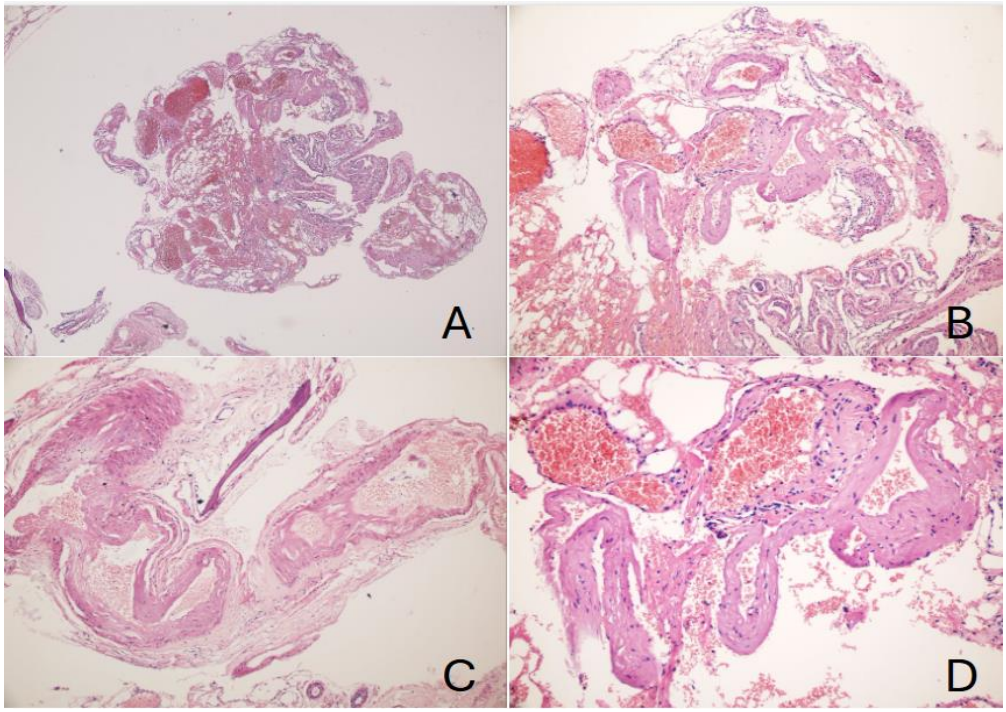

Figure 2. Vascular lumens lined by endothelium and displaying variable wall thickness, forming anastomoses with each other, are observed. (Hematoxylin and Eosin stain; A: ×40, B: ×100, C: ×100, D: ×200)

Histopathological examination of the biopsy specimen revealed a loose connective tissue stroma interspersed with numerous blood vessels of varying calibers. The vascular channels were lined by flattened endothelial cells and exhibited lumens filled with erythrocytes. Some vessels showed marked dilation, forming cavernous-like spaces within mature adipose tissue. These findings were consistent with a hamartomatous vascular lesion (Figure 3).

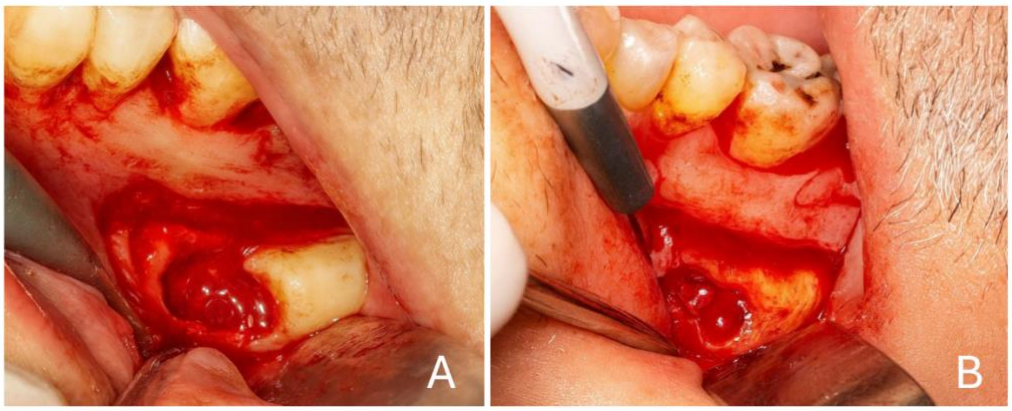

Figure 3. Intraoperative images (A, B) showing the surgical area

The final histopathological diagnosis was hamartomatous vascular lesion. Following the histopathological confirmation, the patient was referred for angiographic evaluation to further assess the vascular characteristics of the lesion and to guide potential treatment planning. Due to the older age of the patient, follow-up was recommended.

DISCUSSION

Intraosseous vascular lesions are rare lesions, and they most frequently occur in the vertebrae, followed by the bones of the skull. Intraosseous hemangiomas of the mandible, however, are exceptionally uncommon and present significant diagnostic challenges both clinically and radiographically.[8] While some cases may exhibit pain and swelling, mandibular intraosseous hemangiomas are typically asymptomatic. However, in the case report of Kumar et al., the patient presented to the clinic with pain and swelling that had been ongoing for five years.[1] Occasionally, patients may report numbness in the lips or mental region.[9] In the case report of Bilgir et al., the cavernous hemangioma of a 28-year-old male patient was asymptomatic and revealed with intraoral swelling.[6] In our case, the swelling and pain were absent.

On OPGs, intraosseous hemangiomas may present with either well-defined or poorly defined borders, depending on the amount of residual bone

Jurnal Radiologi Dentomaksilofasial Indonesia 2025; 9 (3); 183-187 | DOI: https://doi.org/10.32793/jrdi.v9i3.1436

surrounding the lesion. In our case, the lesion observed in the patient had partially well-defined borders, and the internal structure was hypodense. Central hemangiomas typically appear as multilocular radiolucencies with a "soap-bubble" or "honeycomb" pattern. CBCT often reveals these lesions as homogeneous osteolytic areas with well-demarcated borders.[10] In the case report of Bilgir et al., the internal structure of the cavernous hemangioma was found as a homogenous hyperdense lesion, which was thought to be a mandibular torus during the preliminary evaluation.[6] In contrast, the case report by Kumar et al. revealed CBCT findings of a lesion with poorly defined borders and a heterogeneous appearance.[1] In our case report, the lesion was apparent in panoramic radiography and in CBCT images. In addition, in contrast with the case report of Bilgir et al., the spiculated characteristics of the lesion were absent in our case.[6] In addition to these various appearances of hemangioma, in the case report by Jorge et al., a sunburst-like appearance was observed on the obtained CT images.[4] Several reports emphasize the importance of additional diagnostic tools, such as fine-needle aspiration and arteriography, when an intraosseous hemangioma is suspected. In the case report of Oliveira Gómez et al.,a fine needle aspiration cytology was performed to confirm the diagnosis.[2]

Despite their rarity, accurate diagnosis is crucial, particularly in the absence of malignant features and in cases where imaging reveals hypervascularity and strong contrast enhancement.[9] In contrast-enhanced imaging methods, such as (Magnetic Resonance Angiography) MRA, CT, angiography, cavernous hemangiomas may show minimal contrast accumulation on MR angiography. Patchy contrast enhancement can be observed, and these areas may gradually fill in on subsequent post-contrast sequences.[11]

In the differential diagnosis of vascular lesions, arteriovenous malformations, kaposiform hemangioendothelioma, and arteriovenous hamartomas should be carefully considered due to overlapping clinical and imaging features. Arteriovenous malformations are congenital high-flow vascular anomalies that typically lack endothelial proliferation and are instead characterized by progressively ectatic, dysplastic vessels lined with flattened endothelial cells, a pattern observed in the present case.[12] Although these histopathologic features supported a diagnosis of arteriovenous malformation, the patient's medical history of Kaposi sarcoma raised concern for a possible kaposiform or Kaposi-like endothelial proliferation. Kaposiform hemangioendothelioma, however, usually displays spindled or "kaposiform" endothelial cells with infiltrative growth and lymphangioma-like channels, none of which were evident here.[13] Additionally, no Kaposiform cells or hemosiderin-laden macrophages were identified, ruling out kaposiform hemangioendothelioma. Arteriovenous hamartoma, a rare benign vascular malformation composed of an abnormal mixture of arteries and veins without an intervening capillary bed, also presents with flat endothelium but typically exhibits a more organized vascular pattern than seen in this lesion. The rapid blood flow within these vessels poses a significant risk, as rupture may result in life-threatening hemorrhage. This is of particular concern for dental practitioners when the lesion is closely associated with teeth indicated for extraction.[14]

Treatment planning is determined based on the patient's age, medical history, and the specific characteristics of the lesion.[15] According to the literature, surgical intervention is the most commonly employed treatment modality. For instance, in the case report of Bilgir et al., the total excision of the lesion was performed.[6] Other alternative approaches include embolization, radiotherapy, and intralesional steroid injections.[1] In the present case, an incisional biopsy was performed under local anesthesia. A mucoperiosteal flap was elevated, and, based on preoperative CBCT localization, a buccal cortical window was prepared over the molar region. Fine-needle aspiration was performed; however, aspiration was negative (no blood or purulent material). A specimen of loose connective tissue was obtained from the lesion, and hemostasis was achieved. The flap was then repositioned and closed with interrupted sutures. Histopathological examination confirmed the diagnosis, and, given the patient's age and systemic condition, conservative management with follow-up was recommended.

CONCLUSION

Vascular hamartomatous lesions, although extremely rare, should be included in the differential diagnosis of radiolucent jaw lesions, particularly when imaging reveals a partially or completely well-defined, hypervascular area. Their clinical presentation is often asymptomatic, making diagnosis challenging without appropriate imaging and histopathological evaluation. Misdiagnosis can result in inadequate treatment and serious intraoperative complications such as excessive bleeding. Therefore, early recognition and a multidisciplinary approach are essential for accurate diagnosis and effective management. Proper treatment planning is needed, and the lesion's size, location, and vascularity are crucial to ensure both functional preservation and favorable esthetic outcomes.

ACKNOWLEDGMENTS

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FOOTNOTES

Conflict of interest: All authors have no conflict of interest to declare for this article.
Human rights statement: Informed consent was obtained from the patient for being included in this case report.

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