

A RARE LOCALIZATION OF INTRAOSSEOUS HEMANGIOMA: CASE REPORT

NADİR BİR İNTRAOSSEÖZ HEMANJİOM LOKALİZASYONU: OLGU SUNUMU

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ABSTRACT

Hemangiomas are defined as benign neoplasms originating from vascular endothelial tissue. Intraosseous hemangiomas are very rare and account for less than 1% of bone tumors. They are mostly seen in vertebrae, skull bones and long bones. Intraosseous hemangiomas are very rare in jaws. Intraosseous hemangiomas are typically asymptomatic and grow slowly. In this case report, asymptomatic intraosseous hemangioma was found in the mandible and the patient was evaluated clinically, radiologically and histopathologically. A 69-year old male patient was referred to our clinic with a multilocular lesion. CBCT and MRI were evaluated for further examination. Histopathological examination revealed intraosseous hemangioma in the mandible.

Keywords: Hemangioma, Mandible, CBCT, MRI

ÖZ

Hemanjiomlar, vasküler endotelial dokudan kaynaklanan iyi huylu bir neoplazm olarak tanımlanır. Kemik içi hemanjiomlar çok nadirdir ve kemik tümörlerinin %1'inden azını oluşturur. En çok vertebralarda, kafa kemiklerinde ve uzun kemiklerde görülür. Çenelerde intraosseöz hemanjiom çok nadir görülür. İnaosseöz hemanjiomlar tipik olarak asemptomatik ve yavaş büyür. Bu olgu sunumunda mandibulada asemptomatik intraosseöz hemanjiom saptanmış ve hasta klinik, radyolojik ve histopatolojik olarak değerlendirilmiştir. 69 yaşında erkek hasta multiloküler lezyon ile kliniğimize sevk edildi. İleri inceleme için KIBT ve MRG değerlendirildi. Histopatolojik incelemede mandibulada intraosseöz hemanjiom saptandı.

Anahtar Kelimeler: Hemanjiom, Mandibula, KIBT, MRG

INTRODUCTION

Hemangiomas are benign vascular lesions originating from endoepithelial tissue (1). They are usually seen in soft tissues. Intraosseous hemangiomas (IH) are extremely rare and constitute less than 1% of all bone lesions (2). These lesions frequently occur in the vertebrae, long bones, and the skull (3). While it is quite rare in the jaws, it is more common in the mandible than the maxilla (4). They usually occur in female patients and in the fifth decade (5). The most common clinical findings are: painless swelling, facial asymmetry and pulsation. These tumors are usually painless (6).

Central hemangiomas often have a radiolucent internal structure and well-defined or ill-defined borders in panoramic radiography (7). Diagnosis should be supported by Cone Beam Computed Tomography (CBCT), Computed Tomography (CT) and Magnetic Resonance Imaging (MRI). CBCT, which has good hard tissue resolution at relatively low radiation doses, shows the bone structure well and is often chosen to determine the borders of the lesion in the bone. However, CT, which can demonstrate soft tissue pathologies as well and has good bone tissue resolution, is used more frequently. MRI examination should be chosen in the evaluation of the vascular structure of intraosseous lesions. Depending on the amount of venous blood flow in the lesion in MRI examination, the lesion is mostly

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isointense in the T1-weighted sequence, while heterogeneous hyperintensity is seen in the T2-weighted sequence (8). One of the MRI findings of hemangioma is the presence of signal void areas corresponding to calcified areas such as trabeculation or phleboliths (9). They have moderate or high echogenicity in ultrasound images and low blood flow in Doppler imagings (8). Intraosseous hemangiomas grow slowly and treatment is often not necessary. However, if treatment is required for aesthetic or functional reasons, treatment options are excision, embolization, radiotherapy, and curettage. Radiotherapy is useful to reduce the tumor volume, but it has a lot of adverse effects such as damage to healthy tissues, residual scarring and malign transformation. Therefore, radiotherapy is considered an unfavorable therapeutic option (10). However, there are case reports in which radiotherapy was chosen since the hemangiomas were life-threatening (11).

Case Report

A 69-year-old male patient was referred to our clinic with aesthetic and prosthetic complaints. Oral examination revealed endodontic and periodontal problems with missing teeth. A radiolucent lesion with ill-defined borders at the mandibular right premolar-molar toothless region was detected on pre-taken OPG (Figure 1). A CBCT was performed to evaluate the lesion in three dimensions. A non-expansive, 18.5mm x 8.5mm x 17.8mm sized hypodense multilocular lesion with minor trabeculation was observed in the mandibular posterior region (Figure 2). Although the destruction of the lingual cortical bone was smaller than 2.5 mm, there was no clinical condition compatible with paresthesia in the anamnesis taken from the patient. An MRI was performed to evaluate its vascular structure and understand its tumour character. The lesion was seen to be isointense in T1-weighted sequence and predominantly hyperintense with signal void areas in the T2-weighted sequences (Figure 3). These results confirm the radiological diagnostic criteria of hemangioma as defined in the literature.

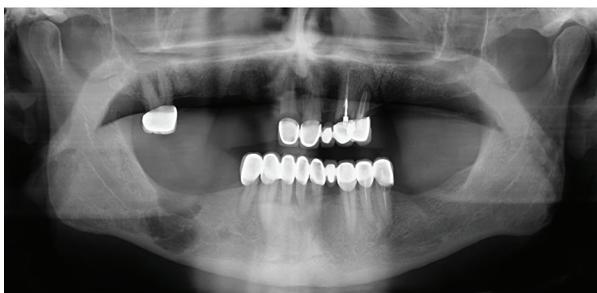


Figure 1: Radiolucent lesion with a soap bubble appearance in the right premolar-molar region of the mandible in panoramic radiograph.

There was no problem in the mucosa in the intraoral examination of the patient before the operation. Mandibular block and buccal infiltration anesthesia were applied to the patient. The lesion was accessed by removing the mucoperiosteal flap over the alveolar crest. Excisional biopsy was decided upon

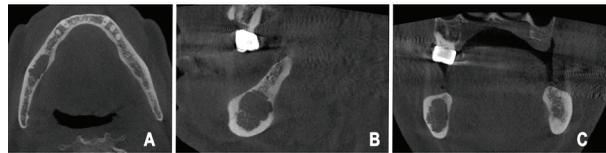


Figure 2: The hypodense lesion with a small amount of trabeculation in CBCT (A: Axial, B: Sagittal, C: Coronal).

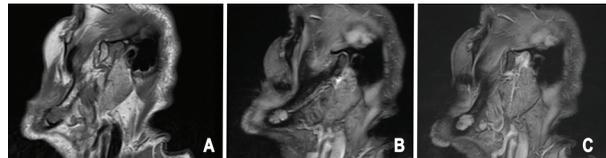


Figure 3: A: The lesion appears isointense in the T1-weighted sequence, B-C: The lesion is heterogeneous hyperintense with signal void areas in the T2-weighted sequence.

because, unlike most hemangioma cases, there was no bleeding in the lesion. In fact, this situation led us away from the pre-diagnosis of hemangioma. The lesion was excised and was sent for histopathological examination, in addition, the cavity walls were cured. There are studies in the literature showing that simple curettage can cause uncontrolled bleeding. As a result of excessive bleeding, insufficient curettage can be performed. The weakness of our case report may be to ignore this situation. However, the absence of bleeding eliminated the risk of incomplete treatment (Figure 4, 5, 6).

Histologic examination of the specimen with a hematoxylin and eosin stain demonstrated trabeculae between the bones, a loose structure, fiber-rich connective tissue lined with many endothelium, and vascular lumens. The endothelium and vas-

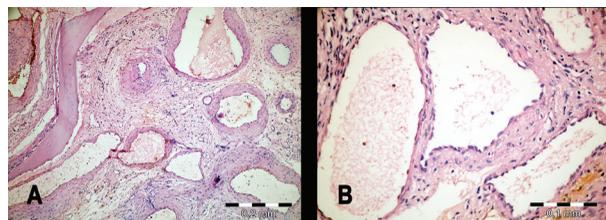


Figure 4: A: Trabeculae between the bone, loose structure, lined with many endothelium in the fiber-rich connective tissue, vascular lumens (H&E X200), B: Vascular lumens lined with endothelium and fibrin particles in them (H&E X400).

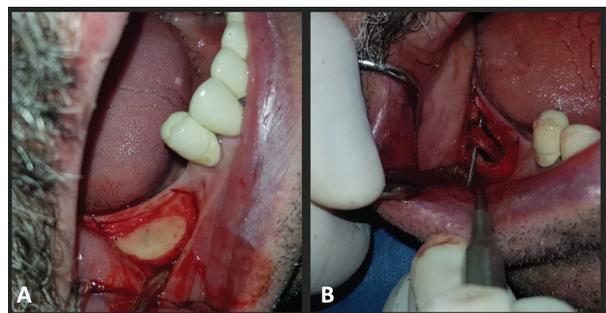


Figure 5: A: Image after removal of the mucoperiosteal flap, B: Excising the lesion



Figure 6: Lesion removed in three parts

cular lumens were observed to be covered with fibrin particles. Thus, the histological diagnosis of the specimen revealed intraosseous hemangioma. On the second postoperative day, ecchymosis developed on the same side of the patient. Ice treatment for ecchymosis was recommended. Ten days later, the patient's ecchymosis disappeared (Figure 7).



Figure 7: Image of ecchymosis in the patient's intraoral mucosa on the second postoperative day.

DISCUSSION

IH are rare benign lesions of the bones. These lesions are accepted by the World Health Organization as being of vasculature or endothelial origin. However, some researchers think they are hamartomas, which later become vascularized and differentiate from mesodermal cells (12). Histopathologically, IH is classified as venous, cavernous and capillary. The cavernous type has large vessels and sinusoids with a thin-single endothelial layer. The capillary type is a vascular network formed by small vessels. Often in hemangiomas, the components of the cavernous and capillary types are mixed (13). Although malignant transformation is very rare, only a few cases have been reported after radiotherapy. It is usually congenital, less often posttraumatic (10). It is also defined as a hamartoma by some researchers because it can occur post traumatically (5).

Intraosseous hemangiomas constitute less than 1% of all bone tumors. In our case, the lesion was in the right premolar-molar region of the mandible (14). Lesions in the jaws appear on

panoramic radiography, as they are used more frequently in a routine examination. Although it is seen in different bones in the skeletal system, it is more than 50% more common in the skull and vertebrae (15). While the localization of the mandible is very rare, it is most common in the mandible in the premolar-molar region (16). Since most patients are asymptomatic, the lesion is often detected incidentally. When it reaches large dimensions, it may cause complaints such as asymmetry for aesthetic reasons. In some cases, painless swelling, facial asymmetry or pulsation may be seen. Our case applied to our clinic for prosthetic reasons; during the radiological examination, the lesion was detected incidentally. CT is often used because it allows us to evaluate soft and hard tissue in diagnosis. An angiogram and MRI can be used to evaluate the vascular structure. The trabeculation of the lesion in CBCT and CT often has the appearance of a soap bubble or honeycomb. Intraosseous hemangioma in MRI is seen to be isointense in the T1 sequence and hyperintense in the T2 sequence. Zlotogorski et al. examined the radiographic features of intraosseous hemangiomas in the jaws of 86 patients (17). In this study, it was reported that these lesions were 96.1% radiolucent, 65.8% multilocular (honeycomb, soap bubble) and with 68.3% diffuse borders. Fernandez et al. reported in their study that intraosseous lesions rarely cause resorption and displacement in the teeth (18). The lesion in our case was located in the edentulous region and radiolucent, multilocular and with ill-defined borders. The differential diagnosis of these lesions often includes osteoid osteoma, fibrous dysplasia, and osteosarcoma. Although a diagnosis cannot be made solely on imaging, there are some key findings to help differentiate intra-osseous hemangiomas from fibrous dysplasia and osteosarcoma. While intraosseous hemangiomas may appear like 'soap bubbles' and 'sun bursts', fibrous dysplasias have a ground-glass appearance. In addition to the sun burst image, osteosarcomas often show a periosteal reaction in the form of 'onion skinning'. This image is not typical for intraosseous hemangiomas (19). Treatment of these lesions is often unnecessary. Surgical intervention or embolization is required when it causes aesthetic and functional problems. They may cause life-threatening bleeding in tooth extractions associated with the lesion. When the lesions are large, they may require reconstruction. However, such a treatment was not needed in our case.

Some precautions should be taken due to the risk of bleeding that may occur during hemangioma surgery. There are frequent cases of embolization hemangioma in the literature. However, since there was no bleeding in our case, such measures were not needed. Nevertheless, it should be performed together with a presurgical embolization in large lesions to minimize the surgical bleeding.

CONCLUSION

Hemangiomas are benign lesions, but their differential diagnosis is critical. Since the excessive bleeding may endanger patients' lives, the differential diagnosis of hemangiomas is of prime importance. The relationship between IHs and adjacent

tissues should be evaluated clinically and radiologically. Moreover, situations that would create a risk of bleeding should be taken into consideration and a treatment procedure should be established accordingly.

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REFERENCES

1. Aksakalli N, Siraneci P, Ramazanoglu M, Ercal O, Gümrü O. PP102: Intraosseous hemangioma of the mandible: An unusual presentation. *Oral Oncol* 2013;49:129.
2. Hansen T, Kunkel M, Katenkamp D, Eletr S, Wagner W. Hemangioma of the mandible: case report with special emphasis on bone degradation. *Oral Maxillofac Surg* 2009;13(4):239.
3. Dereci O, Acikalin MF, Ay S. Unusual intraosseous capillary hemangioma of the mandible. *Eur J Dent* 2015;9(03):438-41.
4. Özdemir R, Alagoz S, Uysal AÇ, Ünlu RE, Ortak T, Sensoz Ö. Intraosseous hemangioma of the mandible: a case report and review of the literature. *J Craniofac Surg* 2002;13(1):38-43.
5. Valentini V, Nicolai G, Lorè B, Aboh IV. Intraosseous Hemangiomas. *J Craniofac Surg* 2008;19(6):1459-64.
6. Perugini M, Renzi G, Gasparini G, Cerulli G, Becelli R. Intraosseous hemangioma of the maxillofacial district: clinical analysis and

surgical treatment in 10 consecutive patients. *J Craniofac Surg* 2004;15(6):980-5.

7. Moore SL, Chun JK, Mitre SA, Som PM. Intraosseous hemangioma of the zygoma: CT and MR findings. *Am J Neuroradiol* 2001;22(7):1383-5.
8. Kose OD, Tanyel C, Kose TE, Erdem MA, Cankaya AB. Tooth extraction from a patient with cavernous hemangioma in maxillofacial region: case report. *J Istanbul Univ Fac Dent* 2016;50(1):51.
9. Moukaddam H, Pollak J, Haims AH. MRI characteristics and classification of peripheral vascular malformations and tumors. *Skeletal Radiol* 2009;38(6):535-47.
10. Cheng N-C, Lai D-M, Hsie M-H, Liao S-L, Chen Y-BT. Intraosseous hemangiomas of the facial bone. *Plast Reconstr Surg* 2006;117(7):2366-72.
11. Park S, Yoon SM, Lee S, Park J-h, Song SY, Lee S-w, et al. Role of fractionated radiotherapy in patients with hemangioma of the cavernous sinus. *Radiat Oncol J* 2017;35(3):268.
12. Eliot CA, Castle JT. Intraosseous hemangioma of the anterior mandible. *Head Neck Pathol* 2010;4(2):123-5.
13. Politi M, Romeike BF, Papanagiotou P, Nabhan A, Struffert T, Feiden W, et al. Intraosseous hemangioma of the skull with dural tail sign: radiologic features with pathologic correlation. *Am J Neuroradiol* 2005;26(8):2049-52.
14. Oliveira GG, García-Rozado A, Rey RL. Intraosseous mandibular hemangioma. A case report and review of the literature. *Med Oral Patol Oral Cir Bucal* 2008;13(8):E496-8.
15. Mutlu M, Yaris N, Aslan Y, Kul S, Imamoglu M, Ersöz S. Intraosseous noninvolving congenital hemangioma of the mandible in a neonate. *Turk J Pediatr* 2009;51(5):507.
16. Drage N, Whaites E, Hussain K. Haemangioma of the body of the mandible: a case report. *Br J Oral and Maxillofac Surg* 2003;41(2):112-4.
17. Zlotogorski A, Buchner A, Kaffe I, Schwartz-Arad D. Radiological features of central haemangioma of the jaws. *Dentomaxillofac Radiol* 2005;34(5):292-6.
18. Fernández L, Luberti R, Domínguez F. Radiographic features of osseous hemangioma in the maxillo-facial region. Bibliographic review and case report. *Med Oral* 2003;8(3):166-77.
19. Brandner JS, Rawal YB, Kim LJ, Dillon JK. Intraosseous hemangioma of the frontal bone. Report of a case and review of the literature. *J Oral Maxillofac Surg* 2018;76(4):799-805.