



Case Report / Olgu Sunumu

Central Hemangioma Involving the Mandible: A Rare Condition: Case Report and Review of Literature

Mandibulayı İçeren Sentral Hemanjiom: Ender Bir Durum: Olgu Sunumu ve
Literatürün Gözden Geçirilmesi

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ABSTRACT

Intraosseous hemangiomas are quite a rare condition and those occurring in the mandible often pose a diagnostic dilemma to the Dental surgeon as they present with variable and atypical radiographic manifestations. Biopsy or surgical excision of the lesion can result in severe haemorrhage, leading even to death; therefore accurate clinical and radiographic diagnosis is essential before any surgical intervention is started. In this paper, we discuss a case of central hemangioma of the mandible occurring in a 17-year-old male patient, angiography was used for diagnosis and was treated with surgical excision.

Keywords: Central hemangioma, intraosseous hemangioma, mandible, facial artery

ÖZET

İntraosseöz hemanjiomlar ender görülen bir durumdur ve mandibulada görülen hemanjiyomlar sıklıkla dişhekimleri için değişken ve atipik radyografik bulgularla ortaya çıktığı için tanı ikilemi oluştururlar. Biyopsi ya da lezyonun cerrahi olarak eksizyonu ciddi kanamalara neden olabilir, ölümlerle bile sonuçlanabilir; bu nedenle herhangi bir cerrahi işleme başlamadan önce doğru klinik ve radyolojik tanı gereklidir. Bu yazıda, 17 yaşındaki erkek hastada mandibulanın santral hemanjiyom olgusu tartışılmıştır. Anjiyografi tanı için kullanılmış ve cerrahi eksizyon ile tedavi edilmiştir.

Anahtar kelimeler: Santral Hemanjiom, intraosseöz hemanjiom, mandibula, fasyal arter

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INTRODUCTION

Hemangioma is a common benign congenital lesion affecting humans. Consisting of three phases: proliferating, involuting and involuted phases, they are present at birth and gradually regress. Although its origin has been debated over the years, World Health Organization (WHO) considers them to be true benign vaso-formative neoplasm; whereas, other authors deem them to be a hamartoma. ^[1,2]

Although cutaneous hemangiomas are common, Intraosseous hemangiomas are a rare condition, comprising less than 1% of all intraosseous tumors. It mainly occurs in the vertebral column and calvaria. Lesions occurring in the Mandible are very rare but when they do occur, they are seen in the body region and often pose a diagnostic dilemma to the Dental surgeons. Biopsy or surgical excision of the lesion can result in severe haemorrhage sometimes leading to death; therefore accurate clinical and radiographic diagnosis is essential before any surgical intervention is started. The following rare case of Intra-osseous hemangioma of the mandible with the facial artery as the feeder artery is being discussed. ^[2-4]

CASE REPORT

A 17-year old male patient reported to us with the mobility of lower right back teeth since 2 months without any associated symptoms of pain. The patient did not give any history of trauma to that region. Past medical, dental, family and personal history were non-contributory.

Extra-oral examination revealed the presence of a diffuse swelling measuring approximately 2 x 3 cms below the angle of the mandible on the right side (Figure 1). The skin over the swelling was normal with no visible pulsations. On palpation, all inspectory findings were confirmed; the swelling was firm, fixed to the underlying structures and non-tender. Lymph nodes were not palpable. On intra-oral examination, there was obliteration of the lower right buccal vestibule (Figure 2). The surrounding mucosa appeared normal. On hard tissue examination, 47 was supra-erupted with grade III mobility; The tooth also showed a pumping movement when the pressure was applied to it and released.

Intraoral periapical radiograph of right mandibular molars showed an ill-defined radiolucency involving the roots of 46 and 47 (Figure 3a). Panoramic radiograph depicted the presence of ill-defined radiolucency, extending from the mesial root apex of 46 up to mid-ramus area measuring about 2 x 3 cm. Resorption of the distal root of 46 and both the roots of 47 was noted (Figure

3b). A provisional diagnosis of an odontogenic tumour and differential diagnosis of a radicular cyst, solitary bone cyst, and central hemangioma were considered.

Computed Tomography (CT) was advised to further evaluate the lesion. Axial section revealed an expansile osteolytic lesion with septae (Figure 4). Inferior border of mandible showed thinning. CT angiogram was performed by intravenous injection of 60 ml of Iomeron 400 mg, which revealed an expansile lesion with multiple well-defined septae having soap bubble-like appearance involving the angle of mandible on the right side, giving an impression of hemangioma; facial artery was traced to be the feeding vessel (Figure 5). With the above CT angiographic feature, an impression of a hemangioma with facial artery as the feeder vessel was given. The mandible was resected beyond the lesion's radiographic boundaries. Since the loss of blood during surgery was anticipated, a provision was made for rapid replacement of blood in case of massive hemorrhage. After surgical preparation of the patient for general anesthesia, a submandibular incision was made extending from the angle of mandible to the symphyseal area. Enbloc resection of the mandibular lesion—including 1 cm of healthy bone—was performed. The mandibular fragment was sent to surgical pathology for histopathological diagnosis. The results confirmed the pre-surgical diagnosis of central hemangioma. The post-operative period was uneventful and the patient was kept on a 6 month follow-up.

DISCUSSION

Hemangioma of bone has been classified by Thoma into two types: peripheral type and central / intraosseous type. Central Hemangiomas can further be classified into cavernous type, capillary type, mixed variant and scirrhou type.^[5] Most central hemangiomas are of the cavernous type. Cavernous type of central hemangiomas consists of large, thin-walled vessels or sinusoids lined with a single layer of endothelium. These are separated by thin septa of connective tissue whereas capillary type of hemangioma comprises of many small capillaries lined by a single layer of endothelial cells, which are supported in connective tissue stroma. Mixed variant as the name suggests has features of both cavernous and capillary type and scirrhou variant consists of abundant proliferating connective tissue.^[4-6]

Most commonly central hemangiomas are seen to occur in the vertebral column and calvarium and rarely in the axial skeleton. Our case was seen to occur in the mandible making it a rare case. The female to male ratio is 2:1, unlike our case, which was seen in a male patient. They are mostly seen to occur between the second and fifth decades of life.

Patients report to the clinician with varied manifestations. The patient may be asymptomatic or may present with swelling, bleeding, bluish discoloration, mobile teeth, deranged arch or early dental exfoliation. A very prominent feature is blood oozing from the sulcus. The teeth involved may also show a pumping movement when pressure is applied to it and released as was seen in the case reported here.^[7, 8]

Being called “The great mimicker”, they also have varied radiologic features. Differential diagnosis based on radiographic picture could include ameloblastoma, odontogenic myxoma, osteosarcoma, fibrous dysplasia, central giant cell granuloma, multiple myeloma, dentigerous cyst and aneurysmal bone cyst. Most commonly this lesion appears as a multilocular radiolucency with small or large loculations giving the appearance of honeycomb or soap-bubble appearance respectively, making the clinician come to a diagnosis of Ameloblastoma, Central giant cell granuloma or Odontogenic myxoma. Phleboliths-like small rounded or sausage-shaped radiopacities with concentric structure and small radiolucent dot in the center may be appreciated. In another variation, it may give a picture of sun-burst or sun-ray appearance resembling osteosarcoma or chondrosarcoma. Sometimes the lesions may also show a unilocular cyst-like radiolucency.^[6-8]

As central hemangiomas have marked variability in its radiological appearance, based on just plain radiographs a definitive diagnosis cannot be made. Other imaging modalities such as computed tomography (CT), magnetic resonance imaging (MRI), ultrasonography (US) and thermography have been used. Intra-arterial angiography remains the gold standard for evaluating vascular lesions. In this case the diagnosis of the facial artery being the feeder artery was made only by CT angiography.^[9]

Numerous treatment options depending on the case are recommended; they include injecting sclerosing agents such as sodium morrhuate and absolute ethanol. Radiation and cryotherapy also have been tried but have high-risk side effects including possible malignant transformation and loss of innervation respectively. Temporary relief with embolization using silicone pellets or isobutyl cyanoacrylate has also been tried. Surgical excision with reconstruction of mandible remains the preferred treatment and was done in this case.^[6, 7, 9]

CONCLUSION

Central hemangiomas have varied clinical and radiographic manifestations and an incorrect diagnosis and treatment could prove fatal. Hemangiomas must always be considered in the differential diagnosis and proper investigations especially imaging studies must always be carried out prior to any surgical intervention.

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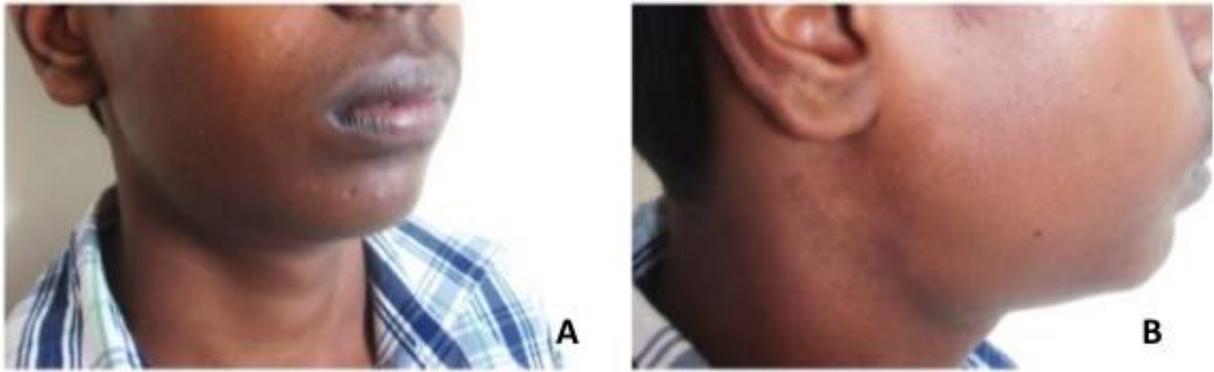


Figure 1. Extra-oral photograph showing diffuse swelling on the right side of the body and ramus of the mandible



Figure 2. Intra-oral photograph showing obliteration of the lower right buccal vestibule



Figure 3a and 3b. IOPAR and OPG showing ill-defined radiolucency at the apex of 46 and 47. Root apex resorption can be appreciated

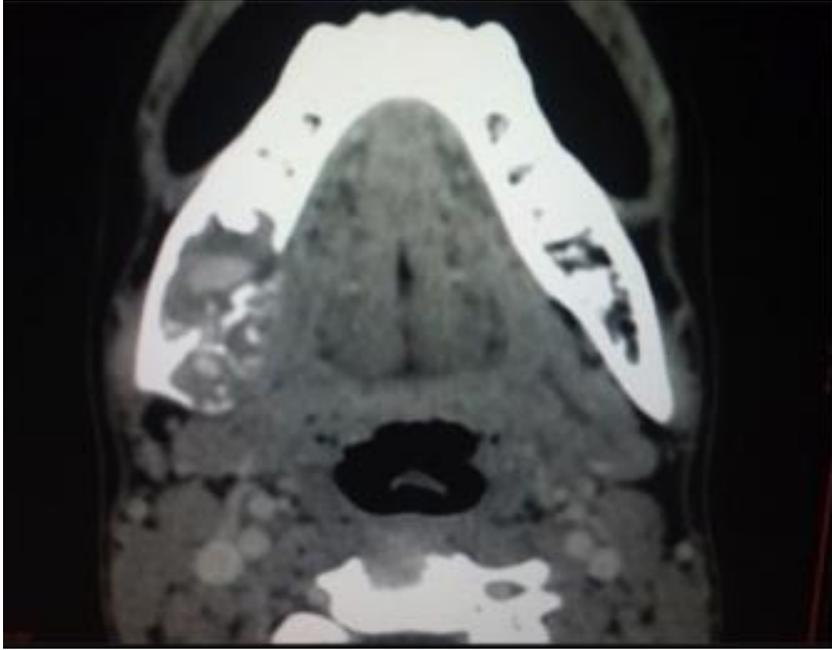


Figure 4. CT –Axial Section Showing perforation of the cortical plate

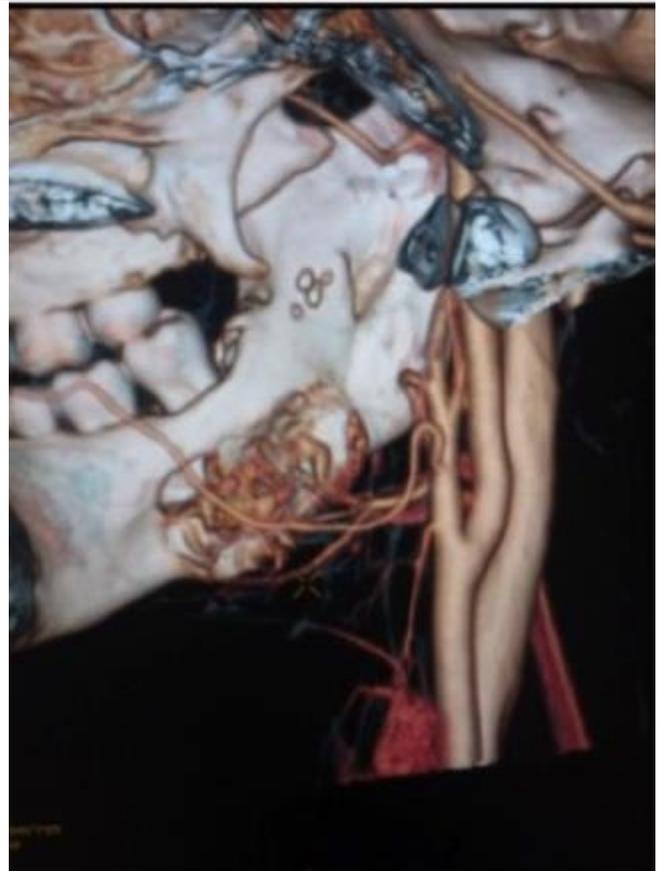


Figure 5. CT Angiography showing facial artery as the feeder vessel.