

Intra-Osseous Hemangioma of The
Mandible: A Case ReportZiegler CM^{1*} and Odegard A²¹Department for Oral and Maxillofacial Surgery, St. Olavs University Hospital, Norwegian University of Science and Technology (NTNU), Trondheim, Norway²Department of Radiology, St. Olavs University Hospital, Trondheim, Norway

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Abstract

Arterio-venous malformations of the jaws are a very rare pathology, but can lead to fatal complications. Presented is the case of a young female patient who was referred to our hospital based on the clinical and radiological diagnosis of an odontogenic cyst related to the first molar in the right mandible. Due to a suspicious mobility of the tooth and root resorption a biopsy and tooth extraction was planned under general anaesthesia. Already during mobilisation of the tooth an enormous threatening bleeding occurred which forced in addition to surgical revision, immediate embolisation and intensive care treatment? The patient recovered well from the treatment without any long-term complications or damages. The later histo-pathological findings supported the intraoperative diagnosis of an intra-osseous haemangioma.

Introduction

Intra-osseous cavernous haemangioma (angiomacavernosum) is a very rare benign vascular malformation of endothelial origin, which represents only 0.5-1 % of all intra-osseous tumours [1]. Localizations are first of all the vertebral column and the cranium. Only few descriptions of an involvement of the facial skeleton like the zygoma, the nose or the mandible are to be found in the literature [2].

The incidence of cavernous haemangiomas is in general twice so high for men than for women, but vice versa in the region of the jaws.

75% appear in the age between 10 and 60 years with culmination between the second and fifth life decades [3].

Possible differential diagnoses are e.g. periapicalodontogenic lesions (granuloma, cyst), giant cell granuloma, multiple myeloma, fibrous dysplasia, and osseous sarcomas [4,5].

Due to the potential fatal complications in combination with the often very uncertain clinical and radiological signs and appearance the diagnosis is a challenge [2,6,7]. Even a biopsy or simple tooth extraction can result in a lethal haemorrhage [5]. In the literature several different treatment options like radical or conservative surgery or embolization (in combination or without) of are described but there is still no final consensus [1].

Case Report

A 15-year old female patient without any other known diseases was referred due to a radiological lucency in the right mandible. The referral diagnosis based on the radiological description was an odontogen cyst related to the first molar (tooth 46). The tooth was mobile with resorption of the root. No extra- or intraoral swelling, fluctuation or pulsation was found, no signs of an acute inflammation. The patient herself had no major subjective complaints. All the teeth in the according area reacted vital to ice-probe, were indolent to percussion and showed no decays.

Beside of the right mandible no other skeletal or extra-skeletal findings were found.

Due to the size of the finding, potential malignancy criteria (root resorption) and the young patient age a biopsy with extraction of the tooth 46 was planned in general anaesthesia.

Intra-operatively the diagnosis had to be changed at once to a cavernous (arterio-venous) haemangioma: Already under carefully mobilisation of the tooth 46 a massive bleeding arose leading to a loss of up to 2 liters blood volume during the complete intervention.

The bleeding could be controlled and stopped intra-operatively by local electro-cauterisation, bone wax and tamponade, so that the patient could be stabilized and received transfusions.

An angiographic radiological intervention with embolisation of the arteria alveolar is inferior dexter with Onyx[®] (EV3/Covidien, Paris, France) was proceeded directly afterwards. The same day, when the patient was still in general anaesthesia, a surgical control with shifting of the intraoral



Figure 1: Preoperative panoramic x-ray with a huge defect in the right mandible and root resorption tooth 46.

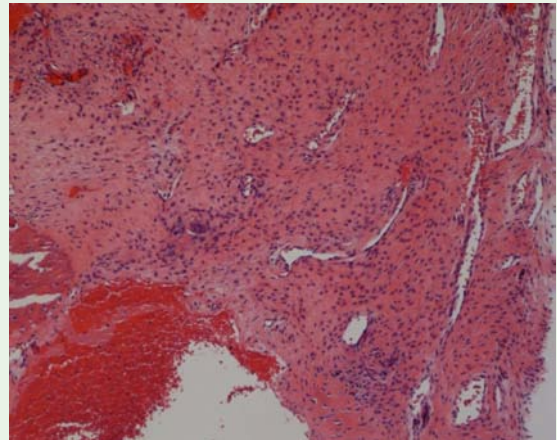


Figure 4: Histological picture no.1 from the biopsies (enlargement x100) shows vessel proliferation and some bleeding, no signs of malignancy.

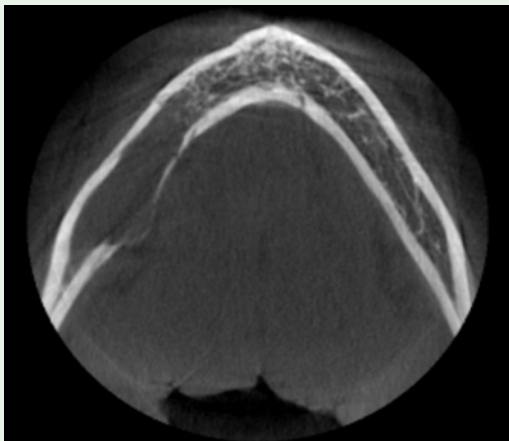


Figure 2: Findings in the MRI and CT described as potential keratocysticodontogenic tumour or ameloblastoma.

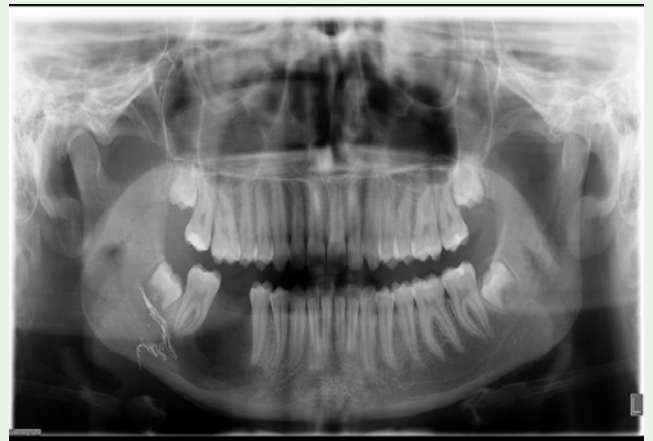


Figure 5: The panoramic x-ray three months postoperative shows already certain bone regeneration.



Figure 3: Preoperative MRI-finding.

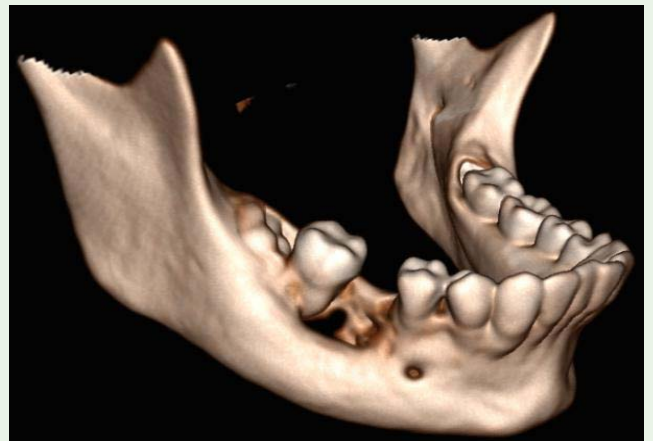


Figure 6: Control-CT 8 months after surgery and embolisation. Still visible bony defect but with regeneration and without signs of tumour recurrence.

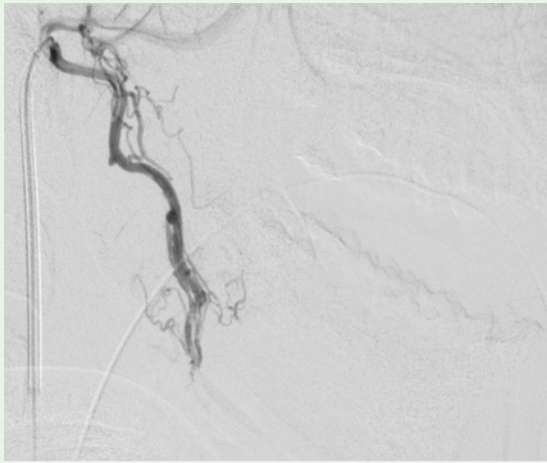


Figure 7: Angiography before embolization with Onyx®.

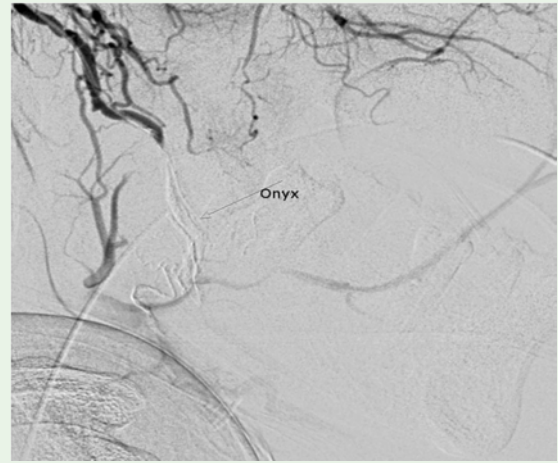


Figure 8: Angiography directly after embolisation.

tamponades was performed to avoid potential problems after awakening of the patient. At this point of time the situation was already stable without any further bleeding. The patient was followed up and treated at the intensive care unit with antibiotic prophylaxis during one week without occurrence of a new bleeding or any other complication.

At the time of dismissal from the hospital one week postoperatively the patient had beside of moderate pain no subjective complains and was in good shape. The clinical investigation showed a moderate swelling in the region of the right cheek without dysphasia, the active mouth opening was over 20 mm.

The later received histological report supported the diagnosis haemangioma together with the clinical and radiological findings.

At ambulatory control two weeks later stitches and some rest of the bone wax, which had come up to the surface, were removed. The wound showed no sign of infection and still no bleeding had occurred since the operation. No hypoesthesia or other signs of nerve damage were registered.

At the last control 8 months after surgery the wound had totally grown, the patient was free of complaints. The CT showed still a defect in the former region of the haemangioma but minor in size and with certain bone regeneration.

Discussion

Compared with the literature our case of a cavernous intra-osseous haemangioma is not typical for the majority of vascular malformations. For this special rare localisation the gender of the patient is described as usual but already regarding the age there is found different reports varying between the second decade of live and the fourth and fifth decade [3,8].

Beside from the neoplasm in the mandible our patient had no other disease pattern, which could be related to general affections of the body or syndromes. In contrast to capillary haemangiomas, which are usually congenital, the intra-osseous cavernous haemangiomas of the facial skeleton are not hereditary and occur in adulthood [8].

Even though CT and MRI are recommended for radiological investigation methods the radiological appearance of a bony lesion with tooth resorption might not lead to a certain diagnosis [1-3,5].

Clinical symptoms, which can be typical for a cavernous haemangioma, were absent in our case [1,4]. Due to the intra-osseous localisation without any visible tissue involvement, swelling, facial asymmetry or vascular pulsation it was not possible to predict a haemangioma in this case based on the intra- and extra-oral investigation.

Nevertheless an uncertain radiological lesion with tooth resorption and without existing infection signs may lead to the suspicion of an intra-osseous cavernous haemangioma or any other kind of tumour [4,5,6].

With respect to the addicted potentially severe complications like first of all a massive bleeding a cavernous haemangioma, even if being a very rare pathology, should be taken in consideration in all kind of similar uncertain findings [1,6,7]. Even if we could treat the occurring bleeding at once under optimal conditions (young patient without general medical risk factors under general anaesthesia, experienced surgical team, intensive care and angio-radiological department directly available) the course of our case shows the potentially life-threatening character of such malformations. Therefore we will not recommend a biopsy under local anaesthesia in an outdoor patient office in case of any distinct suspicion of a cavernous haemangioma or even if this diagnosis cannot be certainly ruled out.

The intra- and postoperative course shows that a local surgical revision combined with an embolization of the feeding vessels can be a sufficient treatment [1]. Therefore invasive and destructive measures such as radical resection of the according jaw or/and ligation of the carotid artery as still discussed in the literature should be preferably avoided not only considering aesthetic reasons [3,4,7,9].

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