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## Centralni hemangiom donje čeljusti: prikaz slučaja

### *Central Hemangioma of the Mandible: Case Report*

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#### Sažetak

Autori opisuju centralni hemangiom donje čeljusti dijagnosticiran radiološki i patohistološki, a kirurški se postupak komplicirao zbog naknadnog krvarenja koje je zaustavljeno dodatnim kirurškim zahvatom. Kako je centralni hemangiom čeljusti dosta rijedak - što potvrđuje i navedena literatura - a moguće komplikacije kod te patološke promjene mogu biti izrazito opasne, svrha prikaza je istaknuti kliničke, radiološke i patohistološke karakteristike te tvorbe. Usporedba s drugim slučajem - koji je radiološki izgledao kao policističan tumor čeljusti, ali je tijekom zahvata počelo obilno krvarenje te je odgođen nastavak zahvata i indicirala se dodatna angiografska i radiološka obrada - pokazala je da nema karakteristične kliničke i radiološke slike koja bi mogla upozoravati na takve promjene. Raznolikost radioloških slika u svjetskoj literaturi upozorava da svaka transparentija u čeljusti među patološkim promjenama može biti i centralni hamangiom. Prikazani slučajevi potvrđuju takvo mišljenje.

Zaprimljen: 2. veljače 2007

Prihvaćen: 15. ožujak 2007.

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#### Ključne riječi

hemangiom; bolesti krvnih žila; donja čeljust

#### Uvod

Hemangiomi su mane ili anomalije u razvoju i položaju krvnih žila nalik na tumore. Još je otvoreno pitanje postoji li prava neoplazma krvožilnog sustava, pa se i danas vode rasprave o tome je li riječ o pravim neoplazmama ili hamartomima (1). Mogu se pojaviti u mekim i koštanim tkivima oralne i maksiofacijalne regije. One u mekim tkivima lakše je prepoznati prema promijenjenoj boji i izgledu sluznice ili kože, a na one centralne u čeljustima važno je posumnjati na temelju radiološkog nalaza kako bi se izbjeglo nekontrolirano krvarenje tijekom vađenja zuba ili kirurškog uklanjanja tvorbe iz čeljusti. Zlatogorski sa suradnicima (2) raščlanio

#### Introduction

Hemangiomas are a defect in the development and position of the blood vessels which resemble tumours. Whether a true neoplasm of the blood vessel system exists remains an open question, and thus today discussion continues on whether they are in fact neoplasms or hamartomas (1). Those in the soft tissues are easier to recognise by the change in colour and appearance of the mucous membrane or skin, while the radiographic findings should be the basis of suspicion for those that are central in the jaws, in order to avoid the possibility of uncontrolled bleeding during tooth extraction or surgical removal of the formation from the jaw. Zlatogorski et al (2) analysed

je 84 slučaja centralnih hemangioma čeljusti iz britanske literature i tome dodao dva nova. Posebnu je pozornost posvetio radiološkoj slici. Tvorbe su se češće javljale u donjoj čeljusti u odnosu prema gornjoj (3,3:1), u 66% slučajeva bile su multilokularne, u 3% unilokularne i u 1% nisu stvarale koštanu šupljinu. Rubovi prosvjetljenja bili su u 32% slučaja jasno ograničeni, a u 68% difuzni. Kod slučajeva u donjoj čeljusti alveolarni kanal bio je zahvaćen u 15% slučajeva. Recentna literatura uglavnom se bavi prikazima novih slučajeva s odgovarajućim specifičnostima i pregledom dosadašnje literature o toj problematici, posebice kad je riječ o dijagnostičkim i kirurškim postupcima. Tako se mogu naći i takozvani „klasični slučajevi“ centralnih hemangioma čeljusti (3), bez obzira na to što to radiološki značilo. Opisan je i hemangiom koji je na rendgenskoj snimci izgledao kao periapikalna promjena (4), zatim slučaj intrakortikalne lokalizacije hemangioma u donjoj čeljusti (5), primjer monocističnog hemangioma poput benigne unilokularne ciste (6) također u donjoj čeljusti, a ostali primjeri opisuju kirurški postupak - pretežito je to bila resekcija dijela zahvaćene čeljusti (7-9) često s imedijatnom rekonstrukcijom, a u slučajevima većih primjera s obavljenom angiografijom i eventualnom embolizacijom krvnih žila. Krmpotić (10) je još 1983. istaknuo koliko je važan dijagnostički postupak da bi se diferencirale sporoprotodne (kavernomi ili kapilarno venske malformacije) od brzoprotodnih promjena (arteriovenske malformacije) te važnost embolizacije prije zahvata u svim upitnim slučajevima. Najvažnije je na temelju kliničke slike i radiološkog nalaza posumnjati i na mogućnost centralnog hemangioma te prije bilo kakvog zahvata zahtijevati dijagnostičke postupke kao što su selektivna angiografija i kompjutorizirana tomografska (CT) obrada (11).

### Prikaz slučajeva

Od mnogobrojnih hemangioma koji su zahvaćali samo meka ili tvrda tkiva maksilofacijalnog područja, ističemo dva slučaja centralnih prosvjetljenja u donjoj čeljusti specifična prema rendgenskoj slici te kliničkom intraoperacijskom i histološkom nalazu.

U prvom slučaju, unatoč kliničkoj slici koja je zbog obilnog krvarenja upućivala na hemangiom donje čeljusti, dijagnoza nije potvrđena, a u drugome se sumnjalo na hemangiom na temelju radiološkog i CT nalaza, a zatim je potvrđena patohistološki.

84 cases of central hemangioma of the jaw from the English literature and added two new cases. Particular attention was paid to the radiographic findings. Formations occurred more frequently in the mandible compared to the maxilla (3.3:1). In 66% of cases they were multilocular, in 3% unilocular and in 1% they did not create a bone cavity. In 32% of cases the edges of the translucency were clearly circumscribed and in 68% diffuse. In the cases occurring in the mandible there was 15% alveolar canal involvement. Recent literature is mainly concerned with the presentation of new cases with corresponding specificity and a review of the literature to date on this problem, particularly with regard to diagnostic and surgical procedures. Thus it is possible to find so-called "classic cases" of central hemangiomas of the jaw (3), whatever this means radiographically. There was a case of hemangioma described as resembling a periapical alteration on the X-ray (4); a case of intracortex localisation of a hemangioma in the mandible (5); an example of a monocystic hemangioma resembling a benign unilocular cyst (6), also in the mandible. Other examples describe the surgical procedure which mainly involves resection of part of the affected jaw (7-9), often with immediate reconstruction, and in cases of larger specimen with previous angiography and eventual embolisation of the blood vessels. In 1983 Krmpotić (10) stressed the importance of diagnostic procedures, with the aim of differentiation between slow flow rate (capillary-venous malformations) and rapid flow rate alterations (arterio-venous malformations), and also the importance of embolisation prior to the operation in all suspect cases. It is most important that the possibility of a central hemangioma should be suspected on the basis of the clinical appearance and radiographic findings, and to carry out diagnostic procedures, such as selective angiography and CT before any kind of operation is undertaken (11).

### Case presentation

Of the numerous cases of hemangiomas which only involve the soft or soft and hard tissue of the maxillofacial region we present two cases of central translucency in the mandible, which are specific according to the X-ray, clinical intra-operative and histological findings.

In the first case, in spite of the clinical appearance, which was indicative of the existence of a mandibular hemangioma due to profuse bleeding, the diagnosis was not confirmed, and in the second case the assumption of hemangioma was based upon radiographic and CT findings and confirmed histopathologically.

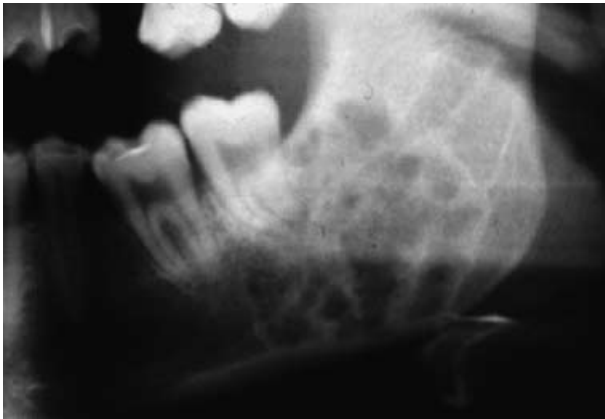
Prvi je slučaj 40-godišnje pacijentice koja je primljena na odjel Klinike za kirurgiju čeljusti i lica zbog slučajnog nalaza koštanog policističnog prosvjetljenja u angulusu donje čeljusti (Slika 1.). Kost nije bila napuhana i pacijentica je primljena s dijagnozom centralnog tumora donje čeljusti, a prema lokalizaciji i policističnosti bila je moguća dijagnoza ameloblastoma. Razmjerno nježne koštane pregrade stvarale su sliku „poput pjene od sapunice“ ili saća, pa se morala uzeti u obzir i mogućnost odontogenog miksoma ili fibroma te neka od gigantocelularnih promjena čeljusti, kao što su centralni reaktivni granulom traumatske ili endokrine etiologije ili aneurizmatička koštana cista čeljusti. Prosvjetljenje je bilo multicistično oštro ograničeno, s lobuliranim rubovima. Tijekom zahvata, nakon što je otvoren kortikalni bukalni stijenke čeljusti, počelo je jako krvarenje i bilo ga je moguće zaustaviti samo čvrstom tamponadom koštane šupljine. Od zahvata se odustalo, rana je sašivena i pacijentica upućena na angiografiju. Angiografijom nije bila potvrđena dijagnoza angioma u donjoj čeljusti, pa je ponovljen zahvat u općoj anesteziji. Tada je pronađena koštana šupljina s nešto mekoga granulacijskoga tkiva i rubno razmekšana kost. Uklonjen je cijeli bukalni kortikalni, a šupljina je izgladana frezom za kost. Mandibularni kanal i manja mjesta krvarenja iz kosti zatvorena su kirurškim voskom. Postavljena je intraoralna sukcija i rana sašivena u jednom sloju. Poslijeoperacijski tijek protekao je bez komplikacija te je rana zacijelila primarno. Patohistološki nije potvrđena dijagnoza hemangioma. Nalaz je pokazao pretežito granulacijsko tkivo s multinuklearnim orijaškim stanicama, nakupinama makrofaga i ponegdje pukotinama kolesterinskih kristala. U drugim dijelovima uzorka nađene su oštećene koštane gredice sa žarištima mononuklearne upalne infiltracije. Dakle, dijagnoza hemangioma, klinički pretpostavljena na temelju obilnog krvarenja tijekom prvog zahvata, nije potvrđena ni angiografijom ni patohistološkom pretragom tkiva.

Drugi pacijent, u dobi od 60 godina, pregledan je u Kliničkom zavodu za oralnu kirurgiju zbog prosvjetljenja u predjelu donje čeljusti. Ono je pronađeno u predjelu donje čeljusti u projekciji vršaka i ispod korjenova donjeg desnog očnjaka i prvog pretkutnjaka i širilo se distalno u područje izvađenih zuba - drugoga pretkutnjaka i prvoga kutnjaka.

Prosvjetljenje je bilo razmjerno oštro ograničeno, lobuliranih rubova, podijeljeno još tanjim pregradama nego u prvom slučaju na više nepravilnih površina i također je stvaralo sliku koja se u radio-

The first case presented as a 40-year-old female patient who was admitted to the Department of Maxillofacial Surgery because of an osseous polycystic translucency in the mandibular angle (Fig. 1). The bone was not distended and the patient was admitted to the Department under the diagnosis of a central tumour of the mandible. According to the localisation and polycysticity, a diagnosis of ameloblastoma was possible. Relatively delicate bone structure created the appearance of “soap foam”, or honeycomb and therefore the possibility of an odontogenic myxoma or fibroma was considered, and also the possibility that it was one of the giant cell changes in the jaw, such as a central reactive granuloma of traumatic or endocrine aetiology or an aneurysmal bone cyst of the jaw. The translucency was multicystically sharply circumscribed, with lobular edges. During the operation, after opening the cortex of the buccal wall of the jaws, profuse bleeding started which was stopped with a firm tamponade of the bone cavity. The operation was postponed, the wound sutured and the patient sent for angiography. The angiography did not confirm the diagnosis of mandibular angioma and the operation was repeated under general anaesthesia. During the second operation a bone cavity was found with relatively soft granulation tissue and marginally softened bone. The buccal cortex layer was entirely removed and the cavity smoothed with a bone bur. The mandibular canal and smaller area of the bleeding from the bone were closed with surgical wax. Intraoral suction was inserted and the wound sutured in one layer. The postoperative course passed without complications and the wound healed primarily. The histopathological findings did not confirm the diagnosis of a hemangioma. The findings showed mainly granulation tissue with multinuclear giant cell accumulations of macrophages and occasional cracks in cholesterol crystals. In other parts of the sample damaged bone trabeculae were found with foci of mononuclear inflammatory infiltration. The diagnosis of hemangioma, which was clinically assumed on the basis of the profuse bleeding during the first operation, was therefore not confirmed either by angiography nor by histopathological examination.

The second case involved a 60-year-old male patient, examined in the Clinical Department of Oral Surgery because of a translucency in the mandibular region. The translucency was found in the body of the mandible in the projection of the apex and beneath the roots of the lower right canine and the first premolar and extended distally into the area of the extracted teeth, which were the second premolar and central incisor.



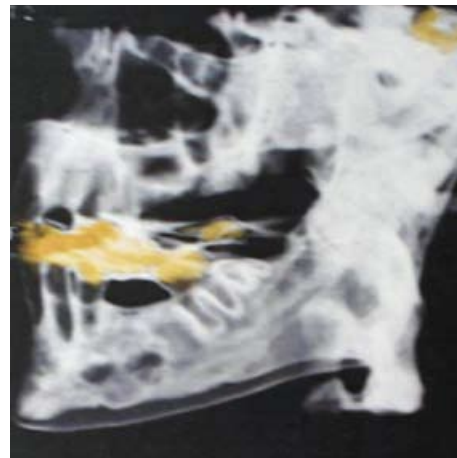
**Slika 1.** Multicistično prosvjetljenje u angulusu donje čeljusti pacijentice  
**Figure 1** Multicystic translucency in the mandibular angle of the presented patient.



**Slika 2.** Prosvjetljenje u donjoj čeljusti drugog opisanog pacijenta  
**Figure 2** Translucency in the mandible of the second presented patient.



**Slika 3.** Trodimenzionalna CT rekonstrukcija pokazuje perforacije čeljusti u predjelu mentalnog otvora  
**Figure 3** Multi-layered CT reconstruction showing perforation of the jaw in the region of the mental foramen.



**Slika 4.** Sagitalni presjek pokazuje opseg promjene i mjesta perforacije čeljusti  
**Figure 4** Saggital cross section showing the extent of the alteration and site of the jaw perforation.

logiji spominje kao prosvjetljenje poput „pjene od sapunice“ (Slika 2.).

Na CT nalazu pronađena je patološka promjena u čeljusti promjera 1,2 x 1,5 cm. Kost je resorbirana i šupljina ispunjena nepravilnim trabekulama. Radiolog je pretpostavio da je riječ o hemangiomu, iako nisu bile isključene ni druge patološke promjene, uključujući sekundarizam. (Slike 3. i 4.).

Zdravstveno stanje pacijenta tražilo je odgovarajuću predoperacijsku pripremu zbog antikoagulantne terapije koju je uzimao zbog preboljenog bakterijskog endokarditisa i mitralne pogreške. Terapija Marivarinom zamijenjena je terapijom Clexaneom 60 mg na dan i protrombinsko vrijeme usklađeno je na vrijednosti od prosječno 80%, APTV (P) na vrijednosti između 25 i 37 sekundi, a trombinsko vrije-

The translucency was relatively sharply circumscribed, with lobular edges, divided by even thinner walls than in the first case and on more asymmetrical surfaces, and also created the appearance known in radiology as translucency resembling “soap foam” (Fig. 2). The CT findings showed pathological alteration in the jaws, 1.2 x 1.5 cm in diameter. The bone was resorbed and the cavity filled with asymmetrical trabecules. The radiologist assumed that it was a case of a hemangioma, although other pathological changes were not ruled out, including secundarism (Figs. 3 and 4).

The patient’s state of health required appropriate preoperative preparation since he was on anti-coagulation therapy, which he was taking for bacterial endocarditis and mitral insufficiency. His



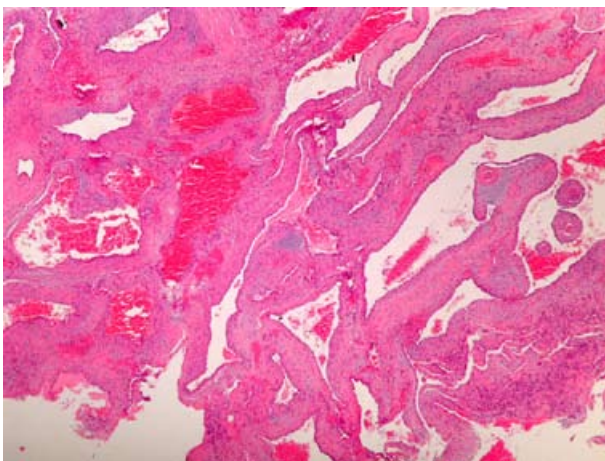
me na vrijednosti između 14 i 21 sekunde. Sat prije zahvata počela je terapija Klimicinom 3 x 300 mg i trajala je dva dana.

Zahvat je obavljen u općoj inhalacijskoj anesteziji i protekao je bez komplikacija. U kosti je nađena nepravilna šupljina s nešto razmekšane spongioze i mekoga tkiva koje je poslano na patohistološku pretragu. Prekutnjak koji je korištenom stršao u defekt uklonjen je, prazna alveola ispunjena mrežicom Surgicel, a neznatna krvarenja iz kosti spontano su prestala tijekom zahvata. Rana je primarno sašivena svi- lenim koncem. Drugi dan nakon zahvata počelo je obilno krvarenje pa je rana revidirana, a krvarenje iz kosti zatvoreno kirurškim voskom. Razvio se je velik podljev lica, brade i vrata sve do prsišta, ali se postupno smanjivao tijekom oporavka i prelaska na an-



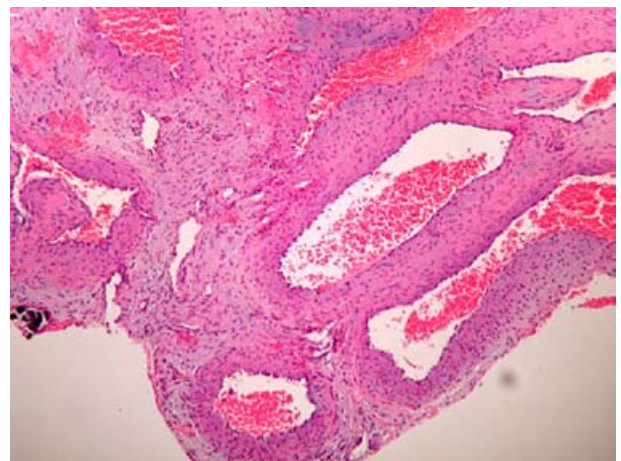
prescribed Marivarin therapy was substituted with 60 mg of Clexane to be taken daily, and the prothrombin time was to be adjusted to a value of approximately 80%, APTV (P) to a value between 25 and 37 s, and thrombin time to a value between 14 and 21 s. Therapy commenced with Klimicin 3 x 300 mg given one hour prior to the operation, and which the patient took for two days. The operation was performed under general inhalation anaesthesia and passed without complications. An asymmetrical cavity was found in the bone with slightly softened spongy and soft tissue, which was sent for histopathological analysis. The premolar, whose root penetrated into the defect was removed, the empty alveole filled with Surgicel haemostat net and slight bleeding from the bone stopped spontaneously during the operation. The wound was primarily sutured with silk sutures. Due to profuse bleeding on the second day after the operation the wound was re-examined, and bleeding from the bone stopped with surgical wax. A large haematoma developed on the face, chin and neck, extending as far as the chest, which gradually decreased during hospitalisation and anticoagulant therapy (Fig. 5). The histo-

**Slika 5.** Poslijeoperacijski podljev lica, brade i vrata petog dana nakon zahvata postupno mijenja boju  
**Figure 5** Postoperative haematoma of the face and neck on the fifth day after the operation, which gradually changed colour.



**Slika 6.** Tumorsko tkivo građeno od celularnog vezivnog tkiva i krvnih prostora različito debelih stijenki, obloženih pravilnim endotelom i ispunjenih eritrocitima. (HE 40 x.)

**Figure 6** Tumorous tissue composed of cellular connective tissue and blood spaces with walls varying in thickness, covered with symmetrical endothelium and filled with erythrocytes (HE 40x).



**Slika 7.** Detalj istog uzorka. (HE 100 x.)  
**Figure 7** Detail from the same sample (HE 100x).

tikoagulantnu terapiju (Slika 5.). Patohistološki nalaz opisuje materijal građen od širokih krvnih prostora umjereno debelih zidova obloženih pravilnim endotelom i ispunjenih eritrocitima, a vezivno tkivo između tih prostora dosta je celularno, mjestimice sluzavo promijenjeno i prožeto sitnim kapilarama. Dijagnoza - Haemangioma cavernosum (Slika 6. i 7.). Daljnji tijek bio je bez komplikacija - pacijent je došao na kontrolni pregled nakon desetak dana i bio je naručen na rendgensku kontrolu za tri mjeseca.

## Rasprava

Dva opisana slučaja pokazuju kako dijagnoza centralnog hemangioma čeljusti nije jednostavna već zbog činjenice što je riječ o ne tako čestoj promjeni. Multicistična prosvjetljenja čeljusti trebala bi redovito potaknuti sumnju na centralni hemangiom, iako ne postoji radiološka promjena koju bismo mogli nazvati karakterističnom za tu tvorbu. Piercell i suradnici (12) smatraju da svaka osteolitička promjena čeljusti može biti centralni hemangiom. Lokalizacija je moguća u objema čeljustima i u kostima lica. Perugini i suradnici (9), od deset opisanih hemangioma, šest su našli u zigomatičnoj kosti, dva u donjoj čeljusti, jedan u gornjoj, a jedan u medijalnom zidu orbite. Autori ističu važnost kompjutorizirane tomografije u dijagnostici. Mansour (13) je godine 1978. dao potpun pregled literature od 1849. do tada i istaknuo kako se mora sumnjati na hemangiom kod svake osteolitičke promjene čeljusti. U prikazu jednoga svojeg slučaja opisao je radikalnu resekciju čeljusti kod osmogodišnje djevojčice te rekonstrukciju s autogenom kosti tibije. Sličan je i prikaz Hoa i Yipa<sup>3</sup> s opsežnim pregledom literature i raspravom o problemima dijagnostike i različitih mogućnosti liječenja hemangioma, iako je vrlo teško složiti se s njihovom procjenom takozvanog klasičnog primjera centralnog hemangioma.

U oba prikazana primjera hemangiom se mogao pretpostaviti na temelju radioloških nalaza. To što u jednom slučaju nije bila riječ o hemangiomu, a intraoperacijski je nastalo obilno krvarenje koje je odgodilo konačan zahvat do nalaza angiografije i CT obrade, a u drugom je postojao hemangiom - što je pretpostavio i nalaz radiologa, ali operacijski nije bilo znatnijeg krvarenja, nego je ono nastalo u poslijeoperacijskome razdoblju - samo upućuje na mnoge mogućnosti razvoja komplikacija kod takvih promjena. Sve se konačno svodi na to da se ispravnom dijagnozom treba omogućiti pravi izbor kirurškoga liječenja ili neka druga metoda, ovisno o naravi promjene (14,15).

pathological findings showed material comprised of wide bloody areas with moderately thick walls covered with symmetrical endothelium and filled with erythrocytes, and between those spaces the connective tissue was relatively cellular, in places mucinously altered and permeated with tiny capillaries. Diagnosis: Hemangioma cavernosum (Fig. 6 & 7). The following course passed without complications and the patient was called back for a follow-up after ten days and an X-ray after three months.

## Discussion

The two cases presented show that the diagnosis of central hemangioma of the jaws is not simple, firstly because of the fact that it is an alteration which is relatively infrequent. Multicystic translucency of the jaw should always arouse suspicion of the possibility of a central hemangioma, in spite of the fact that no radiographic changes can be deemed characteristic unto this formation. Piercell et al (12) consider every osteolytic change in the jaw a possible central hemangioma. Localisation is possible in both jaws and in the facial bones. Perugini et al (9) found six out of ten reported hemangiomas in the zygomatic bone, two in the mandible, one in the maxilla and one in the medial orbital wall. The authors stress the importance of CT in diagnostics. In 1978 Mansour (13) gave a comprehensive review of the literature from as far back as 1849 and emphasised the need to suspect hemangioma when confronted with any osteolytic changes in the jaws. In a presentation of his own case he described the radical resection of an eight-year-old girl's jaw and its subsequent reconstruction with autogenous bone from the tibia. Ho & Yip (3) gave a similar presentation with a comprehensive review of the literature and discussion of the problems in diagnosis and various possibilities for treatment of hemangioma, although it is difficult to agree with their evaluation of a so-called classic example of central hemangioma.

In both of the presented cases the possibility of a hemangioma could have been assumed on the basis of the radiographic findings. The fact that hemangioma was not confirmed in one case and profuse intra-operative bleeding occurred which postponed the final operation until the angiography and CT, and in the other case hemangioma was confirmed, as indicated by the radiologist's findings, although no significant bleeding occurred during the operation, only during the postoperative period, demonstrates the various possibilities for the development of complications in the case of such alterations.

Na temelju osobnih iskustava s opasnim krvarenjima iz centralnih hemangioma čeljusti, koja je prvi autor imao prigodu vidjeti prije više od 30 godina na našoj Klinici i jednoj od europskih klinika za maksilofacijalnu kirurgiju, te na osnovi podataka iz opisanih slučajeva činilo se vrijednim ponovno upozoriti koliko je važna dijagnostika centralnih hemangioma čeljusti kako bi se izbjegle moguće velike i opasne komplikacije.

Consequently, correct diagnosis ensures the correct choice of surgical treatment or choice of some other method of treatment, depending on the nature of the changes within the tissue (14,15).

Based upon personal experience with dramatic bleeding from central hemangiomas of the jaw, which the first author had the opportunity of seeing more than 30 years ago in our Clinic and in a one of the European clinics of maxillofacial surgery, and on the basis of data from the presented cases, the importance of diagnosing central hemangiomas of the jaw should again be emphasised in order to avoid the possible onset of dramatic complications.

### Abstract

The authors present a case of central hemangioma of the mandible which was diagnosed radiographically and histopathologically. The surgical procedure was complicated by subsequent bleeding which was stopped by an additional surgical procedure. As central hemangioma of the jaws is a relatively rare findings, which can be observed from the relevant literature, and possible complications can be extremely dramatic, the aim of the presentation is to emphasise the clinical, radiographic and histopathological characteristics of this formation. Comparison with another case, which radiographically resembled a polycystic tumour of the jaw and which bled profusely during the operation, causing the postponement of the operation and indicating the need for additional angiographic and radiotherapy, demonstrated that there are no characteristic clinical and radiographic appearances which can indicate the possibility of such changes. The diversity of the radiographic appearances presented in the world literature shows that apart from other pathological changes any transparency in the jaws may also be a central hemangioma. The presented cases confirm this judgement.

Received: February 2, 2007

Accepted: March 15, 2007

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### Key words

Hemangioma; Vascular Diseases;  
Mandible

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