C A S E R E P O R T S

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Intraosseous focal venous malformation of the mandibular body: cone beam computed tomography planning followed by piezoelectric knife resection and free bone graft reconstruction

Intraosealna fokalna venska malformacija tela donje vilice: planiranje piezoelektrične resekcije pomoću kompjuterizovane tomografije konusnim zrakom i rekonstrukcija slobodnim koštanim transplantatom

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Abstract

Introduction. Intraosseous vascular malformation could be life-threatening due to uncontrolled hemorrhage after tooth extraction. According to biological behavior of this lesion, adequate diagnostic and treatment strategies are necessary in order to avoid possible complications. We reported cone beam computed tomography (CBCT) planning of an urgent *en bloc* resection of an intraosseous venous malformation by piezoelectric knife. **Case report.** A 55-year-old man was submitted to CBCT planning followed by piezoelectric knife resection of an intraosseous focal venous malformation of the mandibular body. Immediate reconstruction of the defect using iliac bone free graft was performed. The surgical treatment was uneventful and a 2-year follow-up revealed no signs of recurrence. **Conclusion.** Piezoelectric knife could provide precise, safe and bloodless procedure which is especially important in this pathology. Advantages of this technique are: lower risk of damaging soft tissue structures, precise osteotomy and bloodless surgery. Moreover, using piezosurgery bone knife, blood transfusion and blood transmitted diseases could be avoided. This case highlights the importance of CBCT as planning tool for resection of the mandible, using piezoelectric knife as safe method to achieve bloodless surgery.

Key words:
mandibular neoplasms; hemangioma; diagnosis; cone-beam computed tomography; oral surgical procedures; bone transplantation.

Apstrakt

Uvod. Intraosealna vaskularna malformacija vilica može ugroziti život bolesnika zbog nekontrolisanog krvarenja do kojeg može doći nakon ekstrakcije zuba. Shodno biološkom ponašanju tih lezija, primena odgovarajućih dijagnostičkih i terapijskih metoda neophodna je kako bi se izbegle moguće komplikacije. Prikazana je *en bloc* resekcija intraosealne venske malformacije piezoelektričnim nožem nakon prethodno izvršene kompjuterizovane tomografije konusnim zrakom (CBCT). **Prikaz bolesnika.** Kod bolesnika starosti 55 godina resekcija intraosealne fokalne vaskularne malformacije donje vilice piezoelektričnim nožem urađena je nakon prethodne CBCT, a defekt je rekonstruisan slobodnim koštanim transplantatom sa ilijačne kosti. Hirurško lečenje i postoperativni tok protekli su bez komplikacija. U periodu praćenja od dve godine nije došlo do razvoja recidiva. **Zaključak.** Piezoelektričnim nožem može se postići precizna i bezbedna resekcija uz minimalan gubitak krvi, što je od posebnog značaja kada je u pitanju venska malformacija koštanog tkiva. Prednosti ove tehnike su: mali rizik od oštećenja mekotkivnih struktura, mogućnost precizne osteotomije i minimalan gubitak krvi. Korišćenjem piezoelektričnog noža može se izbeći transfuzija krvi kao i transmisija krvnoprenosivih bolesti. U ovom prikazu slučaja posebno je istaknut značaj planiranja resekcije donje vilice piezoelektričnim nožem pomoću CBCT. To je izuzetno bezbedan metod lečenja, kojim se gubitak krvi svodi na minimum.

Ključne reči:
mandibula, neoplazme; hemangiom; dijagnoza; kompjuterizovana tomografija konusnim zrakom; hirurgija, oralna, procedure; transplantacija kosti; lečenje, ishod.

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Introduction

Head and neck vascular anomalies are classified according to the International Society for the Study of Vascular Anomalies (ISSVA) into two groups: vascular tumors and vascular malformations [1]. In 1982 Mulliken and Glowacki [2] proposed the classification of vascular anomalies in order to standardize the nomenclature. Vascular malformations are present at birth and enlarge proportionately with the growth of the child and do not undergo spontaneous involution [2, 3]. Hemangiomas, considered as vascular tumors, have two main types: infantile hemangiomas (IH) and congenital hemangiomas. There are two types of congenital hemangiomas: rapid involuting congenital hemangioma (RICH) and non-involuting congenital hemangioma (NICH). Hemangiomas generally are characterised by spontaneous involution, except non-involuting congenital hemangioma (NICH). Applying the classification of the ISSVA and the classification of vascular tumors of bone of the World Health Organization (WHO), intraosseous cavernous hemangioma (ICH) corresponds to intraosseous focal venous malformation (IFVM) [4–6]. This revised and updated nomenclature is used in this article.

Intraosseous vascular lesions of the mandible are usually asymptomatic, but they can be present as uncomfortable slow growing lesions with hard consistence, spontaneous hemorrhage, pulsatile sensation or tooth mobility [7, 8]. Clinical signs may include swelling of the soft tissues, pain of varying intensity, change in the color of the oral mucosa, toothache, unexplained bleeding of gums and enlargement of the cortical plate that makes the jaw asymmetrical [7, 9]. Among distortion, destruction, hypertrophy, hypoplasia and density change, primary intraosseous vascular lesions were also reported as possible alterations of the skeleton [10]. Radiographically they range from an unilocular rounded lesion, "honeycombed", "sunburst" or radiopaque appearance [8]. Differential diagnoses include odontogenic tumors, ameloblastomas, cystic lesions and fibrous dysplasia [7, 9, 11, 12].

Cone beam computed tomography (CBCT) has been commercially available as diagnostic tool since 2001. It has been used firstly in oral surgery and maxillofacial surgery giving the possibility of adequate visualisation of bony structure and pathology [13–16].

Piezosurgery create bloodless surgical site that makes visibility in the working area much clearer than with conventional bone cutting instruments. Unlike conventional burs and micro saws, piezosurgery inserts do not become hot, which again reduces the risk of postoperative necrosis. Nowadays, piezosurgery is one of the most advanced technologies in dental surgery, but also could become very important in head and neck surgery.

By minimising blood loss during surgery, piezosurgery could be very helpful tool in order to avoid blood transfusion.

We presented an urgent *en bloc* resection of an intraosseous venous malformation by a piezoelectric knife based on CBCT planning.

Case report

A 55-year-old asymptomatic male was referred to the Maxillofacial Surgery Department in Belgrade by his dentist to investigate a radiolucent lesion of the left mandibular body that he accidentally noticed using orthopantomography (OPG) prior tooth extraction (Figure 1). The patient complained of tooth loosening and lower denture instability due to a swelling underneath. There was no reported bleeding and the patient had no other systemic signs and symptoms and no comorbidities.

OPG showed an ill-defined multilocular lesion extending from parasymphiseal region to the angle of mandible (Figure 1).

As additional procedure and in order to minimise patient's radiation, CBCT (Planmeca unit promax 3DS) of the mandible was performed. It showed ill-defined multilocular osteolytic expansive lesion which was associated with mandibular canal. Both of the lingual and buccal mandibular cortical bones revealed signs of invasion (Figure 2).

After administrating of block and terminal (plexus) local anaesthesia (1 : 80,000 lidocaine with adrenaline) and raising of mucoperiosteal flap, non-pulsatile, dark blue soft tissue masses of mandibular body was presented. Biopsy was carried out using No 11 surgical blade. It was followed by excessive intraoperative bleeding. Hemostasis was temporarily achieved with iodoform gauze packing, hemostatic mucogingival sutures and external compressive bandage. Frozen section showed IFVM.

Fig. 1 – Orthopantomographic view showing discrete multilocular radiolucency in the left mandibular body.

Fig. 2 – Core beam computed tomography (CBCT) of the mandible (Planmeca Unit Promax Cone Beam CT 3DS).

After obtaining the histological report, immediate treatment plan was made, using CBCT's prediction (On Demand Software Cybermed Seoul Korea). The patient was prepared and under hypotensive general anesthesia *en bloc* surgical resection of the mandibular body through submandibular approach was performed (Figure 3).

Piezosurgery was used to make precise surgical resection and to prevent excessive surgical bleeding. After achieving radical excision of the IFVM, left iliac bone graft was harvested and precisely remodelled by piezosurgery (Figures 4, 5). Figure 6 shows finding ofter free bone graft reconstruction of the mandible. Postoperative course was uneventful. Final histopathological report confirmed IFVM (Figure 7).

The 2-year follow-up revealed no signs of relapse. The patient was in the process of pre-implant planning.

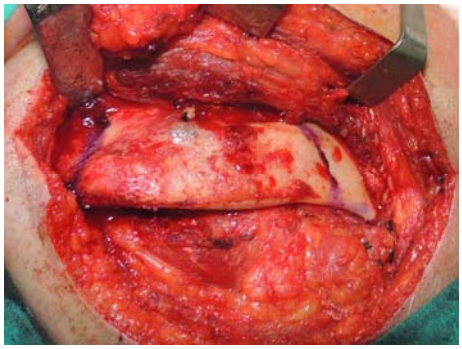

Fig. 3 – Submandibular approach to the mandibular body.

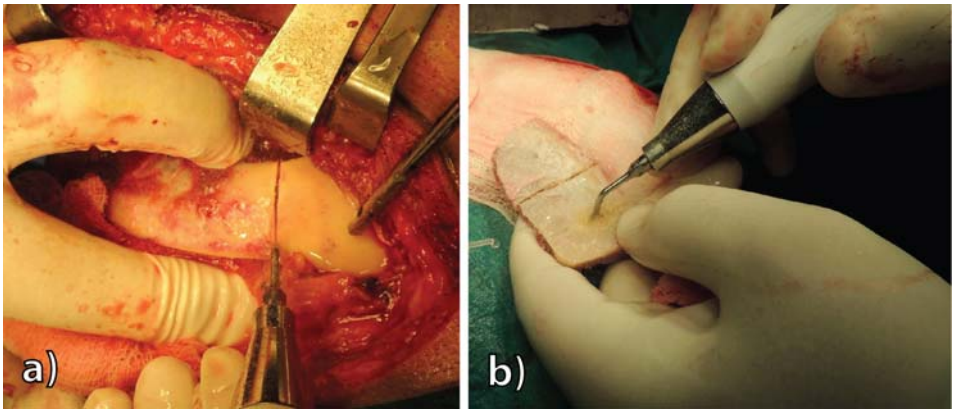

Fig. 4 – a) Use of piezoelectric knife in resection of intraosseous venous malformation (IVFM); b) Conturing iliac bone graft by use of piezoelectric knife.

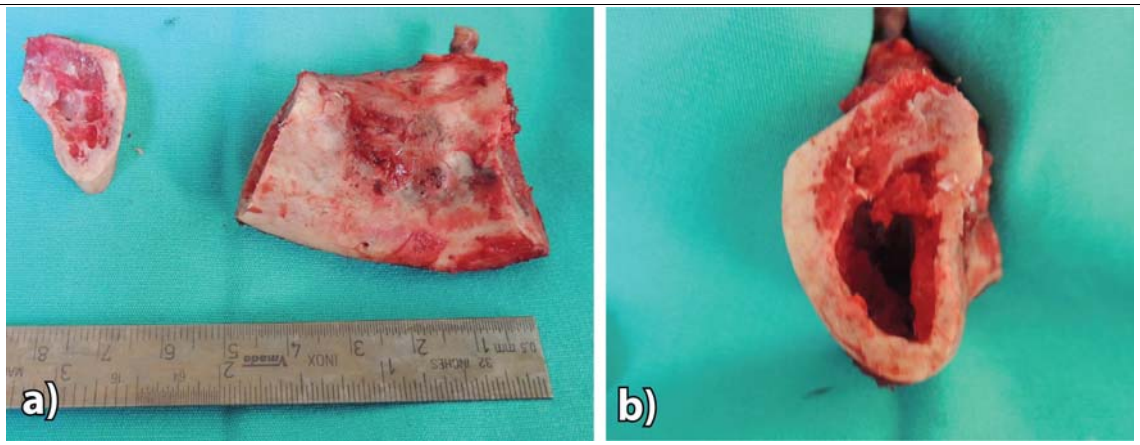

Fig. 5 – a) Surgical specimen; b) Cross section.

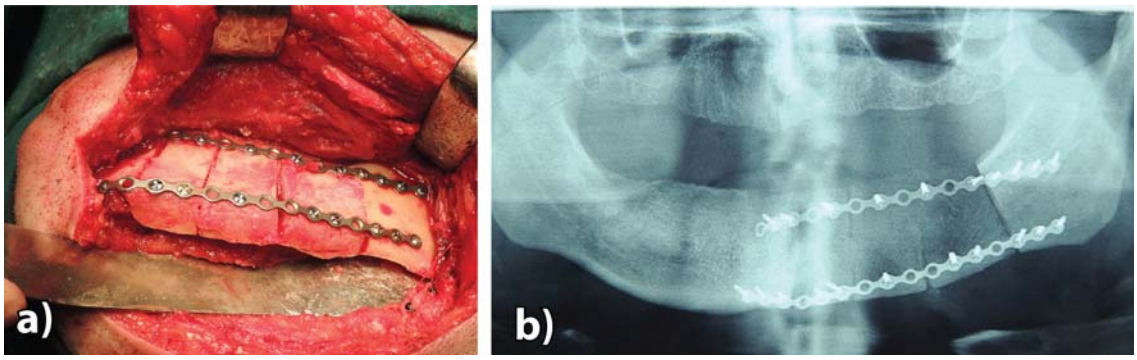

Fig. 6 – a) Iliac bone graft "in situ"; b) Postoperative orthopantomogram (OPG) X-ray.

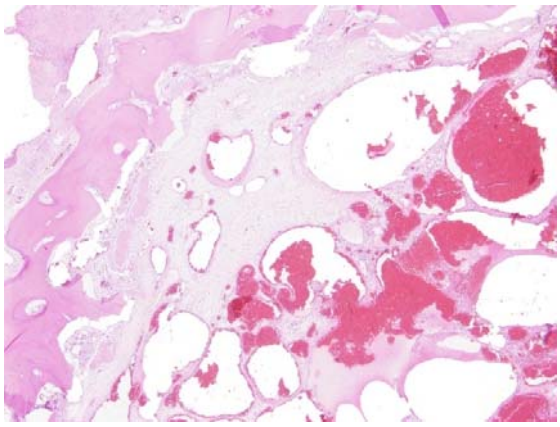

Fig. 7 – Intraosseous focal venous malformation (IFVM), which is composed of anastomosing, ectasic, engorged, thin-walled blood vessels (histologically-capillary hemangioma) (hematoxylin eosin, × 100).

Discussion

Historically "intrabony hemangioma" was presented as peripheral or central intraosseous lesion. It is classified as benign vascular tumor [17, 18] and accounts less than 1% of intraosseous "tumors" [3]. The highest incidence of occurring is in 20–50 years of age and it is almost twice more frequent in female population [8]. These lesions are commonly located in spine and skull bones, rarely in facial bones (zygomatic, orbital, mandible) [7, 12, 18–23].

The term "hemangioma" used in the literature to describe intraosseous vascular anomalies is a source of confusion [24]. Kaban et al. [11] reported that hemangiomas are not localized in the bone and postulated that "intrabony" vascular lesions are mostly venous malformations. Applying the classification of the ISSVA for an effective communication with medical doctors who are dealing with intraosseous vascular malformations, the term of IFVM was used in this article [25].

Usually detailed medical history and examination of a patient are sufficient to establish the clinical diagnosis of vascular anomalies of soft tissue [26]. However, IFVM of the jaws is rare lesion hidden in bone that could be difficult to diagnose. Because of the presence of multilocular radiolucenc, it can be often misdiagnosed as dentigerous cyst, ameloblastoma, central giant cell granuloma, myxoma and metastatic tumor, as well. The following signs and symptoms can be associated with the growth of IFVM: discomfort, pulsatile sensation, numbness, loosening the teeth, bluish discoloration of the

overlying mucosa [7, 8]. The most frequent localization of the IFVM in the mandible is in the premolar-molar region [27].

In this case the patient presented swelling in the mandibular area with no history of bleeding or altered neurological function. Radiologically there was ill-defined radiolucency in the premolar and molar region affecting both of the lingual and buccal mandibular cortical bones. Vascular malformation has to be taken in consideration in the differential diagnosis of any multi- or unilocular radiolucency of the jaws, particularly if there is the presence of "spoke like" and "sun ray" [28]. Clinically and radiologically IFVM could present as many other pathological entities in lower jaw.

Until now there is no single reliable non-invasive imaging technique that is adequate enough to diagnose venous malformation of the bone tissue [29].

Computed tomography (CT) and magnetic resonance (MR) angiography could be used in the diagnosis of vascular malformations (flow and feeding vessels characteristics) [12, 18], but it requires time for patient preparation and usually is not available technique for urgent situations. CBCT technology is clinically introduced in 1998 [29–31], and due to a high resolution imaging possibilities without using contrast became alternative to multislice CT and event better tool than MR [14, 18]. New generation of CBCT presented not only usability in establishing the diagnosis of IFVM but also enable clear visualization of the cortical involvement and relation with surrounding structures [18]. In this case CBCT has been used to plann *en bloc* segmental resection of the mandibular lesion and enable immediate reconstruction of the defect with free bone graft. CBCT was of great importance in giving to the surgeon accurate information about the extension of the lesion and the prediction of the margins of resection's.

With the rapid development of head and neck surgical techniques over the last few decades [32, 33] this surgical technique based on piezoelectric phenomenon seems to have a lot of applications [34, 35], but until now piezoelectric resection of the IFVM of the mandible is not yet presented in the literature, except creating window for embolisation in two cases of arterio-venous malformation [36].

This article reports an urgent treatment of IFVM in the mandibular body because of acute and excessive bleeding after biopsy attempt and usage of piezosurgery enabled surgeon to create almost bloodless surgical field. It also carries minimal risk of bone necrosis due to a constant cooling in comparison to conventional surgical instruments such as burs and micro saws. Beside wide usage in dentistry piezosurgery also became a power tool in head and neck surgery. Using this ultrasonic knife the possibility of damaging blood vessels and nerves is decreased because a piezoknife only cuts hard tissue (bony structures). According to the pathology treated in this case it was very important to provide safe and bloodless procedure as much as possible.

Therapeutic alternatives in the treatment of intraosseous venous malformation include curettage and sclerotherapy [2, 26].

Conclusion

Intraosseal venous focal malformation in orofacial region should be always considered in the differential diagnosis of multilocular intraosseal lesions in oral and maxillofacial region!

The use of CBCT in the diagnosis and radiographic planning of resection of intraosseal venous focal malformation of the mandible is new method and gives more precise information not only about nature of vascular anomaly, but also of extension inside bone structure. This enables surgeon to make precise plan of resection and avoid unnecessary bleeding in potentially life threatening situation.

The advantages of piezosurgery in the treatment of intraosseal vascular malformation of the mandible are: lower risk of damaging soft tissue structures, precise osteotomy and bloodless surgery. Moreover, using piezosurgery, blood transfusion and blood transmitted diseases could be avoided.

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