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THERAPEUTIC AND REHABILITATION APPROACH FOR PATIENTS WITH PRIMARY JAW ARTERIOVENOUS MALFORMATIONS

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Abstract:

Introduction. Issues of diagnostics and treatment of primary arteriovenous malformations (AVMs) of the lower and upper jaws have been underexplored, which is due to the rather rare incidence of this pathology. It is necessary to differentiate primary and secondary lesions of the jaw bones with vascular malformations. It is possible to establish the final diagnosis, to determine the segment of the lesion, the source of bleeding, the nature of local hemodynamic disorders, especially the structure of abnormal vessels, the speed and intensity of blood flow in them by combination of special and interdisciplinary diagnostic measures.

Material and methods. The article presents the experience of treatment of 12 patients with primary arteriovenous malformations (AVMs) of the lower and upper jaws for the period from 2007 to 2017. The algorithm of examination in patients with this pathology includes radiography, computed tomography (CT), multislice spiral computed tomography angiography (MSCT AG), digital subtraction angiography (DSA). As preoperative treatment, endovascular occlusion (embolization) of afferent vessels and vascular cavities was performed; for embolization, ONYX-18 (composition of ethylene vinyl alcohol, tantalum powder and dimethyl sulfoxide, ev3, Irvine, CA, USA) and Histoacryl (2-isobutyl acetate, Aesculap AG, Germany) were used. Surgical approach and the volume of bone resection were chosen based on the localization and size of AVM. Methods of bone grafting, distraction osteogenesis, dental implantation, methods of fixed and removable prosthetics were used for rehabilitation of patients, restoration of the shape and size of the jaws, chewing function.

Results and discussion. The use of modern embolizing materials gives a pronounced hemostatic effect and significantly reduces bleeding during surgery. The main result of treatment in patients with primary jaw AVMs is the cessation of bleeding risk, the lack of continued AVM growth, which is confirmed by the data on dynamic follow-up in this category of patients. The majority of patients with postoperative defects of the lower jaw and dentition underwent multiple reconstructive surgeries.

Key words: *Arteriovenous malformations, upper jaw, lower jaw, embolization, bone grafting, and prosthetics.*

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INTRODUCTION:

Issues of diagnostics and treatment of primary arteriovenous malformations (AVMs) of the lower and upper jaws have been underexplored, which is due to the rather rare incidence of this pathology. Various authors (Dan and Sapelkin, 2008, Bhuyan et al., 2016) described isolated cases of this pathology manifestation.

It is necessary to differentiate primary and secondary lesions of the jaw bone vascular malformations due to the fact that their clinical manifestations are different, which largely affects the choice of examination approach and, fundamentally, the choice of method and scope of surgical treatment.

Primary jaw AVM occurs as a result of pathological arteriovenous fistula formation in the thickness of the spongy substance in lower or upper jaw bone, more often in the area of the mandibular canal, followed by the spread to all bone structures and surrounding soft tissues. Secondary jaw lesions occur when the patient has a long-existing, most often congenital, extensive capillary or venous malformation of the soft tissues on the face and neck or oral mucosa. Changes that occur in the jaw bones may occur in childhood and in patients of the middle and older age groups.

Primary lower and upper jaw AVMs occur only in children and young subjects and almost never in other age groups (Cariati et al, 2018). The first and, as practice shows, the most terrible symptom of this disease is bleeding from the oral cavity of different intensity from slight blood staining of saliva, red border of the lips to massive life-threatening bleeding. These bleedings can be both spontaneous and arising during of dental manipulations (probing, tooth extraction, etc.). It should be noted that spontaneous bleedings in the case of primary jaw AVM are almost never from ulceration on the mucous membrane of the oral cavity and most often from periodontal pockets (Raj et al., 2017).

Primary jaw AVMs can also have a number of indirect signs, the totality of which allows you to timely make the correct diagnosis without special research methods. The typical clinical signs of jaw AVMs are:

- Possible asymmetry of the face, which occurs due to jaw deformation or as a result of a combination of damage of the jaw and surrounding soft tissues;

- increased vascular pattern of the skin or mucous membrane due to the vessels located superficially on the side of the lesion;

- in the projection of the lesion, the pulsation, which is not normally typical for this anatomical zone, is always determined by palpation;

- Diastolic-systolic murmur with a pronounced coarse tone is determined by auscultation.

When examining the oral cavity, there is hyperemia of the mucous membrane on the side of the lesion, an increase in the volume of the gingival margin and angiomatous growths on the mucous membrane. The teeth on the side of the lesion are usually covered with plaque, can be mobile. Violation of oral hygiene is due to the patient's fear of causing bleeding, which can occur when brushing teeth.

It is possible to establish the final diagnosis, to determine the segment of the lesion, the source of bleeding, the nature of local hemodynamic disorders, especially the structure of abnormal vessels, the speed and intensity of blood flow in them by complex of special and interdisciplinary diagnostic measures. Special research methods include X-ray, ultrasound (US), computed tomography (CT), magnetic resonance imaging (MRI). It should be noted that both CT and MRI should be performed using techniques of CT angiography and MRI angiography (Belysheva et al., 2016, Warren et al., 2012).

Multislice spiral computed tomographic angiography (MSCT AG) looks especially significant in this regard. Interdisciplinary methods of research include digital subtraction angiography (DSA) which is carried out in specialized departments in in-patient settings (Theologie-Lygidakis et al., 2015).

MATERIAL AND METHODS:

According to our observations for the period from 2007 to 2017, 72 patients with head and neck AVMs of different localization were treated, 12 patients from total amount were diagnosed with primary jaw AVM (7 women, 5 men, whose mean age was 18 years). Lesions of the upper jaw were observed in 2 patients, of lower jaw in the body and ramus - in 6 patients, and only of lower jaw body - in 2 patients, and 2 patients had localization of AVM in the frontal part of the lower jaw. In most cases, there were no marked vascular changes in the soft tissues surrounding jaw AVM. In 4 patients, typical pulsation from the oral mucosa was clinically determined, due to the fact that there was resorption of the cortical plate in the area of AVM bone lesion.

The leading complaint in patients with the pathology is bleeding from the oral cavity of varying intensity. In 7 patients, spontaneous bleedings from periodontal pockets were noted in history and at admission, and 2

patients had massive bleeding developed as a result of unjustified tooth extraction, and that led to a critical general condition, and in one case, massive bleeding occurred when trying to perform a cystectomy in the area of the frontal group of teeth on the lower jaw.

The algorithm of patients' examination included a clinical method of examination (collection of complaints and medical history, examination, palpation of oral cavity and external skin), orthopantomography (OPTG), CT, MSCT AG. All patients underwent DSA, which in 9 cases was combined with embolization of intraosseous vascular cavities and major afferent vessels from the external carotid artery system. It should be noted that DSA, including that with embolization, was carried out on the right and left sides depending on AVM localization and features of its blood supply. ONYX-18 (composition of ethylene vinyl alcohol and dimethyl sulfoxide, ev3, Irvine, CA, USA) in 6 patients and Histoacryl (2-isobutyl acetate, Aesculap AG, Germany) in 3 patients were used as embolizing agents (Natarajan et al., 2009, Hsiao et al., 2012). In 3 patients, embolization was not carried out due to the fact that in 1 case the patient had a history of external carotid artery ligated to stop bleeding on the side of the lesion and, in 2 patients, the size of AVM in the lower jaw allowed for block resection with preliminary intraosseous administration of Histoacryl.

Surgical treatment was performed in all patients. In 2 patients with AVM of the upper jaw, a block resection of the AVM in the region of the molars was performed; in 4 patients with AVM of lower jaw, block resection of the AVM with the preservation of its continuity was performed, and, in 3 cases, the block resection of lower jaw was combined with its osteotomy to divide the blood supply to the AVM with the contralateral vessels. Half resection of lower jaw with exarticulation was performed in 3 patients. This choice of surgical treatment approach was due to the extensive damage to the body and ramus of AVM lower jaw.

The purpose of the first stage of surgical treatment for jaw AVM was to remove the primary AVM, eliminating the threat of bleeding. Subsequently, these patients underwent multiple surgical interventions to restore the volume of the lost bone. Free bone autograft transplantation was performed in 7 patients, and distraction osteogenesis was performed in 4 patients after autotransplantation. After elimination of the bone tissue defect, dental implantation was performed followed by non-removable prosthetics. In 2 patients, the postoperative defect of the upper jaw was eliminated by using an endoprosthesis with teeth.

RESULTS AND DISCUSSION:

Surgical treatment of primary jaw AVMs still presents significant difficulties, which are primarily due to untimely, incorrect diagnostics, lack of methodologically justified, optimal algorithm of complex surgical treatment. Bleeding from the oral cavity occurring spontaneously or with various dental manipulations can lead to a severe clinical situation up to death in this category of patients. In 5 patients, massive bleeding was provoked by unjustified removal of teeth from the area of the jaw AVM, and in one case, bleeding arose from the incision of the mucous membrane due to cystectomy.

The main result of treatment in patients with primary jaw AVMs is the cessation of bleeding risk, the lack of continued AVM growth, which is confirmed by the data on dynamic follow-up in this category of patients.

Rapid rehabilitation of patients with primary jaw AVMs after their surgical treatment is possible only with the help of removable prosthetics. In 2 patients with AVM of the upper jaw, oroantral fistula was formed after partial resection of the upper jaw with AVM from the tooth 1.3 to 1.8 in the postoperative period, and it could not be eliminated without the use of complex tissue complexes on the microvascular anastomosis. Patient refused from the proposed plan for further surgical treatment for various reasons. In one case, a defect of the upper jaw with an oroantral fistula was eliminated with the help of an endoprosthesis with teeth. The rest 10 patients with postoperative defects of the lower jaw and the dentition required repeated reconstructive operations, including free transplantation of autologous bone, the use of distraction osteogenesis to restore the lost bone volume. The number of surgical interventions ranged from 4 to 6 in different patients. Rehabilitation time (up to the manufacture of fixed prosthetic constructions on implants) was, on the average, 20 months.

According to our observations, during reconstructive surgery in patients after removal of jaw AVM, there are features that appear in the form of increased bleeding from soft tissues surrounding jaw defect and, most importantly, from the bone tissue in the area of previously removed AVM. Obviously, the bleeding is not massive and has the traits of quite abundant capillary bleeding across the surface of the bones aggravated by osteotomy for fixation of compression and distraction apparatus. This is probably due to developed vascular network, which

was formed at the time of the active AVM functioning, and cannot be technically eliminated during surgery. Also prosthetic implants were introduced in these patients after surgical treatment of jaw AVM, and there is increased susceptibility to the occurrence of peri-mucositis and peri-implantitis, which leads to increased bleeding. This circumstance is worrying for patients, who perceive the bleeding as resumption of AVM growth. In this regard, optimal orthopedic design, compliance with oral hygiene, and timely periodontal treatment are of paramount importance.

Clinical case 1

Male patient N. of 21 years old was admitted with a diagnosis of body and ramus AVM on the left side of lower jaw, bleeding (Fig. 1).

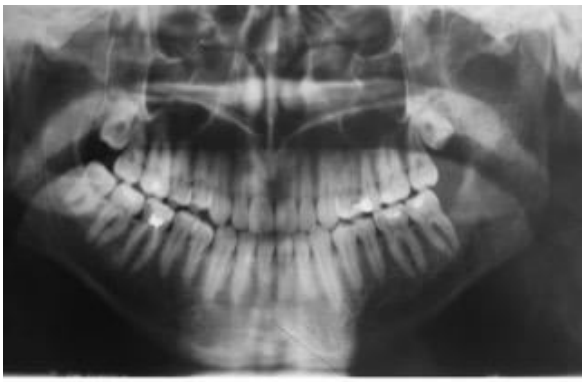

Figure 1. Orthopantomogram of patient N., AVM cavities are revealed in body and ramus of lower jaw

Based on medical history, it is known that the patient consulted to the dentist with complaints of pain and bleeding gum in the left retromolar region. Pericoronitis was diagnosed with impacted tooth 3.8. When trying to excise the hood of the mucous membrane, massive bleeding started, which was stopped by tamponade. The patient was hospitalized in City Clinical Hospital No. 1, Kazan. During examination at the department, bleeding re-emerged and it was decided to remove 3.8 with subsequent tamponade of cavity by iodoform tampon. Blood loss during surgery was 1 liter. The patient was transferred to Moscow by air ambulance. At N. N. Burdenko Research Institute of Neurosurgery, angiography showed AVM of lower jaw body and ramus with the formation of abnormal vascular plexus in pterygopalatine fossa, pterygomandibular and parapharyngeal space. After the analysis of the obtained angiograms, embolization of bone cavity of AVM and hypertrophied pathological rami of the external carotid artery on the left with ONYX 18 was performed simultaneously (Fig. 2; 3).

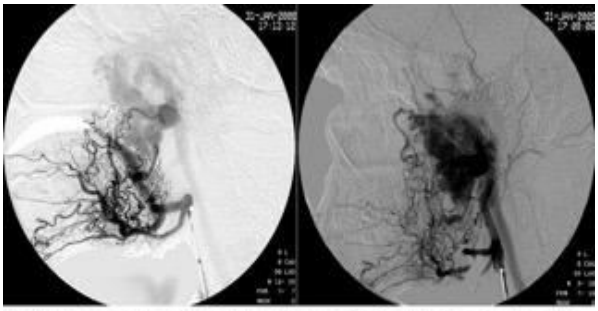

Figure 2. Angiogram of external carotid artery branches on the left side of patient N. in arterial and venous phases

Figure 3. X-ray image after embolization of cavities and vessels of lower jaw AVM with ONYX-18 in frontal and lateral view

Onyx® is a liquid embolizing composition, a solution of ethylene vinyl alcohol co-polymer (EVHO) with tantalum powder in dimethyl sulfoxide (DMSO). The composition is injected through the microcatheter in a liquid state. When injected into the bloodstream, there is a rapid diffusion of DMSO into the fluid in the blood, which results in starting the polymerization reaction of co-polymer, and the final polymer forms a precipitate of soft consistency, which occludes abnormal vessels. Due to its physical properties, Onyx can penetrate directly into the area of the pathological focus. With the correct choice of the embolization zone and compliance with the drug administration technique, most of the vascular abnormalities may be switched off totally from one afferent vessel.

There was no bleeding after embolization. 5 days after the endovascular intervention, the left lower jaw resection was performed with exarticulation. From the features of surgery course, it should be noted that, when exposing the lower edge of the jaw, a lot of non-functioning, empty vessels of arterial and venous types were revealed. Surface of the outer cortical plate of jaw body and ramus was eroded and does not bleed. With skeletonizing lower jaw ramus inner surface, large focus of destruction of the cortical plate was visualized. Hypertrophied empty vessels, mainly of venous type, in the lumen of which there was Onyx 18 in the form of a loose gray-black sponge, were found in pterygomandibular and parapharyngeal

space. Outside the surgical wound, the alveolar process was resected together with the teeth, and the spongy bone and elements of the neurovascular bundle were removed. The orthotopic autograft was modeled from the gross specimen, and its replantation with fixation by titanium plates to the main fragment of lower jaw was carried out. Blood loss during surgery did not exceed 200 mL (Fig. 4).

Figure 4. Stage of replantate formation from resected part of lower jaw body and ramus and its fixation with titanium plates

In six months after the surgery, titanium plates were removed and horizontal osteotomy of the preserved fragment of the lower jaw body was performed with fixation and subsequent activation of the alveolar compression and distraction apparatus (Konmet, RF). The value of distraction was 10 mm. In 5 months after the distraction and removal of the compression and distraction apparatus, in order to equalize the level of the alveolar crest edge, horizontal osteotomy of bone regenerate was performed, and the deformation of the alveolar crest was eliminated with the help of sandwich method (Fig. 5;6;7).

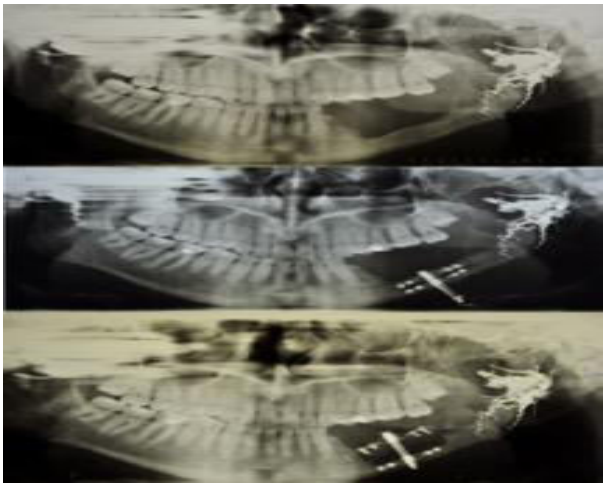

Figure 5. For increased height of alveolar crest, distraction osteogenesis was applied.

Figure 6. Osteotomy of obtained regenerate and increased height of alveolar crest by sandwich method

Figure 7. Orthopantomogram. Lower jaw defect reconstruction.

In 6 months after reconstructive surgery to restore the height and volume of the lower jaw, dental implants were installed followed by the manufacture of non-removable metal-ceramic structure. The duration of rehabilitation for patient after surgery for AVM elimination was 20 months (Fig. 8).

Figure 8. Dental implants were installed and non-removable metal-ceramic constructions were manufactured for patient N. Complete rehabilitation was achieved.

with complaints of cystic mass in area of teeth 4.4, 4.3, 4.2. Mass in the mental area was mistakenly interpreted by oral surgeon as radicular cyst in the area of above mentioned teeth, and teeth were devitalized and sealed before cystectomy (Fig. 9).

Figure 9. Orthopantomogram of patient E. in area of teeth 4.2, 4.3, 4.4, there are multiple bone AVM cavities in mental area.

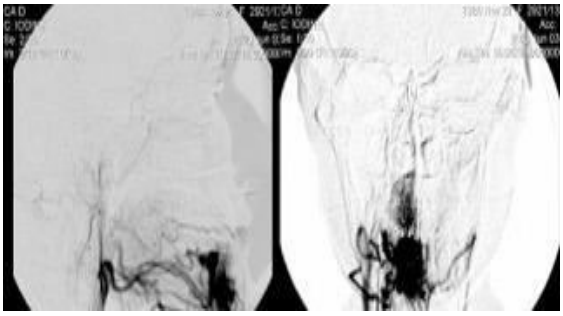

Figure 10. Angiogram (DSA) of patient E. in frontal and lateral view there is filling with contrast agent of AVM cavities in afferent vessels on the both sides.

the incision in the transitional fold, which was stopped due to coordinated actions of medical staff and highly qualified surgeon. In the subsequent clinical examination confirmed by OPTG and CT data, the primary AVM on the right side of mental area was diagnosed. In the oral cavity, in the absence of visible changes in the mucous membrane in the area of oral vestibule, in the area of teeth 4.4, 4.3, 4.2 on the lingual side, soft tissue swelling with a pronounced pulsation in this area was revealed. The teeth in AVM region are stationary. On X-ray in the frontal part of lower jaw on the right side, many cavities of different diameter in the thickness of the spongy bone and on the surface of the outer and inner cortical plates were determined. The entrance to the mandibular canal is dramatically expanded, and the diameter of the canal in the area of the right mental foramen is also significantly increased. According to CT in projection of the teeth 4.2, 4.3, 4.4, there is no internal cortical plate but there is soft tissue mass bulging towards oral cavity floor. The diagnosis of primary lower jaw AVM was made on the basis of clinical examination, X-ray and CT studies. The patient was recommended to undergo DSA with possible embolization of vascular cavities in the bone and afferent vessels. Angiographic examination confirmed the clinical diagnosis of lower jaw AVM, and after that the embolization of AVM cavities and vessels in the immediate vicinity to lesion was performed simultaneously. Onyx 18 was used as an agent for embolization (Fig.10,11).

Figure 11. Control angiogram after embolization with ONYX-18, there is no contrast enhancement of bone AVM in mental area.

unced bone
e jaw when
planning surgical treatment, it was decided not to resect the fragment of the lower jaw, but to limit the block resection with preservation of its continuity. Despite the preoperative embolization during block resection, there was a rather abundant bleeding from the bone and surrounding soft tissues, which can be explained by an extremely developed pathological vascular network, both in bone tissue and in soft tissues adjacent to the lower jaw. Blood loss during surgery was 800 mL (Fig.12).

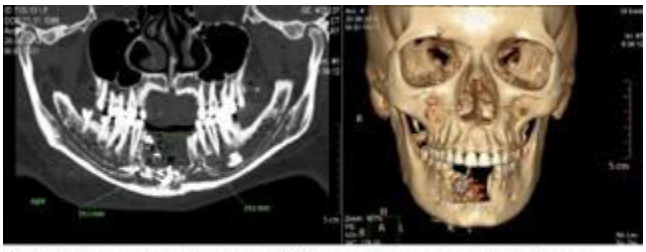

Figure 12. Computed tomography and CT with 3D-reconstruction of patient E after block resection of lower jaw in frontal part area.

The postoperative period was normal. Re-hospitalization took place in 6 months. Horizontal osteotomy of lower jaw in the defect area from 4.4 to 3.1 with fixation of the skeletal distraction apparatus was performed. The value of distraction was 10 mm (Fig. 13).

Figure 13. Increased alveolar crest height: horizontal osteotomy with subsequent fixation and activation of extramedullary alveolar distraction apparatus.

After 7 additional months, surgical removal of the distraction apparatus with simultaneous installation of 4 dental implants was performed. Prosthetics stage according to the standard scheme took place in 3 months (Fig. 14).

Figure 14. After installment of 4 dental implants, non-removable metal-ceramic construction was manufactured, complete rehabilitation was achieved.

The time required for treatment and rehabilitation was 18 months.

CONCLUSION:

1. Approach for examination of patients with primary jaw AVM in addition to the classical survey and examination of the patient must necessarily include orthopantomography, computed tomography, multislice spiral computed tomography angiography and digital subtraction angiography.
2. Surgical treatment of patients with primary jaw AVM consists of resection of the jaw fragment affected by AVM up to half resection of the jaw. Given the non-tumor genesis and nature of the growth of primary jaw AVM, it is advisable to carry out organ-preserving surgery with a possible replantation of the resected jaw fragment. The combination of surgical treatment and preoperative embolization significantly reduces the risk of massive bleeding during surgery.

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All authors were equally involved in the preparation of the material. The investigation was in accordance with the ethical standards of the responsible committee.
Conflict of interest: none declared.

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List of abbreviations:

AVM - arteriovenous malformation

US - ultrasound

CT - computed tomography

MRI - magnetic resonance imaging

MSCT AG - multislice spiral computed tomographic angiography

DSA - digital subtraction angiography

OPTG - orthopantomography

EVHO - ethylene vinyl alcohol co-polymer

DMSO - dimethyl sulfoxide

REFERENCES:

1. Belysheva T.S., Mikhailova E.V., Panferova T.R., Trofimov I.A., Aliev M.D. Sovremennye podkhody k diagnostike sosudistykh mal'formatsii u detei. Sarkomy kostei, myagkikh tkanei i opukholi kozhi. 2016;2:44-53. (In Russ
2. Bhuyan S.K., Birmiwal K.G., Kar I.B., Bhuyan R., Debta P. High Flow AV Malformation (A-V Shunt) Of Mandible: A Rare Life Threatening Entity. *Journal of Clinical and Diagnostic Research.* 2016:10(11):ZD16-ZD18 Available at: https://doi.org/10.7860/jcdr/2016/22221.8862.
3. Cariati P., Marin-Fernandez A., Julia-Martinez M., Perez-de Perceval-Tara M., Sanchez-Lopez D., Martinez-Lara I. Endovascular Treatment of an Intraosseous Arteriovenous Malformation of the Mandible in a Child. A Case Report. *Journal of Clinical and Experimental Dentistry* [Internet]. Medicina Oral, S.L.; 2018;0–0. Available at: http://dx.doi.org/10.4317/jced.5455
4. Dan V.N., Sapelkin S.V. Angiodisplazii (vrozhdennye poroki razvitiya sosudov). M.: Verdana; 2008. (In Russ.) 5.Hsiao, C., Wong, H. and Chen, L. Onyx embolization of a lingual arteriovenous malformation. *Asian Journal of Surgery.* 2012;35(4):159-162. Available at: https://doi.org/10.1016/j.asjsur.2012.04.028
6. Natarajan S.K., Born D., Ghodke B. Britz G.W., Sekhar L.N. Histopathological changes in brain

arteriovenous malformations after embolization using Onyx or N-butyl cyanoacrylate. Laboratory investigation. *Journal of Neurosurgery*. 2009;111(1):105–113 Available at: http://doi.org/10.3171/2008.12.jns08441

7. Raj S. Ch., Mishra D., Praharaj K., Panda S. M. Arteriovenous Malformation of Mandible: A Periodontist Diagnosis. *Indian Journal of Dental Education.* 2017;10(1):57-61. Available at: https://doi.org/10.21088/ijde.0974.6099.101178.
8. Theologie-Lygidakis N., Schoinohoriti O., Tzermpos F., Christopoulos P., Iatrou I. Management of Intraosseous Vascular Malformations of the Jaws in Children and Adolescents: Report of 6 Cases and Literature Review. *Journal of Oral & Maxillofacial Research*. 2015;6(2):e5. Available at: doi:10.5037/jomr.2015.6205.
9. Warren T.A., Gandhi M., Panizza B. Pictorial review: Vascular anomalies of the head and neck. *Journal of Medical Imaging and Radiation Oncology.* 2012;56(1):84-92. Available at: https://doi.org/10.1111/j.1754-9485.2012.02342.x