Prosthetic rehabilitation of combined facial defect resulting after arteriovenous malformation

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Abstract

This clinical report details the rehabilitation of a patient who underwent surgical treatment of arteriovenous malformation located in facial tissues. The treatment led to extensive facial defect including orbital region, nose, zygomatic arch, part of maxilla. Prosthetic rehabilitation was chosen in order to improve esthetics. With the aid of prosthesis retention 5 extraoral implants were placed. In spite of wrong position of 1 implant, complex configuration of the defect, expressed vascular pattern on the skin surround defect, impermanent swelling of the facial tissues, appropriate result was achieved.

Keywords: maxillo-facial prosthesis (P.M.F.); arteriovenous malformation; extraoral implants

INTRODUCTION

There are many diseases and incidents that may result facial defects and deformities: cancer, trauma, burns, congenital anomalies, and autoimmune diseases. Arteriovenous malformation (AVM) is a pathological lesion composed of direct abnormal connections between the arterial and venous vessels and intervening capillary bed is absent,[1] and despite its benign nature surgical treatment of AVMs leads to large unreconstructible facial defects. Head and neck AVMs occur in 0.1% of population.[2] Acquired AVMs can appear from injury, surgical interventions, hormonal changes (puberty or pregnancy).[3] Eerola et. al reported that vascular lesions and AVMs might be genetically determined (RACA1 gen mutation) and associated with Sturge-Weber syndrome, Klippel-Trenaunay syndrome, Parker Weber syndrome.[4]

The diagnosis is usually based on clinical presentation, multiply imaging techniques and histological findings. The clinical symptoms of arteriovenous malformation in head and neck region include a facial asymmetry, pulsation, increased warmth of the skin, red skin

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spot, wind-blowing-like noise. Life-threatening bleeding (for example, uncontrollable bleeding after tooth extraction), severe pain, airway obstruction, bony destruction, tooth mobility often accompany the disease in maxillofacial region.[5,3] Moreover, patient may complain of blurred vision if orbital structures are involved.[6] Arteriovenous shunting may reduce blood supply in the skin, leading to ischaemic necrosis and ulceration.[7] Currently, only multidisciplinary approach including clinical evaluation, histological analysis and radiographic imaging can provide us enough information for definitive diagnosis and comprehensive treatment.[8]

The main aim in treatment of AVM is to reduce the risk of life-threatening bleeding. Pain reduction, function and esthetic improvement also can't be ignored. The treatment of AVM usually needs any kind of surgical intervention: embolization, resection or laser treatment.

Nond Rojvachiranonda MD et al. reported about application of Nd:Y3Al5O12 (Nd:YAG) laser for intralesional treatment of cutaneous AVM in 4 cases. Laser was delivered by fiberoptic that was inserted into lesion via stab incision with 18-gauge needle, at adjacent noninvolved area. Laser energy was set in the range of 20 to 60W/pulse in repetitive mode of 0.1 second off and 0.1 second on. The treatment endpoints was based on subjective surgeon's judgment about visible reduction of mass size and transformation of mass consistency from soft to more solid. As authors see it, Nd:YAG laser is an excellent choice in the treatment of large and deep vascular lesions due to its wavelength specific to hemoglobin and depth of its penetration of up to 5 to 6 mm in soft tissue.[9]

Laser ablation is also useful in treatment low flow vascular lesion. Derby LD and Low DW described their experience of laser treatment of facial venous vascular malformations. Various laser modalities were used: argon laser, yellow dye laser, Nd:YAG\KTP laser (deep intralesional delivery with fiberoptic) in thirty-four patients. These authors reported that lesion bulk and color was generally «improved», but not complete in large lesion. Authors were faced with recurrence in few cases and some complications: cutaneous burn and oral commissure scar contracture.[10]

Nevertheless, this treatment option has shown to be minimal invasive and safe. Laser is a valuable addition to traditional arsenal in treatment of AVM and other vascular lesion in the head and neck region.

For today, there is no consensus what method for management AVMs is the best, however superselective embolization combined with surgical resection is recommended to be used as treatment of AVMs in oral and maxillofacial region.[5,11,12] The main aim of surgery part of the treatment is to remove the nidus and reduce the risk of life-threatening bleeding. But radical resection frequently leads to formation of extensive facial defect: skin, muscles, mucosa, cartilage and bone structures can be removed.[5] Fortunately, free tissue transfer and local flaps may be used to reconstruct defect with acceptable cosmetic and functional results in most cases.[6] However, treatment of AVM is unpredictable and sometimes does not provide defect with free of disease progression margins. Blood supplying of surrounding tissues might be insufficient for surgical reconstruction.[3] It is clear that in this cases prosthetic rehabilitation – the only possible way to improve esthetics.

This clinical report describes prosthetic rehabilitation of patient with extensive facial defect acquired by surgical treatment of an arteriovenous malformation.

CASE REPORT

A 57-year-old man suffered of arteriovenous malformation since 2000, when he had noticed small lesions over medial angle of the right eye. The lesion was gradually increasing, then blindness of right eye have been developed and few episodes of bleeding have happened. First unsuccessful attempt of partial endovasular embolization of AVM was performed in 2014. Then emolization was repeated in 2018 and 2019. Unfortunatly, the last embolization caused inflammation process and tissue necrosis of the nose and severe nose bleed-

ing. Moreover, emboletherapy provoked the growth of collateral blood vessels and additional nidus formation (upper lip). Surgical resection was recommended as appropriate treatment option. Presurgery selected angiography revealed extensive pathological nidus in right orbital region, facial region with afferent vessels from a. maxillaris dextra, a. sphenopalatina dextra, a. infraorbotalis dextra, a. ophthalmica, a. carotis interna dextra (after partial embolisation) and anastomoses from a. carotis interna sinistra. The efferent vessels were presented by v. facialis (v. jugularis externa) and vv. ophthalmica inferior et superior (sinus cavernosus). Then surgical resection of the malformation was performed. Destruction of maxilla, nasal bones and nasal cartilage was revealed during surgery, thereby zygoma, zygomatic arch, part of maxilla were resected, fortunately alveolar ridge, palate were saved. An attempt of primary surgical reconstruction using free thoracodorsal artery perforator flap was made. After a month total necrosis of transferred flap occurred, falp was removed and second attempt of surgical reconstruction using contralateral free thoracodorsal artery perforator flap was performed. Unfortunatly, this attempt of microsurgery reconstruction was unsuccessful too. Finaly, necrotic tissues was eliminated and split thickness skin graft from anterior surface of the tight was harvested and placed on granulation tissue in defect. Postoperative computer tomography revealed resection of nasal bone structures and cartilages, right zygoma, right zygomatic arch and part of right maxilla without loss of alveolar ridge, palate.

One year later the patient was referred to the maxillofacial prosthetist of Institute of dentistry and maxillofacial surgery (Pavlov First Saint-Petersburg State Medical University). Patient demonstrated large combined facial defect including orbital region, nose, zygomatic arch, part of maxilla (Fig. 1). During clinical examination postoperative scars, swelling of right check and deformation of upper lip was revealed, expressed vascular drawing of surrounding skin and cyanosis of upper lip were determinated. Primary facial impression and cast was obtained for preliminary planning of prosthesis' boarders and fixtures positions (Fig. 2). Moreover, primary cast is needed for manufacturing custom-made reinforcing device which is useful during definitive impression.

Five Vistafix extraoral implants were placed according to bone conditions and supposed construction of future prosthesis: two – in the base of piriform opening (frontal aspect of maxilla), two – in lateral aspects of supraorbital rim, one – in medial aspects of supraorbital rim. Because of compromised status of surrounding soft tissues two-stage procedure was chosen. Five month later second stage surgery was performed: three standard abutments (Standart Abutment 4mm) an two console abutments (30 degrees) were fixed (Fig. 3). Then five magnacaps and magnetic impression caps were connected. The defect was large and had complex shape with numerous undercuts, which were blocked out with silicone putty to prevent rupture of the impression (Fig. 4).

To obtain definitive impression we used two-layer technique. First layer of the impression was low-viscosity and low-durometer silicon, which is ideal for reproduction of small surface details. Then second layer of silicone material with higher durometer was used to incorporate the custom-made reinforcing device in impression in order to support. After manufacturing of the definitive cast we fabricated acrylic resin mesostructure with prosthetic magnets and wax pattern. Two try-in appointment was scheduled. First appointment was planned for artificial eye positioning. Unfortunately, the correct positioning of artificial eye was impossible due to wrong position of the fixture (with suprastructure) located in medial aspect of supraorbital rim. The suprastructure was unfixed and this fixture wasn`t used for retention in order to achieve appropriate artificial eye placement and better aesthetic result. Eye lid aperture, shape of opening lid, shape of the nose, contours of the inner surface of the wax pattern, providing breathing, skin texture and marginal adaptation were checked at second try-in appointment.

The traditional "trial-and-error" method of

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mixing a intrinsically colored silicone was used. Moreover, additional portions of different colored silicone were prepared. Laminar painting technique with antislump agent was used (Fig. 5). Blood vessel simulation was achieved by using red tread. When silicone was completely cured, the extrinsic characterization, sealing and deglossing was performed. Generally, patient was satisfied with esthetic and functional aspects, such as breathing and easy insertion and removing (Figs. 6, 7). From time to time patient noticed gap between cheek and edge of prosthesis in spite of correct position of the prosthesis.

DISCUSSION

Communication between surgeon and anaplastologist, creation of detailed plan of prosthetic rehabilitation before the treatment is crucial for successful aesthetic and functional result. In our clinical case one of the fixtures was located in unplanned position (in medial aspect of supraorbital rim) and it made using of this fixture impossible. Moreover, it is reported, that medial aspects of orbital rim is not recommended for fixture placement due to the lateral region provides better bone volume and lower risk of inflammation of peri-implant tissues.[13] Fortunately, another four osteointegrated fixtures are located in prostheticly oriented position and provided sufficient retention. One cannot deny that anatomy-based selection of the location for implant placement and its prostheticly oriented position are equally important in workflow. In order to provide the optimal implant position the guided surgery should be applied. To take into account both bone condition and planned prosthesis configuration during template manufacturing mirror image of the unaffected facial part should be placed on image of the affected side and matched with CT data. Moreover, navigation surgery with special markers and software might be used. It gives opportunity to verify during the operation, in real time, the drilling direction and its depth.[14] We hope that the development of additive surgery such as guide surgery, navigation surgery and augmented reality and mixed reality surgeries will help us to avoid the challenges in fixture positioning and reduce surgical risks.

In cases of extended combined facial defects we usually use technique of reinforced definitive impression. Firstly, the reinforcing device is constructed on diagnostic cast from light curing resin for dental custom trays. Then the custom-made reinforcing device should be placed between two layers of silicon impression material during obtaining of definitive impression. In this way, the impression becomes more rigid and resistant enough against deformation during cast pouring despite its extensive boarders. Consequently, this tip makes pouring of cast more precise and gives us confidence during other stages of prosthesis manufacturing. Rigidness of impression is extremely important in cases of extensive facial defects, because impressions with large borders are more exposed to deformations. Various clinical approaches to obtaining proper facial impression are reported in periodical literature. Kubon et al. suggested to connect the impression copings with prefabricated acrylic resin bars and apply a small amount of uncured acrylic tray resin over the impression. This tips minimize the distortion of the soft tissues and provide accurate capture of implant position.[15] To reduce distortion in case of extensive middle face defect a two-stage impression technique was developed by Coleman et al. The technique involves the custom impression tray preparation: it can be made on the preliminary cast or by adapting uncured resin for trays to the face of the patient. Then the first layer of low-viscosity polyvinyl siloxane impression material should be applied on a custom tray to record the soft tissues that will be contacted by the margins of the prosthesis, after first layer will be cured irreversible hydrocolloid should be applied to the face to record the remaining facial structures (second layer of the impression). The last step is plaster of paris for reinforcement.[16]

At first glance, implementation of digital impression might eliminate shortcomings of traditional techniques related to mechanical distortion, schrinkage and deformation during

cast pouring, but digital technologies have its own disadvantages. Firstly, the implementing of digital treatment pathway requires buying a big amount of additional expensive devices, software and materials. Secondly, the most challenging aspects of digital facial impression is to take into account a wide range of head and facial movements,[17,18] which can be easily recorded by functional movement during obtaining of traditional impression.

Some specific circumstances associated with vascular nature of the primary lesion were revealed during prosthesis manufacturing. Firstly, there is expressed vascular pattern on the skin surround defect. This feature should be considered during coloration of the prosthesis: for extrinsic coloration blue and purple pigments were used to draw venous vessels. In this way, hiding of the prosthesis` edge in areas, where surrounding skin is visibly vascularized was achieved. Secondly, it can be assumed that surrounding soft tissues (not only skin) contain pathological vessels or residual vascular lesion without clinically significant symptoms. In our clinical case patient demonstrated facial asymmetry due to swelling of residual part of the cheek. The swelling was impermanent and increased in the morning. As we see it, the gap between cheek and edge of prosthesis, that patient noticed from time to time, is determined by predisposition of the surrounding tissues to swelling. In order to avoid unsatisfactory edge adaptation the definitive impression should be obtained in the middle of day (not in early morning). Moreover, the horizontal position during impression obtaining is not preferable in case of tissue swelling, the patient's head should be vertically oriented to prevent unwanted soft tissue distortion.[19]

It should also be noted that some authors reported about unsatisfactory margin adaptation in anterior aspect of the cheek even in cases without vascular lesion.[20,21] The skin in the cheek region is more compressible due to absent of any supportive bony structures, therefore the procedure of impression of the defect becomes difficult and unpredictable.

Shigli et al. suggested to overcome this problem by recording the defect with a dermostatic impression using light body consistency material.[20] Sometimes facial prostheses demonstrate compromised margins and poor elasticity during facial expressions and jaw movements, especially in cases of extensive facial defects. In order to achieve better marginal adaptation Kurunmäki et al. have constructed facial prosthesis using a framework of glass fiber-reinforced composite embedded into the silicone body of the prosthesis. Special emphasis was placed on the amount and direction of the fibers, especially at the prosthesis margins, to obtain slight pressure on the skin at these margins, to ensure the margins would adapt to the facial expressions and jaw movements.[21]

CONCLUSION

This clinical report demonstrates the process of facial prosthetic rehabilitation of the patient who underwent radical surgical resection of arteriovenous malformation. We have faced some difficulties:

1. Extent and complex configuration of the defect
2. Lack of supportive bone for fixture insertion in prostheticly oriented position
3. Expressed vascular pattern on the skin surround defect and swelling associated with vascular nature of the primary lesion

Despite these challenges appropriate result was achieved. Currently, anaplastology requires usage of cut-edge technologies, digital planning of the treatment, implementation of individual approach on each step of facial prosthesis manufacturing, but deep understanding of conventional technologies and high level of artistic skill of the prosthetist are still relevant today.

Declarations of Competing Interest

The authors declare no conflict of interest.

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FIGURES

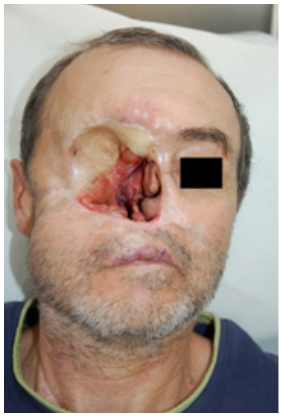

Fig. 1 – Patient with large combined facial defect.

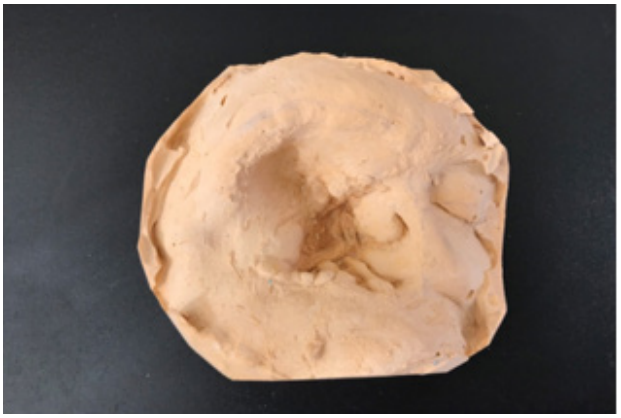

Fig. 2 – Primary cast, that is useful for planning of prosthesis' boarders and fixtures positions.

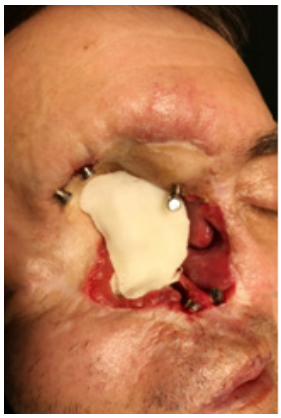

Fig. 3 – Abutments and magnets are fixed.

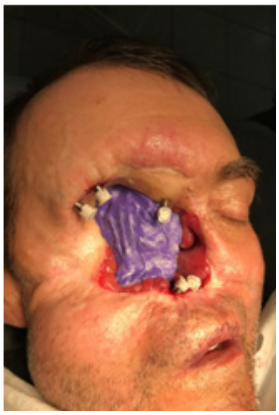

Fig. 4 – Magnetic impression caps are connected and undercuts are blocked out with silicone putty to prevent rupture of the impression.

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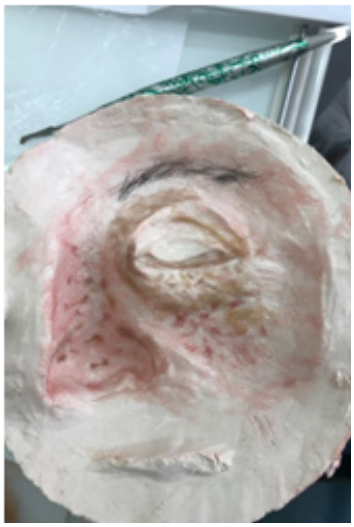

Fig. 5 – Laminar painting in mold with antislump agent.

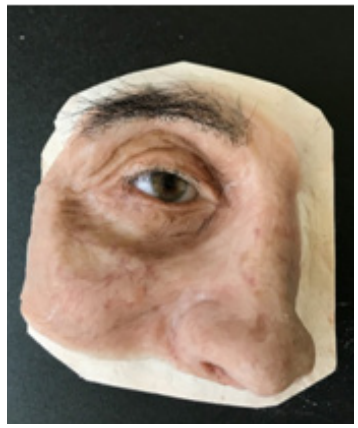

Fig. 6 – Final view of prosthesis.

Fig. 7 – Delivered prosthesis.