Mandibular gingival arteriovenous malformations (AVMs) are rare entity with high potential risk of life threatening complication. Spontaneously loosened tooth result in rapid massive bleeding clinically occurred in some cases[1]. The treatments of mandibular gingival AVMs include extensive surgical resection with mandibular reconstruction and endovascular embolization performing by interventional radiologist [1]. In order to avoid the risk of disastrous bleeding during surgery and shorten the operation time, interventional embolization become the trend to treat these type of vascular malformation [2]. We present this case of left extensive mandibular gingival AVMs which was treated by new embolic agent of Onyx with balloon-assisted technique.

CASE REPORT

This 18-year-old girl had a long history of mandibular arteriovenous gingival malformations with episodes of spontaneous bleeding and referred to our department for emergent interventional therapy. Onyx was chose as embolic agent due to its specific physical properties and infuse under balloon-assisted technique for good penetration of embolic particles into the nidus and feeders. We present this case and share the experience of this technique. We also describe the technical details, properties of embolic agents and review some literatures in public.

Balloon-assisted Onyx Embolization of an Extensive Mandibular Gingival Arteriovenous Malformation: a case report

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ABSTRACT

This is a rare case of mandibular arteriovenous gingival malformations with episodes of spontaneous bleeding and referred to our department for emergent interventional therapy. Onyx was chose as embolic agent due to its specific physical properties and infuse under balloon-assisted technique for good penetration of embolic particles into the nidus and feeders. We present this case and share the experience of this technique. We also describe the technical details, properties of embolic agents and review some literatures in public.

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Onyx embolization of gingival arteriovenous malformation

(Cook Medical, USA) at proximal left external carotid artery to introduce coaxial balloon catheter. A 6-Fr Neuron (Penumbra, USA) catheter was approached into proximal left lingual artery. A 4 x 11 mm Scepter X-tra compliant coaxial balloon catheter (Microvention, USA) was then navigated deeply into left lingual artery crossing over the aneurysm mentioned previously and inflated the balloon (Fig. 2a, arrow). Before embolic agent injection, the Onyx vials were put on a shaker (Vortex-Genie, Scientific Industries, USA) for at least 30 minutes to ensure complete shaking and mixing tantalum for radiopacity. Embolic agent of Onyx 34 (Fig. 2b, arrow) (1.5 ml per vial, 8% EVOH, Covidien, USA) was infused slowly about 0.1 ml per second under balloon-assisted technique. We stopped injecting Onyx immediately and pull back the balloon slightly while the Onyx retrograde reflux near tip of balloon catheter to avoid Onyx entrapment. The upstream aneurysm was then filled up by relatively large volume of Onyx after distal feeders and nidus closure in order to prevent large amount embolic material passing through the shunt into the internal carotid circulation. Another feeder of left inferior alveolar artery was embolized as the same way. At the end of embolization, a total 7.5 ml Onyx 34 was injected. The lateral mandibular plain image revealed Onyx embolic cast

**Figure 1.**

- **a.** The image of lateral view of left external carotid angiogram demonstrated the extensive arteriovenous malformation over left mandibular gingival region with nidus, engorged draining vein and multiple network of arterial feeders mainly from left lingual artery (dotted arrow) and inferior alveolar artery (arrow)
- **b.** The oblique view of left lingual (arrow) angiogram showed a large saccular aneurysm at proximal site (dotted arrow)
- **c.** The post-TAE of left ECA lateral angiogram showed more than 80% blood flow reduction in the nidus of the AVMs and closure of most of the venous lakes.
penetrating deeply into the feeder, nidus, draining vein and the aneurysm and the balloon was located at the proximal lingual artery (Fig. 2c).

After endovascular treatment, the patient recovered well without bleeding and was discharged 1 week after treatment. No recurrent bleeding episode but only mild facial numbness and ecchymosis was mentioned clinically after 13 months follow-up in dental OPD.

DISCUSSION

Mandibular gingival AVMs are rarely documented entities of high flow vascular malformation. It may usually present with abundant vascularity and massive intractable hemorrhage. Mandibular gingival AVMs usually occur in children and probably related to embryologic disturbance of angiogenesis and arising persistence of immature
arteriovenous shunts [3]. The main target of endovascular embolization of the large mandibular gingival AVMs is to decrease blood flow of the shunting and lower risk of spontaneous hemorrhage [2]. Beside, complete obliteration of the nidus is also possible in some smaller AVMs [4]. Surgery is not recommended for extensive mandibular AVMs due to potential hazard of massive bleeding [5].

Sceptor XC balloon catheter used in this case was documented in some reports which describing the utilizing experience of intracranial, head and neck pathologies [2]. In this report, we present our experience of the feasibility and accessibility of this balloon catheter using in mandibular AVMs. The Sceptor XC is a dual lumen catheter with an outer lumen for inflation and an inner 0.0165 inch for micro guiding-wire working lumen. This allows for using 0.014 inch microwires which are more navigable and provide more support for the wire [2, 6]. It is possible to deployed the catheter more distally even the feeding arteries are tortuous.

Embolic materials been generally used in AVMs including Polyvinyl Alcohol Foam (PVA), N-butyl cyanoacrylate (NBCA) and Ethylene-vinyl Alcohol Copolymer (Onyx). In this case, we choose Onyx for embolization due to its physical properties of lower viscosity and more solidification time, which makes the embolic agent travel more distally and penetrate deeper into the AVMs than traditional embolic agent [7]. Onyx 34 with higher EVOH concentration and lower polymerization time was suitable for larger, extensive and high flow AVMs of this patient [8]. Although the flow was decreased under balloon-assisted technique, the risk of embolic agent penetration via dangerous anastomoses from internal maxillary artery into internal carotid circulation is still persisted in this technique [9]. Injecting slowly with 0.1 ml per second and relatively fewer polymerization time of Onyx 34 could prevent embolic agent passing too distally through the shunting into internal carotid circulation [6, 10] and ensure the best infiltration quality of onyx into the AVM nidus [8]. Nevertheless, there are some documented disadvantages of infusion slowly such as requiring more nidal injection time and increasing radiation exposure to the patient and radiologist [4].

In addition, Onyx reflux would harm the normal vasculature. Using Scepter XC coaxial balloon catheter form a proximal plug could avoid Onyx retrograde reflux to the non-target vessel and allowing Onyx travel forwardly which has documented in some reports including cerebral AVMs or arteriovenous fistula [2, 6, 11, 12]. Nevertheless, retrograde reflux and entrapped the tip of balloon catheter by Onyx should be concerned, we injected Onyx slowly and deflated and retrieved the catheter as soon as possible when the Onyx retrograde reflux near the balloon catheter tip. Besides, the inflated balloon assisted technique reducing blood flow allowing greater Onyx injection control, nidal penetration and prevent reflux of the embolic agent.

In conclusion, we present this case based on its uniqueness and rarity with successfully embolization by Onyx under balloon-assisted technique. Good penetration of Onyx into left mandibular gingival AVM’s feeding arteries, nidus and venous lakes depending on its physical feature under balloon-assisted plugging proximally. We consider that Onyx embolization with balloon assisted technique efficiently cure the mandibular gingival AVMs and indicating good clinical prognosis of this patient after 13 months follow up.

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REFERENCES


