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Management of Oral and Maxillofacial Arteriovenous Malformations Using Vascular Ligation and Intralesional Bleomycin Sclerotherapy: A Four-Case Series

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Abstract:
Background: *Arteriovenous malformations (AVMs) are congenital vascular anomalies characterised by abnormal direct shunting between arteries and veins, bypassing the capillary bed. They represent a therapeutically challenging subset of vascular anomalies, particularly in the oral and maxillofacial region where they involve critical anatomical structures. Optimal management necessitates a combination of interventional and surgical modalities.*
Methods: *We present a case series of four patients — ranging in age from 6 to 25 years — treated at the Department of Oral and Maxillofacial Surgery, Mahatma Gandhi Dental College and Hospital, Jaipur, India. All patients harboured AVMs at distinct sites: the base of tongue, anterior mandible, right mandible, and left cheek. Each patient was managed with a staged protocol comprising vascular ligation of the feeding vessels followed by intralesional bleomycin sclerotherapy.*
Results: *Complete resolution of the AVM was achieved in all four patients with no evidence of recurrence on follow-up ranging from 12 to 24 months. Complications were minimal and self-limiting. No patient required repeat surgical excision or developed bleomycin-related pulmonary toxicity.*
Conclusion: *A combined strategy of vascular ligation and intralesional bleomycin sclerotherapy is a safe, effective, and replicable approach for the management of oral and maxillofacial AVMs, including high-flow lesions in anatomically complex regions.*
Keywords: *Arteriovenous malformation; bleomycin; sclerotherapy; vascular ligation; oral and maxillofacial surgery; vascular anomalies*

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I. Introduction

Vascular anomalies encompass a heterogeneous group of lesions derived from blood vessels and lymphatics. The landmark biological classification by Mulliken and Glowacki in 1982, subsequently refined by the International Society for the Study of Vascular Anomalies (ISSVA), divides these lesions into two distinct categories: vascular tumours (predominantly haemangiomas) and vascular malformations.[1,2] Vascular malformations are further subclassified as low-flow (capillary, venous, and lymphatic malformations) or high-flow (arteriovenous malformations and arteriovenous fistulae) based on their haemodynamic characteristics.[3]

Arteriovenous malformations (AVMs) represent the most dangerous subset of vascular malformations.[4] They are structural anomalies resulting from errors in vascular morphogenesis between the 4th and 6th weeks of gestation, establishing aberrant communications between arteries and veins whilst bypassing the normal capillary bed.[5] Approximately 50% of all AVMs involve the head and neck region, with the oral and maxillofacial area being particularly susceptible.[6] Their high-flow haemodynamic nature renders them prone to significant and potentially life-threatening haemorrhage, and their proximity to vital craniofacial structures makes surgical management extraordinarily challenging.[4]

AVMs are present at birth but may not become clinically apparent until later in childhood or adolescence, frequently expanding during puberty or following trauma or infection owing to hormonal influences and vascular recruitment.[5,7] Clinical manifestations range from asymptomatic pulsatile swellings and cutaneous discolouration to throbbing pain, bone destruction, and uncontrolled haemorrhage.[6]

The historical management of AVMs was primarily surgical, but isolated surgical excision is associated with high rates of haemorrhage and recurrence.[8] Modern treatment paradigms favour a multimodal approach, with preoperative endovascular embolisation combined with surgical resection considered the gold standard for high-flow lesions.[5,9] However, in resource-limited settings where interventional radiology facilities may not be readily accessible, surgical vascular ligation followed by intralesional sclerotherapy represents an equally efficacious alternative.[10,11]

Bleomycin, an antibiotic-derived cytotoxic agent from *Streptomyces verticillus*, has emerged as a leading sclerosing agent for the management of vascular malformations, particularly low-flow lesions.[12,13] Its mechanism involves the induction of DNA strand breaks and disruption of endothelial tight junctions, resulting in obliteration of vascular channels and fibrosis.[12] When administered intralesionally following vascular ligation — which converts a high-flow AVM into a low- or no-flow state — bleomycin may exert its sclerosing effect throughout the malformation nidus.[11,14]

We report a case series of four patients with oral and maxillofacial AVMs managed by this combined approach at our institution, demonstrating complete resolution in each case.

II. CASE PRESENTATIONS

Table 1. Summary of Patient Demographics and Clinical Characteristics

Case	Age/Sex	Site	Clinical Features	Investigations	Outcome
1	25-year-old female	Base of tongue	Pulsatile swelling, dysphagia, bleeding episodes	Doppler USG, MRI, CT angiography — high-flow AVM, lingual artery feeder	Complete resolution; no recurrence at 24 months
2	14-year-old male	Anterior mandible	Bony expansion, missing tooth, episodic gingival haemorrhage	OPG, CBCT, CT angiography — inferior alveolar artery feeder with osseous involvement	Complete resolution; no recurrence at 18 months
3	21-year-old male	Right mandible	Firm pulsatile swelling, skin hyperpigmentation, bruit on auscultation	Doppler USG, CT angiography — right facial and internal maxillary artery feeders	Complete resolution; no recurrence at 20 months
4	07-year-old female	Left cheek	Bluish-red soft tissue swelling, thrill on palpation, skin discolouration	Doppler USG, MRI — left facial artery feeder with soft tissue nidus	Complete resolution; no recurrence at 12 months

Case 1 — Base of Tongue AVM in a 25-Year-Old Female

A 25-year-old female presented to our department with a progressive pulsatile swelling at the base of the tongue of 9 months duration. She reported recurrent episodes of spontaneous oral haemorrhage, dysphagia to solids, and an altered voice quality. There was no history of trauma or prior treatment. On intraoral examination, a well-defined, bluish-red soft tissue mass was visible in the posterior floor of the mouth extending to the lingual aspect of the base of tongue, measuring approximately 3.5 × 2.5 cm. Palpation elicited a distinct thrill with visible pulsations. Doppler ultrasonography demonstrated a high-flow lesion with arterial waveforms, and MRI with contrast confirmed a serpiginous vascular nidus without significant deep cervical extension.[4,6]

Surgical management was planned in two stages. Under general anaesthesia with nasotracheal intubation, surgical exposure was achieved through a cervical approach. The feeding facial and lingual arteries were identified and ligated. The lesion was excised with adequate haemostasis, and specimens were submitted for histopathological examination. Hemostasis was achieved using a Gelfoam® pack. A No. 14 drain was placed and secured. The neck was closed in layers.

At 48 hours following ligation, two sessions of intralesional bleomycin sclerotherapy (concentration 1 mg/mL; dose 0.5 mg/kg per session, maximum 15 mg) were administered under ultrasound guidance at fortnightly intervals.[12,13] Postoperative monitoring confirmed progressive lesion regression. At 24 months follow-up, the lesion had completely resolved with full restoration of speech and swallowing function and no clinical or radiological evidence of recurrence.

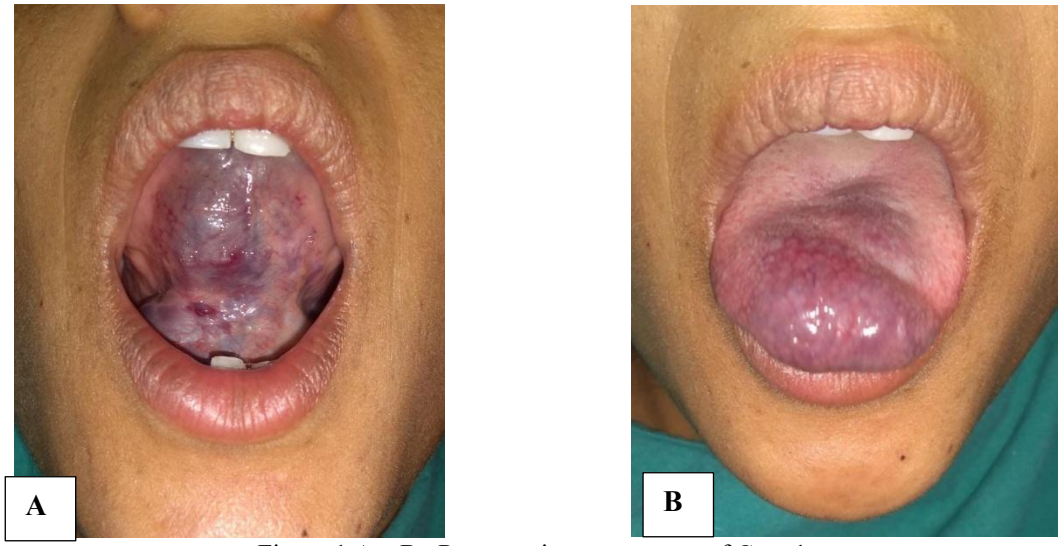

Figure 1 A – B : Preoperative appearance of Case 1.

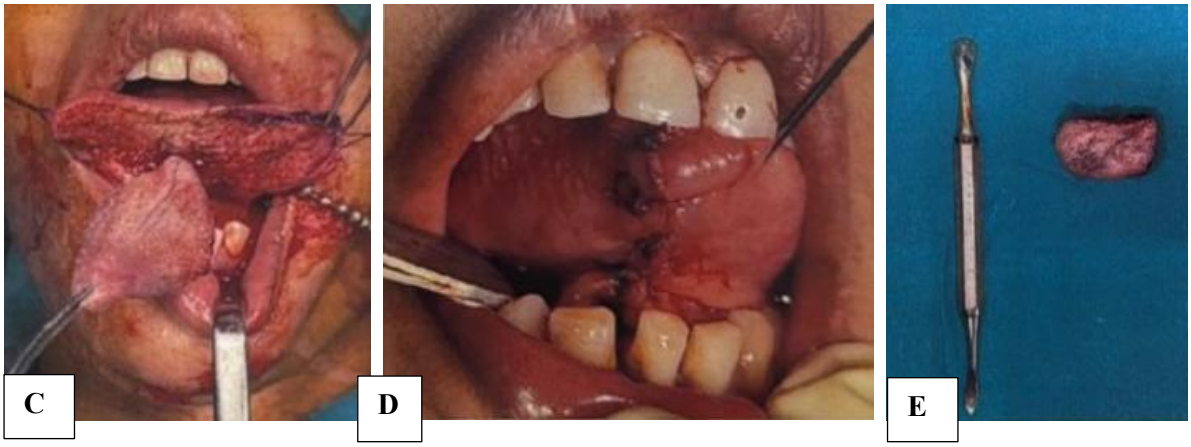

Figure 1C–E: Intraoperative photographs.

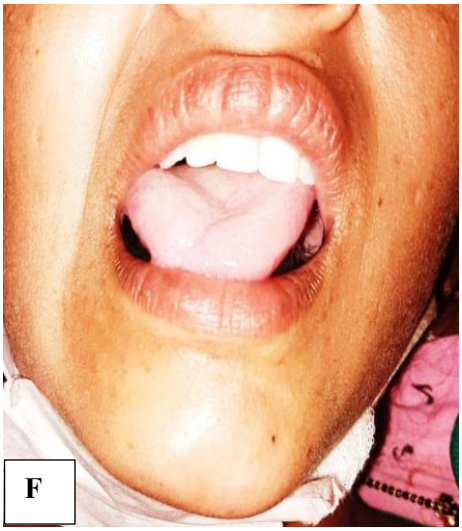

Figure 1F: Follow-up photograph.

Case 2 — Anterior Mandibular AVM in a 14-Year-Old Male

A 14-year-old male was referred with a six-month history of a swelling in the anterior mandibular region, associated with spontaneous gingival bleeding and mobility of the lower anterior teeth. Intraoral examination revealed bony expansion of the anterior mandible with mucosal hyperpigmentation and a bluish pulsatile swelling

in the attached gingiva extending from tooth 33 to 43. Orthopantomogram (OPG) and cone beam computed tomography (CBCT) demonstrated a multilocular radiolucency with poorly defined cortical margins in the anterior mandible with evidence of root resorption of 32 and 42. CT angiography identified an abnormal vascular network in the chin region supplied predominantly by the left facial artery. Venous drainage was observed through a large vein draining into the left internal jugular vein.[8,9]

Under general anaesthesia, bilateral facial vessels supplying the lesion were identified through submandibular approaches and ligated. The vascular malformation was subsequently excised, and wound closure was performed in layers. Tributaries supplying the lesion were isolated, identified, and ligated . The contralateral facial artery and vein were exposed through a pocket incision placed 2 cm below the inferior border of the mandible . Facial artery and vein of right side isolated, identified and ligated. Closure was performed using 3-0 Vicryl sutures and skin staples. Following confirmed reduction in flow on Doppler assessment, three sessions of intralesional bleomycin (0.5 mg/kg, maximum 15 mg per session) were administered directly into the lesion at three-week intervals under ultrasound and fluoroscopic guidance.[14] Progressive osseous healing and lesion involution were confirmed on serial CBCT imaging. At 18 months follow-up, there was complete resolution of the AVM with incipient bone regeneration and no further haemorrhagic episodes.

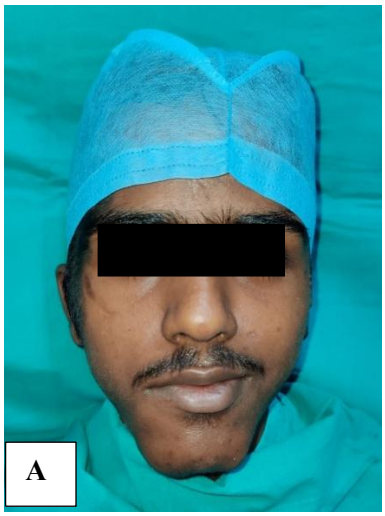

Figure 2 A : Preoperative appearance of Case 2 .

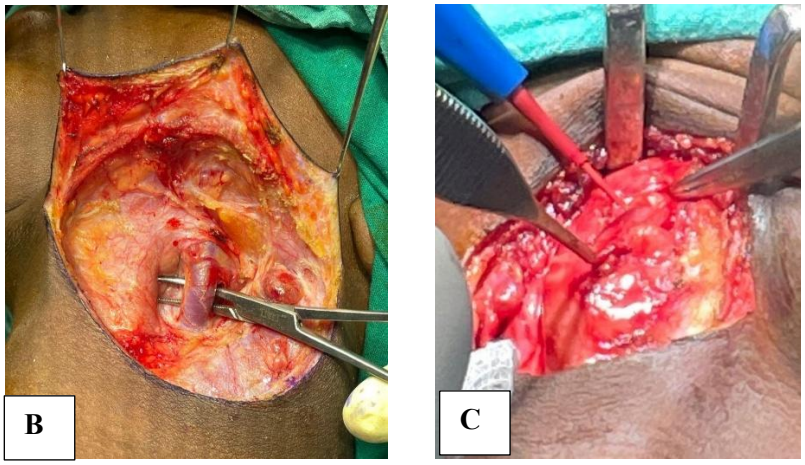

Figure 2 B–C: Intraoperative photographs.

Figure 2 D: Follow-up photograph.

Case 3 — Right Mandibular AVM in a 21-Year-Old Male

A 21-year-old male presented with **an eight-year history** of a progressively enlarging swelling over the right mandibular region .The swelling was associated with blood discharge. The swelling increased in size following episodes of bleeding and subsequently regressed partially over time. The patient reported a history of trauma following a fall from stairs approximately 10 years earlier . A lesion measuring approximately 1 × 3 cm was present in the region of teeth 41–43. Bleeding on probing was present. Doppler ultrasonography confirmed high-flow characteristics, and CT angiography demonstrated ill-defined hypodense lesions seen in subcutaneous plane along anterior and lateral aspect of right hemimandible, along posteromedial aspect of ramus of mandible, superior and right anterolateral aspect of tongue, right parapharyngeal space and bilateral tonsillar regions with phleboliths seen in these regions. Relatively prominent appearing right sided facial, bilateral lingual arteries are seen with gradual increasing nodular filling of contrast seen in venous and delayed scan.

The patient underwent right nasotracheal intubation. Bilateral facial arterial feeders and the right angular artery were surgically identified and ligated through cervical and buccal approaches. Following vascular control, the lesion demonstrated significant flow reduction and was subsequently treated with intralesional bleomycin A left submandibular incision was placed, and dissection was carried out in layers. The facial artery was identified and ligated. A utility incision was subsequently placed in the right buccal region, and layered dissection was performed to identify and secure the remaining vascular feeders. Right angular artery identified and ligated using Ligaclip® clips . Closure was performed in layers.. Vascular ligation was followed within 48 hours by intralesional bleomycin sclerotherapy administered in three sessions (fortnightly intervals; dose 0.5 mg/kg, maximum 15 mg per session) under ultrasound guidance.[11,12] The patient experienced mild post-injection inflammatory swelling that resolved within one week. At 20 months follow-up, there was no residual lesion on clinical or radiological assessment and no recurrence of haemorrhage or swelling.

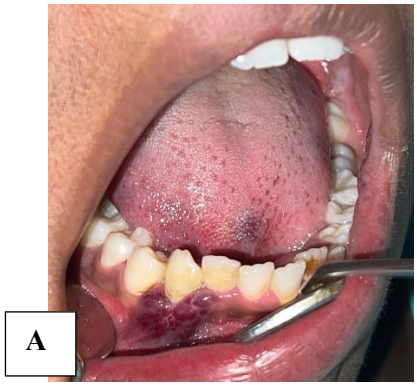

Figure 3 A: Preoperative appearance of Case 3

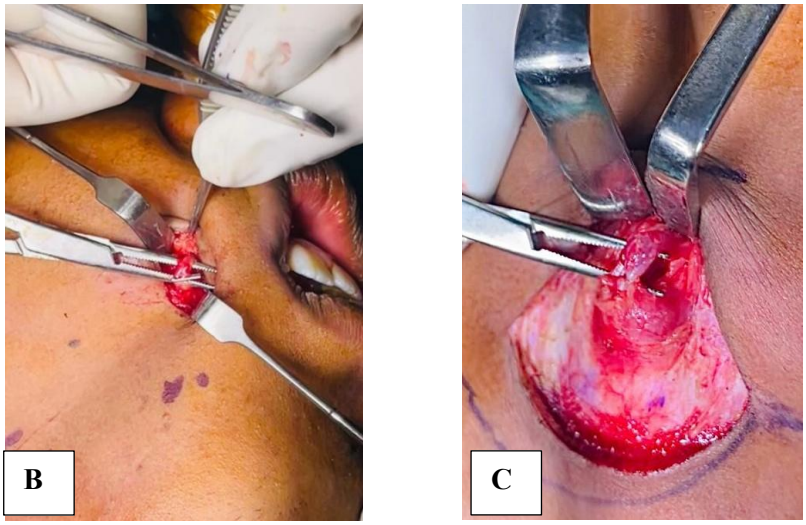

Figure 3 B– C: Intraoperative photographs.

Case 4 — Left Cheek AVM in a 7 - Year-Old Female

A 7-year-old female presented with a three-month history of pain and swelling over the left cheek. The swelling had progressively increased in size over the preceding six months and was associated with intermittent pricking pain. On examination, there was a 1 × 1 cm soft to firm swelling over the left cheek. MRI demonstrated an ill-defined mixed-intensity lesion of size 36x46x55 mm in left maxillofacial region suggestive of phlebolith like hemangioma. CTA showed left masseter muscle appears bulky with ill defined hypodense areas seen along the muscle adjacent subcutaneous plane along its anterior and lateral aspect and in left retroantral fat along the temporalis muscle. The left superficial temporal artery appears relatively prominent. Multiple hypodense foci are seen within the lesion-likely suggestive of phleboliths.[3,5]

Under ultrasound and fluoroscopic guidance, percutaneous access to the vascular malformation was obtained. Contrast-enhanced imaging confirmed intralesional placement, following which sclerosant was administered. Post-procedural imaging demonstrated satisfactory distribution within the lesion Under fluoroscopic guidance, intralesional needle placement was confirmed using contrast injection. Following aspiration, the sclerosant agent was administered with satisfactory distribution throughout the lesion. A final fluoroscopic assessment confirmed adequate intralesional dispersion, followed by saline flush. At 12 months follow-up, complete clinical resolution was confirmed with no residual swelling, discolouration, or recurrence, and cosmetic outcome was satisfactory to the family.

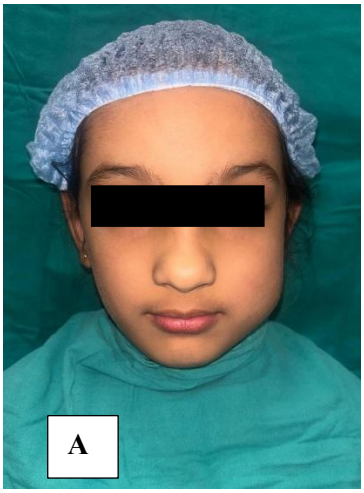

Figure 4 A: Preoperative appearance of Case 4.

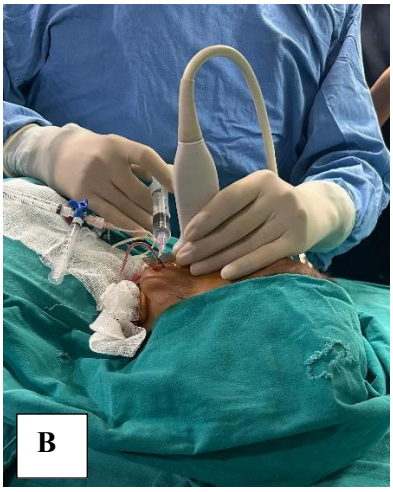

Figure 4 B : Intraoperative photographs

V. DISCUSSION

This case series illustrates the successful management of AVMs across four distinct anatomical subsites of the oral and maxillofacial region — the base of tongue, the anterior mandible, the body of the mandible, and the cheek — using a combined strategy of vascular ligation and intralesional bleomycin sclerotherapy. All four

patients achieved complete resolution without recurrence, supporting the utility of this approach as a viable alternative to the conventional gold standard of endovascular embolisation followed by surgical excision.[5,9]

The fundamental principle underpinning the success of this technique is the conversion of a high-flow AVM into a low- or no-flow state through surgical vascular ligation. High-flow haemodynamic conditions actively wash out sclerosing agents, rendering direct intralesional sclerotherapy ineffective in untreated AVMs.[11] By ligating the feeding arteries — and, where identifiable, the draining veins — the vascular nidus is effectively isolated, prolonging contact time between the bleomycin and the vascular endothelium and maximising the sclerosing effect. This principle has been described by several authors in the context of high-flow lesions of the head and neck.[11,14]

Bleomycin was chosen as the sclerosing agent given its favourable efficacy and safety profile, particularly at the intralesional doses employed in this series. The drug works through oxidative DNA strand breakage and disruption of endothelial tight junctions, producing endothelial cell death and subsequent fibrosis and obliteration of the vascular channel.[12,13] Unlike ethanol — which carries a significant complication rate of 10–15% including skin necrosis, neuropathy, and cardiovascular collapse — intralesional bleomycin at doses not exceeding 15 mg/session and a cumulative dose below 400 units is associated with minimal systemic toxicity.[3,15] Crucially, no cases of bleomycin-induced pulmonary fibrosis were encountered in this series, consistent with published safety data at these dosing levels.[13,15]

The timing of sclerotherapy following ligation is of critical importance. Vascular collaterals begin forming rapidly following feeding vessel ligation, potentially re-establishing high-flow conditions within 48–72 hours.[8] In our protocol, bleomycin injections were initiated within 48 hours of ligation, consistent with the established window employed in post-embolisation surgery protocols. This approach minimised the risk of collateralisation and ensured optimal sclerosant delivery to the devascularised nidus.

The management of the paediatric case (Case 4) merited particular attention. Paediatric AVMs may be approached conservatively in the early years, but progressive enlargement, haemorrhage, and functional compromise mandate active intervention.[7]

Intra-osseous AVMs of the mandible are well described in the literature and carry an appreciable mortality risk if not recognised preoperatively. In our case, the combination of bilateral inferior alveolar artery ligation and image-guided intralesional bleomycin — delivered under fluoroscopic guidance to ensure intra-nidal distribution — resulted in complete osseous healing and lesion obliteration.

Although endovascular embolisation is the globally accepted first-line preoperative intervention for high-flow AVMs, it requires specialised interventional radiology infrastructure and expertise that may not be universally available, particularly in developing healthcare settings.[10] Our experience supports the contention that surgical vascular ligation, when performed by a skilled maxillofacial surgeon with adequate anatomical knowledge, represents a practical and effective surrogate. Furthermore, the use of intralesional bleomycin as the sclerosing agent offers the dual advantage of availability and cost-effectiveness, making this protocol accessible in resource-limited environments.[3,10]

The limitations of this series include its retrospective, single-centre nature and small sample size. Longer-term follow-up data beyond 24 months would be valuable to confirm the durability of treatment. Future prospective, multicentre studies comparing this combined approach to standard embolisation-excision protocols would further validate its role in the management of oral and maxillofacial AVMs.

VI. CONCLUSION

Arteriovenous malformations of the oral and maxillofacial region pose significant diagnostic and therapeutic challenges. The combination of surgical vascular ligation and intralesional bleomycin sclerotherapy represents a safe, effective, and accessible multimodal treatment strategy. In all four cases presented, complete resolution was achieved with an acceptable complication profile and no recurrence on follow-up. This approach is particularly relevant in clinical settings where endovascular facilities are unavailable and merits wider consideration in the management algorithm for oral and maxillofacial AVMs.

Conflict of Interest: The authors declare no conflicts of interest.

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Ethical Approval: Institutional ethical approval was obtained. Written informed consent was obtained from all patients (or legal guardians in the case of the minor patient) for publication of clinical data. Patient identifiers have been removed to protect confidentiality.

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