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Case Report

Dentistry Section

Rare Case of Large Arteriovenous Malformations Arising after a Parotid Tumour Excision

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ABSTRACT

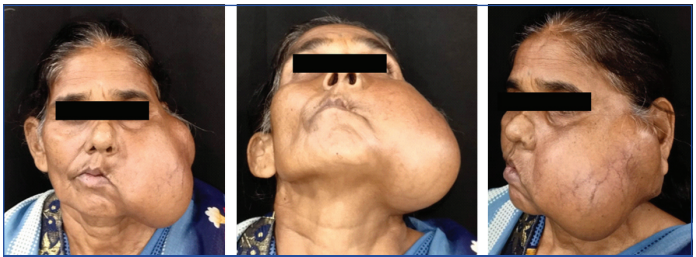
Arteriovenous Malformations (AVMs) of the head and neck are uncommon vascular anomalies characterised by high-flow shunting between arteries and veins without an intervening capillary bed. Among these, AVMs involving the parotid region are exceedingly rare and are often misdiagnosed or mistaken for more common salivary gland tumours. Their presentation may vary from painless swelling to disfiguring facial masses with the potential for life-threatening bleeding. Accurate diagnosis typically requires advanced imaging such as contrast-enhanced Computed Tomography (CT) or Digital Subtraction Angiography (DSA), as clinical signs may be subtle or absent. Management of craniofacial AVMs remains challenging due to complex vascular anatomy, the potential for recurrence, and the risks associated with surgical or endovascular interventions. A multidisciplinary approach involving interventional radiology, surgery, and supportive specialties is essential for optimal outcomes. This report highlights the diagnostic and therapeutic complexities of a large facial AVM discovered in an adult decades after parotid surgery, emphasising the importance of considering vascular anomalies in the differential diagnosis of longstanding facial swellings. The case reinforces the need for early recognition and careful planning in managing such lesions to reduce morbidity and improve prognosis. Given their rarity, each case contributes valuable insights to the evolving understanding and management of parotid and craniofacial AVMs.

Keywords: Diagnostic imaging, Embolisation, Facial asymmetry, Therapeutic, Vascular surgical procedures

CASE REPORT

A 55-year-old female presented with a complaint of a slow-growing, large craniofacial swelling that had been present for approximately 40 years. The patient had undergone partial parotidectomy, according to her history, for a similar swelling 40 years prior. She had previously sought medical advice at multiple tertiary and secondary care centres but was turned away due to the extreme size and high-flow nature of the swelling, which was deemed inoperable or too risky for intervention. Details and documentation of previous treatment were not available.

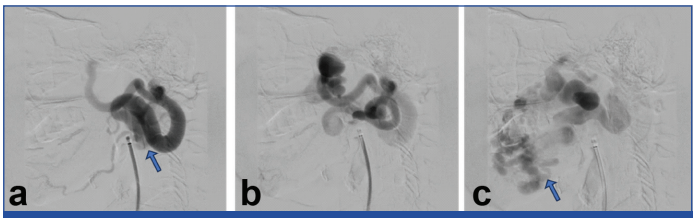
She reported symptoms of fluctuating swelling size, facial disfigurement, and occasional episodes of spontaneous intraoral bleeding. No abnormalities were detected upon systemic examination. On local examination, the patient exhibited extensive swelling on the left side of the face, involving the left mandibular, maxillary, and preauricular regions. There were signs of superficial skin thinning, raising concerns about an imminent risk of rupture [Table/Fig-1]. The extraoral swelling extended anteroposteriorly from the corner of the mouth on the left side to 1 cm posterior to the posterior border of the mandible on the left side, reaching the posterior auricular region, and superoinferiorly from the infraorbital region to 3 cm inferior to the lower border of the mandible on the left side.



[Table/Fig-1]: Initial presentation - frontal, worms view, oblique lateral view (left to right).

The swelling was non-tender, firm in consistency, and had a standing upright appearance with an absence of the Turkey Wattle sign [1]. It was non-tender, non-reducible, but compressible, with pulsations and audible bruit. There was no change in the quantity or consistency of saliva. The differential diagnosis included sarcoma, tumours arising from the parotid gland, and vascular malformations.

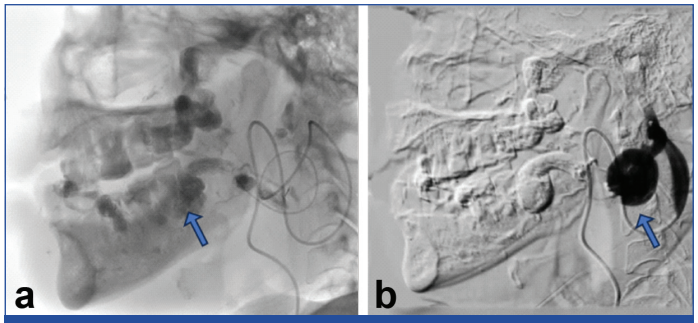
Contrast enhancement and DSA revealed a high-flow arteriovenous malformation with feeding vessels from the external carotid artery and a large venous outflow (left occipital, facial, and internal maxillary branches of the external carotid artery) [Table/Fig-2].



[Table/Fig-2]: a) Pre-operative feeder (occipital branch of external carotid artery - arrow); b) Large branches moving anteriorly; c) AVM shunting vessels over left cheek region.

The interventional radiology team evaluated the patient, and embolisation was planned to reduce vascularity and alleviate symptoms. Super selective cannulation of the occipital branch of the left common carotid artery was performed using a Progreat Microcatheter. Embolisation was completed with 2.5 mL of Inj. N-Butyl Cyanoacrylate and 20 mL of Inj. Lipidol, using a 50% mixture solution along with 255-350 microns and 355-500 microns Polyvinyl Alcohol (PVA) particles in the internal maxillary and facial artery branches. Complete embolisation of the vessels was confirmed with a postoperative angiogram [Table/Fig-3], but there was no decrease in the size of the lesion or resolution of symptoms, raising questions about the true nature of the swelling.

The patient was planned for another embolisation procedure followed by surgery, but she did not return and was lost to follow-up. Although the true nature or origin of the lesion was unclear, the patient was provisionally scheduled to undergo wide local excision of the lesion within 24 hours of another embolisation. This was to ensure embolisation of any new collateral feeding vessels and maintain adequate control of haemorrhage during resection. A locoregional flap was chosen for reconstruction. Since the patient



[Table/Fig-3]: Postoperative: a) PVA particles in shunting vessels (arrow); b) Occlusion of major feeding vessel (arrow).

came from a lower socioeconomic background, she wanted to have the surgery under a government financial support scheme and was discharged to obtain the necessary documents from her native place; however, she was lost to follow-up as she could not be contacted further.

DISCUSSION

Craniofacial AVMs are complex vascular anomalies that exhibit a characteristic high-flow pattern with an absence of a capillary network between hypertrophic feeding arteries and varicose-like dilated veins. They can progressively enlarge, leading to functional impairment, significant cosmetic disfigurement, and life-threatening complications such as rupture or uncontrolled bleeding [2].

The International Society for the Study of Vascular Anomalies (ISSVA) has classified the often-confusing vascular anomalies and provided a framework for distinguishing between haemangiomas, lymphatic malformations, tumours, etc. [3]. This case can be classified as stage 2 in the Schobinger system [4] and type V in the Nair SC et al., classification [5]. The diagnosis requires advanced imaging techniques; in this case, contrast-enhanced CT and DSA provided critical information about the vascular architecture, feeding arteries, and venous drainage. These imaging studies also guided the interventional radiology team in planning the embolisation; however, large AVMs, such as in this patient, are often accompanied by significant collateral vessel formation and multiple feeding arteries, complicating both surgical and endovascular management. The high-flow nature of the AVM also increases the risk of complications, such as the embolic material migrating into normal vasculature or incomplete occlusion leading to revascularisation [6-10].

Vascular malformations in relation to the parotid gland predominantly involve the superficial lobe, although occasional cases of deep lobe involvement have been documented. Patients typically present with a painless, gradually enlarging swelling localised to the preauricular region, without additional symptoms. A clinical feature known as the "turkey wattle sign," characterised by increased prominence or enlargement of the swelling when the head is tilted toward the affected side or during the Valsalva manoeuver, is considered pathognomonic for parotid vascular malformations [11,12]. However, this sign has been observed in only three of the 23 previously reported adult cases of parotid vascular malformation in a study by Saeed WR et al., [1]. Its absence in the present case may be attributed to the thick parotid fascia, which likely prevented the manifestation of this clinical feature.

Vascular malformations of the parotid gland are exceedingly rare, with approximately 50 cases documented in the literature. A study by Beahrs OH et al., identified vascular malformations in only 0.5% of 760 parotid tumours [13], while Byars LT et al., reported a slightly higher incidence of 0.6% among 460 cases [14]. Parotid tumours recorded over a 10-year period from 1998 to 2008 found that parotid malformations constituted 1.6% (10 out of 614 cases) in a study by Achache M et al., [15].

Yee SY et al., documented a 6.4×6.0×13.0 cm parotid and auricular AVM in a middle-aged woman successfully managed with staged embolisation and resection [16]. Gupta M et al., presented two histologically confirmed intraparotid AVMs with slow progression, treated via superficial or total parotidectomy [11]. Similarly, Nowicki TK et al., emphasised diagnostic ambiguity in thrombosed venous malformations that mimic tumours, particularly when cytology yields only blood [17]. Rajput D et al., described a recurrent buccinator muscle AVM in an adolescent treated with compartmentalisation and sclerotherapy, followed by excision [18].

Further expanding this spectrum, Rosbe KW et al., described a series of 12 patients with venous malformations isolated to the masseter muscle, characterised by facial asymmetry and pain during jaw clenching, diagnosed primarily via MRI. Sclerotherapy was used to treat 10 patients, resulting in sustained symptom relief in seven cases, demonstrating it as a safe and effective treatment option for these lesions [19]. Nagarajan K et al., described four maxillomandibular AVMs, including high-flow intraosseous variants, managed primarily with endovascular embolisation using particles or glue, depending on flow characteristics and arterial feeders [20]. Bhat VS et al., presented a post-traumatic cheek AVM treated surgically via lateral rhinotomy following embolisation, underscoring the role of trauma as a potential etiology in otherwise unexplained cases [21].

The exact mechanisms behind the formation and progression of AVMs remain a topic of ongoing debate, including factors such as congenital abnormalities, haemodynamic stress, trauma, genetic mutations, and hormonal influences. This case may represent a congenital AV malformation that was incompletely removed during parotidectomy and has recurred, or it could be a result of surgical intervention during pubertal years, during which hormonal changes might have stimulated the growth of the AVM. These lesions are considered the most persnickety and menacing type of vascular malformation for treatment. The recurrence rate can be as high as 80% after multimodal management [22].

In a large retrospective study by Liu AS et al., analysing 272 patients with extracranial AVMs, the recurrence rate after treatment was found to be extremely high—98% after embolisation alone and 81% even after resection [23]. Pedreira R et al., conducted a retrospective study at Johns Hopkins evaluating adult facial AVMs treated with embolisation followed by delayed surgical resection. Over an average follow-up of nearly three years, only two out of 11 patients had recurrences, and both had lesions larger than 6 cm. This suggests that larger size, rather than timing or extent of surgery, increases the risk of recurrence [24]. Thus, the management strategy in our case—planned delayed surgical resection following embolisation—echoes the approach validated in Pedreira's study, although unfortunately, the patient's follow-up was lost before the resection could occur.

Among vascular anomalies, AVMs are considered some of the most aggressive and damaging. Their management often requires a combination of embolisation, surgical resection, and hybrid treatment approaches. While low-flow vascular lesions can frequently be managed with conservative treatments or laser therapy, AVMs typically demand more advanced interventions due to their potential to cause severe complications [25]. A multidisciplinary approach involves the roles of radiologists, pathologists, vascular surgeons, maxillofacial surgeons, and plastic and reconstructive surgeons.

CONCLUSION(S)

AVMs of the parotid gland are exceedingly rare in adults, as documented in the global literature. Early diagnosis, advanced imaging, and the use of interventional radiology techniques, such as embolisation, play a crucial role in mitigating the risks associated with the surgical management and prognosis of these high-flow vascular anomalies. This case highlights the complex nature of diagnosing and managing craniofacial AVMs, emphasising the importance of awareness, precise documentation, and a multidisciplinary approach in diagnosis and treatment.

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