

*Case Report***A Rare Case of Submandibular and Submental Arteriovenous Malformation in a Female Patient**Kunal Shahi<sup>1</sup>, Sanjay P<sup>2</sup>, Vishal NS<sup>3</sup>, Rani Baby<sup>2</sup>

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*Received: 18/02/2014**Revised: 03/05/2014**Accepted: 12/05/2014***ABSTRACT**

Vascular malformations of the submandibular and submental region can lead to various complications. Lesions of the submandible region are rare and potentially life threatening entities. However, the clinical course of malformations involving the mandible and the submental region remains unclear and unpredictable. Treatment may be surgical or non-surgical.

**Keywords:** Submandibular, submental malformation, arteriovenous, female.

**INTRODUCTION**

Arteriovenous malformations (AVMs) are high-flow lesions of intercommunicating veins and arteries formed by abnormal vessel morphogenesis. [1]

Most of the AVMs discussed in previous review articles are congenital lesions that became clinically apparent later in life, whereas acquired AVMs are rare and usually form post traumatically. [1]

Both types of AVMs may produce symptoms of pain, swelling, or discoloration, whereas some AVMs present solely as a mass lesion. The 2 types differ in that congenital AVMs often have multiple communications between arteries and veins, whereas acquired lesions may be solitary arteriovenous communications. [1]

The arteriovenous malformation is an uncommon lesion that is considered to be part of the spectrum of hemangiomatous anomalies. Specifically, it represents a mass of intercommunicating, variously sized arteries and veins. [2]

The AVM is classified as congenital or acquired (traumatic). The congenital type represents vascular development arrested at the retiform state; the acquired type results from penetrating wounds or blunt trauma. [2]

Lesions of the mandible are rare and potentially life threatening entities that can present as innocuous episodes of gingival bleeding, slow-growing expansile masses, or severe haemorrhage. A biopsy or even a simple tooth extraction can cause a catastrophic bleeding that may even lead to death.

However, the clinical course of malformations involving the mandible and the maxilla remains unclear and unpredictable. [3]

Vascular malformations are frequently seen in the skin, but rarely affect the visceral organs or bones; approximately 51% occur in the head and neck, with a male female ratio of 1 to 1.5. [4]

Treatment may be surgical or non-surgical. The latter includes intravascular embolization with coil and/or sclerosing solutions. Surgical resection is reserved for lesions that are extensive and/or refractory to endovascular therapy. Cure is defined as the complete eradication of disease or permanent resolution of symptoms with complete devascularisation. [4]

Although literature discussing the complications of these treatments is limited, complications that have been described include reperfusion bleeding, ischemic ulceration and necrosis, infection, numbness of the inferior alveolar nerve, and temporary facial nerve paralysis. [5]

Most cases of extracranial AVMs in the literature are cited in female patients with very few cases presenting in males. Furthermore, only 8 cases of submandibular AVMs have been cited in the literature, with all cases presenting in females. [5]

Here in, we report the case of a 28-year-old woman with an AVM of the submandibular and submental region.

## CASE REPORT

A 28 year old female patient came with complaints of swelling in the left submandibular and submental region since 10 years. The swelling was initially small in size and started to increase in size from past one year.

The swelling measures approximately 5 x 2cm in size (Figure 1). The swelling is soft in consistency, does not change in size on swallowing or valsalva maneuver (Figure 2).

On auscultation bruit was heard on the swelling. There was no local rise of temperature in the swelling. The swelling increased in size during early morning and the patient complains of difficulty in speech in early morning due to increase in size of swelling which pushed the tongue upwards and to the opposite side. The swelling later subsides as day progresses.

The patient had no other complaints. The patient was advised ultrasonography of the submandibular region.

Ultrasound showed increased soft tissue echogenicity in submental and submandibular region with dilated and tortuous vessels seen bilaterally. (Figure 3 and 4)



Figure.1



Figure: 2

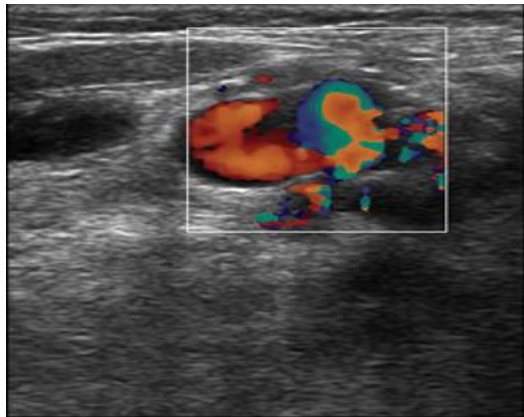


Figure: 3

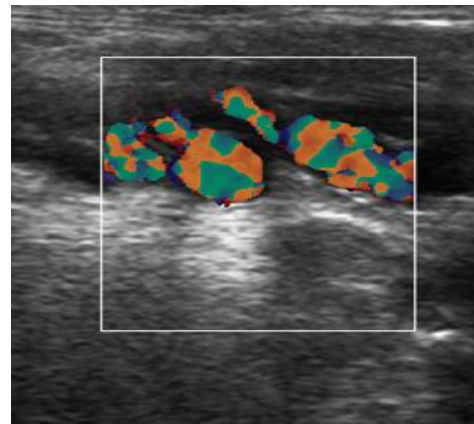


Figure: 4

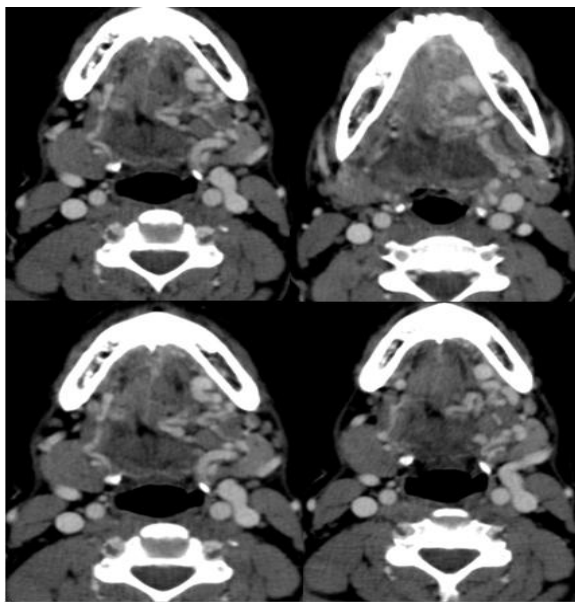


Figure: 5

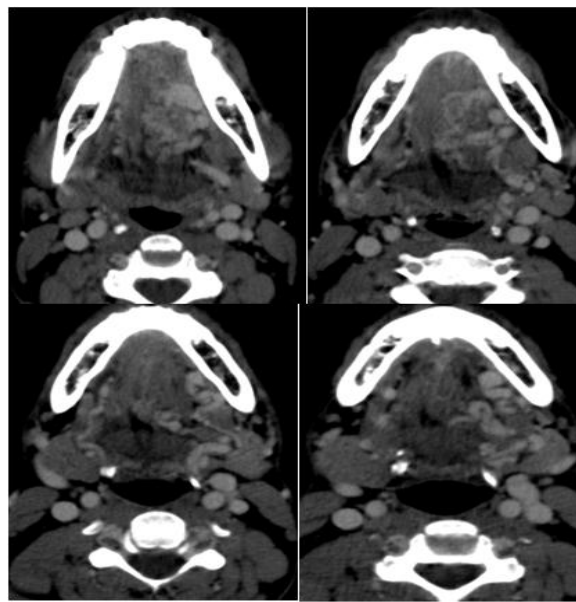


Figure: 6

Arterial phase of CT scan of the neck region showing tuft of tortuous vascular channels (L>R) in submandibular & submental regions bilaterally (Figure 5)

Venous phase of CT scan of the neck region shows the lesion draining into bilateral internal jugular veins (Figure 6).

CT neck plain and contrast was performed which showed tuft of tortuous vascular channels (L>R) in submandibular & submental regions bilaterally (Figure 5 and 6).

The lesion measured about 54 X 32mm on left side.

The vascular lesion was fed by branches arising from bilateral external carotid arteries just after their origin, the lesions were draining into bilateral internal jugular veins. CT features suggested arterio-venous malformation.

## DISCUSSION

The first case of a submandibular AVM was noted in the literature in 1985 and described in a 57-year-old female patient. [2] Since that time, very few cases of extraosseous submandibular AVMs have been reported. [3]

AVMs are extremely rare entities that can be life threatening if left untreated. [4]

Although CT, MRI and MRA may localize the arterio-venous shunt lesion, super selective arteriography remains an essential tool for diagnosis and planning of treatment. [4]

AVMs usually present with non-specific symptoms including bruit, dental loosening, swelling of soft tissues, change in skin and mucosal colour and dysesthesia of the lower lip or chin.

Management of AVMs is usually complex and requires a multidisciplinary team for successful outcome. Observation may be used as a temporary measure in special situations, such as extreme age, pregnancy or refusal of therapy. No spontaneous regressions have been documented. On the other hand, it has been reported that the volume of the lesion may gradually increase. [4]

Arterial ligation was used in the past as a purely symptomatic treatment or before surgery. At present, it is well known that ligation of external carotid artery should not be performed, firstly as many anastomoses promote the rapid appearance of a collateral circulation, and secondly because future embolization would be impossible. [4]

At present, superselective angiographic embolization is considered first line treatment, alone or in combination with surgical approach to reduce intraoperative bleeding. Occlusion of the lesion is obtained using movable balloon, coils or liquid glue. Endovascular therapy as definitive treatment for the AVMs of the mandible has been reported to have a success rate of 70%. [4]

However, serious complications after embolization (e.g., occlusion of pulmonary or cerebral vessels) or recurrence of AVMs should be considered. In some cases, the procedure has to be carried out in several

sessions such as previous history of arterial ligation or high flow characteristics of the lesion or microshunts, which are invisible during hyperselective catheterization and therefore inaccessible to treatment. [4]

Vascular malformations of the head and neck can be classified into low-flow and high-flow lesions with low flow lesions including hemangiomas and capillary and venous malformations. High-flow lesions such as AVMs are rarer and may not become clinically apparent until later in childhood, with the majority located in the head and neck region, particularly in intracranial locations. Extracranial lesions often appear to have osseous involvement, most notably in the mandibular and maxillary bones. Lesions in the submandibular triangle that do not involve the bone are uncommon and appear to have a glandular or nodal origin. [5]

Our patient appears to be a rare reported case of an extraosseous and extraglandular submandibular AVM in a female patient. Furthermore, based on her CT findings, which demonstrated multiple arteriovenous communications within the lesion and her lack of a traumatic history, this AVM is most likely a congenital lesion. Presentation of a congenital lesion at 28 years of age is also very unusual because most lesions present by late childhood.

The female predominance of reported submandibular lesions has not yet been explored. Most of the documented cases have been in middle-aged females, suggesting that hormonal changes may affect endothelial cells and vascular formation. In one study, 33% of female patients were noted to have exacerbation of symptoms with their menstrual cycles and with pregnancy. This hormonal influence may help explain why the AVMs seen in these patients presented late and with no trauma history. [5]

Therefore, additional research is needed to explain the novel etiological factors involved in the development of submandibular AVMs and their predilection to affect women. [5]

In our case, surgical excision of AVM involving soft tissues in the submandibular space was achieved without complications.

## CONCLUSION

In conclusion, considering the present case and literature data, we can define the following general principles of treatment.

Arterial ligation should not be performed, even in emergency settings. Super selective arterial embolization is the treatment of choice and can be repeated in the case of relapse.

In AVMs of the mandible, surgery should be reserved only for cases that are refractory to endovascular therapy and/or for therapeutic complications that are not otherwise treatable (bone fractures and/or necrosis).

In AVMs of the soft tissue, surgery could be used if there are no significant side effects.

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