

Arteriovenous hemangioma of left submandibular region: A case report and review of literature

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Abstract:

Arteriovenous malformations (AVMs) of the jaws are relatively rare, with fewer than 200 cases reported in the literature. Fifty percent of all intraosseous AVMs occur in the maxillofacial region and are extremely infrequent in the mandible. Arteriovenous hemangiomas of submandibular region are extremely rare conditions that can result in disastrous complications, if handled carelessly. Various diagnostic aids like MRI, CT scan and Angiography are used for the diagnosis of arteriovenous malformation.

Only six cases of extra osseous submandibular AVMs have been reported earlier in the literature. Here we represent a case of male patient aged 21 years diagnosed with Arteovenous Hemangioma of Submandibular region. This case report highlights the importance of correct diagnosis of Arteovenous hemangioma.

Keywords: AVM, MRI, CT scan.

Introduction

An arteriovenous malformation (AVM) may be defined as a condition in which arteries communicate with veins through channels other than the normal capillary network.¹ This includes angiomatous malformations, arteriovenous aneurysms, arteriovenous shunts, hemangiomas (cavernous, capillary, sclerosing and cirroid) and arteriovenous fistulas.² The common unifying clinical element is severe bleeding which can result from this type of lesion. Clinical and histopathologic differences exclude the aneurysmal bone cyst from this group. AVM's may be either congenital or acquired. The acquired malformations are derived from injury (penetrating wounds), spontaneous rupture of an aneurysm or ligation en masse of an artery and vein. Congenital AVMs are thought to result from failure of the common embryologic analogue to differentiate into normal capillary networks. An evaluation of all vascular malformations, including simple capillary hemangiomas indicates that more than 50 percent occur in the head and

neck region, even though this anatomic location accounts for only 14 percent of the body area.³ Large vascular malformations with altered venous shunting are considered to be rare lesions. Gomez's view of AVMs from 1955 to 1965 at the Mayo Clinic revealed that the vast majority of them were congenital.

Case Report

Patient aged 22 years reported with swellings below chin on left side since 2 years. Swelling was gradually increasing in size. Patient gives history of deciduous tooth extraction from left lower jaw one year back and the post extraction period was uneventful. There was no history of pain, paresthesia, blood discharge or local trauma. There was no change in size during meals and remained asymptomatic other than the obvious facial asymmetry due to the evident swelling. On general examination, patient's gait and posture were normal, well-built and nourished and vital signs were within normal limits. On inspection, a solitary swelling of approximately 3 x 2 cm size noticed on left submandibular region. On palpation, the swelling was non-tender, bimanually not palpable, uniformly firm, non-compressible, non-reducible, and non-pulsatile in nature. There was no local rise in temperature. Intraorally no relevant findings noted. A provisional diagnosis of submandibular lymphadenopathy was made. Differential diagnosis included submandibular sialadenitis, sialocele and benign salivary gland tumors of submandibular gland. Panoramic radiograph was within normal limits except for the mesioangular impacted 38. Occlusal radiograph also revealed no pathologies. Color doppler USG of the submandibular region revealed increased vascularity in the left submandibular region compared to the contralateral side. Bilateral submandibular and parotid glands were normal.

Enlarged level Ib lymph nodes were also noticed bilaterally. CECT of mandible shows a heterogeneously enhancing soft tissue density lesion noted on inner margin of left angle of mandible measuring 1.6 X 2.7 X 2 cm. Lesion shows vascular channel within it. Adjacent cortex of mandible show irregularity and thickening. Bilateral Parotid and submandibular glands were normal. A few subcentimetric nodes noted at bilateral level Ib and right level II and III. These CT finding indicated vascular malformation. MRI showed a 3 x 2.1 x 1.8 cm lobulated T1, T2 heterogeneously hyperintense lesion showing few flow voids and moderate to good enhancement with prominent feeders from lingual artery on left side noted in the submandibular space with mass effect on left digastric and mylohyoid muscles. Adjacent mandible shows hypertrophic changes. MRI confirmed the diagnosis of Arteriovenous Hemangioma of left submandibular region with internal AV shunting. Patient was referred to Sree Chitra Tirunal Institute of Medical Sciences & Technology, Thiruvananthapuram for further management.

Discussion

Majority of the swellings in the submandibular region are either lymph node swellings or cases of chronic sialadenitis. An Arteriovenous hemangioma is a benign vascular abnormality involving complex grid of interconnecting arterial and venous structures.¹ Mulliken and Glowacki in 1982 proposed a simplified classification system that divided the vascular anomalies into haemangioma and the vascular malformations based on their endothelial cell characteristics and clinical behaviour.² Arteriovenous hemangiomas can be divided into two types: one occurring in deep locations, and associated with

varying degrees of arteriovenous shunting, the other type superficial, occurring in the dermis with no significant shunting. The deep form occurs mostly in young persons and is regarded by some as an arteriovenous malformation (Enzinger FM and Weiss SW 1995).³ It is thought to be due to partial persistence of fetal capillary bed, causing abnormal connections between the arteries and veins. These lesions are commonly seen in the head, neck and the lower extremities. Hemangiomas are found to occur more frequently in females. It has been observed that the left side is more frequently involved (Batsakis JG, 1986), a finding supported by the case in study.⁴ CT and magnetic resonance imaging are useful techniques for diagnosis of hemangioma. CT angiogram act as a gold standard for diagnosis of Arteriovenous hemangioma. In our case, left side of submandibular region is involved. In addition to submandibular hemangioma, the differential diagnosis of a vascular mass in the submandibular area includes intramuscular hemangioma, lymphangioma, and angiosarcoma.⁵⁻¹⁰ Intramuscular hemangiomas are very rare; only 2 cases have been reported, both in the mylohyoid muscle. The treatment of choice for intramuscular hemangiomas is surgical excision. Compared with intramuscular hemangiomas, lymphangiomas are more common in the head and neck, but they are diffuse in nature and rarely arise from salivary gland tissue. Angiosarcomas of the submandibular gland are malignant tumors that arise from endothelial cells, and they require surgical excision along with postoperative radiotherapy. Only a very few cases of sub mandibular arteriovenous hemangioma have been reported till date.¹¹⁻¹² Surgical excision is the treatment of choice for submandibular hemangiomas. Other treatment options described in

the literature include laser ablation, cryosurgery, sclerosing injections, and vascular ligation.

Conclusion

Hemangiomas and vascular malformations are endothelial lesions that can present with a number of serious medical problems. Knowledge of the differentiating clinical features and characteristic imaging findings of these lesions is essential for providing appropriate monitoring. This case report shows that correct diagnosis plays an important role in the management of arteriovenous hemangioma. If arteriovenous hemangioma of submandibular region is not diagnosed at right time, it may become fatal.

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Legend Figure



Figure 1: Swellings below chin on left side.



Figure 2: Solitary Swelling of approximately 3x2 cm size on left submandibular region.

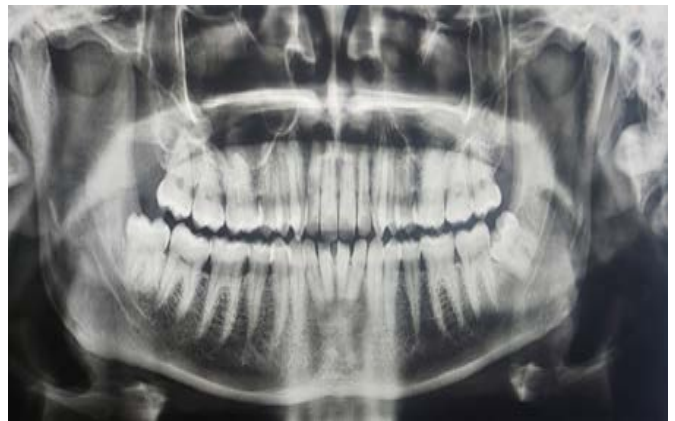


Figure 3: panoramic radiograph shows mesioangular impacted 38.



Figure 4: CECT of mandible shows a heterogeneously enhancing soft tissue density lesion noted on inner

margin of left angle of mandible measuring 1.6x2.7x2cm. Lesion shows vascular channel within it. Adjacent cortex of mandible show irregularity and thickening.

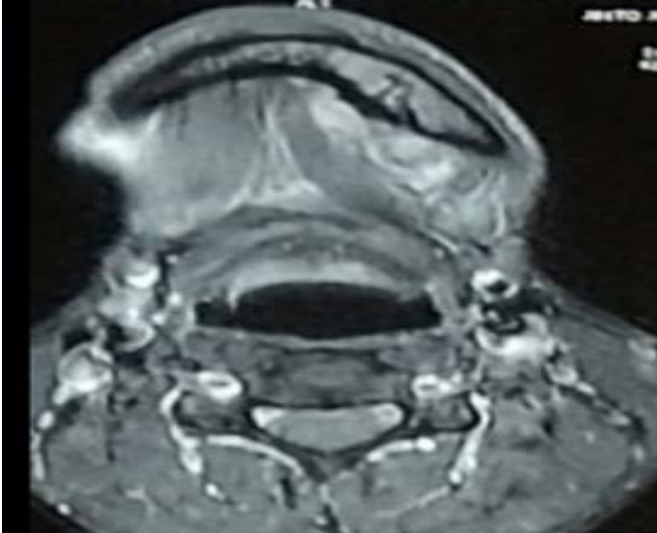


Figure 5: MRI showed 3x2x1.8 lobulated T1, T2 heterogeneously hyper intense lesion showing few flow voids and moderate to good enhancement with prominent feeders from lingual artery on left side noted in submandibular space with mass effect on left digastric and mylohyoid muscles.