Arteriovenous malformation within left masseter muscle with arterial feeders from branches of left external carotid artery and venous drainage into retromandibular vein: A rare case report

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Abstract

Arteriovenous malformation (AVM) of the head and neck region is one of the rare vascular anomalies. It is a lethal benign disease, which is persistent and progressive in nature. With the increase in interest and recent advances in the diagnosis, management, and molecular characterization of these lesions have improved the treatment strategies. Because of its wide variability in its presentation, difficulty in diagnosing, and managing such malformation is a challenging task. We report an unusual case of AVM within left masseter muscle with arterial feeders from branches of left external carotid artery and venous drainage into retromandibular vein. The present case was of importance to diagnose at an early stage where the condition was asymptomatic and its association with high morbidity and mortality rate. This paper reviews the importance of early diagnosis and the complications associated with AVM of the head and neck region.

Keywords

Arteriovenous malformation, external carotid artery, retromandibular vein

Introduction

Anomaly of angiovascular or lymphovascular structures is grouped as “vascular malformation.” These are usually present at birth and constitutes around 1% and many of which not presenting for treatment. Arteriovenous malformation (AVM) is characterized by vascular abnormality that occurs where arterial vasculature connects with the venous vasculature. In the case of trauma as an etiology, the AVM usually involves a single vessel. In the congenital form, AVM involves multiple vessels. Vascular malformations are usually present at birth. These grow commensurably with the patients age, usually do not show any clinically significant features until later in childhood. AVMs have the same frequency of involvement in both sexes. AVMs may progress acutely due to activating stimuli like trauma, pregnancy, puberty, infection, or even iatrogenic trauma (biopsy, proximal feeder ligation, or subtotal excision). Complex cervicofacial malformations may be more suited to a combination of preoperative embolization, resection, and sclerotherapy, and if needed followed by ingenious reconstruction using skin grafts, local tissue flaps, or free flaps.

We report an unusual case of AVM within left masseter muscle.

Case Report

A 23-year-old male patient came with a chief complaint of swelling over the left cheek region since 6 months and desires to get it treated. Swelling was gradual in onset, started as small swelling and gradually reached the present size. Swelling subsides on its own and appear on exposure to the cold environment. Clinical examination revealed a diffuse swelling on the left lower and middle third of the face measuring approximately 5 cm × 6 cm in dimension anteroposteriorly and superoinferiorly. It extends from the ala tragal line superiorly to the lower border of the mandible inferiorly. Anteriorly, it extends from 2 cm away from the corner of the mouth and posteriorly till the posterior border.
of ramus. Skin over the swelling was stretched, and there was no change in the color. Swelling was soft to firm in consistency and non-tender on palpation [Figures 1 and 2]. Intraoral examination revealed no significant findings other than a generalized grayish black pigmentation over the attached gingival [Figure 3].

A provisional Diagnosis of Intra masseteric lipoma was given. AVM and Hemangioma of left cheek were considered in the differential diagnosis. Investigations included complete hemogram, orthopantomograph, ultrasound, computed tomography, and magnetic resonance imaging.

Complete hemogram showed a normal blood count. Orthopantomograph revealed no abnormalities. Ultrasound revealed a heterogenous echotexture area and on Doppler ultrasound color flow was noted suggestive of vascular malformation. Computed tomography scan of mandible with contrast showed a heterogenous enhancing soft tissue lesion in the left submandibular, peri mandibular, and lower buccal region [Figure 4]. The lesion as a whole measures 4.5 cm × 1.6 cm with multiple dilated vascular channels within and adjacent to the above-described lesions. The left submandibular salivary gland is in close proximity to the lesion.

Magnetic resonance imaging revealed heterogenous signal intensity lesion noted in the left masseter muscle with multiple dilated vascular channels within muscle. On angiogram, there was prominent ascending pharyngeal artery with multiple tortuous dilated arterial feeder lateral to left submandibular gland and enlarging the left masseter muscle. Few small arterial feeder are seen arising from left facial artery posterior to neck of the mandible. Features are suggestive of AVM within left masseter muscle with arterial feeders from branches of left external carotid artery and venous drainage into retromandibular vein [Figure 5].

A final diagnosis was given as AVM within left masseter muscle with arterial feeders from branches of left external carotid artery and venous drainage into retromandibular vein. Patient was referred to a plastic surgeon for further treatment. The patient did not take any treatment, and he was reviewed after 6 months, there was no change in the size of the lesion was observed.

Discussion

An AVM of the head and neck region can be defined as an abnormal fistulous connection that forms between the feeding arteries and respective draining veins, without an intervening capillary bed in the subcutaneous layer. Various terminologies

Figure 1: Frontal view

Figure 2: Lateral view

Figure 3: Intraoral view

Figure 4: Contrast-computed tomography showing heterogenous enhancing soft tissue lesion in the left submandibular peri mandibular and lower buccal region
Arteriovenous malformation within the masseter muscle

Magnetic resonance imaging showing heterogenous signal intensity lesion in the left masseter muscle with multiple dilated vascular channels within muscle

are in vogue; arteriovenous aneurysm, cirsoid aneurysm, racemose aneurysm, aneurysm by anastomosis, plexiform angioma, aneurysmal varix, AV fistula, abnormal arteriovenous communication and have been known for centuries. Hemangiomas and vascular malformations are grouped as congenital aberrancies of vascular development. These may present as identifiable birthmarks of the skin and mucosa and a varying degree of underlying soft tissue abnormalities. When the vascular anomalies are considered under the global heading, these lesions commonly occur within the head and neck with an incidence of one in 22 children. AVM is a structural vascular abnormality where the arterial vasculature connects with the venous vasculature. In 1982, Mulliken and Glowacki introduced a classification based on the clinical and histological characteristics of the lesion. This classification divides vascular masses into two groups: (i) Tumors caused by endothelial cell proliferation (e.g., hemangiomas) and (ii) vascular malformations that do not show endothelial cell proliferation. In 1992, Jackson et al. reclassified vascular anomalies as (i) hemangiomas, (ii) vascular malformations, and (iii) lymphovenous malformations. Vascular malformations were further subdivided into two groups, based on the vascular flow characteristics, comprising (i) low-flow lesions (capillary, lymphatic or venous malformations), and (ii) high-flow lesions (AVMs).

Among the series of cases reported by various authors, the largest series of AVMs was reported by Kohout et al. It was reported that out of 81 cases in the head and neck region, majority of the cases were located in the cheek (31%), the second most common site being ears (16%) followed by the forehead, upper lip, the mandible, the neck, the scalp, and the maxilla. The present case was localized to the cheek region, which was in accordance with the case series of Kohout et al. Vascular malformations are usually present at birth, and they commensurably grow with patient’s age. These lesions usually non-significant clinically until later in childhood. Both the males and females are affected equally by these lesions. AVMs may progress acutely due to activating stimuli like trauma, pregnancy, puberty, infection, or even iatrogenic trauma (biopsy, proximal feeder ligation, or subtotal excision).

Vascular malformations are characterized by features which never involutes, unlike hemangiomas. This principle difference between the hemangioma and vascular malformation makes the diagnosis challenging and thereby further treatment and prognosis are influenced by this.

In the present case, patient noticed the swelling 6 months back which is asymptomatic and gradually increased to the present size. Ultrasound revealed heterogeneous echotexture area noted and on Doppler ultrasound color flow was noted suggestive of vascular malformation. Magnetic resonance imaging revealed heterogenous signal intensity lesion noted in the left masseter muscle with multiple dilated vascular channels within muscle. On angiogram, there was prominent ascending pharyngeal artery with multiple tortuous multiple arterial dilated tortuous arterial feeder lateral to left submandibular gland and enlarging the left masseter muscle, suggestive of AVM within left masseter muscle with arterial feeders from branches of left external carotid artery and venous drainage into retromandibular vein. These findings confirmed the diagnosis.

The most common presenting event of AVMs is hemorrhage, which occurs in about half of the patients. The rupture rate of AVM is between 2%/y and 4%/y. After the initial hemorrhage, the risk of rehemorrhage is 6%/y for the first year. The risk then returns to baseline. The risk of serious morbidity or mortality after hemorrhage is 30% to 35%, and the mortality rate after the first, second, and third hemorrhagic events is 10%, 15%, and 20%, respectively. Other common presentations include seizure (25%), headaches (20%), and focal symptoms (15%). In the present case, there were no such complications for 2 years after the diagnosis, and patient was reviewed every 6 months.

We conclude that AVM can be considered under differential diagnosis of the long-standing asymptomatic swelling in the facial region, and early diagnosis with prompt management can reduce the facial disfigurement and complications.

References

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