Scholars Journal of Medical Case Reports **ISSN 2347-6559 (Online)**
Sch J Med Case Rep 2017; 5(10):655-660 **ISSN 2347-9507 (Print)**

(An International Publisher for Academic and Scientific Resources)

Cavernous Hemangioma of Masseter in a Kid- A case report with review of literature.

Dr. Praveena Raman, BDS, MDS
Senior lecturer, Department of Oral Medicine and Radiology, Sathyabama Dental College and Hospital, Chennai, Tamil Nadu, India

***Corresponding author**
Dr. Praveena Raman

Article History
Received: 13.10.2017
Accepted: 19.10.2017
Published:30.10.2017

DOI:
10.36347/sjmcr.2017.v05i10.016

Abstract: Hemangiomas are benign vascular hamartomas and abnormal development of embryonic vasculature is the suspected cause. They are the most common benign tumours of the infancy and occur in about 10% of infants. Hemangiomas can be present at birth, or shortly after birth and grow rapidly during the first year of life, with slowing of growth in the next 5 year and involution by 10-15yr of age. The larger forms of infantile cavernous hemangiomas are visible displeasingly, frequently have an accelerated growth, and may cause significant functional disturbances and existential complications. Vascular lesions of the face are not very common. These lesions are clinically & histopathologically differentiated from other soft tissue lesions. The hemangiomas, have considerable importance as sometimes if they are intraosseous & risk of severe bleeding on attempting for extractions. So knowledge of these is important for appropriate clinical management. Here we report a case of cavernous hemangioma in a 11 year old male child.

Keywords: Vascular lesions, Hemangioma, Hamartoma, Masseter muscle, Cavernous

INTRODUCTION

Hemangiomas are innocuous vascular lesions comprising of heterogeneous group of lesions that share similar histological features [1]. The terminology hemangioma was first coined by Sznejder *et al* in 1973 & was called "Haemorraghic Hemangioma [2]. Hemangiomas are benign proliferation of dilated blood vessels & capillaries [1-3]. They are the most common soft tissue tumours of head & neck in children, increases in size till puberty or adulthood & then regresses [3-6]. But few lesions can persist & are seen in older individuals. These lesions are commonly located on skin, lips, deeper tissues (intramucosal & intramuscular) & in bones (maxilla or mandible) [1, 4, 9-11]. Females are predominantly affected than males [1-4]. Mullikan & Glovack in 1982 proposed a classification to classify the vasoformative errors into hemangiomas & vascular malformations [1-3]. Hemangiomas are further classified based on size of vascular spaces into capillary, cavernous, mixed & sclerosing. Based on vessel type it is classified into arterial and arteriovenous [1-4]. Here we report a case of Cavernous Hemangioma in an 11 year old male child.

CASE DESCRIPTION

A 11 year old male patient came to the department of Oral medicine and Radiology, with chief complaint of forwardly placed upper front tooth since 2 years and a swelling at his left middle half of face since birth and associated restricted mouth opening since birth. Patient was apparently healthy 2 yrs back after which patient started noticing mal-alignment of his teeth. Not associated with masticatory and speech difficulties. Patient gives a history of a small, reddish blue coloured swelling over the left mid and lower part of the face which was slowly increasing in size with intermittent pain as age advances. Earlier, doctors had suggested about the involution of the swelling as the age advances and patient was diagnosed as having congenital cavernous hemangioma over the left side of face and was taking homeopathic medications from 10months of age which is prescribed for pain relieval and for the swelling over the face to subside. Swelling is associated with restricted mouth opening since birth which is slowly improving on homeopathic medications. No history of trauma to face.

On extraoral examination, facial asymmetry evident on inspection (Fig 1). A single diffuse hemispherical shaped swelling measuring about 5cm×6cm in size is evident at left middle and lower part of face approximately extending medially 1cm away from left commissure of the lip, laterally 1cm away from the angle of the mandible, superiorly 3cm below the lower eye lid and inferiorly involving and extending

1cm below the left lower border of mandible. Colour and skin over the swelling appears normal. No visible pulsations**.** On palpation, the inspectory findings are confirmed. The swelling is firm in consistency, tender, not warm, fluctuant and compressible but not reducible. Translucency negative. Skin over the swelling is pinchable. No bruits or murmurs heard in auscultation. Tender on palpating left masseter muscle. On intraoral examination a single lobulated hemispherical swelling was noted on the left side maxillary buccal mucosa measuring about 2x3cm in size with normal mucosal appearance. Evidence of three small palpable nodules within the swelling on palpation, measuring about 1cm, 0.5cm and 0.3cm in size. The swelling was firm in consistency, non-tender, no warmth, freely movable, compressible but not reducible with no secondary changes. Tender on palpating left pterygomandibular raphae region.

On correlating the patient's history and clinical examination, a provisional diagnosis of Congenital Vascular malformation was made with differential diagnosis as Hamartoma- Hemangioma or Lymphangioma. Haematological investigations were apparently normal. On performing, Ultrasound examination [Fig 2 (a,b)], a heterogenous mass lesion was evident exhibiting cystic or anechoic spaces on a background of diffuse heterogeneity with bulkiness in the entire left masseter with minimal increased vascularity, which was in correlation with a benign low flow vascular malformation. On performing Magnetic Resonance Angiography [Fig 3 (a,b,c)], left side peri-maxillary and peri-mandibular soft tissue hyperintense lesion was evident measuring 6.9×3.7cm with few small cystic components and specks of phleboliths, with lesion extending to the retromaxillary and pterygoid region. Mild scalloping of left posterolateral wall of left maxillary sinus was also noted. Incisional biopsy (Fig 4) revealed congested fibro collagenous tisuue admixed with clusters of mature adipocytes, muscle tissue and fibroblast, features suggesting of hamartoma.

On correlating all the above findings, a final diagnosis of Cavernous hemangioma involving the left Masseter muscle was made. Surgical management was done under general anesthesia, preauricular skin incision was made, dissecting lateral to the parotid gland, and skin flaps were raised. Within the masseter the bulging mass, was evident. The branches of the facial nerve were preserved. The external carotid artery was looped and proximal vascular control was achieved; small feeding vessels were individually ligated and blood loss during the procedure was minimal. The mass was completely removed with a margin of normal surrounding muscle to prevent recurrence. Primary closure was done. Postsurgically patient was prescribed with antibiotics and analgesics for 5 days. There was mild postoperative facial edema, which subsided within twenty days with no evidence of pain and significant cosmetic problem. Patient is kept under regular follow up.

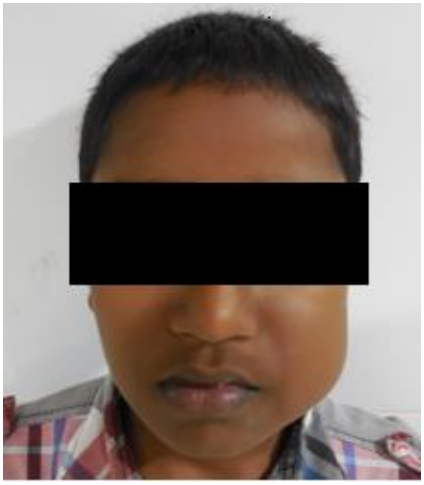
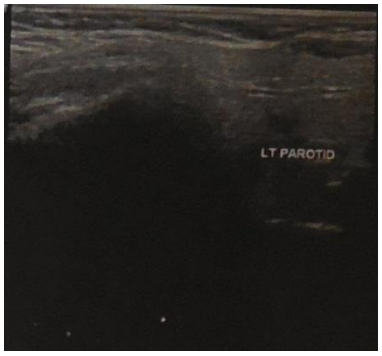
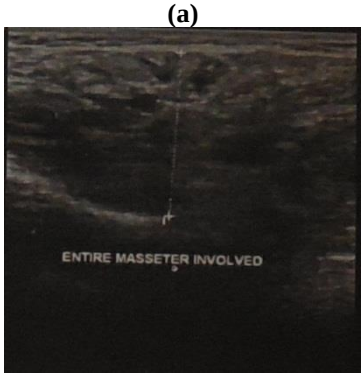


Fig-1- Extraoral: Profile

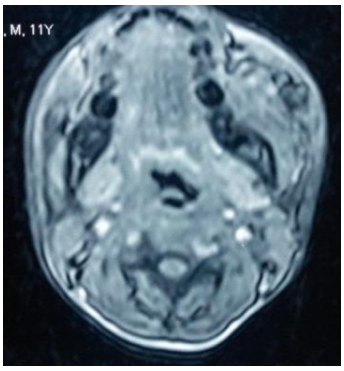


(a)

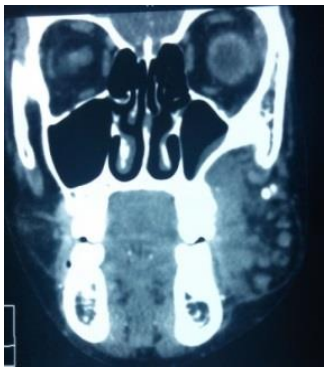


(b)

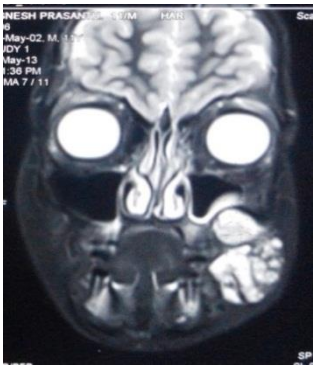
Fig-2: **(a, b) USG: Cystic or anechoic spaces on a background of diffuse heterogeneity with bulkiness in the entire left masseter with minimal increased vascularity.**



(a) ***Axial view***



(b) Coronal view



(c) Coronal view

Fig-3: **(a,b,c)- Magnetic resonance angiography: Left perimaxillary and erimandibular soft tissue hyperintensity with few small cystic component and specks of phleboliths extending into the retromaxillary and pterygoid regions.**

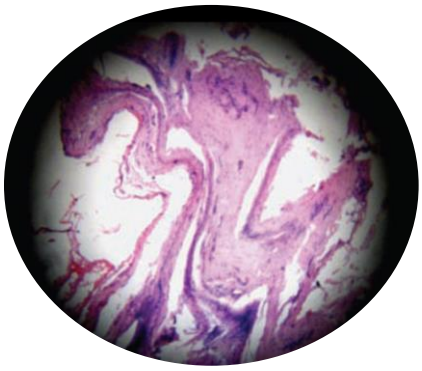


Fig-4: **Histopathology picture**

DISCUSSION

Hemangiomas are the most common tumours of infancy [12]. The incidence in newborns is 1-3% and this increases to 10% by the age of 1year [13]. 70% of hemangiomas initially appear in the first several weeks of life. Rapid growth during the neonatal period is the historical hallmark of hemangiomas [18]. It is known that hemangiomas of infancy occur more frequently among female infants (male/female ratio 1:3) [19] and are most commonly located on the head and neck (<60% of cases) [18]. The exact etiology is not known but few authors have hypothesized that angiogenesis plays a vital role [1]. Cytokines like basic fibroblast growth factor (bFGF) & vascular endothelial growth factor (VEGF) are acknowledging inducing the process of angiogenesis [1, 2]. The earliest sign of a hemangioma is blanching of the involved area. This may be followed by fine telangiectasias and then a red crimson macule. If the hemangioma is located in the subcutaneous tissue, the overlying skin may be completely normal. Most hemangiomas reach a maximum size of 0.5-0.5cm, but they can range from the size of a pinhead to greater than 20cm in diameter [13, 16]. Hemangiomas are composed of proliferating plump of endothelial cells. Early in proliferation, the cells are in disarray, but, with time, they form vascular spaces, channels and replete with blood cells [14]. When lesional vascular channels are considerably enlarged, the term cavernous hemangiomas have traditionally been applied. This differs from capillary hemangioma as a well circumscribed, larger and is usually deeper in submucosal tissues. Sluggish blood flow may result in organized or dystrophically calcified thrombi within dilated vessels. The vessels may be arranged in a haphazard or a somewhat lobular pattern and there may be areas with fibrosis of the background stroma [14, 15]. Hemangioma can also occur at extra cutaneous sites including liver, gastrointestinal tract, larynx, central nervous system, pancreas, gall bladder, thymus, spleen, lymph nodes, lung, urinary bladder and adrenal glands.

Finn *et al*. [13] in a large series found that 60% of hemangiomas occurred on the head and the neck, 25% on the trunk and 15% on the extremities. Whereas 80% of patients have a single hemangioma, others have multifocal ones [17]. In our case the patient had only a single hemangioma. With respect to the gender, Alparslan Dilsiz *et al.* Krishna Kripal et al and Nadeem Jeddy et al found that females are more commonly affected than males, but in our case the patient is a young male of 11 yrs of age [26].

Takahashi et al hypothesized that during the third trimester of fetal development, immature endothelial cells coexist with immature pericytes which

maintain their proliferative capacity for a limited period during post-natal life [20]. Yoshikawa F, et al reported a rare case of pedunculated cavernous hemangioma of the oral mucosa. In our case the hemangioma was presented with multiple phleboliths. Triona McNamara et al reported a series of 5 facial cavernous hemangiomas in pediatric patients illustrating their diverse clinical presentations and their dental perspective in identifying the location and extent [22].

Tarkan Califianeller et al reported a rare case of intramuscular hemangioma involving the temporalis muscle and emphasized that radiological methods are generally insufficiency for the correct diagnosis of intramuscular hemangiomas [23]. Kocer U, Acikgoz A suggested that hemangiomas commonly occur at head & neck region and the most common intraoral sites are gingiva followed by lip, tongue and palate [26].

Alparslan Dilsiz *et al.* [2], Krishna Kripal *et al.* Nadeem Jeddy *et al.* in their study on hemangiomas noted that clinically these lesions were soft, sessile or pedunculated, smooth or lobulated, painless, purple or deep dark blue in color with size varying from small size to a larger lesion. Our observations were not similar to the findings noted by above authors [26].

Small and uncomplicated hemangiomas in children can be managed conservatively with observation and follow- up alone. Complicated lesions and those involving eyelids, ears, tongue or lip require active management. Superficial ulcerated lesions can be managed with laser treatment. If the lesion is less than 2.5cm diameter, sclerotherapy, intralesional steroids or Bleomycin injections are recommended. Systemic treatment is needed for a lesion larger than 2.5cm and also for complications such as cardiac failure and coagulopathy [24]. Corticosteroids and interferon form the first line of treatment. Vincristine and Bleomycin are considered for problematic hemangiomas in infants, which fail to respond to steroids. Surgery is indicated in small, well localized lesions of the eyelid, lip and neck or other parts of the body proliferating and residual vessels from hemangiomas. The flash lamp-pumped pulsed dye laser has become the most widely employed laser for selective ablation of vascular tissue in childhood [24].

CONCLUSION

The identification of hemangioma is challenge as these lesions clinically resembles other entities like the vascular malformation, pyogenic granuloma, epulis, varicosities & oral squamous cell carcinoma. Extensive work up has to be done to arrive at the accurate diagnosis taking into account the differential diagnosis and the latest technology in diagnosis of the lesions. For clinical practitioners who has limited knowledge about the diagnosis of these lesions, it is a uphill task & it involves lot of risk during the treatments like the extractions & periodontal surgery as it results in profuse bleeding creating an emergency situation. In conclusion, the best approach in management of hemangioma should be individualized according to the age of patient, location, size of the lesion, and presence of complications. Once the decision to treat hemangioma is made, the main issues are the choice of the most appropriate time and method of treatment. Early steroid therapy is associated with better results [25]. The surgical plan should be individualized according to the patient's age, symptoms and the existence of cosmetic, functional or neurological deficits, the depth of invasion and the vascular structure of the tumour [23]. In depth analysis, clinical growth pattern and the histological type has to be kept in mind to yield targeted therapeutic treatment & abbreviate social embarrassment to the individual.

REFERENCES

1. Vipin Bharti & Jagmohan. Capillary hemangioma of palatal mucosa. J Indian Society of Periodontol. 2012. jul-sep: 16(3):475-478.
2. Dilsiz, Alparslan, Tugba Aydin, and Nesrin Gursan. "Capillary hemangioma as a rare benign tumor of the oral cavity: a case report." Cases journal 2.1 (2009): 8622.
3. Krishna Kripal, Senthil Rajan, Beena Ropak, Ipsita Jayanti. Cavernous Hemangioma of the tongue. Case reports in dentistry volume.2013.
4. Nadeem Jeddy, Jeva Pardha Duraiyan, Shivkumar Palanivelu, Ananthalaksmi Ramamoorthy. A giant compound hemangioma of lower lip. Journal of Oral & Maxillofacial Pathology. 2012;16(3):438-440.
5. Thomas R, Ramalingama SJ, Samuela J, Kumara KD. Cavernous hemangioma of the uvula–A Case Report. Sri Ramachandra Journal of Medicine. 2009 Jan; 1:36-7.
6. Sznajder N, Dominguez FV, Carraro, JJ, Lis G. Hemorrhagic hemangiomas of gingiva: report of case. J Periodontol.1973; 44:579- 582.
7. Enzinger FM, Weiss SW: Soft tissue tumor. 3rd edition. Mosby; 1995:581-586.
8. Silverman RA: Hemangiomas & vascular malformation. Pediatr Clin North Am.1991, 38:811-834.
9. Kocer U, Ozdemir R, Tiftikcioglu YO, Karaaslan O: Soft tissue hemangiomas formation within a previously excised intraosseous hemangioma site. J Craniofac Surg; 2004,15:82-83.
10. Acikgoz A, Sakallioglu U, Ozdamar S, Uysal A rare benign tumors of oral cavity capillary hemangiomas of palatal mucosa: a case report. Int J Paediatr Dent 2000,10:161-165.
11. Yoon RK, Chussid S, Sinnarajah N. Characteristics of a pediatric patient with a capillary hemangioma of the palatal mucosa: a case report. Pediatric dentistry. 2007 May 1;29(3):239-42.
12. Edgerton MT, The treatment of hemangiomas with special references to the role of steroid therapy. Ann Surg.1976,183;517-532.

13. Finn MC, Glowacki J, Mulliken JB. Congenital vascular lesions; clinical application of a new classification. J Pediatr Srg,1983,18;894
14. Taksande AM, Vilhekar KY. Cavernous hemangioma of the buccal mucosa in child. Journal of Chinese Clinical Medicine. 2008 Feb;3(2).
15. Fetch JF, Weiss SW. Observations concerning the pathogenesis of epitheliod hemangioma (angiolymphoid hyperplasia). Mod Pathol. 1991, 4:449-455
16. Boon LM, Enjolras O, Mulliken JB. Congenital hemangioma: evidence of accelerated involution. J. Pediatric. 1996, 128(3):329-335
17. Cohen RC, Myers NA. Diagnosis and management of hepatic hemangiomas in childhood. J Pediatr Surg, 1986,21;6
18. Antaya RJ. Infantile Hemangioma. In: JP Ortone. eMedicine Dermatology. St. Peterburg, eMedicine Corp, 2001/eMedicine Dermatology (journal serial online) 2002.
19. Frieden IJ, Eichenfield LF, Esterly NB. Guidelines of care for hemangiomas of infancy. American academy of Dermatology guidelines/ outcomes committee. J Am Academy Dermatol, 1997, 37; 631-637
20. Takahashi K, Mulliken JB, Kozakewich HP. Cellular markers that distinguish the phases of hemangioma during infancy and childhood. J Clini Invest, 1994, 93:2357-2364
21. Yoshikawa F, Nishiyama T, Sakuda M. Pedunculated cavernous hemangioma. A case report. Oral Surg Oral Med Oral Path, 1993, 75(6):688-689
22. Triona McNamara, Carroll-Ann Trotman, Antonia Hewson, Matthew Walshe. Facial developmental vascular anomalies. Special Care in Dentistry.1995;15(3).
23. Calişaneller T, Ozdemir O, Yildirim E, Kiyici H, Altinoers N. Cavernous hemangioma of temporalis muscle: report of a case and review of the literature. Turkish neurosurgery. 2007;17(1):33-6.
24. Taksande AM, Vilhekar KY. Cavernous hemangioma of the buccal mucosa in child. Journal of Chinese Clinical Medicine. 2008 Feb;3(2).
25. Snezhana Murgova, Chavdar Balabanov. Conservative treatment of cavernous hemangioma on eyelids. Journal of IMAB - Annual Proceeding (Scientific Papers) 2007, vol. 13, book
26. Abidullah M, Hussain J, Karpe T, Gaddikeri K. Cavernous hemangioma of buccal mucosa: A rare case report with a reappraisal of differential diagnosis and review of literature. Br Biomed Bull. 2014;2:638-43.