Neonatal vascular malformation of parietal bone

ABSTRACT

Vascular malformations (VaMs) are congenital lesions and are present at birth. They grow commensurately with age. These lesions often affect the soft tissues with intraosseous involvement of the scalp being rare. Here, we discuss a case of intraosseous VaM in a 2-month-old infant which was involving the parietal bone.

Keywords: Scalp, vascular anomaly, venous malformation

INTRODUCTION

Vascular malformations (VaMs) are fallacies of vascular morphogenesis that are present at birth and grow with the child.^[1] A great majority of congenital VaMs are recognizable in childhood with incidence being 1.5%. Both the sexes are equally effected.^[2] They can cause significant morbidity and even mortality in both children and adults.^[3] In the head and neck, VaMs of the skull bones are exceedingly rare with the mandible and maxilla being most common bones that are affected.^[4] Here, we report a case of a VaM of scalp involving the parietal bone in a 2-month-old male infant.

CASE REPORT

A 2-month-old male infant was referred to the department of oral and maxillofacial surgery from the Department of Neurosurgery with a slow-growing swelling of right parietal bone since birth. History revealed that the child is the first child born to parents of nonsanguineous marriage by vaginal delivery in hospital. The swelling was noticed 1 week after birth and grown to the present size for last 2 months. Otherwise, the child is healthy and active without any comorbidity. Clinical evaluation revealed a diffuse, firm, nontender, nonpulsatile swelling of right parietal bone around 6 cm in diameter [Figure 1]. Auscultation of the area was not significant. The scalp tissue over the lesion was healthy. There was a solitary

Access this article online	
	Quick Response Code
Website:	
www.njms.in	
DOI:	
10.4103/njms.NJMS_253_20	E1-20-26-245

firm and nontender lymph node palpable in the right occipital region. Corroborating the history and clinical findings our clinical impression was expansile lesion of the skull. All the hematological and biochemical parameters were within normal limits. Ultrasound and color Doppler revealed heterogeneously hypoechoic area with multiple anechoic sinusoidal spaces with mild vascularization and venous abnormality. Magnetic resonance imaging (MRI) and computed tomography (CT) were carried out under general anesthesia. MRI ruled out any intracranial involvement. CT scan revealed bicortical expansion of parietal bone filled with irregular sinusoidal spaces [Figure 2]. Expansion of the outer cortex was more than inner cortex. The surface of the lesion was pitted like a golf ball with multiple communicating channels. There was no evidence of increased intracranial pressure. With these radiological findings, we could deduce the provisional diagnosis as a low flow VaM. The child was operated under general

N. K. SAHOO, SUDARSHAN BHAT¹, SANJAY KUMAR²

Department of Oral and Maxillofacial Surgery, Army Dental Centre (R and R), ¹Army Dental Centre (R and R), New Delhi, ²Department of Neurosurgery, Armed Forces Medical College, Pune, Maharashtra, India

Address for correspondence: Dr. Sudarshan Bhat, Army Dental Centre (R and R), New Delhi - 110 010, India. E-mail: sudarshan_in@yahoo.com

Received: 24 November 2020, Revised: 02 February 2021, Accepted: 31 July 2021, Published: 13 December 2021

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Sahoo NK, Bhat S, Kumar S. Neonatal vascular malformation of parietal bone. Natl J Maxillofac Surg 2021;12:431-4.

© 2021 National Journal of Maxillofacial Surgery | Published by Wolters Kluwer - Medknow



Figure 1: Swelling on parietal region (right side)



Figure 3: Surgical exposure of the lesion

anesthesia for excision of the lesion. Marking was done around the lesion and 1:100,000 adrenaline solution was injected along the incision line. A full thickness pericranial flap was raised to expose the lesion [Figure 3]. Perimeter of the lesion was marked with the sharp end of periosteal elevator. The craniectomy was completed with the help of pizzo cutting instrument, and the lesion was removed in toto. The underlying dura was intact and healthy. The blood loss was within the acceptable limits, and no blood transfusion was required. Gross examination of the specimen revealed intact outer and inner cortex. There were multiple indentations of the inner cortex. Coronal sectioning of the specimen revealed bicortical expansion filled with multiple irregular empty spaces lined with thin lining and some with fatty tissue [Figure 4]. The specimen was sent for histopathological evaluation which revealed VaM demonstrating saccular blood vessel expansions without endothelial proliferation [Figure 5]. Postoperative healing was uneventful. The child was followed up for 2 years without any recurrence [Figure 6].



Figure 2: Computed tomography scan (three-dimensional image) showing pitted golf ball appearance in the right parietal region



Figure 4: Coronal section of the specimen revealed bicortical expansion filled with multiple irregular spaces

DISCUSSION

Vascular anomalies were classified originally by Mulliken and Glowacki in 1982 based on clinical features and biologic behavior which was later accepted by the International Society for the Study of Vascular Anomalies into two basic types: (a) Hemangiomas and (b) VaMs. VaMs have been classified based on the different types of involved vessels (arterial, venous, lymphatic, and capillary) and hemodynamic characteristics (low flow and high flow).^[5]

VaMs are benign, nontumorous lesions that are always present from birth, although they may not always be visible until weeks or months. They grow commensurately with the child. The incidence is 1.5% and approximately two-thirds are predominantly venous malformations. They are evenly distributed according to sex and race. They never proliferate or involute and remain present throughout the patient's life



Figure 5: Histopathology demonstrating saccular blood vessel expansions without endothelial proliferation (H and E stain, ×100)

and tend to expand following puberty, trauma, or attempted subtotal excision.^[6,7] In our case, the VaM was present since birth and slowly increased in size and was involving the right parietal bone. VaMs can occasionally be completely intraosseous. Mandible is the most common bone involved, although maxillary, nasal, and frontal lesions have also been reported.^[8] With regard to skeletal changes, VaMs effect 35% of the cases. They may lead to secondary changes in bone size, shape, and density.^[9] Clinical evaluation in our case revealed a diffuse, firm, nontender, nonpulsatile swelling of right parietal bone with no change in the scalp tissue over the lesion. A prudent imaging plan is essential for the evaluation of these lesions and when such lesions are covered by skin the role of imaging becomes indispensible for diagnosis. Ultrasonography and MRI techniques should be the initial choice being nonradiation methods, and ultrasound plays a major role in differentiating the lesion into high- and low-flow lesions. If bone is involved, CT scan becomes inevitable as it precisely demonstrates the involved bones.^[1,10] In our case, ultrasonography and color Doppler were done which revealed no soft-tissue abnormality. MRI and CT were also carried out under general anesthesia. MRI ruled out any intracranial involvement and CT scan showed bicortical expansion of parietal bone filled with irregular sinusoidal spaces. Expansion of the outer cortex was more evident than the inner cortex, and the surface of the lesion was pitted like a golf ball with multiple communicating channels. These radiographic findings revealed that the lesion was not aggressive and was a slow flow lesion. Kaban and Mulliken also reported that most lesions that are described as "intraosseous hemangiomas" of the maxillofacial skeleton are in fact venous malformations.^[1,10] Venous malformations are the most common lesions of head-and-neck region after hemangiomas.^[10] The choice of treatment depends on the depth, extent, and anatomical location of the lesion. Gresham



Figure 6: Two year follow-up without recurrence

T. Richter and Adva B. Friedman in their study have suggested that almost all VaMs eventually require intervention.^[11] There are various methods available for treatment of VaMs, namely, laser therapy, excision, sclerotherapy, and combinations.^[5] Our patient was operated under general anesthesia for excision of the lesion. The underlying dura was intact and healthy and very minimal bleeding was encountered during the procedure. A confirmatory diagnosis of VaM was revealed following histopathological evaluation. Postoperatively, the patient is being followed up for 2 years without any recurrence.

CONCLUSION

Hemangiomas and VaMs are the most commonly occurring vascular anomalies of the head-and-neck region. Clinical presentation of VaMs being extremely variable imaging plays a major role in the diagnosis of such lesions, chiefly when they are intraosseous lesions. Treatment of vascular anomalies is complex. Correct diagnosis is imperative for appropriate treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Vargel I, Cil BE, Kiratli P, Akinci D, Erk Y. Hereditary intraosseous vascular malformation of the craniofacial region: Imaging findings. Br J Radiol 2004;77:197-203.
- Syed NM. Vascular lesions of head and neck: A literature review. Indian J Dent Sci 2016;8:176-82.
- Donnelly LF, Adams DM, Bisset GS 3rd. Vascular malformations and hemangiomas: A practical approach in a multidisciplinary clinic. AJR Am J Roentgenol 2000;174:597-608.
- Nagy CZ, Cantrell S, Wu X, Arturo Solares C. Vascular malformation of the sphenoid and temporal bone: A diagnostic dilemma. Am J Otolaryngol 2019;40:334-6.
- Ethunandan M, Mellor TK. Haemangiomas and vascular malformations of the maxillofacial region – A review. Br J Oral Maxillofac Surg

2006;44:263-72.

- Redondo P. Vascular malformations (I). Concept, classification, pathogenesis and clinical features. Actas Dermosifiliogr 2007;98:141-58.
- Arneja JS, Gosain AK. Vascular malformations. Plast Reconstr Surg 2008;121:195e-206e.
- Senapathi SV, Pradeep. Vascular lesions of oral and maxillofacial region. J Med Sci Clin Res 2014;2:524-31.
- El-Sheikh MM, Zeitdoun IM, El-Massry M, Medra AM. Vascular malformations of the jaw bones. Report on nine patients. J Craniomaxillofac Surg 1993;21:9-14.
- Bhat V, Salins PC, Bhat V. Imaging spectrum of hemangioma and vascular malformations of the head and neck in children and adolescents. J Clin Imaging Sci 2014;4:31.
- 11. Richter GT, Friedman AB. Hemangiomas and vascular malformations: Current theory and management. Int J Pediatr 2012;2012:645678.