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Case Report

Successful Management of a Vascular Malformation of Oral Cavity using Sclerotherapy in a Pediatric Patient – A Case Report with Literature Review

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Abstract

Congenital vascular malformations in the oral cavity is an uncommon clinical entity encountered in children significantly affecting function, aesthetics and speech. Vascular malformations are congenital deformities of the vascular system that may affect different anatomical regions even including the oral and maxillofacial region. Children afflicted with vascular malformations involving the oral cavity/orofacial region, if left untreated can pose complications of pain, functional imbalance, and bleeding finally disturbing patient self-esteem and aesthetic appearance. Various management strategies have been described in the literature based on nature and type of the vascular malformation. Early diagnosis and timely intervention is essential to provide overall benefit to a child patient avoiding risks and complications. The purpose of the present research paper is to illustrate the occurrence of vascular malformation in a 4-year-old Indian patient which was successfully managed using conservative sclerotherapy treatment procedure.

Keywords: *Differential diagnosis; Pediatric patient; Sclerotherapy; Sodium tetradecyl sulphate; Vascular malformation.*

Introduction

Vascular malformations are congenital anomalies arising from disturbances in vasculogenesis and angiogenesis which frequently seen associated with neonatal dysmorphogenesis. Congenital vascular malformations in children usually affect about 60% the head and neck region. Pertaining to the oral cavity, vascular malformations most commonly affect the buccal mucosa, gingiva, tongue and palate area [1]. The reported clinical features of these lesions are cosmetic impairment, feeding problems, ulceration, pain, bleeding and airway obstruction [2]. The risks and complications associated with vascular malformations in children are infection, purpura, scarring and sometimes fire hazards following laser treatment. Congenital vascular malformation lesions basically categorized into two types based on flow dynamics of blood as low-flow and high-flow lesions, each exhibiting different clinical manifestations and distinct therapeutic challenges [3-5]. Although oral vascular malformations in children are rare, but are highly important when they occur due to their high-flow nature and potential for spontaneous or trauma induced haemorrhage encountered during dental procedures such as extractions or biopsy procedure [3]. As a result, it is essential that every pediatric dental specialist should have in-depth knowledge about congenital vascular malformations occurring in children and also it is warranted to establish interdisciplinary co-ordination involving multiple medical and dental specialists to arrive at cautious procedural treatment planning.

Presence of vascular malformations pose significant diagnostic and therapeutic challenges mainly in pediatric dentistry. Various theurapeutic approaches have been suggested for the management of vascular malformations in children [6-8]. Treatment options required for small and peripheral vascular lesions are conventional surgical excision, laser therapy, cryotherapy, selective embolization, sclerotherapy, and medical treatment using beta blockers or steroids [7, 8]. Among these, sclerotherapy is a safe and effective treatment for head and neck vascular malformations in children. Regular post-operative follow-up is required to minimize airway and thromboembolic complications [9-10]. Following sclerotherapy, reduction in swelling size and functional improvement can be noticed. In this research article, occurrence of congenital vascular malformation in a 4-year-old child patient has been narrated enlightening the literature pertaining to clinical manifestations, prevalence, diagnostic criteria, differential diagnosis and different treatment strategies of congenital vascular malformations in children. The current article also highlights the rarity of the condition and successful management of the case following conservative treatment approach using intralesional sclerotherapy.

Case Report

A 4-year-old girl reported to the outpatient Department of Paediatric and Preventive Dentistry with a chief complaint of a reddish-brown swelling involving her lower left cheek region since birth with no history of pain or ulceration in the area. The parents also mentioned that the lesion affected the child's appearance, impacting her mental well-being. The patient's medical history revealed that the condition had been small at birth and had continued to grow to attain the current size. On clinical examination, a 20-mm diameter sized diffuse swelling was observed in the left angle of the mouth. The swelling spread to the lips and intraorally into the buccal mucosa, extending to the left mandibular deciduous first molar region [Figs. 1(a) and 1(b)]. On palpation, the lesion felt non-tender, soft, non-pulsatile and did not blanch under pressure. No history of spontaneous pain or bleeding was elicited. Patient was subjected to ultrasonographic scan evaluation. The Color Doppler ultrasonographic scan confirmed the lesion's vascular origin and supported the diagnosis of a vascular malformation measuring around 20 x 21.6 mm. [Fig. 1(c)]

Based on these findings, a conservative treatment approach using intralesional sclerotherapy was planned for the paediatric patient, using 3% Sodium tetradecyl sulfate. During the first visit, 0.5 ml dosage of the drug was injected into the centre and 1 ml into the periphery. Clinical presentation at 1 week follow-up revealed regression in the size of the lesion. [Fig. 2(a) and 2(b)]. Additionally, four doses of the drug was injected at intervals of 1 week, where the lesion showed a size reduction and eventually resolved significantly at the follow-up of 6 weeks. Following consultation with the Department of Oral and Maxillofacial Surgery and Dermatology, further intervention was discontinued after the fifth dose. At every visit, a prescription for 100 mg of ibuprofen, a non-steroidal anti-inflammatory drug, was given to alleviate the pain at the injection site. Upon 6-month and 1year follow-up, the patient remained asymptomatic and showed no indications of recurrence [Fig. 2(d) and (e)] which was again confirmed by ultrasonography [Fig. 2(c)].

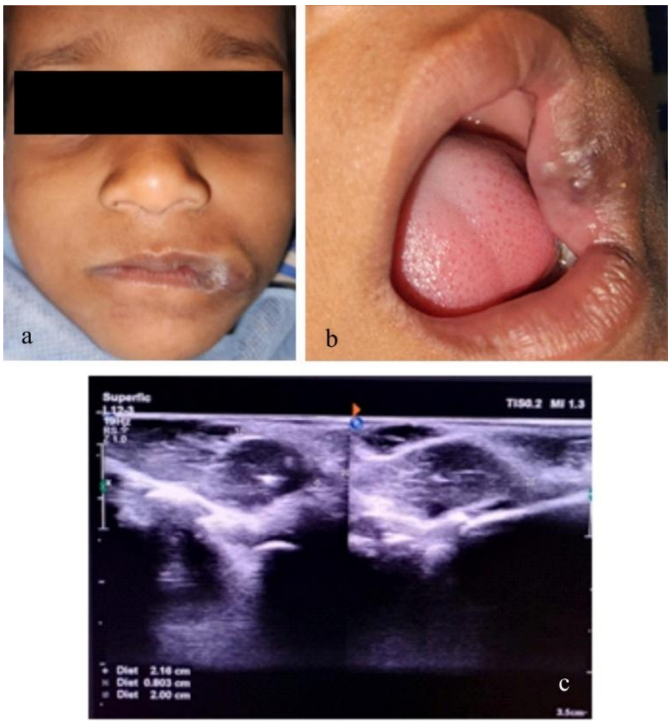

Figure 1. (a) and (b) Preoperative clinical photographs showing well defined reddish-brown swelling involving left angle of mouth, lips and buccal mucosa, (c) Color Doppler Ultrasonography presenting predominantly hyperechoic soft tissue lesion and confirming the vascular origin.

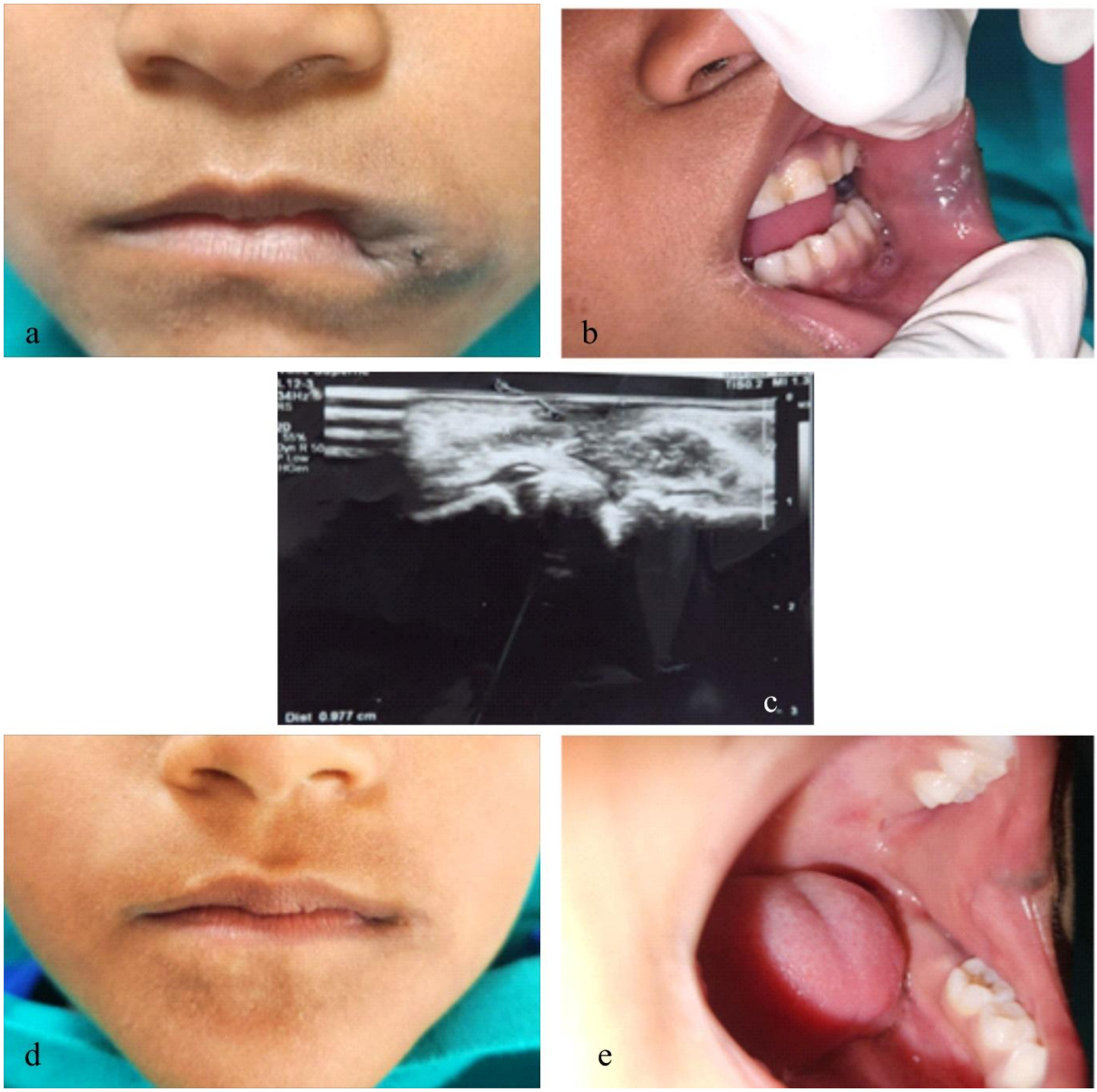

Figure 2. (a), (b) Clinical follow-up (1-week follow-up). 1 year follow-up revealed satisfactory healing with no signs of recurrence (d) and (e). Ultrasonographic scan assessment revealed regression in size of the lesion (c).

Discussion

Pediatric dentistry is a specialised branch of dentistry dealing children with different art and science right from birth through adolescence. Pediatric dentist can come across an array of soft and hard tissue lesions, tumors and developmental abnormalities as evident in the pediatric dental literature [11-14]. Vascular malformations are congenital abnormalities resulting from disturbances in the embryologic development of the vascular system including lymphatic system, venous or arterovenous system. The most common is the lymphatic malformation (58%), followed by venous (31%), and arterovenous malformation (4.5%). The mean age of diagnosis was 2.2 years and regarding gender predilection, females are more predominantly affected compared to males especially in children with arterovenous malformation [4].

Various terminologies have been explained to describe these lesions as this will greatly help in correct diagnosis and management of the lesion [15-17]. Correa et al estimated the prevalence of vascular malformation, oral hemangioma and varix in a Brazilian population using retrospective data. Oral hemangioma and oral varix were found more in the ventral surface of the tongue and oral vascular malformation were observed in the lips. Regarding treatment, oral hemangiomas were treated with sclerotherapy (54%) and vascular malformations were managed with sclerotherapy and surgery in 19% cases [4].

The International Society for the Study of Vascular anomalies (ISSVA) in 2018 classified these lesions into two types based on biological nature and endothelial activity of the lesion as vascular malformations and vascular tumors [5]. This classification is strongly recommended for proper diagnosis and to delineate appropriate treatment. Based on this standard classification, and considering history and clinical manifestations of the lesion (such as presence of lesion since birth, gradual expanding nature, exhibiting low-flow features on ultrasound diagnostic method), the condition was diagnosed as a vascular malformation in our case.

In differential diagnosis, other similar conditions should be considered that include mucoceles, benign soft tissue tumors, ranulas, hemagiomas and lymphangiomas [18-20]. Sometimes syndromic conditions like neurofibromatosis and Sturge-Weber syndrome should also be ruled out as vascular malformations can occur either isolated form or in association with syndromes [15-17]. In comparison, hemangiomas are proliferative lesions that typically appear post-natally and regress spontaneously. Whereas, vascular malformations are seen at birth, are non-proliferative and tend to persist or enlarge gradually over time. The similar features of vascular malformations were observed in the current case strongly indicating diagnosis towards a vascular malformation [8].

Oral involvement of these lesions can cause functional impairment, aesthetic concerns and increased risk during routine dental procedures [15]. Therefore, proper diagnosis and early intervention is required to avoid future complications. For diagnostic evaluation of these lesions, different diagnostic techniques/tools are employed such as Doppler ultrasound, Computed Tomograph (CT) angiography and Magnetic Resonance Imaging (MRI). Correct diagnosis of the vascular malformation type is important using these mentioned diagnostic tools to provide optimal treatment and for appropriate care of patients [8].

Treatment of congenital vascular malformation varies based on the complexity of the condition, and various malformation types [8]. Different therapeutic strategies suggested in the literature are laser treatment, cryotherapy, embolization, sclerotherapy and surgery approach [21-26]. Low-flow lesions are easily managed using conservative treatment approach. Whereas high-flow lesions are treated by embolization followed by surgical excision. In majority, non-symptomatic cases, a ‘wait and watch approach’ is followed till intervention is required [8].

In children, most of the time, conservative treatment approach is followed to avoid complications and to preserve function [21-23]. However, in children with vascular malformations interventional treatment is also suggested. Sometimes, in majority cases more than one treatment is required. Surgical treatment is usually indicated for lesions causing significant functional or aesthetic impairment. However, it is always wise to consider clinical manifestations, esthetic concerns and potential complications, before undertaking surgical approach [8, 18]. Effective management of vascular malformations of the oral cavity in children requires interdisciplinary collaborative approach among various specialists like dermatologists, vascular specialists, radiologists, pediatrician and pediatric dentists. Such multidisciplinary care is required not only patient safety but also for governing psychosocial concerns, parental anxiety and long-term quality of life. Pediatric dentists play a significant role in the multidisciplinary team, demanding expertise in preventive care, minimally invasive techniques and behaviour management that complement medical interventions.

Intra-lesion sclerotherapy has emerged as a promising alternative to surgical treatment for small, benign oral and facial vascular malformations specifically located where esthetical conservation is required [24]. Sclerotherapy can partially or almost entirely regress the lesion and is an effective treatment for symptom relief. It is a simple, less invasive, and inexpensive procedure, but should be performed with caution to avoid complications like pulmonary embolism, anaphylaxis, nerve damage, and increase of pain.[6] However, this procedure is not the definitive treatment, and proper patient selection and evaluation are crucial to prevent any adverse effects. Recently image-guided sclerotherapy is becoming the preferred treatment for low-flow vascular malformations in head and neck region [24]. For children with low-flow vascular malformations in head and neck region, sodium tetradecyl sulphate foam or ethanolamine is used for venous malformation and doxycycline for lymphatic malformations as primary scerosants [24, 25]. Following this, dexamethasone 0.2 mg/kg thrice daily is administered to decrease post-sclerotherapy swelling and single dose intravenous mannitol of 0.5 g/kg is given to minimize thromboembolic complications [3]. In addition to this, lauromacrogol foam assisted sclerotherapy guided with ultrasound has also been investigated which found effective and safe for children with lip venous malformations [23]. Laser therapy is an alternative treatment approach in some cases of venous malformations in children. Most commonly Neodymium-Doped Yattrium Aluminium Garnet laser is used which has shown a promising results in pediatric patients with inoperable venous malformations in the oral cavity [22]. However, mild to moderate scarring is the most common complication associated with laser therapy. This particular case was planned for sclerotherapy using sodium tetradecyl sulphate instead of a surgical approach, considering that the affected region was an aesthetic part and there was a lower risk of bleeding and scarring following this procedure [25]. In addition, the patient and her parents were apprehensive about undergoing any surgical treatment.

Besides above treatments, additional dental treatments including preventive dental care consisting of dietary counselling, oral hygiene maintenance, minimally invasive treatments and education are essential in these patients which can profoundly decrease complications and risks thereby improve treatment outcomes [3, 15]. Because dental care in children

associated with vascular malformations necessitates heightened vigilance. Risks of pain, poor cooperation from pediatric patients, and bleeding may worsen routine dental treatments. Considering the high risk of haemorrhage particularly in vascular-rich areas, pediatric dentist or general dental practitioners must be extremely careful during dental treatment. Inadvertent trauma to the lesion or sudden movements by the children may cause life-threatening haemorrhagic episodes [3, 15].

Conclusion

In clinical practice of pediatric dental care, pediatric dentist can come across variety of congenital, developmental malformations, deformities, cysts and tumors with each clinical condition requiring challenging diagnostic evaluation followed by appropriate treatment. Every clinical condition exhibit array of clinical manifestations, pathognomonic features demanding difficulty to arrive at correct diagnosis. Hence, thorough knowledge on occurrence of such pediatric orofacial conditions is essential among every pediatric specialist to provide holistic dental and oral health care.

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