CASE REPORT

Unilateral Lingual Hemangiomatosis

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Abstract

Hemangioma are characterized by hyperplasia of blood vessels, usually veins and capillaries, in a focal area of submucosal connective tissue.We hereby report a case of unilateral lingual hemangiomatosis in a 57 years old female who presented to us with abnormal speech and blueish patches over the tongue.

Key Words

Hemangioma, Benign Tumor, Lingual Hemangiomatosis

Introduction

Hemangioma (Greek: Haima-blood; angeion vessel, omatumor) by definition can be defined as "a benign tumor of dilated blood vessels." Hemangioma of head and neck appear a few weeks after birth and they grow rapidly. They are characterized by hyperplasia of blood vessels, usually veins and capillaries, in a focal area of submucosal connective tissue. (1,2) Hemangiomas are lesions that are not present at birth. They manifest within the first month of life, exhibit a rapid proliferative phase, and slowly involute to near complete resolution. Hemangiomas exhibit both a proliferating phase and an involuting phase, whereas vascular malformations are more stable and fail to regress.

Case Report

We report a 57 years old female who presented to us with abnormal speech and blueish patches over the tongue. Examination of the tongue revealed multiple small blueish spots over the right half of the tongue,which were soft and compressible and would fill again after compression (*Fig 1*). They were more prominent after valsalvas manouver, after crying,and placing the tongue in the dependent position. The borders were well defined and there were no ulcerations over the lesions. The lesions were not attached to the tongue muscles, With these findings she was diagnosed as multiple small unilateral hemangiomatosis of the tongue. She did not have any other hemangioma over the body at a different site. She was investigated and she had normal hemogram, liver function tests, renal function tests, thyroid profile, electrocardiogram. The color doppler of the tongue revealed multiple dilated vascular channels over the right half of the tongue

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Fig. 1 Showing Tongue with Multiple Small Blueish Spots

Discussion

Hemangiomas are the most common benign tumours of the head and neck in children, but their occurrence on the tongue is extremely rare. The tongue requires special consideration because of its susceptibility to minor trauma and consequent

bleeding and ulceration, swallowing difficulties, and breathing problem, although the major concern is cosmetic in most cases. The hemangioma appears as soft mass, smooth or lobulated, and sessile or pedunculated and may vary in size from a few millimeters to several centimeters (1,2). They are usually deep red and may blanch on the application of pressure and if large in size, it might interfere with mastication (3,4). The superficial hemangiomas are often lobulated, and blanch under finger pressure and the deeper lesions tend to be dome-shaped with normal or blue surface coloration, and they seldom blanch. A lesion with a thrill or bruit or with an

obviously warmer surface, is most likely a special vascular malformation, called arteriovenous hemangioma (arteriovenous aneurysm, A-V shunt, arteriovenous malformation), with direct flow of blood from the venous to the arterial system, bypassing the capillary beds. Cavernous hemangiomas are composed of large, irregular, deep dermal and subcutaneous blood-filled channels that impart a purplish discoloration to the overlying skin. They are typically soft, poorly defined, and readily blanch with compression, giving them a characteristic "bag of worms" feel. The lesion may expand and darken with crying, when agitated, or when placed in a dependent position. Often, a capillary component overlies a cavernous component, and it may be difficult to distinguish these components histologically. Cavernous and mixed hemangiomas demonstrate the same patterns of proliferation as those of capillary lesions. However, involution is often incomplete, depending on the location and the presence of associated arteriovenous malformations. Various syndromes that are associated with the vascular malformation include the Osler-Weber-Rendu syndrome, Sturge-Weber syndrome, and blue rubber bleb nevus Histologically, the appearance of these lesions depends on the stage of the evolution. Early lesions may be very cellular with solid nests of plump endothelial cells and little vascular lumen. Involuting lesions show increased fibrosis and hyalinization of capillary walls with luminal occlusion (5).

Management of hemangioma depends on a variety of factors, and most true hemangioma requires no intervention. However, 10-20% requires treatment because of the size, exact location, stages of growth or regeneration. Individualized therapy depends on the age of the patient, the size and the exact location of the lesion, the stage of growth or regression, and the functional compromise. In general, the treatment of small hemangiomas that do not compromise function is observation. Conservative management consists of periodic visits, parental support, and photodocumentation. The ultimate result of involution for capillary hemangiomas is far superior to primary excisional therapy. Excision can be justified under certain conditions, especially when function is compromised. The two

primary medical treatments are steroids and beta-blocker therapy (2)

There are many treatment modalities reported in the literature for head and neck hemangiomas, including wait and watch policy, for spontaneous involution, intralesional and systemic corticosteroid treatment, embolization, excision, electrolysis and thermocautery, immunomodulatory therapy with interferon alfa-2a, and laser photocoagulation. Recent interest has centered on interstitial delivery of laser energy to photocoagulate vascular lesions. Currently, sclerotherapy is employed largely because of its efficiency and ability to conserve the surrounding tissues. Among the sclerosing agents available, excellent results have been reported for sodium morrhuate, sodium sulfate tetradecyl, polidocanol and ethanolamine oleate, and hypertonic glucose solution (6). Growing hemangioma can be treated effectively by systemic drug therapy, sclerotherapy, laser therapy or combined therapy. Transcutaneous and contact applications of laser energy have been studied with the argon and Nd: YAG lasers while the 585-nm flashlamp - pulsed dye laser can be used in cutaneous and subcutaneous hemangiomas.

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