

Case Report

Intramuscular hemangioma with phleboliths of the tongue

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Abstract

Intramuscular hemangioma (IMH) is relatively rare benign tumor of vascular origin. Phleboliths are calcified thrombi found in the presence of hemangioma. The main treatment of the hemangioma is a surgical extirpation based on location, accessibility, and cosmetic considerations. We herein report a rare case of IMH with phleboliths of the tongue with clinical, imaging, and histopathological findings.

Key words: Intramuscular hemangioma, phleboliths, tongue

INTRODUCTION

Intramuscular hemangioma (IMH) is a relatively rare benign tumor of vascular origin, accounting for less than 1% of all hemangiomas.^[1] Phleboliths are calcified thrombi found within vascular channels, often in the presence of hemangioma or vascular malformation. They are most frequently found in the pelvic veins, where they are subject to stress injury during defecation.^[2] The next most common site for a phlebolith is the head and neck area.^[3] In this area, the masseter and/or buccinator muscles are the most common site. We herein report a rare case of intramuscular hemangioma with phleboliths of the tongue.

CASE REPORT

A 51 year-old man was referred for the evaluation of a swelling of the tongue. The condition had been first noticed by the patient 2 years earlier and had increased slowly in size. There had been no hemorrhage in or around the lesion. These vascular tumors exhibiting a deep red color about 1 cm in diameter were noticed on the right border, lower surface and root of the tongue [Figure 1a and b].

These tumors seemed to disappear upon pressure and to reappear when pressure was released. Computed tomography (CT) shows multiple phleboliths in the tongue [Figure 1c]. Contrast enhanced T1-weighted magnetic resonance (MR) imaging shows strongly enhancing mass with smooth borders [Figure 1d]. The diagnosis of an IMH with phleboliths was made based on these findings. IMH of the tongue were surgically excised under general anesthesia. Several hard bodies within the mass were observed [Figure 2a].

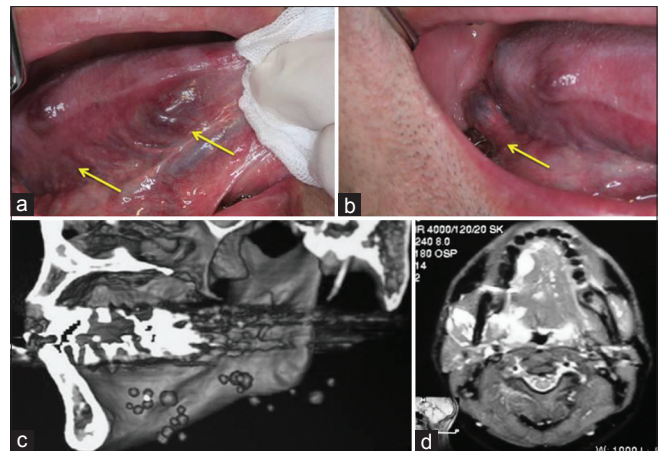


Figure 1: Hemangiomas on right border (a), lower surface and root of the tongue (b). CT showing a mass with multiple phleboliths (c). Contrast enhanced T1-weighted MR imaging shows strongly enhancing mass with smooth borders (d)

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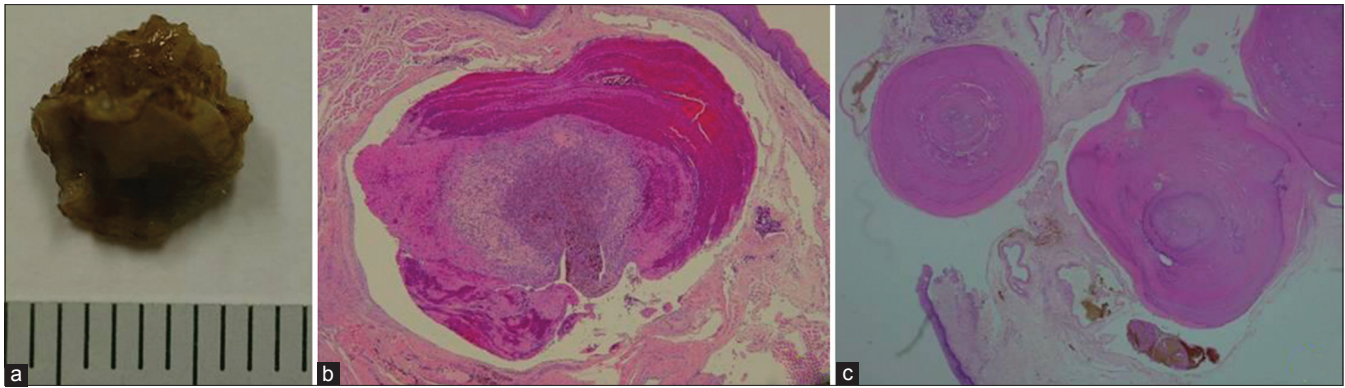


Figure 2: Phleboliths taken from the intramuscular hemangioma of the tongue (a). Microscopically, specimen demonstrates hemangioma of the tongue (HE original magnification 3200) demonstrates laminated structure of thrombus (b) and phleboliths (c)

Histopathological examination demonstrated cavernous hemangioma with thrombus [Figure 2b] and phleboliths [Figure 2c] containing dysplastic venous channels with flat endothelium and irregularly attenuated walls with deficient smooth muscle cells. One year follow up showed no evidence of recurrence.

DISCUSSION

The diagnosis of the head or neck phlebolith requires its differentiation from other calcifications that occur in the same area. Sialoliths, tonsilloliths, calcified lymph nodes, atherosclerotic plaques in the carotid artery, healed acne lesions, cysticercosis, and miliary skin osteomas all serve to muddy the diagnostic waters.^[4]

The pathogenesis of phlebolith is thought to involve thrombi produced by slowing of peripheral blood flow becoming organized and mineralized. Initially, calcification the thrombus occurs, forming the core of the phlebolith. The fibrinous component then undergoes secondary calcification and becomes attached. Repetition of this process causes enlargement of the phlebolith.^[5] In this case, long term presence of hemangioma and stagnant blood flow caused a large number of phleboliths.

Management of hemangioma varies depending on the individual case. Attention needs to be given to the tumor location, extent, growth rate, accessibility, patient's age, and esthetics.^[6] The ideal treatment is complete excision of the tumor with a surrounding margin of normal muscular tissue, due to the infiltrative

nature of the tumor. Minor feeding vessels and residual tumor are responsible for recurrence.^[1] Excessive blood loss during surgery may result in significant surgical morbidity. Complete tumor resection could be performed with less bleeding if a wide surgical field exposing the whole tumor and surrounding area was obtained.^[7] In this case, bleeding during operation was little due to wide resection of the tumor.

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