BUCCAL PHLEBOLITH: REVIEW OF THE LITERATURE AND REPORT OF TWO CASES

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INTRODUCTION

Pathological calcification is a degenerative process characterized by the deposition of calcium salts in diseased tissues\textsuperscript{1}. It is an uncommonly encountered pathology that may occur in any part of the body including the oral cavity. A variety of diseases notably tuberculosis, parasitic/worm infestation, filariasis and histoplasmosis may lead to the formation of these mineralized deposits in the affected tissues\textsuperscript{2-4}. Phlebolith or calcified thrombus also represents a distinct example of pathological calcification that occurs in venules, veins and sinusoidal vessels of cavernous haemangiomas\textsuperscript{3, 5}. They may occur singly or as multiple calcified bodies that are often small and round/circular in shape. Radiographically, they may appear as uniformly radiopaque or laminated bodies with radiopaque or radiolucent centre\textsuperscript{2-5}. X-ray diffraction studies and infra-red spectrometry have identified calcium phosphate and calcium carbonate as the principal components present in these structures\textsuperscript{6}. Histologically, these calcified bodies have a laminated configuration i.e. dark and light concentric rings corresponding to the alternating high and low mineral content present, giving them an onion skin appearance\textsuperscript{6, 7}.

Phlebolith formation is on the whole uncommon and its occurrence in the buccal soft tissues is decidedly rare. The following 2 cases are reported here because of their obscure aetiology.

ABSTRACT

Pathological calcification refers to the deposition of calcium salts in tissues affected by disease. Phlebolith formation or calcification of thrombus is one example of pathological calcification. It is however rarely encountered. Two such cases, one affecting the buccal mucosa of a 6-year-old Malay, and another in a 29-year-old Chinese, both females, are described. A collective review of 11 previously reported and current 2 cases yielded 9 female and 4 male patients whose ages ranged from 6 to 57 years (mean age, 27.9 years) and who presented with a mean onset duration of 12 years. A brief discussion on the differential diagnosis of pathological calcifications of the cheek mucosa is also included.

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and also because the case in the 6-year-old Malay female most probably represents the youngest example of such an occurrence.

CASE REPORTS

Case 1

A Malay female, age 6 years, presented with a small firm swelling located deep in the right buccal mucosal soft tissues opposite the molar and premolar teeth. She first noted this swelling about 3 years previously. It was painless, non-tender to palpation and had remained the same size since. A small cystic and haemorrhagic lesion with a firm mass in its centre was excised under local anaesthesia. The specimen was submitted for histopathological examination with a provisional diagnosis of a cystic swelling.

Ten years postoperatively, the patient, now 16 years of age, presented with a diffuse right cheek swelling that extended from the lower border of the mandible to the maxillary prominence on the same side. She also complained of occasional pain and limitation of mouth-opening. Radiographs revealed 2 small radiopacities within the soft tissues of the right cheek. The clinical impression was thrombophlebitis. These were excised under local anaesthesia and submitted for histopathological evaluation.

Case 2

A 29-year-old Chinese female presented with a complaint of a small painless nodule located below her left cheek near the ramus. She first noted this lesion 3 years ago. Clinically, it appeared attached to the underlying structures. The overlying oral mucosa was however unremarkable. Radiographs revealed a small radiopacity at the complaint site. This was excised under local anaesthesia and submitted for histopathological examination with a provisional diagnosis of choristoma.

Pathological findings

The macroscopic appearances of the first phlebolith in Case 1 and bisected specimen in Case 2 are shown in Figs. 1 and 2 respectively. Histological examination of the first specimen in Case 1 showed a round haemorrhagic, calcified mass composed of alternating concentric dark and light rings (Fig. 3). It was surrounded by bits of haemorrhagic connective tissue. A mild diffuse chronic inflammatory cell infiltrate was also present. The subsequent 2 specimens examined also presented microscopically as darkly-staining haemorrhagic laminated calcified bodies (Fig. 4).

Fig. 1 Gross appearance of first phlebolith as a haemorrhagic mass in Case 1.

Fig. 2 Gross appearance of bisected specimen in Case 2.

Fig. 3 Photomicrograph of the first phlebolith removed from the right buccal soft tissues in Case 1. The mass is darkly-staining and haemorrhagic and surrounded by bits of also haemorrhagic connective tissue (Haematoxylin & eosin stain. Original magnification x 7).
these calcified bodies remained obscure. Clinically in both cases neither a haemangioma nor phlebolith formation was initially suspected. In Case 1 the fact that a cystic swelling, haemorrhagic in nature, with clot and a firm mass in the centre was removed during operation, seems to suggest that the calcified mass at least developed in haemorrhagic tissues. Nevertheless the cause of the diffuse right cheek swelling in this case 10 years later remained unknown. It has been suggested that in instances where there is no evidence of a vascular lesion, the possibility of the phlebolith representing the residual sign of a childhood haemangioma in an adult need to be considered. The other plausible explanation relates to organization and dystrophic calcification of a haematoma following some form of injury that the patient is unaware of. The possibility that the current 2 cases represented residual childhood haemangiomas or calcified haematomas need to be considered.

Although phleboliths are established pathological entities, they are infrequently found in the soft tissues including the buccal soft tissues. A review of the English-language literature yielded 11 instances of buccal phleboliths (Table 1). A collective analysis of these previously reported and current 2 cases revealed a wide age distribution (6-57 years) with a mean age of 27.9 years, a marked female preponderance (male: female ratio, 1:2.3) and a mean onset duration of 12 years (range, 1-15 years). Most of these cases occurred in association with haemangiomas. In 2 other cases there was concurrent masseteric hypertrophy. Neither a haemangioma nor masseteric hypertrophy was observed in our cases.

In view of the obscure aetiology of the current 2 cases, pathological calcifications other than phlebolith formation were also considered in their differential diagnosis. The principal entities considered were lymph node calcification, sialolithiasis, myositis ossificans, ectopic tooth germs, miliary osteomas and calcified parasites. Calcification in lymph node tends to be large and usually occurs secondary to tuberculosis, histoplasmosis, filariasis, lymphomas and metastatic diseases. In the present 2 cases, neither lymphoid tissues nor infective organisms were found in the sections examined. Sialolith-like phleboliths may also present with a concentric laminated configuration, but unlike the latter, they are usually elongated in shape, appear...
as filling defects in sialograms and may present with symptoms of salivary obstruction. There was no histological evidence that the calcified bodies of the current cases were related to salivary tissues. Myositis ossifican of the masseter muscle may also present as calcified masses. In this condition, a linear striated morphology or zonal phenomenon is the characteristic diagnostic hallmark. This feature was however not seen in the 2 cases described here. In the other types of pathological calcifications namely ectopic tooth germs, miliary osteomas and calcified parasites, each of these has its own distinct histological presentation, and can be easily differentiated from phleboliths on microscopic grounds.

In summary, 2 cases of buccal phlebolith are described here along with a literature review and a brief discussion on the differential diagnosis of pathological calcification of the soft tissues of the cheek.

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REFERENCES


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<th>Diagnosis and Comments</th>
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<tr>
<td>Thoma et al</td>
<td>1948</td>
<td>57/M/F-</td>
<td>Slowly-growing lump in (R) cheek x 15 years</td>
<td>RO material like a mass of buckshots</td>
<td>Surgical excision</td>
<td>Haemangioma with phlebolith</td>
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<tr>
<td>Schwartz and</td>
<td>1955</td>
<td>17/M/Cew</td>
<td>Painless mass (L) cheek present since birth</td>
<td>Doubtful opacities in (L) masseter</td>
<td>Surgical excision</td>
<td>Carvenous haemangioma with phlebolith</td>
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<td>Salz</td>
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<td>Deighun and</td>
<td>1956</td>
<td>31/F/Negro</td>
<td>Gradually increasing swelling of (R) cheek x 15 years</td>
<td>Multiple, small concentrically calcified bodies 1-5mm in size lying anterior to ascending</td>
<td>Surgical excision</td>
<td>Carvenous haemangioma of buccal fat pad with phlebolithsis</td>
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<td>Barton</td>
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<td>ranus</td>
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<td>Gray</td>
<td>1957</td>
<td>32/M/-</td>
<td>Swelling (R) cheek x 16 yrs</td>
<td>Multiple shadows in soft tissue of cheek</td>
<td>Surgical excision</td>
<td>Carvenous haemangioma of cheek with phlebolith formation</td>
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<td>Quinn</td>
<td>1965</td>
<td>46/F/-</td>
<td>Several movable masses in (R) buccal tissue. Asymptomatic</td>
<td>One large calcified body and several smaller, irregular circumscribed ones</td>
<td>Surgical removal</td>
<td>Idiopathic calcified bodies/phlebolith</td>
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<tr>
<td>O’Riordan</td>
<td>1974</td>
<td>27/F/-</td>
<td>Carvenous haemangioma of left side of face and lips. Symptom-free</td>
<td>Multiple RO bodies mainly in substance of the (L) cheek</td>
<td>No treatment</td>
<td>Phlebolith</td>
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<td>Four RO bodies in soft tissues of cheek</td>
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<td>Sano et al</td>
<td>1988</td>
<td>17/M/-</td>
<td>Swelling of (L) cheek since birth, several hard, round nodules</td>
<td>Several normal calcified foci in (L) buccal region</td>
<td>UK</td>
<td>Phlebolith</td>
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<td>Zachariades</td>
<td>1991</td>
<td>8/M/-</td>
<td>(L) buccal swelling and slight spontaneous pain. H/O extirpation of a (L)</td>
<td>Multiple calcified bodies, varying in size, in the cheek</td>
<td>Surgical excision of tumour</td>
<td>Carvenous haemangioma with phlebolith</td>
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<tr>
<td>et al</td>
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<td>buccal tumour 16 years ago</td>
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<td>Present study</td>
<td>1995</td>
<td>6/F/Malay</td>
<td>Firm swelling (R) cheek. Painless x 3 years</td>
<td>NA</td>
<td>Surgical removal</td>
<td>Phlebolith. Recurrence x 10 years later. 2 other phlebolith excised</td>
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<td>29/F/Chinese</td>
<td>Nodular growth (L) buccal mucosa</td>
<td>Small radiopacity at site of lesion</td>
<td>Excisional biopsy</td>
<td>Phlebolith</td>
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**RO** - Radiopaque  **NA** - Not available  **UK** - Unknown