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

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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.

INTRODUCTION

Pathological calcification is a degenerative process characterized by the deposition of calcium salts in diseased tissues[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[2-4]. Phlebolith or calcified thrombus also represents a distinct example of pathological calcification that occurs in venules, veins and sinusoidal vessels of carvenous haemangiomas [3, 5]. They may occur singly or as mutiple 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[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[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[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

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 thrombophleboliths. These were excised under local anaesthesia and submitted for hitopathological 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).

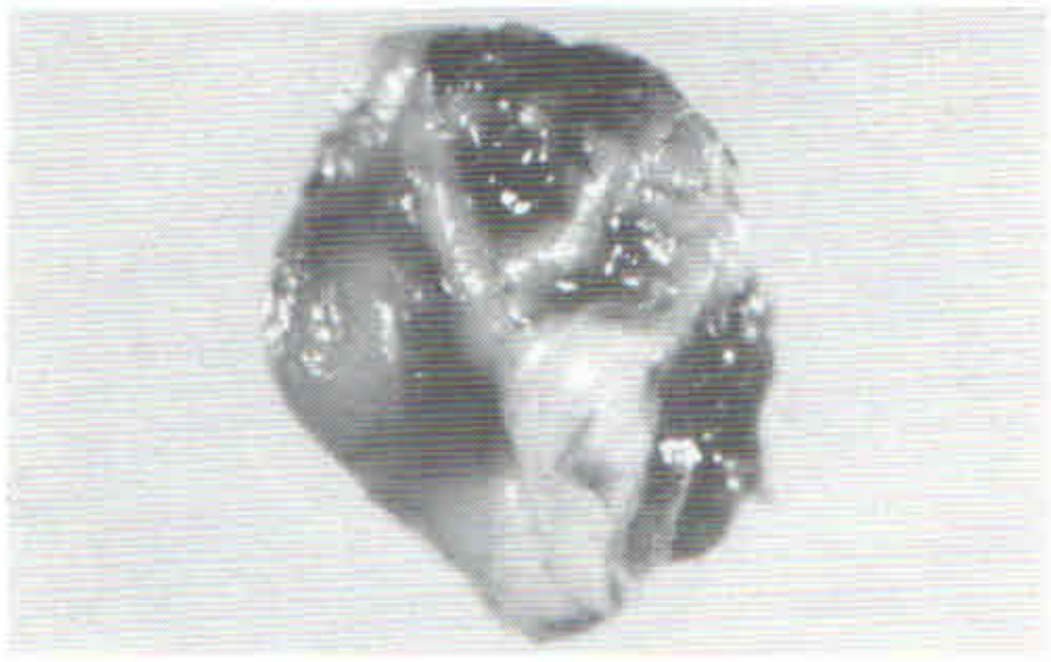

Fig. 1 Gross appearance of first phlebolith as a haemorrhagic mass in Case 1.

Fig. 2 Gross appearance of bisected specimen in Case 2

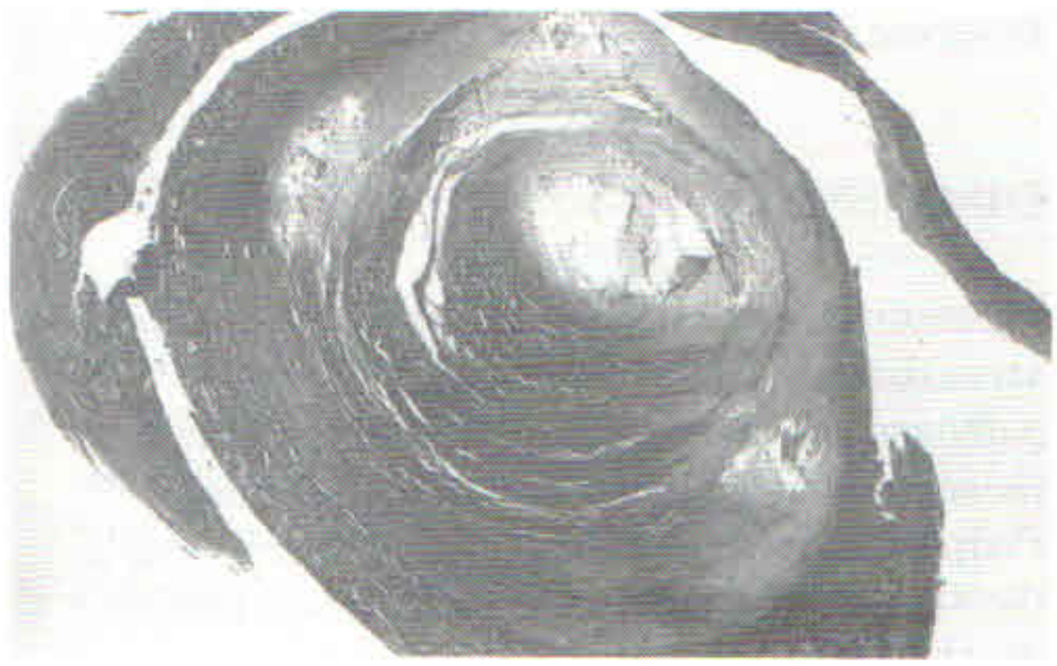

Fig. 3 Photomicrograph of the first phlebolith removed from the right buccal soft tissues in Case 1. The mass is darkly-staining and haemorrhagic and surrounded by bits of also haemorrhagic connective tissue (Haematoxylin & eosin stain. Original magnification x 7).

Fig. 4 Photomicrograph of one of the 2 darkly-staining, haemorrhagic phleboliths removed from the same site (Case 1) 10 years later. (Haematoxylin & eosin stain. Original magnification x7).

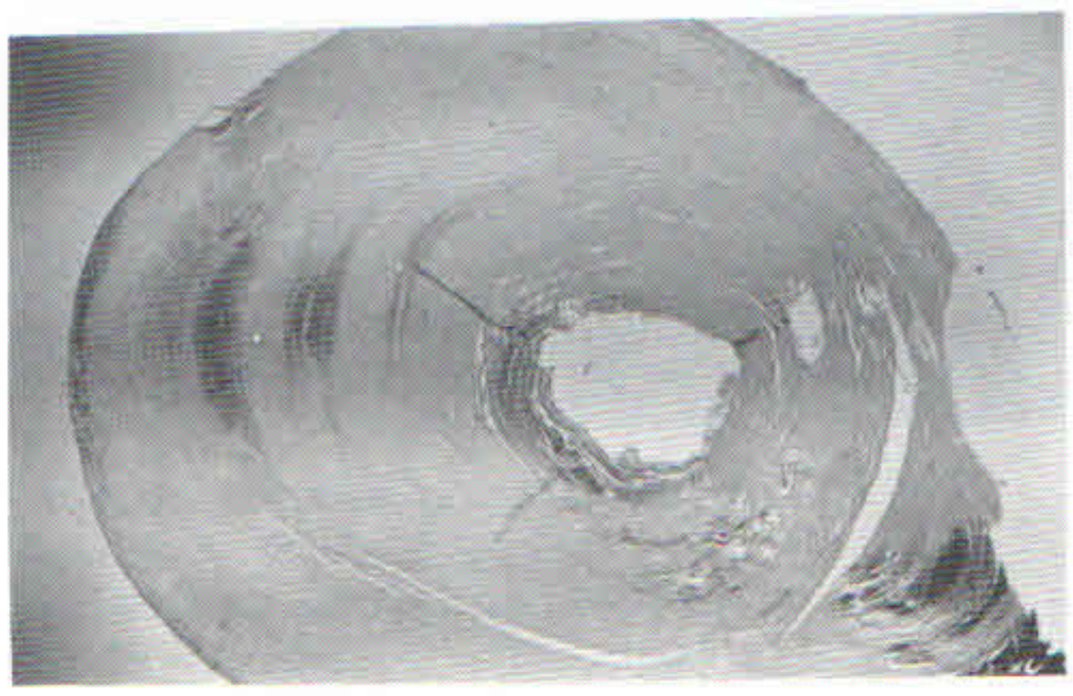

Fig. 5 Phlebolith removed from the cheek of patient in Case 2. (Haematoxylin & eosin stain . Original magnification x7).

In Case 2, a single calcified mass with a similar concentric laminated configuration was also observed (Fig. 5).

DISCUSSION

Phleboliths are calcified thrombi. When these structures occur in haemangiomas, the pathogenetic events leading to the formation of these calcified bodies are well-understood [6,7]. Peripheral slowing of the blood produces stasis in the sinusoidal spaces of haemangiomas resulting in thrombus formation, organization and secondary calcification. It is believed that a calcified core within the thrombus acts as a nidus initiating concentric mineralization to form a phlebolith. In both the cases described here, the underlying cause leading to the development of 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 unkown. 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[3]. The other plausible explanation relates to organization and dystrophic calcification of a haematoma following some form of injury that the patient is unaware of[8]. 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) [2,3,6,8-12]. 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[3]. 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[2]. 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.

ACKNOWLEDGEMENTS

We are grateful to Afidah and other staff of the Division of Stomatology for their secretarial and technical assistance, and to Dr. Mak Joon Wah , Director, Institute for Medical Research, for his permission to publish this paper.

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Table 1. Reported cases of phlebolith of the buccal soft tissue

Reference	Year	Age/Sex/Race	Clinical Presentation	Radiographic appearance	Features	Diagnosis and Comments
Thoma et al[9]	1948	57/F/-	Slowly-growing lump in (R) cheek x 15 years	RO material like a mass of buckshots	Surgical excision	Haemangioma with phlebolith
Schwartz and Salz[10]	1955	17/M/Jew	Painless mass (L) cheek present since birth	Doubtful opacities in (L) masseter	Surgical excision	Carvenous haemangioma with phlebolith
Deighan and Barton[11]	1956	31/F/Negro	Gradually increasing swelling of (R) cheek x 15 years	Multiple, small concentrically calcified bodies 1-5mm in size lying anterior to ascending ramus	Surgical excision	Carvenous haemangioma of buccal fat pad with phlebolithiasis
Gray[12]	1957	32/M/-	Swelling (R) cheek x 16 yrs	Multiple shadows in soft tissue of cheek	Surgical excision	Carvenous haemangioma of cheek with phlebolith formation.
Quinn[8]	1965	46/F/-	Several movable masses in (R) buccal tissue. Asymptomatic	One large calcified body and several smaller, irregular circumscribed ones	Surgical removal	Idiopathic calcified bodies/phlebolith
O'Riordan[2]	1974	27/F/-	Carvenous haemangioma of left side of face and lips. Symptom-free	Multiple RO bodies mainly in substance of the (L) cheek	No treatment	Phlebolith
		47/F/-	Acute swelling (R) cheek	Four RO bodies in soft tissues of cheek	UK	Phlebolith
Sano et al[6]	1988	17/M/-	Swelling of (L) cheek since birth, several hard, round nodules	Several normal calcified foci in (L) buccal region	Subtotal resection of tumour and surgical extirpation of phleboliths	Carvenous haemangioma with phleboliths
		37/F/-	(L) buccal swelling and slight spontaneous pain. H/O extirpation of a (L) buccal tumour 16 years ago.	Multiple calcified bodies, varying in size, in the cheek	Surgical excision of tumour	Buccal haemangioma with phleboliths
Zachariades et al[3]	1991	8/M/-	Moderately hard, painless swelling (R) mandible area x 4 years	Multiple mineralised bodies within hypertrophic masseter	Surgical removal	Phlebolith and normal muscle
		9/F/-	Moderately hard, painless swelling (R) mandible x 1 year	(R) masseteric hypertrophy and discrete radiopacities	Surgical removal and trimming.	Normal muscle and phlebolith
Present study	1995	6/F/Malay	Firm swelling (R) cheek. Painless x 3 years	NA	Surgical removal	Phlebolith. Recurrence x 10 years later. 2 other phlebolith excised
		29/F/Chinese	Nodular growth (L) buccal mucosa	Small radiopacity at site of lesion	Excisional biopsy	Phlebolith

RO – Radiopaque NA – Not available UK – Unknown