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PHLEBOLITHS AND SALIVARY CALCULI

B. O'RIORDAN, B.D.S., N.U.I., F.D.S., R.C.S.
Central Middlesex Hospital, London NW10 7NS
and Mount Vernon and King's College Hospital

Summary. Four cases of facial haemangioma containing phleboliths are described. The differential diagnosis between swellings due to obstructive salivary gland disease and haemangiomata is described and the distinguishing features of the radiopaque calcification associated with those swellings outlined. The early use of sialography in the diagnosis of these conditions is encouraged.

HAEMANGIOMATA in the region of the major salivary glands account for approximately 8 per cent of all salivary gland lesions, including mumps, in children under 15 years of age, and represent 20 per cent of all benign tumours in this age group (Krolls *et al.*, 1972). The haemangioma is thus the second most common benign tumour of salivary tissue, but when adults are considered as well the incidence falls to approximately 1 per cent. It is of some interest to note that 90 per cent of all haemangiomata in the region of the salivary glands are noted in the first year of life (Thoma, 1970). Although the majority of haemangiomata are hamartomata and not true tumours they have been included with benign tumours in many studies. They are always included in the differential diagnosis of swellings occurring in the area of the major salivary glands.

Because of their tendency to fluctuate in size, they may mimic salivary gland swelling due to inflammation or obstruction. Since skull radiographs, and in particular sialograms, play a large part in the differential diagnosis of swellings in the region of the salivary glands, it is important to consider a radiological feature of vascular malformations that may lead to confusion, namely the occurrence of phleboliths in the cheek or floor of the mouth.

Phleboliths arise when an intravascular thrombus becomes calcified and are not uncommon in the veins of the pelvis and in cavernous haemangiomata. They are rarely, if ever, seen in intra-bony haemangiomata (Parker & Frommer, 1964). Radiologically, phleboliths are classically described as radiopacities, generally circular, with a laminated morphology and a radiopaque or radiolucent centre (Fig. 9). The following four cases illustrate a number of the features which must be considered in the differentiation between swellings due to vascular malformations and salivary gland disease.

CASE REPORTS

Case 1. Miss R., age 23 years, was referred to hospital by her dental surgeon for the excision of a haemangiomatous nodule on the right side of her lower lip, as this was unsightly. This vascular malformation had been present from birth and also affected her neck. The haemangioma had initially been treated with *Thorium X* when the patient was a child.

On examination, a cavernous haemangioma was found affecting the right lower lip, right side of the neck down as far as the clavicle and also the tongue and right side of the floor of the mouth, mainly in the form of a bluish skin blemish, dilated veins and

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haemangiomatous nodules. The nodule on her lower lip was unsightly and it was therefore decided to remove it.

A radiograph taken to exclude involvement of the mandibular bone in the haemangioma showed multiple radiopaque bodies varying from 1-3 mm in diameter in the soft tissues. Further radiographs taken in planes at right angles, such as postero-anterior

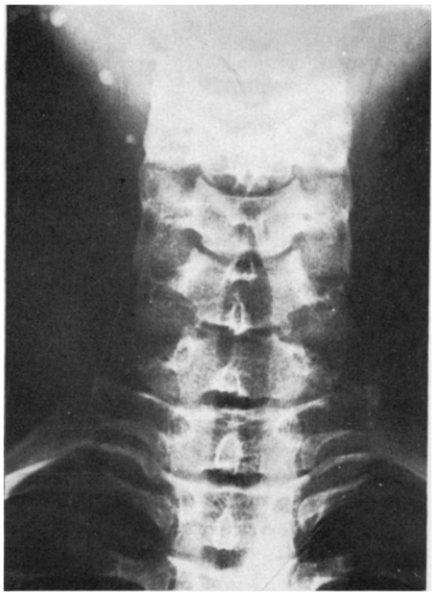

FIG. 1

Fig. 1.—Case 1. P.A. cervical spine and mandible showing phleboliths extending from angle of right mandible to 4th cervical vertebra.

Fig. 2.—Case 1. Central true occlusal view of floor of mouth showing multiple radiopaque bodies of homogenous density and varying size.

Fig. 3.—Case 1. Sialogram shows phleboliths to be outside the duct system. Similar appearance in submandibular region on oblique lateral view (not shown).

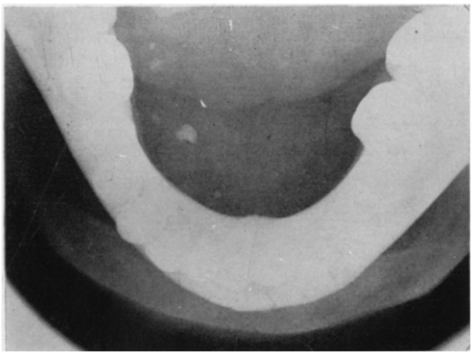

FIG. 2

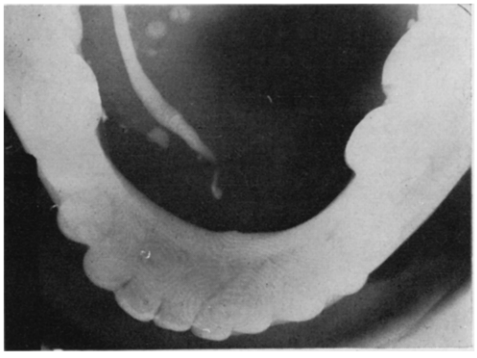

FIG. 3

and lateral views of the skull and occlusal views of the mandible, localised the radiopaque bodies to the floor of the mouth, right cheek in the region of the parotid gland and the right side of the neck down to the level of the 4th cervical vertebra (Figs. 1 and 2). In appearance, only one showed the 'classical' features of a phlebolith.

On questioning, the patient gave a history of episodes of swelling of the floor of the mouth and neck not related to meals but sometimes to posture; for example, her neck felt swollen after lying down in bed. All the swellings began suddenly with slight pain, and were not related to the thought, sight or intake of food. The swellings in the floor

of the mouth lasted from minutes to a couple of hours, but one over the larynx persisted for 2 weeks.

A sialogram of the right submandibular gland was performed to exclude salivary calculi or other gland abnormality as the cause of these swellings. It showed that the

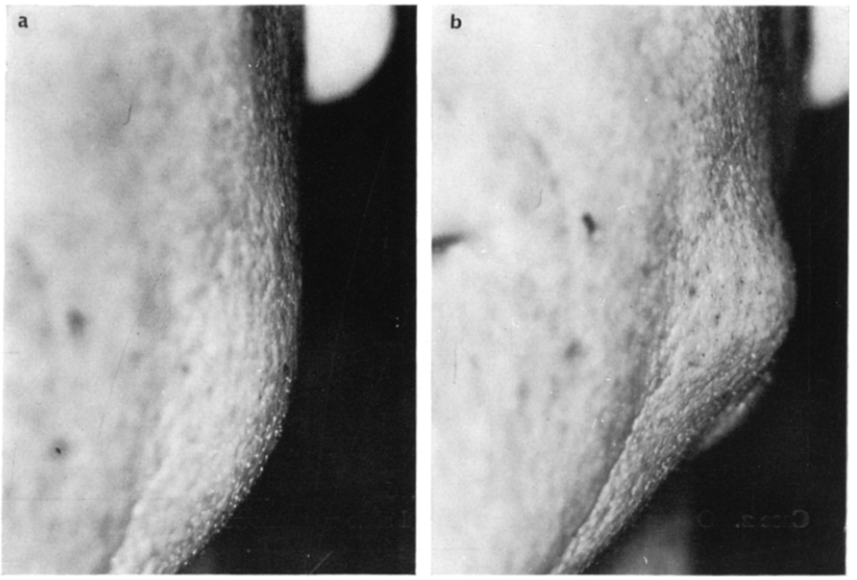

FIG. 4 (Case 2)

A, Swelling of left cheek at rest. B, Swelling erect and lobulated on contracting masseter.

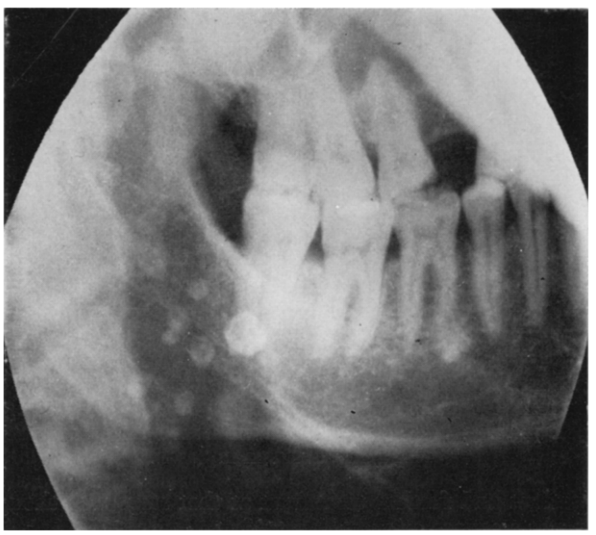

FIG. 5

Case 2. Left oblique lateral view showing multiple phleboliths with laminated structure.

gland was normal and that the radiopaque bodies were outside the duct system (Fig. 3).

The unsightly haemangiomatous nodule on the right lower lip was excised under local anaesthesia. The nature of the floor of the mouth and cervical swellings was

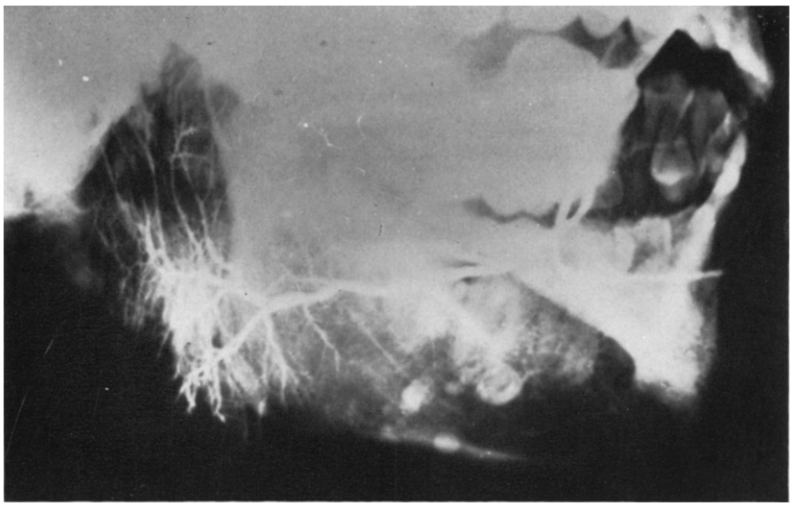

FIG. 6

Case 2. Oblique lateral jaws. Parotid sialogram shows radiopaque bodies outside duct system and gland.

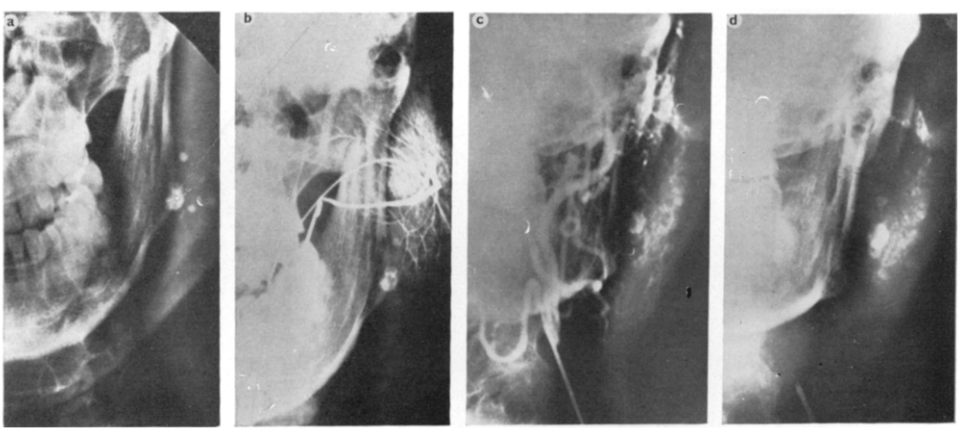

FIG. 7 (Case 2)

A, P.A. jaws showing phleboliths, some homogenous, some laminated. B, P.A. jaws. Parotid sialogram shows phleboliths outside duct system and normal duct pattern. C, P.A. jaws. External Carotid angiogram shows filling of haemangioma fed from above and below. D, P.A. jaws. External carotid angiogram (emptying phase) shows delayed emptying of haemangioma.

explained to the patient and further treatment at that stage was though unnecessary as the swellings were not a source of great discomfort.

Case 2. Mr F., age 43 years, was seen in 1973 complaining of swelling in his left cheek which had recently become painful. This was not affected by eating, but the

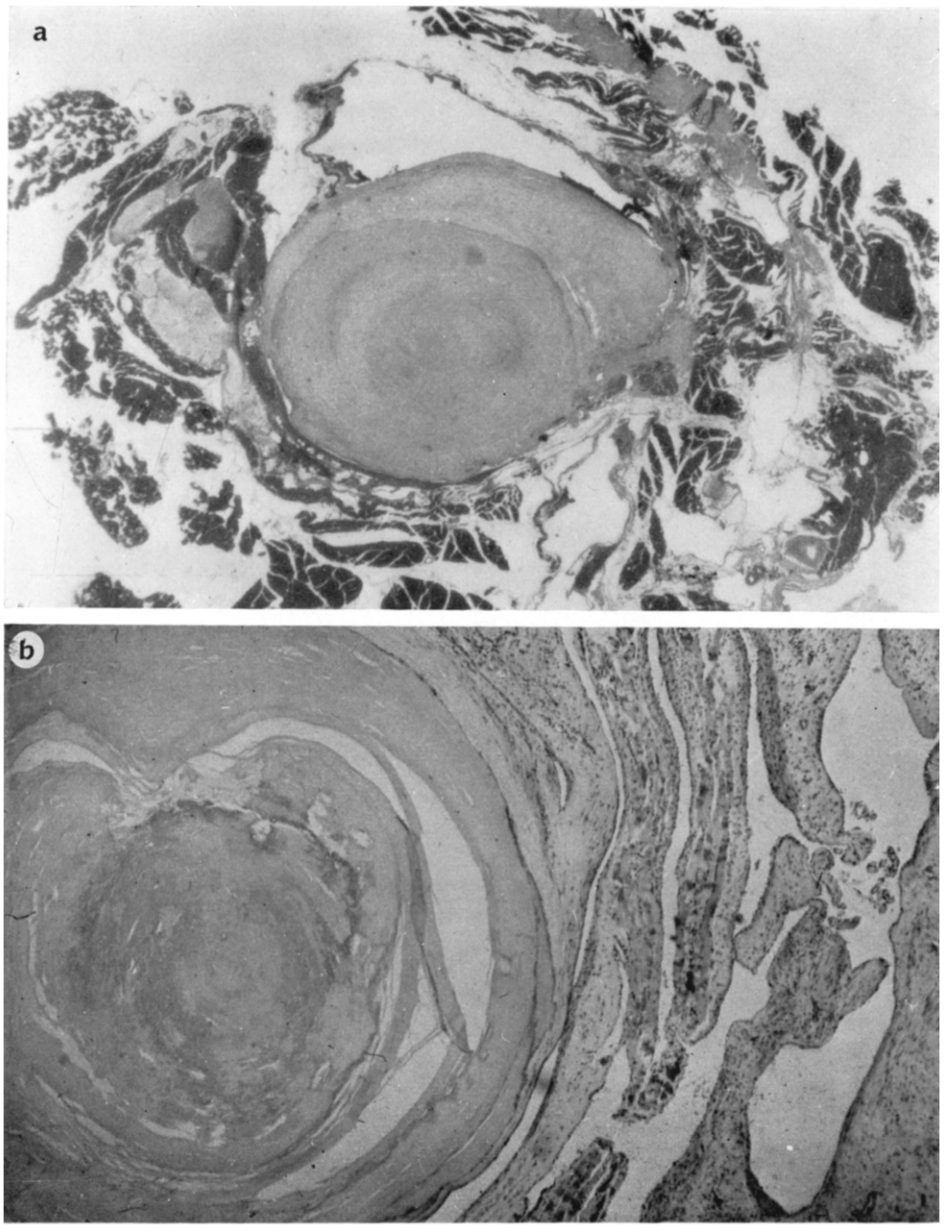

FIG. 8 (Case 2)

A, Histology. Laminated structure of phlebolith is clearly visible. Note large vascular spaces between striated muscle bundles. Some vascular channels are filled with blood and others with organised thrombus. H. & E. × 12. B, Phlebolith showing gap between 'nucleus' and outer lamina. This would appear as a radiolucent zone on a radiograph. (Courtesy of Dr R. Browne.)

lesion did swell up on bending forward or straining. The swelling was first noticed 10-11 years before this visit and seemed to follow two months after a kick to that part of the face while playing football. Swellings occurred about three times a year and were painful.

In the nine months before presentation his cheek was almost continuously swollen with intermittent increase in size. The presenting swelling had come up 3 days before consultation, after eating lunch, was very painful, but was slowly resolving.

On examination there was an ill-defined soft tender swelling in the left cheek, $1\frac{1}{2}$ cm in diameter over the anterior aspect of the lower pole of the parotid gland (Fig. 4A). There was no indication in the skin or mucous membrane of a haemangioma, such as skin blemish or dilated veins. It was easier to palpate bi-manually with one finger on the cheek and one in the buccal sulcus. No stone could be palpated. On performing the Valsalva manoeuvre—forcible expiratory effort with both nose and mouth closed—the

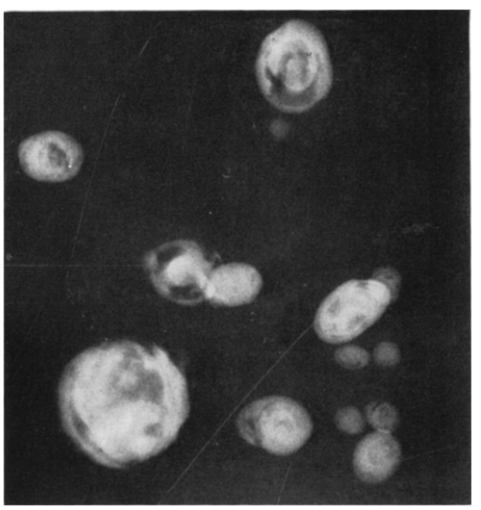

FIG. 9

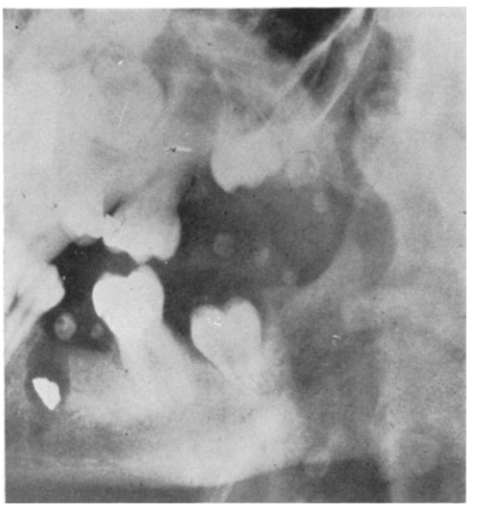

FIG. 10

Fig. 9.—Case 2. Radiograph of excised haemangioma. Classical radiologic features of phleboliths, multiple, round or oval, laminated with either radiolucent or radiopaque centres.

Fig. 10.—Case 3. Multiple laminated phleboliths in a haemangioma (incidental finding). These may be the residual signs in the adult of a childhood haemangioma (Thoma, 1970).

swelling increased in size. In addition when the patient contracted his masseter by biting his molar teeth together, the swelling became much more prominent and then appeared to be bi-lobed (Fig. 4, B).

Radiographs of the face showed a collection of radiopaque bodies in the cheek at the site of the swelling (Figs. 5, 7, A and 13) and sialography showed that these were outside the duct system (Figs. 6 and 7, B).

A diagnosis of cavernous haemangioma with phleboliths was made and arrangements were made to excise the lesion. An angiogram was performed in an attempt to identify the vascular feeders and this showed a haemangioma being fed inferiorly and superiorly by branches of the facial and external maxillary arteries respectively (Fig. 7, C and D).

At excision, under endotracheal anaesthesia, the haemangioma was found to be deep in the substance of the masseter muscle.

The histology report was as follows: '*striated muscle containing cavernous vascular spaces filled with calcified, organised thrombus and blood. Appearances are those of a*

cavernous haemangioma with phleboliths' (Fig. 8, A). Some of the phleboliths were up to 0·5 cm in diameter (Figs. 8 and 9). (Dr D. Lovell, Central Middlesex Hospital.)

It is possible that this lesion was an arterio-venous fistula resulting from direct trauma to the masseter muscle and blood vessels in the area, or a dormant congenital haemangioma re-activated by the direct trauma to that area.

Case 3. Miss C., age 27, who was a dental nurse, attended for the removal of an upper lateral incisor tooth. It was noted that she had a cavernous haemangioma which affected the left side of her face and lips. Radiographs were taken to exclude bone involvement in the haemangioma. The left oblique lateral view of the mandible showed multiple radiopaque bodies (Fig. 10). A postero-anterior view showed these to be mainly in the substance of the left cheek, extending from the lower border of the mandible

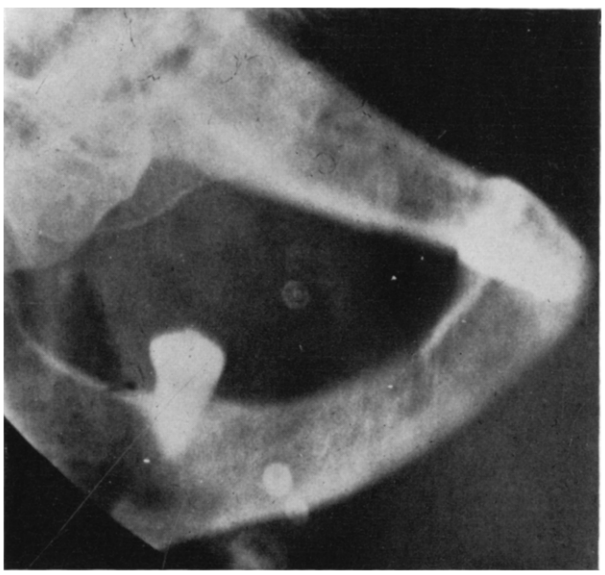

FIG. 11

Case 4. Multiple laminated phleboliths. Intermittent swelling of the haemangioma lasted for up to 2 weeks.

to the zygomatic arch. These radiopacities were unevenly calcified, and most had a laminated morphology. They were mainly circular and varied from 2 to 6 mm in size, and were considered to be phleboliths.

As this was an incidental finding and the patient was symptom-free no treatment was instituted.

Case 4. Mrs C., age 47, presented with acute swelling of the right cheek. Bluish dilated vessels were noted on the inside of the cheek. She gave a previous history of a number of swellings, which began suddenly and resolved slowly over a period of approximately 2 weeks.

A radiograph showed four radiopaque bodies in the soft tissue of the cheek (Fig. 11). One had the classical appearance of a phlebolith—laminated pattern with a radiopqaue centre and periphery—looking like a shooting target. A diagnosis of thrombosis in a haemangioma was made.

DISCUSSION

It is particularly important that adequate pre-operative information be obtained of lesions in the area of the parotid gland as any surgery in that area will involve dissection of the facial nerve, to a greater or lesser extent.

Haemangiomata mimic salivary gland swellings in that they can swell intermittently and can contain small radiopaque bodies. One of the lesions reported here did not show any surface vascular markings and hence it was easy to confuse it with swellings of salivary gland origin (Case 2).

Intermittent Swellings. Intermittent salivary gland swelling is classically related to the thought, sight or intake of food. Submandibular swellings usually resolve soon after a meal and may be painless or only slightly painful whereas obstructive parotid swellings last much longer and more often become infected. A careful history will usually show that swellings due to haemangiomata occur at times not necessarily related to gustatory stimulation and will, in general, be related to intravascular pressures.

Swelling of a haemangioma can occur when the venous return to the heart is restricted as in the Valsalva experiment. This restriction of the venous return is the cause of transient enlargement of juvenile facial haemangiomata when the child strains or cries. Backus and De Felice (1957) described a 6-year-old boy who could produce his swelling by holding his breath or pressing on his neck veins. The same little boy had one episode of acute prolonged swelling, which resolved when he had a general anaesthetic for an urgent sialogram. The authors postulated that venospasm, in preventing outflow of blood from the lesion, was the cause of that swelling, and was relieved by general anaesthesia.

Another case of haemangioma in the face in a female, not reported here as there were no phleboliths, used to enlarge at menstruation and became dramatically swollen during the delivery of the patient's first child, presumably again due to restricted venous return as a result of the muscular straining in labour. A haemangioma will also enlarge when the head is held in a dependent position. In Case 1 the patient reported that her neck felt swollen on lying down. The photographer when preparing this patient for a photograph asked her to remove her polo necked sweater. The bending over and muscular effort in doing this was enough to restrict the venous return and produce the swelling. Patey (1968) used the term 'turkey-wattle sign' to describe the enlargement of submandibular haemangiomata when the head is held dependent.

The transient swellings, lasting a few minutes in the first case reported here, may have been due to thrombosis in a vessel. The blockage so formed could lead to a swelling, which would resolve when the blood found other vessels through which to empty. Pain often accompanies the thrombotic episodes. More prolonged swellings are possibly due to thrombophlebitis in the haemangioma, as in Case 1 and Case 4.

A haemangioma involving the masseter muscle may fluctuate in size when the muscle contracts as in eating (Fig. 4, B), (Parker & Frommer, 1964). This is exemplified in Case 2 and Chipp and Weiler (1950) described a similar erectile haemangioma occurring in the masseter muscle. They also noted a time lag of approximately 2 sec. between contraction of the muscle and erection of the haemangioma, postulating that muscle contraction constricted the efferent vessels.

Radiopaque Bodies. The second area of confusion between salivary gland swellings and haemangiomata is in the radiographic appearance of radiopaque bodies. It is important, therefore, to consider the distinguishing features between calculi occurring in salivary glands and phleboliths occurring in a haemangioma.

As long ago as 1843, phleboliths were described in the splenic vein by Canstatt. They also occur frequently in the pelvic veins, and for some time in the early part of this century caused much confusion. However, their differentiation from uretheric calculi is now well known.

It has been said in many articles that phleboliths can be recognised by their concentric laminations, and radiopaque or radiolucent centres. However, many salivary calculi, especially submandibular calculi are similar in their radiographic appearance in that they show a laminated structure (Fig. 12, A and B), but it

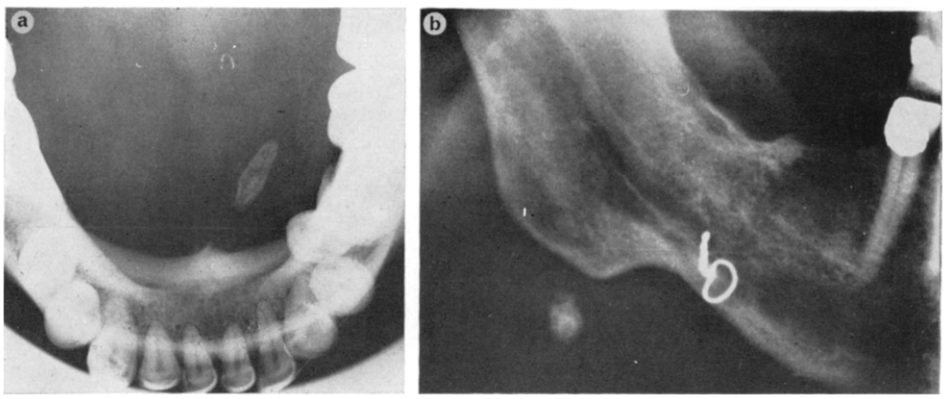

FIG. 12

Salivary calculi. A, Central true occlusal view of the mandible showing an elongated laminated salivary calculus in the anterior part of the duct. B, Section of a radiograph showing a circular laminated salivary calculus in the submandibular gland.

must be pointed out that parotid calculi are seldom large enough to show this concentric lamination. Seward (1968) in his series of articles on anatomical surgery for salivary calculi, showed a radiograph of a parotid stone which demonstrated all the classical morphological features of a phlebolith. However, sialography showed that this was indeed a salivary calculus, and was within the duct system causing an obstruction. It, must, therefore, be said that the shape of a 'phlebolith' as seen on a radiograph is not diagnostic, but is only suggestive that a haemangioma is present. This latter fact may be important as in many cases there is no superficial sign of the haemangioma, such as bluish discolouration as in Case 3 and the radiological findings may be the first indication of its presence as in Case 4. Thoma (1970) states that a phlebolith may be the only residual sign, in the adult, of a childhood haemangioma.

The Table shows the distinguishing radiological features between salivary calculi and phleboliths. A salivary calculus is uniformly radiopaque though it can be laminated. A phlebolith is usually laminated with a radiopaque centre or nucleus, and sometimes a radiolucent centre. Small phleboliths are usually

uniformly radiopaque, whereas the larger a phlebolith is the more likely it is to be laminated. A salivary calculus is usually shaped by the duct—therefore is usually elongated—but a phlebolith is generally circular, although intra-glandular duct as opposed to extra-glandular duct salivary calculi are often circular.

These latter details are best seen on non-screened films and where possible x-ray views should be taken using either a periapical film or an occlusal dental film. When the lesion occurs in the cheek or lips a periapical film can be held inside the mouth against the mucous membrane and the x-rays projected from outside. An occlusal film can be used for lesions in the floor of the mouth and of the skin over the lower border of the mandible. Tangential views of lesions at the angle of the mandible are also possible by holding an occlusal film under the angle (Fig. 13). Dempsey and Murley (1970) in a well illustrated article reported three cases of haemangiomata containing phleboliths where one group of phleboliths in the cheek was very well shown on an intra-oral film.

TABLE

RADIOLOGICAL FEATURES

Salivary calculus	*Phlebolith*
Uniformly radiopaque. Can be laminated if large, especially in Submandibular Gland.	Usually laminated with radiopaque centre or nucleus, sometimes radiolucent centre. Uniformly radiopaque if small.
* Usually shaped by the duct and so are elongated.	Usually circular.
*** Number—1 to 2.	Number—multiple.
* Sialography—shows filling defect at site of calculus.	Sialography—shows phleboliths to be outside the duct system.

Radiological features of phleboliths and salivary calculi. The number of asterisks indicate the degree of importance of a particular feature. No one of these radiological features is, of itself, diagnostic of a phlebolith. It is a combination of these appearances, or a combination of clinical and radiological features that confirms a diagnosis

In number salivary calculi rarely exceed one to two, whereas phleboliths are multiple. This is one of the most significant differences between the two types of radiopacities.

Sialography shows a filling defect at the site of a salivary calculus, whereas it demonstrates that a phlebolith is outside the duct system. It may also show whether the phlebolith, and by implication, the haemangioma is deep to the salivary gland. In the parotid region 90 per cent of haemangiomata occur superficial and 10 per cent deep to the gland. Thoma (1970) suggests that the former probably develop from cutaneous lesions and infiltrate deeply, and the latter develop from muscle.

Phleboliths on analysis consist of calcium carbonate and phosphate and in histological section are laminated concentrically (Fig. 8). They may even be ossified in part (Thoma 1969). Multiple haemangiomata containing phleboliths are sometimes seen in association with dyschondroplasia (Ollier's Disease) and in this circumstance is known as Maffuci's Syndrome (Shanks & Kerley, 1971).

Other Dystrophic Calcifications. Other calcifications with which a phlebolith might be confused are calcified lymph nodes which are probably the

commonest calcification seen in soft tissues of the head and neck. These are usually very large and consist of a mass of calcifications and do not look at all like a sialolith and like tonsoliths, which can also be superimposed on the parotid region in the oblique lateral view, will be seen to be lingual to the mandible on the postero-anterior view. This and the following descriptions are taken from an excellent review of calcifications in the facial soft tissues in *Dental Roentgenology*, (Ennis, Berry & Phillips, 1967). In cysticercosis, small oval calcifications are seen. These are multiple and represent calcified parasites. They are oval when found in muscle because of the pressure from the muscle fibres, but are usually round in the viscera.

FIG. 13

Tangential view of swelling in Case 2 taken with an occlusal (non-screened) film held under the angle of the mandible with the haemangioma 'erect'.

Calcifications may occur in epithelial cysts, in sub-cutaneous tissue, in epidermal cysts and sebaceous cysts and, infrequently, in cysts of sweat glands. These are superficial, however, and this can be confirmed by tangential radiography.

Calcified arteries are typical in their appearance. Myositis ossificans has a striated linear morphology and is not likely to be confused with calcifications in blood vessels.

Calcified acne lesions are multiple and Ennis (1967) describes their shape as being like snow flakes.

Multiple miliary osteomata of skin may look like phleboliths as they are doughnut shaped.

None of the above lesions, however, produce swellings which mimic salivary gland lesions and, therefore, should not cause the diagnostic problems related to phleboliths.

Additional Diagnostic Features. In addition to the differences already described the following features have often been put forward as useful in distinguishing between salivary swellings associated with calculi and haemangiomata associated with phleboliths.

When obstructive salivary gland disease occurs in the presence of a calculus, protein analysis of the excreted saliva will reveal a pattern consistent with infection —potassium and protein levels raised—whereas saliva would be normal when the swelling was due to a haemangioma. In addition, in the presence of infection, the saliva may be frankly purulent—'snow storm' effect.

It has been stated (Gorlin & Goldman, 1970) that phleboliths cannot be palpated. Nevertheless, it is feasible that when they are near the surface—either mucous membrane or skin—they could be felt (Thoma, 1963; Ennis *et al.*, 1967; Hopkins, 1969). Salivary calculi are said to cause pain whereas phleboliths do not. It is quite true that a calculus directly brings about a painful swelling due to obstruction of a salivary duct, and a phlebolith, since it is a calcification in an organised clot and therefore represents an historical event, does not. However, this is not entirely helpful in the clinical situation as the swelling under examination may be painful whether it is due to a salivary gland obstruction by a radiopaque calculus or to an acute enlargement of a haemangioma in which x-ray examination happens to reveal a phlebolith. In both instances, the presenting clinical feature is a swelling and in both an opacity or opacities are seen on the radiograph. One still has to decide whether the opacity is a phlebolith or a salivary calculus.

On palpation a cavernous haemangioma may have an elastic consistency, may empty on pressure and slowly fill up again or be pulsatile, and may also feel lobulated. Auscultation should always be performed in the case of suspected haemangiomata to detect the presence of bruit. The performance of the Valsalva manoeuvre will in many cases reproduce the swellings if it is due to a haemangioma.

Sialography still remains the one most important investigating procedure in distinguishing salivary gland lesions from others occurring in the same area. It is possible, of course, for the haemangioma to infiltrate the gland and appear as a space-occupying lesion on sialography. It is also possible that despite the presence of phleboliths, a swelling may be due to an obstruction of the salivary gland by a radiopaque calculus, the calculus showing up in the midst of the phleboliths. A sialogram would show a filling defect in a duct and the phleboliths would, therefore, be just an incidental, although confusing, finding. This problem has arisen in the pelvis when uretheric calculi have occurred in the presence of uretheric phleboliths.

Angiography (Figs. 7, C and D) is of limited diagnostic help. Although it shows the presence of a haemangioma, the contrast medium may not enter the whole of the lesion as channels may be blocked by thrombus formation, and those areas with phleboliths may even appear to be outside the lesion as they represent parts of the hamartoma which have undergone thrombosis in the past. The feeding vessels may, of course, be identifiable and this is of considerable practical importance when surgery is being considered.

The purpose of this paper was to consider the differentiation between

phleboliths and salivary calculi. Of prime importance in the differential diagnosis between intermittent vascular and salivary gland swellings is the history, which will usually show that swellings of intra-glandular origin are related to food. The haemangioma, if superficial, will cause a skin blemish but if deep may not. On physical examination, parotid swellings will be restricted to the anatomical outline of the gland whereas haemangiomatous swellings may occur within the gland periphery or outside but close to it. Inflammatory lesions due to duct obstruction will often be accompanied by a flow of purulent saliva—'snow storm' effect—whereas those due to vascular malformation will have clear saliva.

In addition to the above clinical methods radiology has much to offer in distinguishing between lesions of salivary gland or vascular origin. Radiopacities should be accurately localised by taking, in addition to the usual periapical and oblique lateral x-ray views, postero-anterior and lateral skull and true occlusal views. Sialography should always be performed as it is extremely helpful in separating radiopacities in the salivary glands from those in the substance of the cheeks or floor of mouth (Deignan & Barton 1956; Hopkins, 1969; Backus & De Felice, 1967).

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