A CLINICAL AND RADIOLOGICAL SURVEY OF 192 CASES OF RECURRENT SWELLINGS OF THE SALIVARY GLANDS

Hunterian Lecture delivered at the Royal College of Surgeons of England on 26th March 1953 by

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INTRODUCTION

IT IS PROPER and fitting in a Hunterian Lecture to pay tribute to the works of the man whose name we honour to-day. What greater tribute can I pay than to say that it was the application of Hunterian principles of research that led to the work that has culminated in this paper.

It was the challenge offered by the repeated mis-diagnosis of salivary calculus in the recurrent parotid swelling that first stimulated my enquiries and eventually clinical observation and investigation were rewarded by a measure of understanding of the conditions encountered.

Considering the attention that has been paid to the salivary gland tumours, it is surprising that the non-neo-plastic conditions have escaped more detailed examination.

Knowledge of some of the factors influencing secretion of saliva, however, dates back to the Arab physicians and there are abundant references to the effect of fear and appetite on salivation from this time forward. Indeed, one of the trials by ordeal was the enforced taking of a liberal portion of dry flour. This was washed down easily by the unhindered salivation of the innocent—and provided they were not too

TABLE 1

ANALYSIS OF 192 CASES OF RECURRENT SALIVARY GLAND SWELLINGS

	Cases	Male	Female
Parotid gland:—			
Congenital sialangiectasis	10	9	1
Duct obstruction :—			
(*a*) Papillary	26	10	16
(*b*) Buccal	9	2	7
(*c*) Calculous	3	2	1
" Idiopathic " duct dilatation	4	1	3
Recurrent parotitis :—			
(*a*) Streptococcus viridans	28	10	18
(*b*) Pneumococcus	7	2	5
(*c*) Miscellaneous infections	6	1	5
(*d*) Parotid abscess	5	2	3
Sjögren's syndrome	16	—	16
Mixed infection and duct obstruction	4	1	3
Submandibular gland:—			
Duct obstruction :—			
(*a*) Calculous	72	46	26
(*b*) Non-calculous	2	1	1
TOTAL	192 cases		

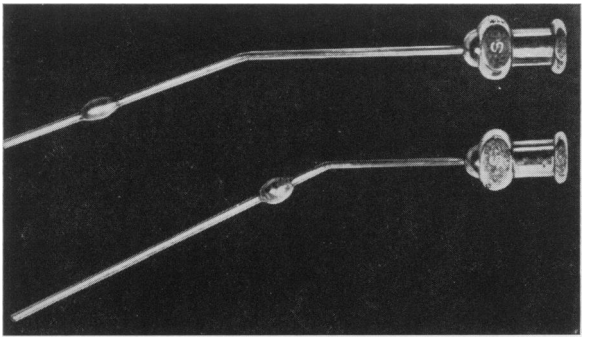

Fig. 1. The cannulae.

impressed with the rigours of the examination they were acquitted. Nowadays, public speaking forms an efficient substitute for this ordeal.

My remarks to-day, will be confined to these conditions of the salivary glands which are characterised by *recurrent swelling,* and are based on the personal experience of 192 documented cases (Table 1).

ANATOMICAL CONSIDERATIONS AND SIALOGRAPHY

The diagnosis of the recurrent swellings of the salivary glands has been aided in comparatively recent years by the visualisation of the sialodochal tree by means of the introduction of radiopaque media.

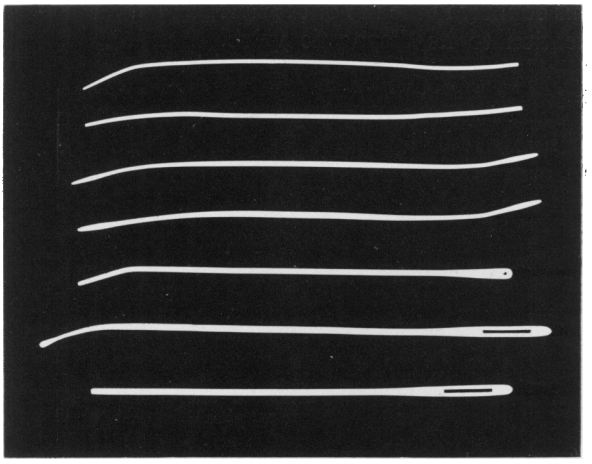

Fig. 2. The lacrimal dilators.

Although sialography had been used experimentally for many years in the cadaver it was not until 1925 that Barsony described its application as a diagnostic procedure. Jacobovici *et al.* (1926), however, attributes the first sialogram to Charpy. Barsony used a solution of 20 per cent.

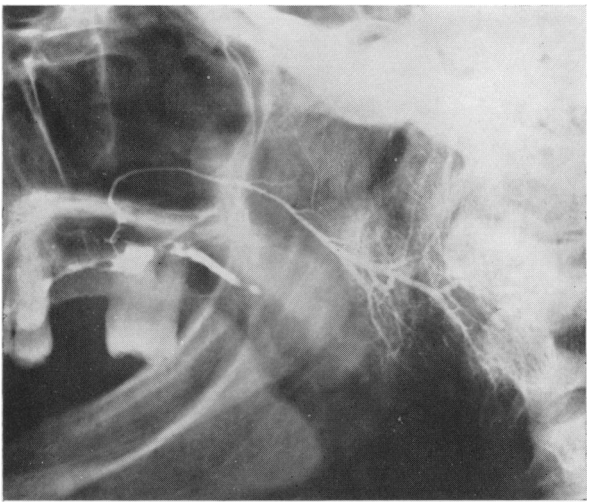

Fig. 3. Normal parotid sialogram. Lateral view.

potassium iodide in a case which he described as an idiopathic dilatation of Stenson's duct, but discarded this medium because of the considerable pain and reaction produced. A year later Carlsten (1926) reported a case in which lipiodol had been used and since this date the iodised oils have replaced all others in this field. R. T. Payne (1931) was the pioneer in this country in the use of the sialogram and was the first worker to draw attention to its diagnostic value.

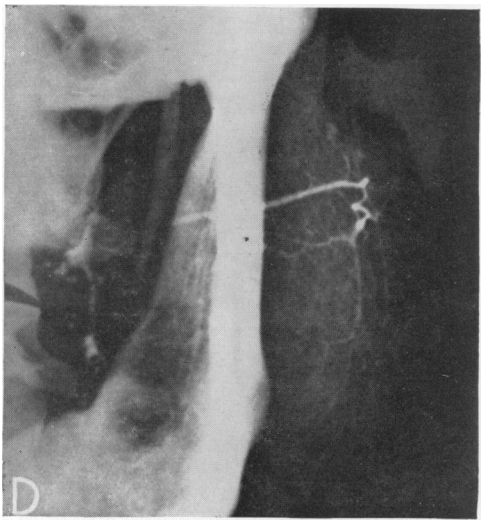

Fig. 4. Normal parotid sialogram. Anteroposterior view.

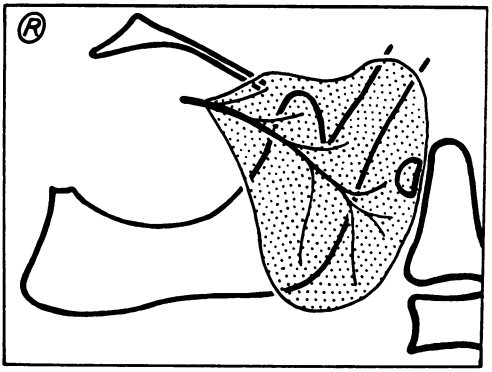

Fig. 5. Diagram of lateral parotid sialogram. Note the four superior ducts—the anterior of which is the duct of the socia parotidis when present and the two inferior ducts. This is the usual distribution and angle of incidence of the secondary ducts with the main Stenson's duct.

Other authors, notably Pyrah and Allison (1931), Barsky and Silberman (1932), Leroux (1947), Schulz and Weisberger (1947), have made valuable contributions in this field.

From time to time the iodised oils have been criticised on account of their tendency to break up into globules when they come into contact with a watery phase. This is certainly true when large cavities are to be outlined, but as the amount used in sialography is generally small, this objection does not arise.

Furthermore it has been found repeatedly, on attempting to use the water soluble radiopaque materials such as viskiosol that the absence of a fluid interphase results in the rapid emptying of the injected gland with consequent loss of radiographic definition. As our technique (Rose, 1950), does not usually involve injection on the X-ray table with its attendant inconveniences and delays, it is important that the fluid should remain *in*

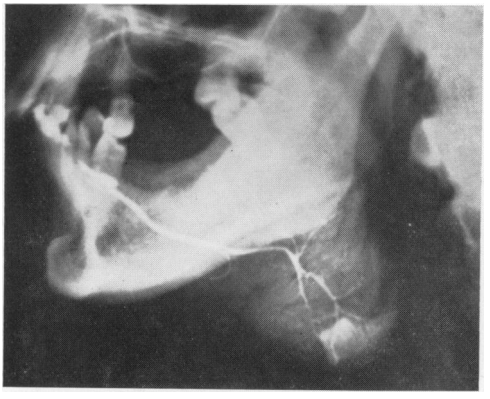

Fig. 6. Normal submandibular sialogram. Lateral view.

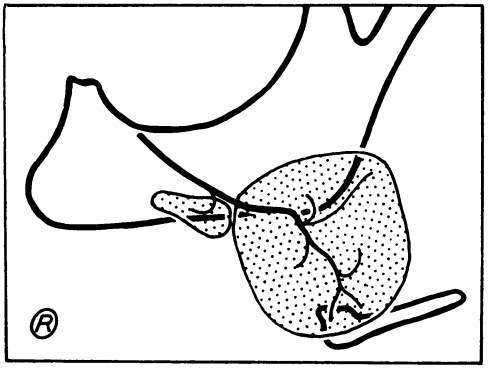

Fig. 7. Diagrammatic representation of the lateral submandibular sialogram. Note the right angle made by the duct as it descends into the gland. The retromandibular branch is a constant feature.

situ for 10 or 15 minutes and be capable of giving a clear picture at the end of that time. Neohydriol *fluid* is admirably suited for this purpose and is not so viscous that it is difficult to inject. In over 200 sialograms, I have observed no untoward effect.

Technique

Two ccs. of *Neohydriol fluid* are injected into the appropriate duct orifice through a specially constructed cannula (Fig. 1). A blob of solder is fused on to a blunted 3in. No. 20 gauge angled intravenous needle about $\frac{1}{4}$in. and $1\frac{1}{4}$in. from the end to produce a parotid and submandibular cannula respectively. The cannula thus cannot be inserted too far and reflux during injection is prevented.

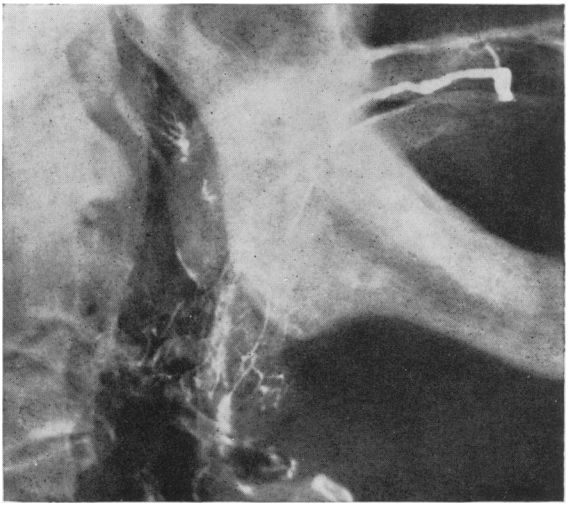

Fig. 8. Malignant parotid tumour showing break-up of duct pattern.

A set of modified lacrimal dilators are to hand if their use should become necessary (Fig. 2). The discomfort is minimal if the injection is carried out slowly and the X-rays are taken after the cannula is removed, a swab being kept in place over the orifice while the patient is positioned.

Anteroposterior and lateral views are taken in the case of the parotid—while lateral and oblique views are used for the submandibular gland ; only using the occlusal and basal views when especially indicated to

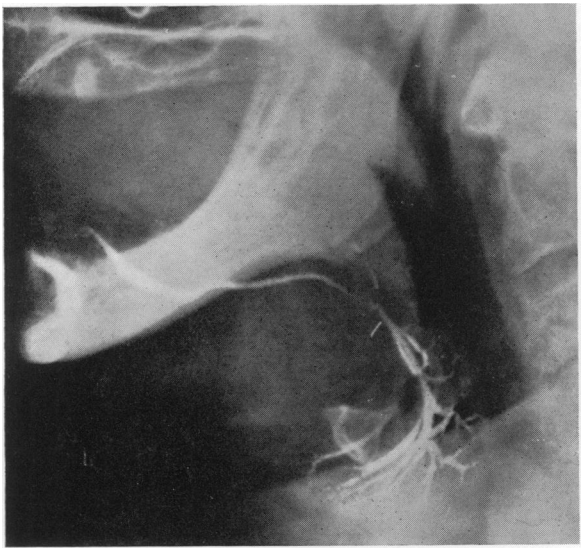

Fig. 9*a*. Space occupying lesion of submandibular gland showing duct distortion.

elucidate some special point. Normal sialograms produced in this way are illustrated and diagrammatically represented by the accompanying Figs. 3–7.

The points to be noted on the sialogram are :—

1. The duct pattern.
2. The duct character.
3. The presence of filling defects.

The duct pattern in both glands is remarkably constant and follows the plan demonstrated by Figs. 3-7. Gross distortion of this pattern, especially with alteration of the angle of incidence of the secondary ducts with the primary ducts means a space occupying lesion (Figs. 8-10).

The duct character

The duct is normally of even calibre, fairly narrow and its subdivisions may be followed branching in a racemose manner until they are lost to X-ray visualisation.

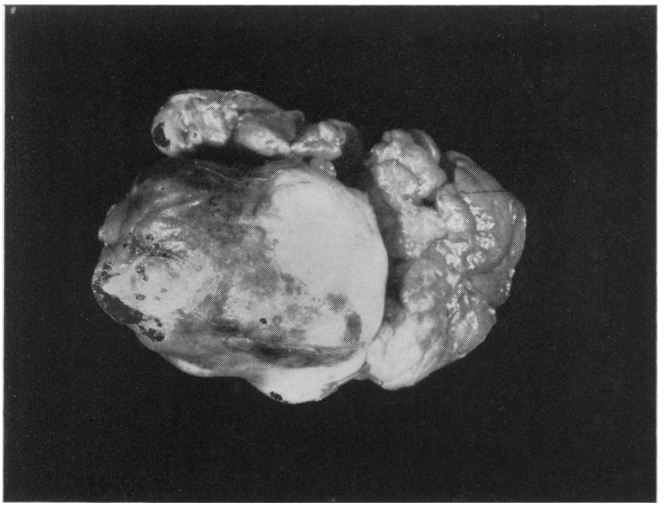

Fig. 9*b*. Lipoma.—For comparison with the sialogram shown in 9*a*.

The presence of filling defects

In addition to distortion of the sialodochal tree, a space occupying lesion may produce a *filling defect* in a gland which contains enough radiopaque medium to give acinar filling—*i.e.*, to produce a diffuse shadow of the surrounding parenchyma. This is well shown in Fig. 10.

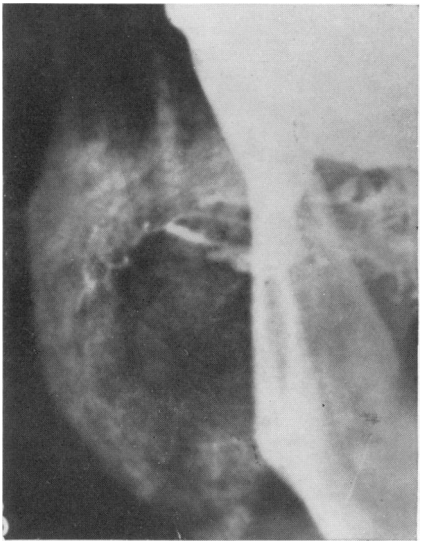

Fig. 10. Space occupying lesion of parotid gland. Filling defect with good filling of the surrounding gland.

Samuel (1950) has referred to this outlining of the gland as the sialoacinar reflux and poses the question of its significance.

In a previous communication (Ollerenshaw and Rose) it was shown by the injection of post mortem glands *in situ* and of living glands under controlled pressure that the sialoacinar reflux is entirely due to overfilling of the gland and varies directly with the pressure and volume of fluid used. Furthermore it is important to note that only the normal glands and normal parts of pathological glands will fill in this way. Of the histology of the glands, there is little to be added to the standard text book descriptions but it is worthwhile observing that the parotid alone possesses a well marked narrow intercalary duct connecting the acinus with the intralobular ducts, a factor which may be of contributory importance in the comparatively high incidence of infection in the parotid gland.

Congenital Anomalies

The developmental anomalies encountered in the salivary glands are few in number and are usually present as accidental findings in association with the particular pathological process under investigation. Anomalies

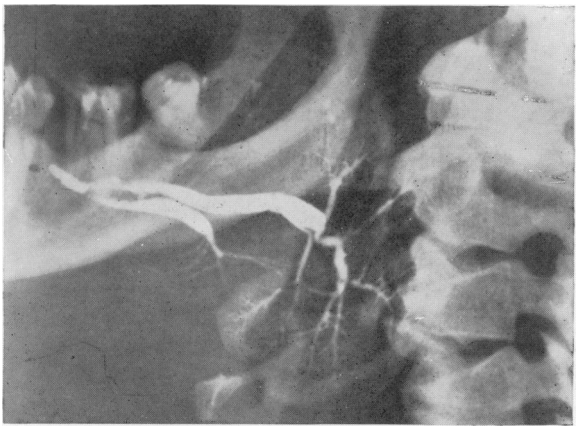

Fig. 11. Congenital hypertrophy of submandibular glands.

of the ducts of both glands however are sometimes found and Fig. 11 illustrates a case of congenital hypertrophy of both submandibular glands.

The patient, a woman of 42, who had noticed a bilateral submandibular swelling for as long as she could remember, visited her dentist complaining of an aching tooth. She was referred to hospital where a sialogram was carried out and a bilateral duct anomaly was discovered in addition to the unusually large size of the glands.

Congenital dilatation of Stenson's duct is occasionally reported and is of special interest in that Barsony's original case fell into this category (Fig. 12). More often, however, the finer parotid ducts are affected (Pearson). Advanced changes are found in the sialograms of children

with a short and comparatively mild history of recurrent parotid infection and it is believed that these cases are examples of secondarily infected congenital sialangiectasis. The condition may be localised or diffuse in one gland or affect both sides. Fig. 13 depicts this condition in a child of three and a half. Ten cases in this series fall into this group. They all occurred in children between the ages of three and 15, with a history of from six to 12 months recurrent attacks of swelling and pain in the parotid gland, bilateral in two cases. The infecting organism was the streptococcus viridans in every case and the precipitating factor seems to have been an erupting tooth. Sialograms showed appearances comparable with the long-standing streptococcus viridans infections seen in adults. These changes were also noted in the clinically normal opposite gland. The saliva on this side was quite normal.

In two of the cases seen, the inspissated secretion and salivary debris had become partially calcified and appearances resembling a sialogram were obtained on the plain X-ray film shown here (Fig. 14). This was a

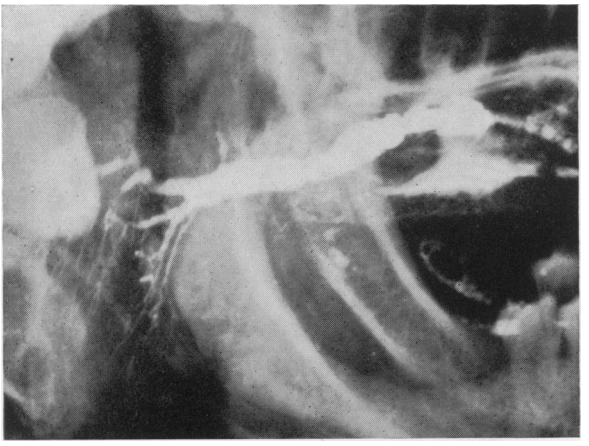

Fig. 12. Congenital dilatation of the parotid duct.

woman of 28 with a history of recurrent swelling of both glands for as long as she could remember. The sialogram confirmed the acinar distribution of the calcification and is shown side by side with the plain film.

Physiological Considerations

Before proceeding to discuss the conditions affecting the glands, I would like to survey briefly what we know of the secretion of saliva and its composition. The saliva is a mixed secretion of parotid, submandibular, sublingual, palatal and buccal glands. The total amount varies between one and $1\frac{1}{2}$ litres a day in health—rather less than one-third of this amount being the parotid contribution to the total. The parotid secretion is thin and serous, having a slow solid content of ·5 per cent. or less and a relative viscosity of under four. The submandibular saliva is thicker with a higher

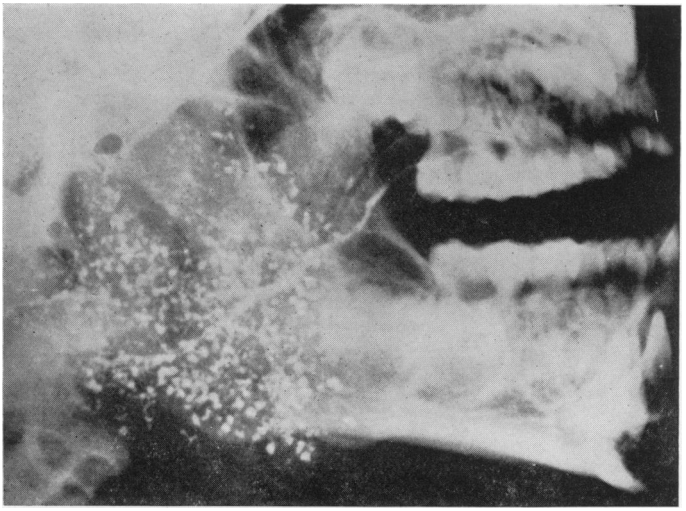

Fig. 13. Congenital sialangiectasis.

solid content averaging 2 per cent. but with a possibility of concentrating up to 6 per cent. It is a mixed serous and mucous gland, the relative viscosity being just under 6. The sublingual, palatal and buccal glands are entirely mucous in their secretion.

The factors controlling the secretion of saliva are local and central. Local reflexes arise through smell and taste, by means of the taste buds or *through* the nerves of common sensation and the character of the saliva produced is profoundly influenced by the nature of the stimulus. For example a bolus of meat produces an immediate flow of thick mucinous submandibular saliva as a lubricant, the parotid secretion

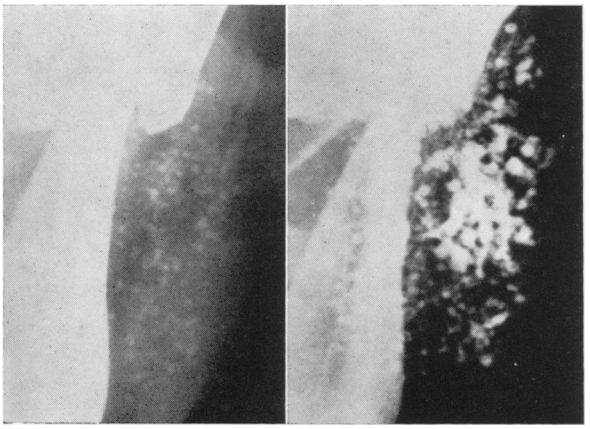

Fig. 14. Congenital sialangiectasis with calcification.

following after an appreciable lag. On the other hand a piece of the same meat dried and powdered produced a profuse flow of watery saliva from the parotid, to give a rinsing action. This is a response to common sensation, for a ground-up pebble produces a similar watery flow, while the intact pebble is inert in this respect. Central stimuli may be just as powerful in producing or inhibiting the secretions and the ease with which these alterations are accomplished has made possible the fascinating study of the conditioned reflex. During their classical work on this subject Pavlov and Babkin showed among other things that simple laparotomy and rectal feeding considerably diminished the salivary flow and increased the virulence of the normal oral flora.

More recently the study of the alarm reaction by Hans Selye has produced ample evidence that stress alters the amount and character of the saliva.

Further information can be obtained with regard to the relative activity of the two main salivary glands by comparison of their respective functions. They may be studied under the following headings :—

1. *Lubricant.*—The lubricating action of saliva is required for two purposes : (*a*) for maintaining the resting moisture of the tongue, lips and the oral mucosa for the purpose of articulation, and (*b*) for facilitation of the mixing and swallowing of food. The lubricant properties of saliva depend on its mucin content and therefore the parotid plays little part in this activity. The remaining glands are responsible for this function and the part played by the palatal and buccal glands must not be minimised. Indeed, in senile xerostomia it is customary to see a dry mucosa with quite good secretion from the parotid and submandibular glands and I believe that atrophy of the small mucous salivary glands is responsible for this condition.

2. *Rinsing activity.*—Lavage of the interdental crevices and consequent maintenance of oral hygiene is normally dependent on a brisk flow of serous saliva and one has only to recall the sordes of pyrexia and dehydration to realise the importance of this rinsing action. Any diminution of the parotid flow, for it is the parotid that is important in this respect, will tend to increase the incidence of oral sepsis.

3. *Digestive function.*—The amylase ptyalin is produced in the parotid and its action in initiating the digestion of carbohydrate is well known. It is not quite so well known, however, that it is activated by the presence of chloride ion, and if the digestive function of a specimen of saliva is required, chloride must not be added or an abnormally high reading will be obtained. The influence of the chloride ion on enzyme activity may be appreciated by observing the effect of the addition of salt to a carbohydrate meal on the blood sugar curve. An appreciable rise is obtained under such conditions.

A mucolytic enzyme, lysozyme, is also present in normal saliva and is of particular interest in view of its undoubted bacteriostatic action. It is secreted mainly by the parotid and acts on the polysaccharide capsule of many organisms such as Staphylococci, Streptococci, Pneumococci and Meningococci.

In conditions in which *hyposecretion* of parotid saliva occurs, for example recurrent parotitis and Sjögren's syndrome there *is both reduction in the chloride content and total enzyme content of the saliva so that the digestive and bacteriostatic activity is considerably diminished.* The opportunity for infection is then greatly increased.

4. *Buffer activity.*—The presence of mucin and salts such as sodium bicarbonate, the acid or alkaline sodium phosphate and sodium and potassium chloride makes saliva second only to blood as a buffered solution. The pH of the mouth can only be momentarily changed by the introduction of acids and alkalies as the rapid secretion of saliva soon restores the *status quo.* The pH of mixed saliva averages at a level of 6·5. My figures for parotid and submandibular saliva collected separately show that the parotid secretion is rather more acid than the submandibular—the former averaging 5·8 and the latter averaging 6·8.

5. *Taste.*—The function of *taste* requires solution of the food in saliva and the presence of the appropriate end organs. In xerostomia the lack of taste is due to impairment of both these faculties, the end organs taking part in the general atrophy.

6. *Dehydration.*—Dehydration by depressing the secretion of saliva rapidly produces the sensation of thirst and constitutes an early warning. Unfortunately, if relief is not immediately forthcoming, the stage is set by this inhibition for ascending infection, and this is the main cause of acute suppurative parotitis. This condition, however, does not precede recurrent pyogenic parotitis and is not discussed in this lecture.

Clinicopathological Considerations

In the introduction to this lecture, I referred to the type of case which stimulated the original work on this problem, that is the recurrent parotid swelling with the all-embracing diagnosis of ? parotid calculus.

Many surgeons, however, recognised that the condition was often inflammatory in origin and we are indebted in particular to R. T. Payne for the first comprehensive account of recurrent pyogenic parotitis.

In the course of investigation of the present series, however, it became apparent that some of the patients could not be classified under the heading of recurrent infection. They presented a somewhat different clinical picture ; there was a striking difference in the sialogram and this investigation alone often brought about a cure. It became rapidly obvious that they suffered from duct obstruction alone.

The following classification has, therefore, been put forward as a basis for discussion.

TABLE 2

CLASSIFICATION OF RECURRENT SALIVARY SWELLINGS

(1) *Swellings due to obstruction of the salivary flow*
A.—Parotid gland :—
(*a*) Stenotic duct obstruction :—
(i) Papillary
(ii) Buccal.
(*b*) Calculous duct obstruction.
B.—Submandibular gland :—
(*a*) Calculous duct obstruction.
(*b*) Non-calculous duct obstruction.
(2) *Swellings due to diminished salivary flow*
(*a*) Congenital sialangiectasis.
(*b*) Acquired sialangiectasis :—
(i) Recurrent pyogenic parotitis
(ii) Sjögren's syndrome.
(3) *Swellings of mixed origin*
Swellings due to obstruction of the salivary flow :—
A.—Parotid gland.
(*a*) Stenotic duct obstruction.

In the parotid gland the common conditions encountered are stenotic in origin, designated as papillary or buccal according to the part of Stenson's duct involved in the stenosis. This occurs in two age groups,

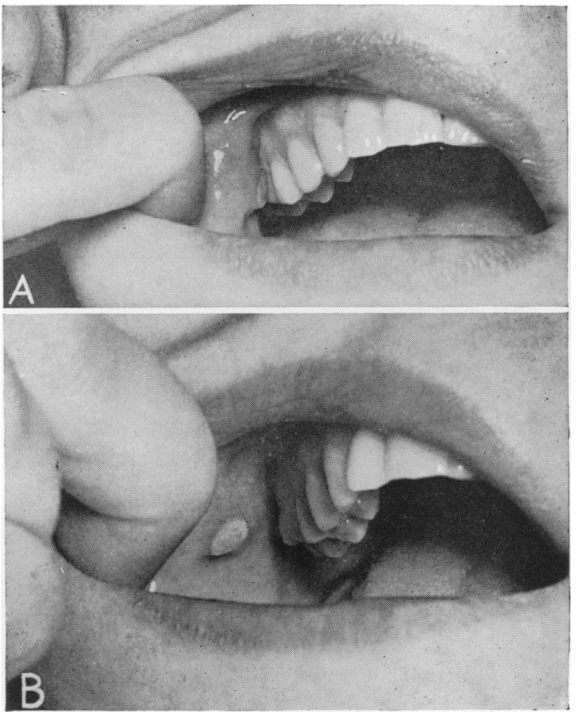

Fig. 15. Traumatic ulcer of the parotid duct orifice.

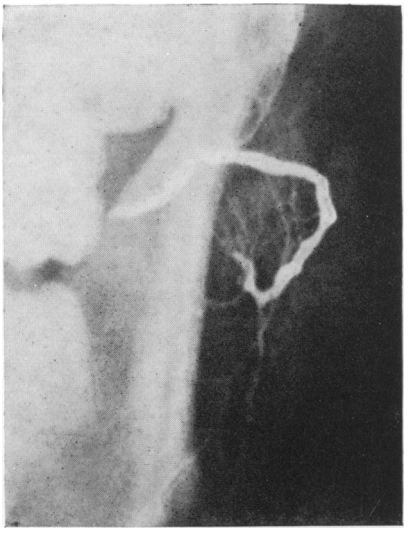

Fig. 16. Papillary duct obstruction.

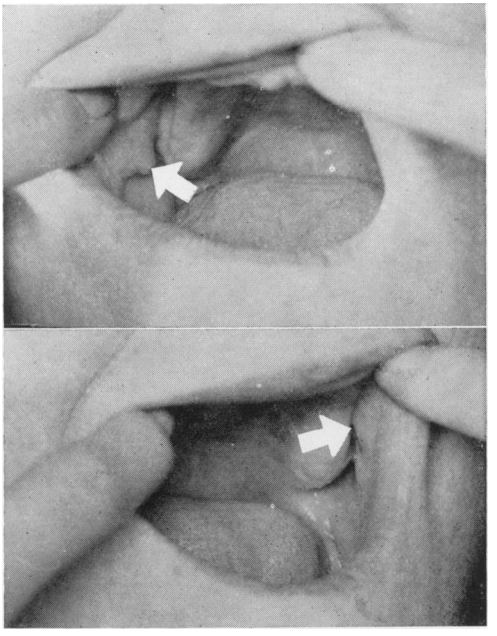

Fig. 17. Hypertrophied papilla of parotid duct.

namely the 18-25 group and the over 40 group. The papillary type of obstruction may be present in both these groups but the buccal type occurs predominately in the older age group as it depends upon the presence of dentures for its formation.

In the younger age groups the obstruction occurs about the time of eruption of the third molar tooth and it is believed to be due to alteration in the normal mechanism of mastication. The food is chewed on the opposite side and the papilla of Stenson's duct is sucked in so that it is traumatised by the bite. I have seen in four patients what I consider to be the earliest lesion. Fig. 15 shows a traumatic ulcer situated over the orifice of the right parotid duct occurring in a girl of 21 years. The relationship of this ulcer to the molar teeth is seen in the upper picture. She also had considerable oedema and tenderness of the gum overlying a partially erupted left lower third molar, *i.e.*, on the side opposite the lesion.

Six months after this ulcer was seen and treated she developed an attack of swelling of the right parotid which recurred two months later. When seen she showed the clinical picture of a classical duct obstruction. She gave a history of swelling lasting for two days occurring with meals and subsiding in the intervening periods. The parotid was diffusely outlined with a soft enlargement. This was confined to the anatomical limits of the gland and the edge was imperceptible. Pressure over the gland produced a sluggish flow of clear saliva without any of the normal pouting of the orifice. A culture of saliva was sterile. Some difficulty was experienced in cannulation of the duct while attempting a sialogram and minimum dilatation was necessary. The resulting appearance is shown in Fig. 16. This demonstrates the stenosis at the end of the duct with a fair degree of proximal dilatation. The gland itself is normal in outline and architecture *and there is no evidence at all of acinar dilatation.*

The pain of the obstructed gland may be described as salivary colic and may be reproduced by raising the pressure in Stenson's duct. The history is usually short and the onset dramatic so that the doctor is consulted in the first or second attack. In this series of 38 cases the average length of history was six months. The attack usually lasts no more than 48 hours and disappears as suddenly as it came. It is usual to find that the patient has had a recent cold or other upper respiratory tract infection and it is likely that the final occlusion is by oedema.

DUCT OBSTRUCTION IN THE OLDER AGE GROUP

These patients are all edentulous. Either the dentures are ill-fitting from the start or have become so as the alveolar margin has absorbed. Examination of the mouth reveals evidence of ulceration in the upper alveobuccal sulcus which may extend downwards onto the buccal mucosa. Occasionally a large hypertrophied duct papilla is seen which either abuts onto the bite or lies in relationship to the abnormally large gap

between the upper dentures and the corresponding alveolar margin (Fig. 17). The papilla may be caught in either of these situations giving rise either to a papillary duct obstruction or to a buccal duct obstruction if the area involved extends posteriorly. Fig. 18 shows the buccal type of obstruction, the fibrotic stenosis having extended backwards along the duct.

On three occasions I have seen what must be the end result of this type of trauma (Fig. 19). The whole of the buccal mucosa overlying the parotid duct has become fused to the upper alveolar margin after long-standing ulceration of the contiguous surfaces. The duct orifice (here indicated by an arrow overlying the alveolar margin and pointing outwards towards the cheek) become involved in scar tissue and eventually stenosis occurs

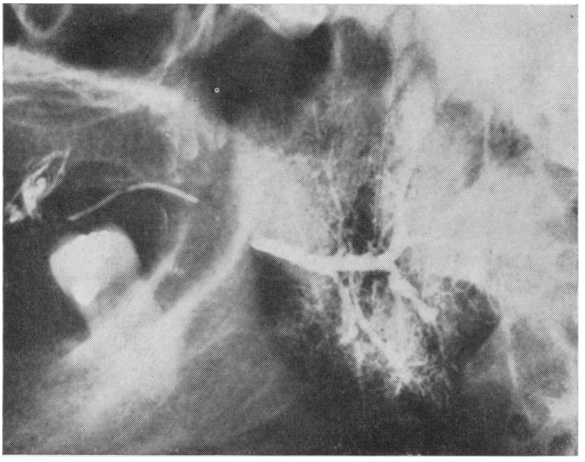

Fig. 18. Buccal duct obstruction.

with consequent parotid swelling. This patient presented in this way. This process takes many years to develop and the three patients mentioned were 76, 78 and 81 respectively.

In this series of 38 duct obstructions, 26 were papillary and nine were buccal, the remaining three being due to calculi.

This leads us to consideration of *calculous duct obstruction* and the *salivary calculus*. Stone in the parotid gland is comparatively rare. In this series of 75 calculi, only three were parotid in origin. The factors accounting for this difference in incidence are numerous. Firstly, there is the *acid-base relationship*. We have seen that the submandibular saliva is more alkaline. Calcium ions are present in saliva and as the alkalinity increases there is a tendency for deposition of the insoluble calcium carbonate and phosphate. If submandibular saliva is left to stand in an open dish, a crust of these salts develops around the edge as carbon dioxide is given off and the pH rises. The mechanism of formation of

dental tartar is similar. Secondly, we must consider the *solid content.* Submandibular saliva has a much greater solid content than parotid saliva, 40 per cent. of the solids being made up of inorganic salts and 60 per cent. are composed of organic constituents, the glycoprotein mucin predominating. The physical deposition of Ca salts has already been referred to, but for a calculus to form *in vivo* it is a general rule that an organic nidus is first deposited. This nidus is produced by the so-called clumping phenomenon in which cellular debris and coagulated mucin became fused to form a salivary thrombus. Micro-organisms may or may not be present. If submandibular saliva is examined these salivary thrombi are usually present, but they are scanty in the parotid saliva, only becoming prominent when the gland is infected.

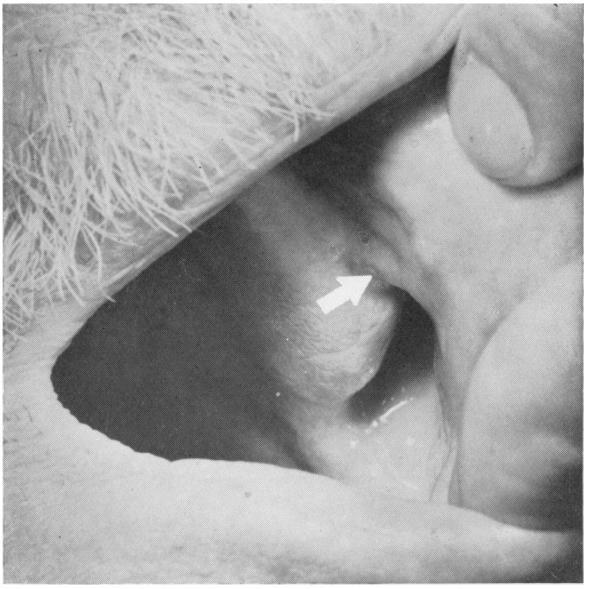

Fig. 19. Fusion of papillary region to the alveolar margin.

The deposition of calcium on to the organic framework of a salivary thrombus produces a microcalculus and at first the process is reversible.

It is likely that many microcalculi are pressed without symptoms from the great majority of submandibular glands. *Foreign bodies.*— Many foreign bodies have been incriminated in the production of calculi, but anyone who has tried to cannulate a salivary duct will realise the odds against a chance entry by a stray particle. Even if such a fortuitous origin were possible it would hardly explain stones in the depths of a gland. The presence of a streptothrix in the centre of the submandibular calculus has *not* been confirmed in this series.

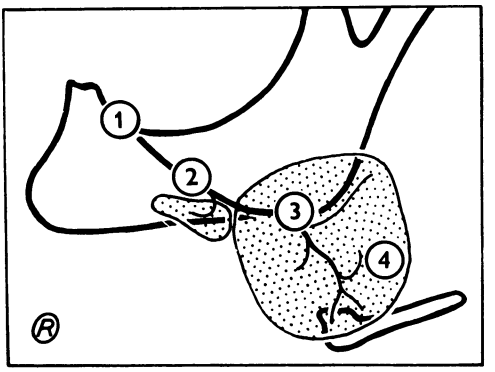

Fig. 20. Sites of submandibular calculi.—(1) Papillary, 50 per cent.; (2) Date-Stone, 7 per cent.; (3) Comma, 27 per cent.; (4) Intraglandular, 16 per cent.

Role of infection

If pus is seen emerging from Wharton's duct, the diagnosis of calculus can be made with absolute certainty in every case. If pus is seen emerging from Stenson's duct, the diagnosis is one of recurrent parotitis—and a stone is rarely found. In the submandibular gland, infection is always secondary to the stone and will clear up when the obstruction is relieved. In the parotid, however, the comparative frequency of infection and rarity of stone leads one to the conclusion that infection alone is not an

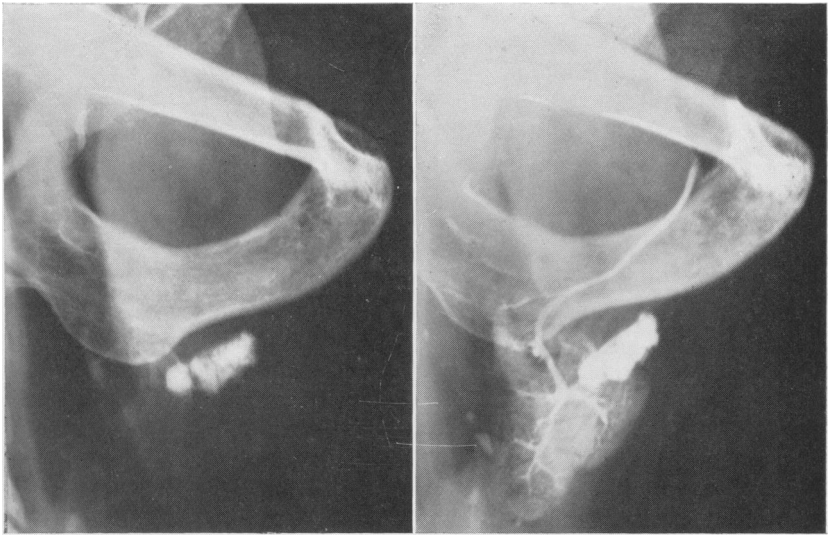

Fig. 21. Comparative plain films and sialogram of a submandibular calculus.

important factor in the production of stone. On the other hand the presence of infection is certainly an aggravating feature. I do not believe that a normal parotid gland can produce a stone in the same way as the submandibular gland. In the three cases I have seen, all have a grossly dilated Stenson's duct system, extending into minor ducts, but not into the acini, thus differing from the recurrent parotitis. In two other cases with grossly dilated ducts of the idiopathic type, the saliva shows the presence of a number of salivary thrombi.

These dilated ducts also have the complicating factors of secondary infection and Sir Cecil Wakeley has pointed out the importance of ammoniacal organisms producing deposition of calcium by raising the pH of the saliva.

Mechanical factors

The free flow of parotid saliva is not only aided by its low viscosity but also by the massage of Stenson's duct afforded by the contraction of the buccinator and movement of the cheeks. Wharton's duct on the other hand, has the drawback of its slightly uphill course with minimal mechanical propulsion.

Clinical features

In the parotid, stones are often multiple, usually form in the duct and are made up of a loosely knit collection of calcium salts, cellular debris and inspissated secretion. They do not seem to form the hard and compact type of stone seen in the submandibular gland but tend to crumble away when rolled between the fingers. They are never more than a few millimetres in diameter.

The symptoms of a parotid calculus are not simply those of a duct obstruction, but depend also on the degree of associated duct dilatation and infection. They resemble in every respect the syndrome of recurrent infection and may persist even after the stone has passed or been removed unless the infection is treated.

Submandibular gland

On the other hand *the submandibular calculus* is primarily responsible for the underlying infection and its removal is followed by complete resolution providing that there are no other calculi. It may be single or multiple and tends to occur in certain sites (Fig. 20). These are :—

(1) *At the papilla*—where a small round stone may impact.

(2) *In the floor of the mouth*—where an elongated stone is gradually built up—the so-called date stone type.

(3) *At the angle of Wharton's duct*—the so called comma calculus with its tail dipping down towards the gland ; and

(4) *In the gland itself*; usually large and irregular.

The sequence of swelling at the commencement of a meal, followed by pain, at first due to glandular distention and later to secondary infection relieved by a discharge of saliva or pus into the mouth is well known and requires no further elaboration.

The plain X-ray is diagnostic but it must be remembered that 20 per cent. of these stones are radioluscent, and require sialography for diagnosis (Feuz). In addition a sialogram may be necessary to ascertain whether a stone lies in the duct or the gland from the point of view of management (Fig. 21).

Swellings due to inhibition of salivary flow

I place recurrent pyogenic parotitis under this heading for I believe this to be the etiological factor in producing this condition. Clinically, the cases of recurrent parotitis form a distinct group and *46* have been studied. The condition occurs more frequently in women, the ratio in this series being 2 : 1. The age of onset is usually the middle forties and is rare below 35. Cases occurring in the adolescent and in early adult life are probably examples of the congenital conditions already described, and may be distinguished by the age of onset. The onset is insidious rather than dramatic and the time between onset of symptoms and consultation is much longer than in the duct obstruction, averaging two years. The swelling is usually confined to one parotid, although there is a marked tendency for both glands to become involved as the disease progresses.

The earliest symptom is slight aching pain in the preauricular or retromandibular region accompanied or followed by a slight degree of localised parotid swelling. The swelling is only related to meals in that the meal has a " trigger " action at the commencement of the attack and the close relationship seen in a simple duct obstruction does not exist.

The swelling may take a day or two to reach its maximum size and the length of attack varies from 48 hours in the early to six weeks or longer in the later cases. The gland at first tends to regain its normal size in between attacks but as these become more frequent, a little residual thickening becomes obvious. The gland then feels distinctly lobulated with a firm palpable edge. The swelling may remain confined to a localised part of the gland, and become so discrete that it is mistaken for a salivary tumour especially when just behind the angle of the jaw in the classical site. The diagnostic difficulties arising from this clinical similarity are readily settled by sialography.

The attacks are infrequent at first, occurring perhaps only once in six months to a year, but the intervals of remission become shorter as time goes on. The end of an attack is often heralded by a discharge of mucopus into the mouth, the patient complaining of an unpleasant salty taste. The saliva, when examined by the smear technique contains masses of mucin, desquamated cells, pus cells and bacteria ; the number of pus cells indicating the severity of the infection. The infecting organism is usually the S. Viridans—in 65 per cent. of the cases in this series. This is often found in pure culture but it is occasionally combined with the Staphylococcus pyogenes. Other infecting organisms are the Pneumococcus, Strep. Haemolyticus, Bacillus Proteus, B. Fusiformis and B. coli,

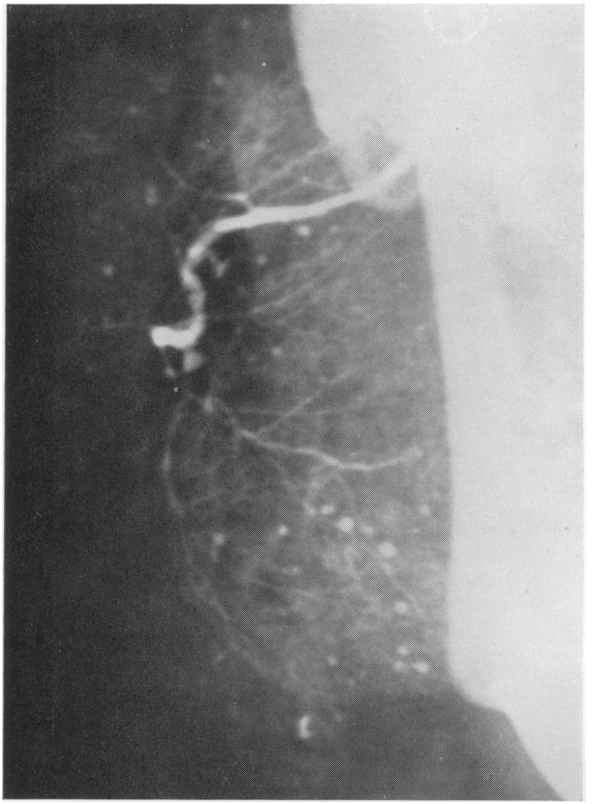

Fig. 22. Early sialangiectasis.

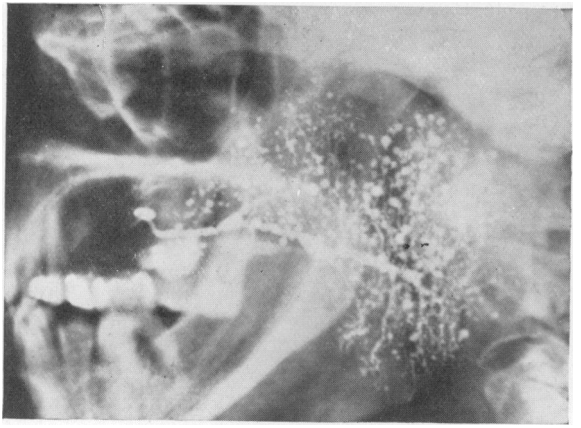

Fig. 23. Fully developed sialangiectasis. Lateral view.

and Diphtheroid in approximate order of frequency. Of these, the Pneumococcal type occurs with sufficient regularity to warrant an additional clinical description—20 per cent. in this series.

These cases are much more severe in their manifestations, presenting a clinical picture which suggests a much more acute type of parotitis than that already described. The degree of pain, swelling and general malaise produced is reminiscent of the acute Staphylococcal parotitis and the similarity may be carried further in the formation of an intraglandular abscess (Payne, 1940). In the S. Viridans infection the saliva during an attack is turbid and contains globules of mucopus and epithelial debris graphically described by Payne as the " snowstorm " saliva. The Pneumococcal cases, however, produce a more frankly purulent saliva with a characteristic greenish tinge. The cellular content of the debris in these cases is much greater and is indicative of the more advanced destruction occurring in the gland. There is also a tendency towards a reversal of the viscosity, the parotid saliva approaching or even surpassing the submandibular saliva in its consistency.

Sialographic appearances

The sialograms in these cases of recurrent infection show characteristic appearances (Figs. 22-26). The S. Viridans infection in its early stages shows a fine globular dilatation of the terminal ductules and acini either localised to one area or scattered throughout the gland, with intervening areas of normal filling—so-called sialectasis or more accurately sialangiectasis (Fig. 22) (Swinburne, 1940).

The main ducts in all early cases are normal and show no evidence of obstruction, but later they become beaded and as infection spreads along them, the lining epithelium is desquamated plugging the larger ducts at

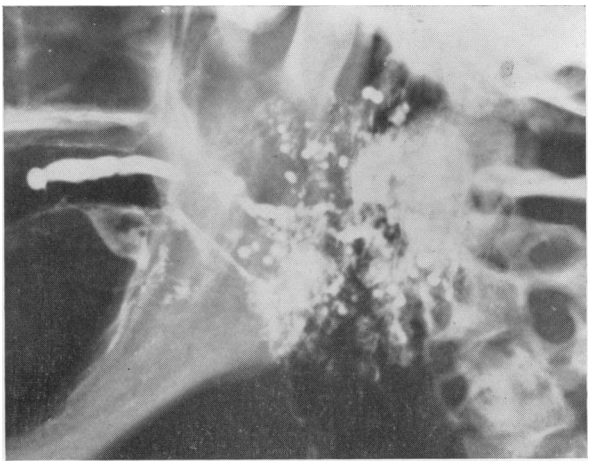

Fig. 24. Pneumococcal parotitis.

intervals and causing dilatation. This gives the appearance seen in Fig. 23, namely a lengthened widened tortuous duct with frequent dilatations and strictures along its course and *marked sialangiectasis.*

The pneumococcal cases show much greater gland destruction than is seen in the S. Viridans case (Fig. 24). Terminal dilatation and duct irregularity is of the grossest character. In these cases there is a tendency for the dilated terminal ducts and acini to coalesce and a solitary abscess cavity may be formed (Fig. 25). Five cases of chronic parotid abscess were found in this series and all were pneumococcal in origin. It will be noticed that there is a clear unfilled area round the abscess cavity due to the surrounding cellulitic infiltration, while the peripheral parts of the gland fill normally.

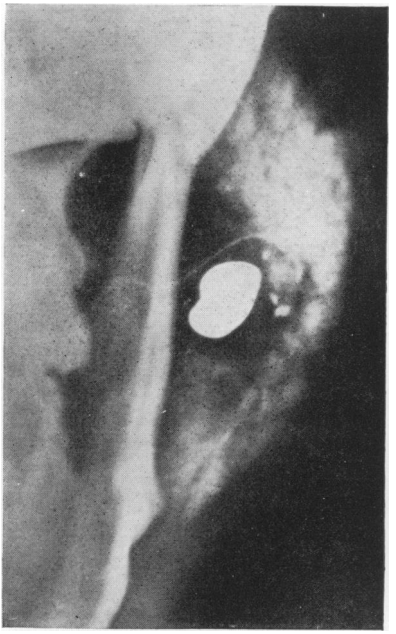

Fig. 25. Parotid abscess.

It is convenient at this point to discuss Sjögren's syndrome because of the light thrown on the etiology of these conditions. It has long been recognised that the syndrome referred to as Mickulicz disease with enlargement of lacrimal, parotid and submandibular glands has included a number of different entities and it remained for Hendrik Sjögren in 1943 to describe a condition in which keratoconjunctivitis sicca and salivary gland swellings were associated. In a masterly monograph on the subject he ascribed the condition to a widespread parasympathetic hypo-activity resulting in dry eyes, xerostomia, achlorhydria, atrophic glossitis and pharyngitis. Scleroderma, Raynaud phenomena and

associated mild rheumatoid lesions of the fingers and hands were also found and the term uveoparotitic polyarthritis was used to describe the condition. A prominent feature of this syndrome is recurrent bilateral parotid swelling with remissions during which the gland fails to return to its normal size. Xerostomia precedes the parotid enlargement and pressure over the gland produces a thick glutinous mildly turbid jelly-like saliva from Stenson's duct which on culture frequently contains S. Viridans. The sialogram shows gross dilatation of the terminal ducts and acini after the manner of the recurrent infection—but with greater accentuation of the picture (Fig. 26). The persistance of globules of Neohydriol after a contralateral sialogram two months previously indicated a gross degree of secretory stasis. In our series of 16, *all* were women in the 45 to 60 age group.

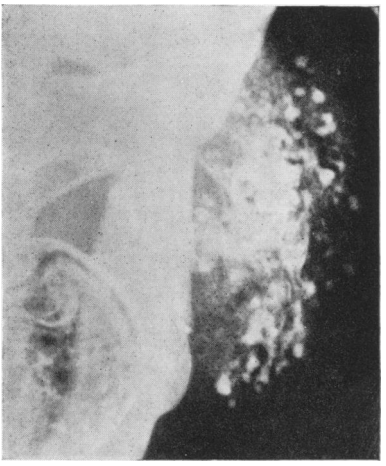

Fig. 26. Sjögren's syndrome.

From the point of view of parotid swelling, it would seem that the sequence of events was an inhibition of parotid secretion with diminution of chloride and enzyme content and a gross increase of viscosity. This considerably diminishes the bacteriostatic activity of the saliva and ascending infection follows. The problem of terminal dilatation is of further interest. While it might be reasonably assumed that this is secondary to ascending infection it might be asked why the acini are first involved and not the ducts. We have seen that back pressure does not cause acinar dilatation in the duct obstruction and also drawn attention to the existence of sialangiectasis of a mild type in the normal or I should say asymptomatic gland of the child with what was considered to be congenital dilatation. This phenomenon of sialangiectasis without clinical infection was repeated in two cases referred from the Ophthalmic Department on account of a xerostomia associated with keratoconjunctivitis sicca.

There was no history of parotid swelling—nor was there any infection of the saliva. A sialogram, however, revealed early dilatation.

This suggests that the dilatation of the acini may appear very early in the disease and is probably due to atrophy of the acinar cells. Once infection is established a vicious circle occurs because of destruction of neighbouring acini with further depression of secretion and further infection. All that has been said of Sjögren's syndrome may be said of recurrent pyogenic parotitis with regard to the formation of dilated acini and it seems probable that the etiology of the latter condition is similar. This is further suggested by the study of a series of cases which contains at one extreme a recurrent swelling of one parotid gland in an otherwise normal patient through cases of bilateral gland involvement which have in addition a hypochlorhydria, or a mild rheumatoid condition or a tendency to sticky eyes and so on—to the fully developed cases of Sjögren's syndrome. Finally, biopsy of the affected glands both in Sjögren's disease and recurrent parotitis give an identical picture of atrophic acini, fibrosis and round cell infiltration (Fig. 27).

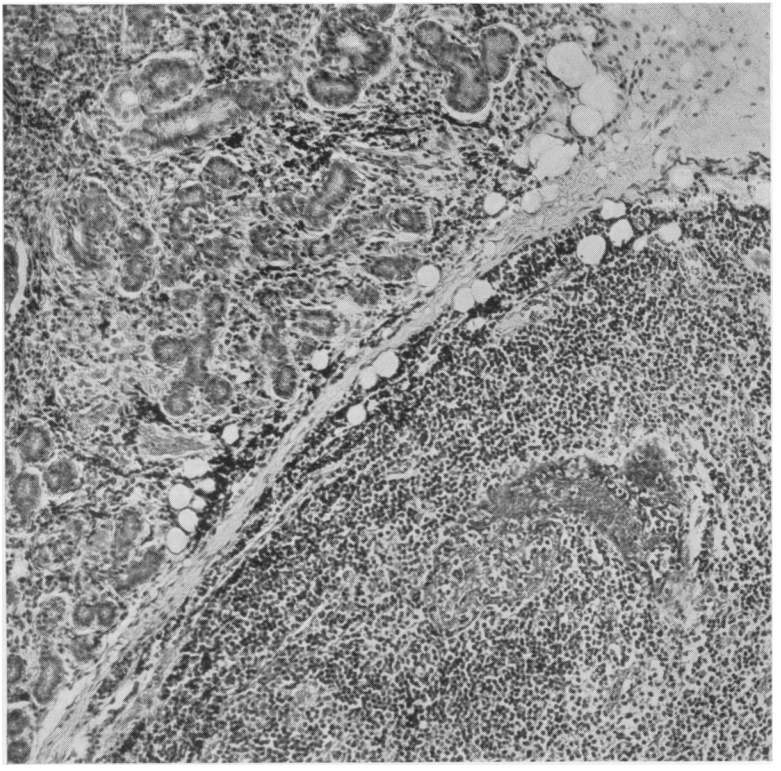

Fig. 27. Sjögren's syndrome. Photomicrograph.

One distinguishing feature of the pure recurrent parotitis is that it tends to occur 10 years earlier than the fully developed Sjögren's syndrome and it may occur in men.

Two questions remain to be answered: What causes the inhibition of secretion ? and what stimulates the recurrent attacks of infection ? The first question is the most difficult and it is probably best ascribed to a central effect, *i.e.*, an abnormally long and intense reflex inhibition as one aspect of the " alarm " reaction using alarm in its broadest sense. Recurrent infection is of course, much more dependent on local conditions and *oral sepsis, reaction to ill-fitting dentures, catarrhal infection* and *any condition which causes drying of the mouth—e.g., prolonged nasal obstruction,* all play their part. The actual precipitating factor is almost certainly the *blocking of a duct*, major or minor with cellular debris or oedema.

CAUSES DUE TO A COMBINATION OF OBSTRUCTION AND INFECTION

A third small group of cases should be included as they are distinguishable clinically. These occur in the aged (70 plus), where there appears to be a combination of buccal or papillary duct obstruction and infection. There is a distinctive history of duct obstruction type with obvious hypertrophy of the papilla with local signs of trauma—but the pain is more severe than expected and the saliva is frankly purulent. A sialogram necessitates dilatation of the duct and shows a dilated duct with the beading of chronic infection—extending into the minor ducts but with no acinar dilatation.

Treatment

(1) *Calculi.* (*a*) *Parotid.*—The danger of fistula and damage to the facial nerve by operative removal of these calculi has actuated a preliminary conservative regime in these cases although their rarity makes it difficult to speak from any great experience.

The first essential is to control the associated infection by the method to be described in relation to recurrent parotitis. In all three cases in this series this was sufficient to bring about a considerable improvement.

Impressed as I was with the loosely knit character of the parotid stone, I was prompted to try the effect of daily lavage of the parotid duct with *10 ccs.* of full strength Suby's solution G. in one case (2 ccs. were injected at a time over a 30 minute period). I was rewarded with a discharge of calcareous debris after two days' treatment and the stone ultimately passed spontaneously about 10 days later. I feel this worth while repeating.

(*b*) *Submandibular calculi.*—So far I have had no success with conservative treatment here and believe that all these stones should be removed. Calculi palpable in the floor of the mouth should be removed through this route provided that it is certain that they are not in the gland (*Vide supra*).

All stones lying in the submandibular gland generally require removal of the gland itself as the recurrence rate in glandular stones is high.

The comma calculi occasion most difficulty but with good intratracheal anaesthesia they can usually be removed through the mouth.

(2) *Duct obstruction.*—These are the cases which were reputedly cured by the sialogram and a single dilatation with the dilators shown in Fig. 2 usually suffices. Dentures are attended to if need be. The treatment chart illustrated here (Table 3) shows that of 38 cases only four required a second dilatation and these were after five and four years respectively. All the others are asymptomatic—the majority having been followed up over a four-year period.

TABLE 3
RESULTS OF TREATMENT

		No.	I.S.Q.	Improved	No Symptoms
Recurrent parotitis:—					
(*a*) Intraduct penicillin	(1 course)	5	—	4	1
	(2 courses)	4	2	2	—
(*b*) Intraduct streptomycin	(1 course)	25	—	2	23
	(2 courses)	6	—	6	—
Sjögren's syndrome:—					
Intraduct streptomycin	(1 course)	14	—	12	2
	(2 courses)	2	—	2	—
Duct obstruction:—					
One dilatation		34	—	—	34
Two dilatations		4	—	—	4

Recurrent infections and Sjögren's syndrome

These conditions may be considered under the same heading as much of the treatment is common to both conditions.

General treatment is directed towards the correction of anaemia, achlorhydria and oral sepsis. A dental opinion is sought if indicated. Patients are advised not to take their food dry and the use of condiments is encouraged. Ordinary chewing gum is of great help particularly in cases of xerostomia and loss of taste. I have found no adequate sialogogue and iodides, although effective in large doses, are not practicable over a long period.

The greatest single benefit to these patients is a daily intraduct injection of 1 *gramme of streptomycin in* 2 *ccs. of solution for six consecutive days*, given a sensitive organism. In addition to its antibiotic effect, it acts by bouginage and lavage of the duct. There may be a slight exacerbation of symptoms after the first injection but simple analgesics are sufficient to relieve the pain. Subsequent injections cause less disturbance and this routine may be carried out in the most severe cases. I now use this treatment in acute *pyogenic parotitis* with good results. I have not incised a postoperative parotitis for three years. It is also used in the *infected congenital sialangiectasis*. Sjögren's syndrome is treated in the same way

always with considerable relief—although there is a much greater tendency to relapse. Residual thickening is often benefited by short wave diathermy. I also find it beneficial in these cases to give small doses of oestrogens, *e.g.*, Stilboestrol 1 mgm. daily. The treatment chart shows the follow-up results.

SUMMARY

The technique of sialography has been described and the normal radiographic anatomy of the salivary glands has been demonstrated.

A classification of the conditions causing recurrent swellings of the salivary glands has been put forward based on the experience gained from a study of 192 cases.

The aetiology of salivary calculi; duct obstruction and recurrent pyogenic parotitis has been reviewed in relation to the physiology of salivary secretion.

The different clinical pictures have been distinguished and a routine treatment established.

ACKNOWLEDGMENTS

I should like to thank the Honorary Surgical Staff at the Manchester Royal Infirmary for sending me their cases so freely when they knew of my interest.

I must also thank my colleagues at the Royal Eye Hospital and Dental Hospital at Manchester who have referred many cases. I am indebted to the Department of Medical Photography at the Manchester Royal Infirmary for putting so much work into the reproductions.

Much credit is due to the Department of Radiology and in particular to Miss E. Hill who has done all the radiography. Finally, I must express my gratitude to Professor A. M. Boyd for his constant stimulation and advice.

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