RECURRENT SWELLING OF THE PAROTID GLANDS

BY

R. S. BRUCE PEARSON, D.M., M.R.C.P.
(From the Hospital for Sick Children, Great Ormond Street, and Guy's Hospital.)

In the following paper a series of cases is described of recurrent swelling of the parotid gland of a type which hitherto has not been clearly recognized. Recurrent swelling, the result of infections of the gland or its ducts is well known as in the cases reported by Payne7 and Pyrah. In the eleven cases now to be described infection seems to play but a small part in the etiology of the condition, which differs in several features from the recurrent pyogenic parotitis of Payne.

Clinical records.

The main features of the cases, summarized in table I, are as follows:—

The majority are children, seven of the series being under twelve years of age, eight are males and four females. The onset usually occurs without obvious predisposing causes. The swelling affects one or both parotid glands: it may be bilateral from the start, may affect one side before the other or remain unilateral, and always appears to involve the entire gland. It varies in size, not only in different individuals but in the same patient in different attacks, from a slight fullness of the face to a swelling sufficiently prominent for a mistaken diagnosis of mumps to be made. It may occur daily over a long period or recur at intervals of months or years, and may last for only a half an hour or persist for several days. The swelling develops quickly and the onset not infrequently coincides with the taking of food. Acid foods in particular are often mentioned as precipitating factors. In two patients (no. 5 and 8) the consumption of certain definite foods was thought to be associated with attacks but in the remainder there was no apparent connection with specific articles of diet, and the association of swelling with eating seems merely to depend on the increased secretory activity accompanying mastication. In this respect the history resembles that of stone in Stenson's duct, which must always be excluded.

The development of the swelling is usually accompanied by a feeling of tension and this may give rise to actual pain. In many cases, however, pain and local tenderness are absent, and pressure can be applied over the swollen gland without evoking signs of distress. It is uniformly firm and smooth to the touch and the skin over it is neither reddened nor hot. The
<table>
<thead>
<tr>
<th>Case</th>
<th>Age in years and sex</th>
<th>Duration of symptoms in years</th>
<th>Unilateral or bilateral</th>
<th>Frequency</th>
<th>Precipitating factors</th>
<th>Infection preceding first attack</th>
<th>Parotid secretion</th>
</tr>
</thead>
<tbody>
<tr>
<td>F. E.1</td>
<td>M.</td>
<td>7</td>
<td>Bilateral</td>
<td>2-3 annually. Recently every month</td>
<td>Colds sometimes</td>
<td>None</td>
<td>NON-INFECTED Clear saliva containing occasional epithelial cells during attacks</td>
</tr>
<tr>
<td>F. C.2</td>
<td>M.</td>
<td>1½</td>
<td>Bilateral</td>
<td>Monthly</td>
<td>Acid food or fruit</td>
<td>None</td>
<td>Clear during attacks: not microscoped</td>
</tr>
<tr>
<td>M. C.3</td>
<td>F.</td>
<td>19</td>
<td>Bilateral</td>
<td>Daily now occasionally</td>
<td>None</td>
<td>None</td>
<td>Not seen during attack, clear in interval</td>
</tr>
<tr>
<td>C. P.4</td>
<td>M.</td>
<td>3½</td>
<td>Bilateral</td>
<td>Every 3 wks. in summer. Occasionally in winter</td>
<td>None</td>
<td>None</td>
<td>Clear during attack: not microscoped</td>
</tr>
<tr>
<td>M. P.5</td>
<td>F.</td>
<td>8</td>
<td>Bilateral</td>
<td>Severe every 4 weeks. Slight in winter</td>
<td>Taking food especially fish and acids</td>
<td>None</td>
<td>Clear saliva containing plugs packed with eosinophils during attacks</td>
</tr>
<tr>
<td>P. P.6</td>
<td>M.</td>
<td>3</td>
<td>Bilateral</td>
<td>3 or 4 a year. Formerly more frequent</td>
<td>None</td>
<td>With pneumonia</td>
<td>Clear during an attack. Dry mouth</td>
</tr>
<tr>
<td>D. L.7</td>
<td>M.</td>
<td>½</td>
<td>Bilateral</td>
<td>2 attacks</td>
<td>None</td>
<td>None</td>
<td>No secretion seen, mouth dry</td>
</tr>
<tr>
<td>J. M.8</td>
<td>F.</td>
<td>6</td>
<td>Left</td>
<td>Monthly, more often in summer</td>
<td>None</td>
<td>None</td>
<td>Clear fluid while swelling is present. Not microscoped</td>
</tr>
<tr>
<td>L. H.9</td>
<td>M.</td>
<td>5</td>
<td>Left</td>
<td>2 swellings only</td>
<td>None</td>
<td>None</td>
<td>Clear during attacks. In interval a few epithelial cells</td>
</tr>
<tr>
<td>M. H.10</td>
<td>M.</td>
<td>3</td>
<td>Right</td>
<td>3 swellings</td>
<td>None</td>
<td>None</td>
<td>Clear and scanty during attack. Not microscoped</td>
</tr>
<tr>
<td>J. F.11</td>
<td>M.</td>
<td>9</td>
<td>Right</td>
<td>1 attack 2 weeks ago, for last 3 wks. daily</td>
<td>None</td>
<td>None</td>
<td>Clear during attacks. No cells</td>
</tr>
<tr>
<td>N. K.12</td>
<td>M.</td>
<td>6</td>
<td>Bilateral</td>
<td>1 or 2 annually</td>
<td>Acid foods</td>
<td>Measles</td>
<td>INFECTED Pus and debris and strept. Purulent during attacks. Clear during interval</td>
</tr>
<tr>
<td>L. L.13</td>
<td>M.</td>
<td>1½</td>
<td>Bilateral</td>
<td>5 attacks</td>
<td>Sore throats</td>
<td>Scarlet fever</td>
<td>Mouth dry and no secretion seen</td>
</tr>
<tr>
<td>J. P.14</td>
<td>M.</td>
<td>½</td>
<td>Bilateral</td>
<td>Recently daily, formerly only on occasions</td>
<td>Food</td>
<td>None</td>
<td>Turbid during attack. Pus and epithelial cells</td>
</tr>
<tr>
<td>F. S.15</td>
<td>M.</td>
<td>2</td>
<td>Bilateral</td>
<td>Every fortnight</td>
<td>Cold temperature</td>
<td>None</td>
<td>Pus and debris. Epithelial cells</td>
</tr>
<tr>
<td>B. H.16</td>
<td>F.</td>
<td>1½</td>
<td>Left</td>
<td>Daily variations</td>
<td>Worse on waking</td>
<td>None</td>
<td>Clear. Epithelial and occasional pus cells seen under microscope</td>
</tr>
<tr>
<td>S. B.17</td>
<td>M.</td>
<td>2</td>
<td>Left</td>
<td>Irregular from 2 wks. to 1½</td>
<td>None</td>
<td>Followed otitis media on same side</td>
<td>Purulent during attack. Clear in interval</td>
</tr>
<tr>
<td>CASES.</td>
<td>Signs of infection</td>
<td>Allergy</td>
<td>Sialogram</td>
<td>Remarks</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>--------</td>
<td>-------------------</td>
<td>---------</td>
<td>-----------</td>
<td>---------</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Personal</td>
<td>None</td>
<td>Injection of lipiodol caused immediate swelling of glands which remained swollen for some hours</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Family</td>
<td>Dilatation of terminal ducts</td>
<td>Eosinophilia 10 per cent. Swelling of left side preceded that of right by some mths</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Grandfather asthma</td>
<td>Dilatation of terminal ducts</td>
<td>Sister of Case-J.P. Not observed during attacks.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Grandfather asthma</td>
<td>Dilatation of terminal ducts</td>
<td>Pap. urt. in summer only and is then often coincident with parotid swelling. 9 per cent. eosinophilia</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patulous</td>
<td>None</td>
<td>Asthma, angioneurotic oedema, hay fever</td>
<td>Dilatation of terminal ducts</td>
<td>The severe attacks coincide with the severe attacks of asthma. Sialogram see figure 1, a</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Asthma</td>
<td>Dilatation of terminal ducts, Bilateral</td>
<td>Pyrexial with attacks—mouth dry in attacks—swelling lasts 3 days; one side usually precedes the other</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Mother migrane</td>
<td>Normal</td>
<td>Tonsils removed 3 mths, before first attack. Mouth dry and pyrexial when seen in attack</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Asthma, eczema</td>
<td>Swellings are often associated with colic and are then more severe. Sialogram see figure 1, a</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Mother eczema. Uncles rhinorrhoea</td>
<td>Normal (L).</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Grandmother asthma</td>
<td>Swellings persist for a week. Child feels ill during swelling and is pyrexial. Right side only was affected for first 4 years</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Mother hay fever</td>
<td>Normal (L).</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CASES.</td>
<td>Not seen in attack</td>
<td>Personal</td>
<td>None</td>
<td>Swellings persist for a week. Child feels ill during swelling and is pyrexial. Right side only was affected for first 4 years</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Reddened, patulous duringattack</td>
<td>Tender</td>
<td>Pap. urticaria, pork idiosyncasy</td>
<td>Normal</td>
<td>Pyrexial during attacks. Dry mouth and cracked lips. Attacks last a week and associated with sore throat. 9 per cent. eosinophilia</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>Slight heat and tenderness over glands</td>
<td>Personal</td>
<td>None</td>
<td>Glands palpable between attacks. Brother of Case 3. 5 per cent. eosinophilia. Sialogram, see figure 1, c</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Reddened duringattack</td>
<td>Local heat and tenderness</td>
<td>Personal</td>
<td>None</td>
<td>Attacks often associated with colds</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>None</td>
<td>Pap. urticaria</td>
<td>Dilatation of terminal ducts, Bilateral</td>
<td>Swelling persisted with fluctuations for 6 weeks in first attack</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>Redness and tenderness reported by mother</td>
<td>Personal</td>
<td>None</td>
<td>Swellings usually last for several days. 7 per cent. eosinophilia</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
regional lymph glands are not enlarged and trismus is not present. The parotid gland itself cannot be palpated between the attacks. The condition of the mouth is healthy, and only two patients complained of dryness during attacks (no. 6 and 7). The openings of Stenson's ducts are not reddened: they are sometimes difficult to detect, as in some normal subjects, but they can always be rendered obvious by introducing lemon juice or an acid sweet into the mouth for a few moments. Clear saliva is seen to escape from the duct orifices in the majority when the swelling is present, and this may sometimes be increased by pressure over the affected gland. In other cases, however, no flow of saliva from the parotid duct was seen at the time of the swelling.

Sufficient observations have not yet been made to decide whether these variations depend on the stage at which the swelling was observed, that is during its development, at its height or during subsidence, or whether the findings are constant in the same individuals on different occasions or not. Microscopical examination of the fluid obtained from the duct while the swelling was present was carried out in several instances and showed that the saliva was either quite clear or contained a few epithelial cells, and differed in no way from fluid obtained similarly between the periods of swelling. Case no. 5 is an exception and the findings in her case are described fully later. Mantoux tests (o.t. 1/1000) were carried out in four of these cases and a positive result was obtained in one (no. 6).

In every case radiological examination of the parotid duct and its branches was carried out after the injection of lipiodol. A hypodermic syringe fitted with a blunt ended 20-gauge needle or a lachrymal duct cannula was employed. Lipiodol was injected until a feeling of tension was experienced in the region of the parotid gland: this generally occurred after 0·3–0·6 c.c. had been injected, but one patient (no. 5) retained over 3 c.c. At this point when more was injected, the lipiodol could be observed escaping from the duct along the side of the needle. On withdrawing the latter, escape of lipiodol was prevented by placing a finger inside the mouth over the opening of the duct, and exerting counter-pressure against it on the outside of the cheek with the thumb. The finger was removed from the mouth immediately before exposing the x-ray plate: in those patients in whom the swellings were bilateral both sides were examined in this way, usually on separate occasions, and bilateral examinations were also carried out in those cases of unilateral swelling in which the appearance on the affected side was thought to be abnormal. In cases of unilateral swelling in whom the x-ray appearance on the affected side appeared to be normal, the other side was not examined.

Of the eleven cases forming the group under discussion, bilateral swellings were present in seven. Five of these showed small dilatations of the terminal ducts or acini (see fig. 1). One (no. 5) showed gross dilatations of the main duct and its branches: the finer ducts either failed to fill or appeared normal (see fig. 1). In one case (no. 7) the appearance was regarded as normal. Of the remaining five cases, in which the swelling
RECURRENT SWELLING OF THE PAROTID GLANDS

SIALOGRAMS.

FIG. 1.

a. Case no. 8. Sialogram showing normal appearance.

b. Case no. 4. Sialogram showing dilatation of terminal ducts.


d. Case no. 5. Sialogram showing gross dilatation of main and branch ducts.
was limited to one side, the terminal ducts were dilated in one (no. 11), while the appearance on the other side was normal: the other four cases showed no abnormality (see fig. 1). It is clear that apart from the fact that the appearance of the ducts is frequently abnormal, particularly in cases with bilateral swellings, no constant radiological features are shown.

In order to exclude the possibility of over-distension with lipiodol being responsible for the small terminal dilatations referred to, radiological examination of the salivary ducts was also carried out by the same method, in ten control cases between the age of five and twelve years, and one aged thirty years. In all these the findings corresponded with what has been regarded as normal by other authors, notably Pyrah and Allison who state that they too have investigated a number of controls.

A striking feature of these cases is the frequent association with conditions usually referred to as ‘allergic.’ Thus eight of the eleven patients in the series are or have previously been the subject of some allergic manifestation: asthma occurs in four cases (no. 5, 6, 8, and 9), papular urticaria* in two (no. 4 and 11), hay fever in three (no. 5, 8, and 10), while spasmodic rhinorrhoea (no. 2), angioneurotic oedema and urticaria (no. 5), and eczema (no. 9) are each present in one case. In one patient (no. 8) attacks of colic following the consumption of certain foods was thought to be due to sensitization. In five of these cases there was in addition a family history of allergy and this was also obtained in one other (no. 7) so that nine patients had a personal or family history of allergy. A raised eosinophil count in the blood was observed in several cases. In the majority there was no evidence of any relationship between the development of the swellings and the occurrence of the allergic manifestations, but in three cases, referred to later the two forms of attack frequently coincided.

These are the main features of this group of cases, and the following case histories of individual patients serve to illustrate them more fully.

**Case no. 1.** F. E. was a boy aged eight-and-a-half years. There was nothing of interest in his family history and his health has been good except for an attack of measles. Since the age of one year he has had swellings in the region of the parotid glands on both sides; these occur once or twice a year. The swelling is sometimes limited to one side. It develops suddenly and may last for a day or more. Colds appear to predispose to attacks. On examination there was bilateral swelling of the parotid glands (see fig. 2), which were firm but not tender to the touch. There was no heat or reddening of the skin. The mouth was clean and clear saliva was seen to issue from the duct orifice, which appeared normal: microscopical examination of the saliva obtained from the duct at the time of the swelling, revealed the presence of a few epithelial cells but no other abnormality.

* Papular urticaria and migraine are for the purposes of this paper regarded as allergic states, though it is recognized that there are other factors probably of greater importance in their etiology.
RECURRENT SWELLING OF THE PAROTID GLANDS

One month later the mother reported that the swelling had occurred every morning for the past week subsiding again during the day. The week following, no swelling was apparent and bilateral radiological examination following injection of lipiodol into Stenson's ducts, revealed bilateral terminal dilatations. This procedure was followed within ten minutes by rapid swelling of both glands, which subsided within the next half hour but remained visible for two days. When seen five months later slight degrees of swelling had occurred several times, but had caused no inconvenience. The boy's general health was good.

FIG. 2.

F. E. Photograph illustrating appearance during moderate swelling of parotid glands.

The following three cases are those already referred to, in whom there was a clearly recognizable relationship between the parotid swellings and the associated allergic conditions.

Case no. 4. C. P. was aged five-and-a-half years. A grandfather suffered from asthma. The swelling was bilateral and of moderate size but was more pronounced on the right side than on the left. It began at the age of one year and ten months and
Case no. 3. J. M. was aged 26 years. His mother suffers from eczema, and several members of his father's family have spasmodic rhinorrhea. He is an only son and lives on a farm. At the age of fifteen, he had eczema of the arms and axillae. He has had hay-fever in the early summer for ten years, and for the past three-and-a-half years has suffered from asthma. As a child he suffered from 'bilious attacks' and these have continued into adult life. They may come on after eating pork or dripping, but also occur without any recognized precipitating factor. The attacks begin with colicky pain in the left iliac fossa, and lead to vomiting and sometimes diarrhoea, with the passage of much mucus. At the same time the left parotid gland swells and may remain up for two or three days. The swellings also occur apart from these attacks, and are particularly frequent during the hay-fever season. He gives no skin reactions to foods, but gives large reactions to horse dander, and smaller ones to chicken feathers, cat hair, dog hair dust extracts and pollens (intradermally).

Examination of this patient was possible on one occasion only. At this time there was slight swelling in the region of the left parotid gland. The orifice of Stenson's duct was normal and clear saliva was seen to pass into the mouth. The x-ray appearance of the ducts was normal.

Case no. 5. M.P. was aged 52 years. A brother has eczema and several cousins hay-fever. The patient has had hay-fever for many years. The parotid swellings which are bilateral have occurred intermittently for eight years; she has had attacks of asthma for a slightly shorter period. The latter are usually accompanied by the swellings which become hard like cricket balls and are painful. During the time that these severe attacks are present, the pain is greatly increased if pressure is applied over the gland; after a few hours however a sensation of subjective improvement is experienced, and at this time pressure causes the expulsion of a stream of saliva into the mouth with further relief of symptoms. In between the major attacks, which occur at intervals of several weeks, minor degrees of swelling are frequently noticed, and mastication of food or even the thought of it may be sufficient to precipitate these. The patient has observed that eating fried fish is particularly apt to be followed by an attack of asthma, with swelling of the floor of the mouth and associated parotid swellings. Occasionally the tissues around the orbit become swollen in addition. This appears to be due to angioneurotic oedema rather than any swelling of the lachrymal glands. Recently she had noticed swelling on both sides in the floor of her mouth. These have not been seen but from her description would appear to be swellings of the sub-lingual glands.
RECURRENT SWELLING OF THE PAROTID GLANDS

When first examined there was a soft swelling of moderate size involving both parotids; the orifices of Stenson's ducts which were slightly patulous were clearly seen on opening the mouth, and this act caused the ejection of clear saliva. On several occasions pressure over the glands caused a plug of viscid material to be ejected, followed by a gush of several cubic centimetres of clear fluid.

Examination of the plugs showed that these were about three cm. in length and the thickness of a piece of string. When spread on a slide and examined microscopically, they were found to consist of a clear matrix closely packed with cells. These were almost entirely mature, perfectly formed, large granulated eosinophils. A count kindly carried out by Dr. F. H. Knott gave the following picture:—Eosinophils, 96 per cent.; polymorphs, none seen; endothelial cells, 2 per cent.; and small round cells, 2 per cent. Epithelial cells were seen in small numbers, but no organisms could be detected in a direct smear.

The parallel between these parotid plugs and the Kuschmann's spirals seen in asthmatic sputum seems to be close.

Discussion.

It is clear from what has been said that these cases differ from those previously described by Pyrah and Payne. In the first place their patients were considerably older; the average age at onset in Payne's nineteen cases of recurrent pyogenic parotitis is twenty-nine years, the earliest being eleven years, while Pyrah's four cases are between sixteen and forty-seven years of age. Secondly, the parotid swelling was accompanied in their cases by clear signs of inflammation, such as heat, redness and pain, reddening of the orifice of the duct, a foul taste in the mouth and the passage of purulent or turbid saliva. There was also induration of the gland which rendered it palpable in the intervals between periods of swelling. Moreover in these cases the condition almost invariably began unilaterally and later spread to involve the opposite side. In five of the present series of cases the condition was bilateral from the beginning. Lastly, neither Pyrah nor Payne report association of the parotid swellings with allergic conditions though they do not state if this association was sought for.

It therefore seems justifiable to seek some explanation other than infection for the origin of the recurrent swelling in the present cases. From this point of view the frequent association with allergic conditions in the personal and family history is of special interest. This frequently is so high as to point to more than a chance relationship. In addition the intimate associations between the occurrence of the swellings and other allergic phenomena in three of the cases make it reasonable to suggest that the recurrent swellings of the parotids may themselves be an allergic manifestation while the presence of eosinophil-containing plugs in the parotid secretion makes this likelihood still greater in one case.

The conception of allergy as an explanation for parotid swellings is not new. Vogeller in 1922 described a case of recurrent parotid swelling
occurring in a woman of forty-three. When this began seven years before, it was sudden in onset and painless: relief was obtained by expressing a white plug from the duct. Later the swelling became associated with fever and pain, and enlargement of neighbouring lymph glands. A blood count showed an eosinophilia of 11 per cent. He comments that the original incidence of swelling indicates some obstruction: the plug when removed was not followed by copious secretion or immediate subsidence of the swelling. Obstruction, therefore, does not entirely explain the condition. He believes that the swelling of the gland occurs as a result of a process similar to angioneurotic oedema of the subcutaneous tissues. He also suggests that later, secondary infection from the mouth has occurred leading to destruction of the glandular tissue with sialodochitis. The condition is likened by him to asthma. Burton Fanning in 1925 described a case of recurrent parotid swelling in a man aged sixty-two years associated with a dry mouth and attacks of rhinorrhea; on one occasion an injection of adrenaline cut short an attack. More recently Meyer has described under the title of 'chronic sialodochitis' the case of a child of six years in whom parotid swellings were associated with passage of ropy saliva. The swelling was usually unilateral but affected opposite sides in different attacks. There were no signs of inflammation but various organisms were grown from cultures of the parotid saliva; it is difficult to see that these were obtained under strictly aseptic conditions. Positive skin tests to spinach, bacon and paprika were obtained, and removal of these from the diet together with chocolate, which was suspected on clinical grounds, was followed by freedom from the swellings for the following six months. While under observation, sialography showed dilatations of the minor ducts. The mother and great-grandmother of this patient suffered from similar swellings. Although it has not been possible to find other records in which the authors have specifically cited allergy as a possible cause, several cases are recorded in which it seems possible that the condition was allergic in origin. One of the most interesting is the case described by Kussmaul in 1879, and referred to by him as a case of 'fibrous sialodochitis,' a term he was the first to use. The patient was a woman of thirty-two years who had a four years' history of unilateral recurrent swelling of the parotid. Her only previous history was one of frequent pain in the jaw on that side following trauma as a result of removal of a tooth. The swellings were at first painless, and might last for a period of half-an-hour only, but later they became hot and painful and drops of pus exuded from Stenson's duct, as though infection had been superadded. The swelling subsided with expulsion of what is called a 'fibrous plug.' This was packed with 'masses of round cells' which resembled 'fresh pus cells.' Larger epithelial cells were present in smaller quantities and the presence of a Charcot-Leyden crystal was also noted. The extrusion of this plug was followed by a gush of clear saliva. Kussmaul argues that the gland itself was clearly not infected and regards the condition as similar to a chronic fibrous bronchitis. This case resembles in many ways the cases of Vogeller and Meyer and of M. P. (no. 11) in our series. It would appear that as in Vogeller's case, infection was superadded after the condition had become established. It may well be wondered whether many of the cells seen in the plugs of this case were not in fact eosinophils—the use of the word 'fresh' (frisch) applied to pus cells, the presence of a Charcot-Leyden crystal, and the fact that Ehrlich's method of differential staining was not then in use make it likely. Ipscher in the same year records a similar condition affecting the submaxillary duct. Von Reuss describes a patient, of sixteen years in whom parotid swellings occurred during the monthly period and in whom there was an eosinophilia, and in addition a girl of eight in whom parotid swellings were associated with abdominal colic. Londe and Pelz also describe the
RECURRENT SWELLING OF THE PAROTID GLANDS

case of a child in whom the appearance of unilateral parotid swelling, and attacks of abdominal colic were common features though they are not described as occurring coincidently.

Even among cases presenting signs of infection, an association with allergy is not uncommon. In addition to the eleven cases described, six others have been observed in whom there was evidence of infection (see Table). In four of these, pus cells were found in the parotid secretion which was clearly purulent in three (no. 12, 15 and 17), turbid in one (no. 14), and apparently clear in the fourth (no. 16). In the fifth case (no. 18) swelling always coincided with tonsillitis and the lips were dry and cracked: these attacks lasted for a week at a time and the patient was pyrexial during this time. No saliva could be obtained from the parotid duct during the presence of the swelling. In the last case (no. 12) there was a close but not invariable association with exacerbations of otitis media. Other signs of infection such as reddening of the duct orifice and heat and tenderness in the neighbourhood of the parotid gland were noticed in some of these cases.

Three of these infected cases (no. 14, 16 and 17) suffered from papular urticaria, one of them having in addition an idiosyncrasy to pork which was said invariably to cause vomiting. Two of these patients and one other gave a family history of allergy (no. 15, 16 and 17). The following cases show further points of special interest in this connection, and suggest that factors other than infection play a part.

Case no. 15. F. S., gave a history of swellings which were of two quite distinct kinds, namely those which persist for several days, were painful and tender and associated with an unpleasant taste in the mouth, and those which lasted for periods as short as twenty minutes and were not accompanied by any of these signs. Exposure to cold appeared to precipitate the short attacks: this factor is mentioned by Osler and Macrae in their brief reference to acute swellings of the parotid gland. The saliva obtained from the left parotid duct during one of the severe periods was found to contain numerous pus cells, streptococci and clumps of eosinophils. A further specimen obtained three days after one of the short attacks contained a very few epithelial cells only. It is thought that the presence of eosinophils in the saliva, and the history of the brief periods of swelling, taken in conjunction with the presence of asthma in a younger brother offer strong support for the belief that infection has been superimposed on an allergic basis.

In the following patient there is also evidence that infection may not be entirely responsible for his condition:

Case no. 16. J. P., has had swellings for over a period of four years. These have altered in character recently, having become more painful, and tender, and lasting for longer periods so that some degree of swelling is almost constantly present. The saliva was clear when the patient was first seen, but on one later occasion was turbid and contained pus cells. A blood picture showed the presence of 5 per cent. eosinophils. Moreover his sister, now a married woman of twenty-six years (no. 3) has suffered from a similar condition since childhood. The swellings have been slight.
and infrequent for some years in her case, and she presents no signs of infection. A familial factor is thought to be present. It will be recalled that in Meyer's case a family history was also obtained.

Sialographic examination of these patients gives findings similar to those of the uninfected group, except that in two cases (no. 12 and 14) in addition to the terminal dilatations, there is some dilatation of the main and branch ducts. It is suggested that this is due to the infective process. It is also of interest with regard to patient no. 16, that although the swelling of which she complained was limited to the left side, terminal dilatations were revealed on both sides. In another case, in whom there was a unilateral parotid swelling associated with signs of inflammation and secretion of pus from the duct, bilateral dilatations were also observed.* This may well indicate a pre-existing bilateral condition of obstruction of the smaller ducts, on which infection has supervened on one side.

It is clear that radiography is of no value in determining the presence or absence of infection, since similar appearances are obtained in each group.

It seems probable that infection in at any rate some of these cases, occurs secondarily, and may well be superimposed on a condition of an allergic origin. This sequence of events has not been excluded in the cases of Pyrah and Payne, and may have been present in some of them.

Consideration of recorded cases, together with the observations carried out on the series, discussed in this paper, afford strong support for the belief that there exists a group of patients in whom recurrent swelling of the parotid gland is of allergic origin. At present this conception must remain a suggestion only, since as yet there is little understanding of the conditions called allergic, nor is there clear evidence as to the mechanism of the parotid swellings. It is also recognized that other causes of recurrent swelling undoubtedly occur. Apart from those cases in whom infection appears to be responsible, others are recorded in whom neither infection nor allergy seems to play a part. Such a case is that reported by Wolff12 of an infant with enterospasm in whom manipulation of the stomach and intestines was associated with the sudden development of swellings: the author's suggestion of reflex spasm offers the most reasonable explanation. Von Steinitz11 also records a case in which he believes increased viscidity of saliva may be responsible for the development of the swelling. Nevertheless, it is believed that, cases belonging to the present group are by no means uncommon. The frequent association of allergy in the patients and their relatives, the apparent absence of infection, the coincidence of the attacks of swelling with the associated allergic phenomena in three cases, and the presence of eosinophil-containing plugs in one of them are the chief grounds upon which the association with allergy is believed to be important. The

---

* This case is not included in the present series because the swelling had occurred on one occasion only.
frequent bilateral onset of the condition, the rapid development of the
swelling, the presence of two cases in one family, and evidence that, even
in six cases with infection, other factors were also present, are minor points
of similar significance.

A number of possible factors come into mind as the immediate cause for
recurrent parotid swellings. The enlargement may occur as a result of
recurrent infections of the gland tissue itself, of the lymphoid tissue within
it, or as a result of angioneurotic oedema. There may be obstruction of the
duct by spasm, oedema of the lining membrane, or the presence of
abnormally viscid saliva, mucous plugs, plugs of pus and epithelial debris
or stone. Achalasia of the duct must be considered. If spasm occurs it
may be due to the direct effect of some locally-formed substance on the
muscle fibres, or as a result of a reflex mechanism. In the group of eleven
cases described in this paper, there is reason to believe that infection plays
no part. If this is excluded, the remaining mechanisms, with the exception
of achalasia and stone, for neither of which is there any evidence, might all
occur as the result of allergic processes. It seems probable that obstruction
to the escape of saliva in the course of the parotid ducts is in most cases the
immediate cause of the swelling. The tendency for it to develop during meal
times, and the frequent presence of dilatation in some part of the course of
the ducts, as shown by x-ray examinations after injection of opaque material
support this contention.

Summary and conclusion.

(1) Eleven cases of recurrent swelling of the parotid gland are described,
these are characterized by the absence of evidence of infection, and the
frequent history of allergy.

(2) In three of the patients allergic manifestations frequently coincided
with the development of the swellings. In one of these the passage of plugs
containing large numbers of eosinophils from the parotid duct is described.

(3) It is suggested that these swellings may be allergic in nature.

(4) The possible mechanism of the swellings is discussed and obstruction
of the ducts is thought to play a prominent part.

(5) Five cases in whom there is evidence of infection are also described.

(6) It is suggested that infection may in some cases be superimposed
on an allergic basis.

My thanks are due to the physicians and surgeons of the staffs of
Guy's Hospital and the Hospital for Sick Children, Great Ormond Street,
who have allowed me to make use of their cases.
REFERENCES.

6. Osler, W., & Macrae, T., Modern Medicine, Lond., 1926, III, 278.