Recurrent unilateral swelling of the parotid gland

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SUMMARY The clinical features of 109 patients with recurrent unilateral parotid swelling (24 patients with Sjögren’s disease were excluded) have been analysed to explore the best system of management. The cause was definitely a parotid duct calculus in 36 patients, and evidence is presented that the same diagnosis probably applied to another 59 patients. Features suggesting a diagnosis of calculus included age (greater than 29 years); duration of the attacks of pain (less than 24 hours); cessation of salivation on the affected side; and a spurt of saliva heralding the relief of symptoms. Only three patients in the definite calculus group (8.3%) had no physical signs. However, had physical examination not included inspection and palpation of the parotid duct and its orifice from within the mouth 75% of the proven calculi would have been missed. The intraoral and anteroposterior plain radiographs are likely to be helpful, and sialography even more so. A sialographic appearance of a stricture in the main duct with proximal dilatation is usually due to a calculus. It would appear that calculi are the cause of recurrent unilateral parotid swelling (after exclusion of Sjögren’s disease) in an overwhelming proportion of patients with this symptom.

The first attack of swelling of the parotid gland is customarily managed by the family practitioner. The diagnosis usually made is mumps, especially in a child, and the patient is managed accordingly; should there be no more trouble there is no need to question this diagnosis. However, those who suffer a recurrence of the parotid gland enlargement, or those who have a prolonged attack require referral.

At hospital it will be noted whether or not the symptoms and signs are bilateral or unilateral. If bilateral or other salivary glands are involved, particularly if the patient complains of dry mouth and eyes, he should be investigated for the possibility of Sjögren’s syndrome. If unilateral, and provided that the swelling affects the whole gland and is not a lump in the parotid region (Hobsley, 1970), the condition may be referred to as chronic (or recurrent) unilateral parotitis. The following communication is concerned with this group of patients; 24 patients with Sjögren’s disease (benign lymphoepithelial disease of the parotid gland) are excluded.

Methods

Patients

During the period 1967–78 133 patients with unilateral recurrent parotid swelling were referred to

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one of us (M. H.). All patients had a full history, examination and radiology, including sialography. When Sjögren’s disease was suspected, serum antibodies were studied (including specific salivary gland antibodies) and parotid secretion studies were performed by the technique described by Curry and Patey (1964). When parotid tissue had been excised, the histological material was reviewed. The case material was collected and analysed and the 24 patients who were suffering from Sjögren’s disease (benign lymphoepithelial disease) were excluded as they form the material of another paper. The remaining 109 patients are presented here.

Results

After reviewing all the evidence, the 109 patients were grouped into the diagnostic categories shown in Table 1.

Group 1: Proved Calculi (36 patients)

They were patients in whom a parotid duct calculus was definitely proven by recovering the calculus. It had either passed spontaneously into the mouth, or been removed by operation.

The male to female ratio was 21:15. The right parotid gland was affected in 28 and the left in eight. The ages ranged from 29 to 69 years with a mean of 50.5 years; 92% of the patients were aged between 45 and 65 years.
The presenting symptoms are shown in Table 2. The attacks lasted for less than a day in 69% of patients and for less than a week in all. Particular attention is to be paid to the fact that pain and swelling were intermittent rather than continuous and that the patients had frequently noticed a spurt of saliva heralding the relief of the symptoms.

By the time they reached hospital only 18 had parotid enlargement and the whole gland was affected; the rest had no swelling, although the family doctor had detected an enlargement weeks earlier. Stones were palpable along the parotid duct in 26 patients (72-2%). In all these cases they were palpable from within the mouth, and in three from the outer aspect of the cheek as well. The stoma of Stensen's duct was deformed in 11, and a turbid discharge was expressible through the stoma in six. Three patients had no positive physical signs (8.3%).

Plain radiology, including an intraoral view (Hobsley, 1970) (Fig. 1) was performed on 35 patients and reported as positive in 25 (71-4%). When the radiographs of 31 patients were reviewed, the success rate of the intraoral view was 22 (71%), of the anteroposterior view, 17 (54-8%) (Fig. 2), and of the lateral view only six (19-4%). Sialography by Pattinson's (1969) technique was done in 34 patients; filling defects within the duct system were shown in 29 (85-3%), dilatation of the main duct system proximal to a localised region of apparent narrowing in four (11-8%) (Fig. 3), and a sudden cut-off of the duct with no penetration of the contrast medium beyond that point, in one.

Figure 4 shows the subsequent management of all the cases. One patient developed a parotid abscess after sialography; on incising the abscess one stone discharged itself. Two weeks later a second calculus

Table 1  Diagnostic groups of 133 patients managed for recurrent unilateral swelling of the parotid gland (24 patients with Sjögren's disease excluded)

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>No. of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Proved calculi</td>
<td>36</td>
</tr>
<tr>
<td>2 Presumed calculi</td>
<td>59</td>
</tr>
<tr>
<td>3 Parotitis secondary to other causes</td>
<td>3</td>
</tr>
<tr>
<td>4 Traumatic parotid duct stricture</td>
<td>2</td>
</tr>
<tr>
<td>5 Pending diagnosis</td>
<td>9</td>
</tr>
<tr>
<td>Total</td>
<td>109</td>
</tr>
</tbody>
</table>

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By the time they reached hospital only 18 had parotid enlargement and the whole gland was affected; the rest had no swelling, although the family doctor had detected an enlargement weeks earlier. Stones were palpable along the parotid duct in 26 patients (72-2%). In all these cases they were palpable from within the mouth, and in three from the outer aspect of the cheek as well. The stoma of Stensen's duct was deformed in 11, and a turbid discharge was expressible through the stoma in six. Three patients had no positive physical signs (8.3%).

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Table 2  Symptoms of 36 patients with proven parotid calculi

<table>
<thead>
<tr>
<th>Patients</th>
<th>Symptom</th>
<th>Pain Intermit</th>
<th>Continuous Pain</th>
<th>Swelling Intermit</th>
<th>Continuous Swelling</th>
<th>Cessation of salivation</th>
<th>Rush of saliva</th>
<th>Passing stones</th>
<th>Unpleasant taste</th>
<th>Pain in ear</th>
</tr>
</thead>
<tbody>
<tr>
<td>No.</td>
<td>33</td>
<td>3</td>
<td>32</td>
<td>6</td>
<td>24</td>
<td>22</td>
<td>6</td>
<td>5-6</td>
<td>11-1</td>
<td></td>
</tr>
<tr>
<td>Percentage</td>
<td>91-7</td>
<td>8-3</td>
<td>88-9</td>
<td>16-7</td>
<td>66-7</td>
<td>61-1</td>
<td>16-7</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 3  Symptoms of 59 patients presumed to have had calculi

<table>
<thead>
<tr>
<th>Patients</th>
<th>Symptom</th>
<th>Pain Intermit</th>
<th>Continuous Pain</th>
<th>Swelling Intermit</th>
<th>Continuous Swelling</th>
<th>Cessation of salivation</th>
<th>Rush of saliva</th>
<th>Passing stones</th>
<th>Unpleasant taste</th>
<th>taste</th>
</tr>
</thead>
<tbody>
<tr>
<td>No.</td>
<td>52</td>
<td>7</td>
<td>50</td>
<td>12</td>
<td>15</td>
<td>16</td>
<td>3</td>
<td>5</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>Percentage</td>
<td>88-1</td>
<td>11-9</td>
<td>84-7</td>
<td>20-3</td>
<td>25-4</td>
<td>27-1</td>
<td>5</td>
<td></td>
<td></td>
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</tr>
</tbody>
</table>
passed through the fistula, the latter then healed and the patient had no further trouble. Stomatoplasty was performed when the symptoms were persistent and the stone was at or near the duct orifice. Restomatoplasty was carried out if the stoma narrowed down. Superficial parotidectomy (Fig. 5) was performed if the symptoms were persistent and the stone was far away from the stoma, or the restomatoplasty had failed. After superficial parotidectomy all patients developed varying degrees of facial paresis, lasting for a few weeks in the majority of patients and up to nine months in two. One patient had a minor residual paresis of the upper lip for up to one year after operation: this was not noticeable in repose, and the patient did not complain of it. Another patient had a second intraoral operation to remove the parotid duct remnant containing a tiny calculus.

GROUP 2: PRESUMED CALCULI (59 patients)
In this group (Fig. 6) a presumptive diagnosis of calculus was made because of evidence that was
suggesive, but never confirmed by recovery of a stone. The evidence was one or more of the following features: (1) a definite past history of passing parotid calculi (three patients); (2) palpation of a stone within the duct (34 patients); (3) radiographic appearances diagnostic of a stone in 44 patients (18 positive on plain radiographs, all 44 on sialography; (4) histology of a parotidectomy specimen reported as typical of the large duct sialectasis produced by a stone (seven patients).

In 22 patients features (2) and (3) coexisted, while all seven patients with histological evidence had both a stone that was palpable and radiological evidence. All 34 palpable stones were felt from within the mouth and three of them were also palpable from the external aspect of the cheek. In seven patients histological material was obtained. The sections showed microcalculi, dilatation of the ducts, and periductal inflammation. No stones were found by the surgeon; but radiographs of two out of three excised parotid specimens examined showed localised calcification.

The symptoms of the 59 patients are shown in Table 3: it is evident that they experienced sharp pain in relation to meals, with cessation and rush of saliva in many. These features were very suggestive of calculi being the underlying cause. Moreover, the outcome of the management (Fig. 6) was in favour of calculi, as the attacks stopped spontaneously in most of the patients, presumably after a calculus had been passed.

GROUP 3: PAROTITIS SECONDARY TO ASCENDING INFECTION DUE TO OTHER CAUSES (three patients)

In this group of patients there was an oral identifiable cause of recurrent parotitis. In two, the attacks started immediately after a tooth extraction, while the third patient was in the terminal stages of disseminated lymphosarcoma with poor mouth hygiene. In all three patients the condition was apparently precipitated by ascending infection and progressed to abscess formation in the parotid gland. Surgical drainage was required in one patient: in the other two, pus was noted to be draining from the orifice of Stensen’s duct, and conservative management

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**Fig. 4** Management of 36 patients with parotid calculi.

36

3 Spontaneous passage of stones

1 Stones discharged on draining abscess

19 Stomatoplasty

13 Unsuitable for stomatoplasty

3 Stones removed

3 Stones subsequently passed

1 Stone removed

12 + 13

25 Superficial parotidectomy
succeeded. Once the infection was controlled, they remained symptom free. Subsequent sialography was normal in two but the patient with lymphosarcoma died without a sialogram.

**GROUP 4: TRAUMATIC PAROTID DUCT STRICTURE** (two patients)
These had lacerations of the face. On sialography a parotid duct stricture was shown in both. One had extravasation of the contrast medium at the site of the stricture and therefore required a superficial parotidectomy. The other responded to dilatation of the duct.

**GROUP 5: DIAGNOSIS PENDING GROUP** (nine patients)
In this group no final diagnosis has been made.
Seven of the nine patients had normal radiology including sialography. Of these seven, two had a history suggestive of calculi with a definite story of a gush of saliva, after which they have remained symptom-free for more than four years. It is possible that they had passed tiny calculi. Four of the remainder had recurrent attacks of intermittent pain and swelling of the parotid gland in relation to meals, but no history of cessation or gush of saliva; their parotid gland enlargement was never confirmed in hospital. They have now been symptom-free for more than one year, and it is possible that they, too, had had parotid calculi. The remaining patient was a woman aged 40 years who had positive parietal cell and thyroid antibodies and could have been a case of Sjögren's disease. She remained symptom-free for nine months after our investigation and then emigrated. The last two patients of this group had multiple strictures in their parotid ducts on sialography. Their symptoms were so severe that they required superficial parotidectomy, which cured them. The histological appearances were of chronic inflammation without the large duct dilatation suggestive of stone or any features indicative of Sjögren's disease, so the aetiology remains unknown.

**Discussion**

While there have been some reports of recurrent swellings affecting all the salivary glands—for example, Rose (1954) and Patey (1965)—many have concentrated on recurrent parotid gland enlargement (Payne, 1940; Pearson, 1961; Maynard, 1965). The present series is the only one devoted solely to unilateral recurrent swelling of the parotid gland, and the conclusions drawn from this study of 109 patients should be worthwhile.

It is clear that parotid stones are a real rarity in the group below 30 years, so recurrent enlargement of the parotid gland in that age group is most unlikely to be caused by a calculus.

The typical history in patients with a parotid calculus is of more or less intermittent pain and swelling, usually with meals or when brushing the teeth. The feature of brief attacks, lasting for minutes or hours, while often encountered, is not as dominant as it is in patients with submandibular salivary gland stones, because the submandibular duct is wider, the stone dislodges more easily, and relief of pain is faster (Thomson and Hobsley, 1973). Thus the attacks often last a day, rather than hours. Nevertheless, the distinction from the periods of weeks of dull aching in Sjögren's disease remains fairly clear cut. The patient may notice that the ejection of saliva ceases on the affected side and that a gush or spurt of saliva heralds the relief of his symptoms.

With regard to radiology, the value of the intraoral view is clearly shown and was formerly stressed by Hobsley (1970), and the anteroposterior view also helps. Sialography in expert hands can be expected to be positive in most cases.

However, the fact that a small stone may not
Recurrent unilateral swelling of the parotid gland reveal itself as a filling defect is as true here as in the biliary and urinary tracts. The contrast medium should therefore be as thin as possible. Even so, the medium may overwhelm a tiny stone which, if it has caused obstruction, expresses itself as an apparent area of narrowing with a sudden dilatation of the duct system proximal to it. These appearances are often wrongly interpreted as indicating a stricture, but in all four patients in whom they were present, a stone at that site was subsequently proved. A sudden cut-off of the duct with no contrast medium going further is a rare expression of the presence of a calculus.

A group of patients referred to a surgeon with a special interest in salivary glands may show bias towards the severe cases, yet when the groups of proved and presumed calculi are scrutinised (95 patients) it is clear that 46 patients (48.4%) cured themselves by passing their stones. Stomatoductoplasty, a minor operation (Hobsley, 1978), was successful in 10 out of 29 cases (34.5%). Superficial parotidectomy was necessary in 32 patients (33.7%). Although the superficial parotidectomy is known to be difficult in such cases (Conley, 1975), with patience during the operation and insistence on following the duct to its termination, very good results are to be expected. Certainly we do not advise this procedure as one for the occasional parotidectomist. Results in the group of patients with presumed calculi reflect the milder nature of the majority of parotid duct calculi and the fact that they may either pass, or trouble the patient very little. The fact that in seven patients with presumed calculi no stones were detected by the surgeon in the parotidectomy operation might have been related to the lag of eight months between sialography showing filling defects and the operation: perhaps the stones had passed, leaving the gland a seat of sialadenitis, a phenomenon well known with calculi at other sites.

Judging by the patients referred to us for a second opinion from other centres, the clinical and radiological examination of this problem is often incomplete. Oral inspection and palpation of the termination of Stensen's duct were frequently omitted, plain radiology often did not include the anteroposterior and intraoral views, and sialography was rarely used.

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Fig. 6 Management of 59 patients presumed to have had calculi.
performed. Without these examinations, 75% of our patients with stones would have remained incorrectly labelled as ‘recurrent parotitis—cause obscure’.

Turning to the patients with ascending parotitis secondary to some other cause; tooth extraction depresses mastication and the flow of saliva is reduced. Poor mouth hygiene in debilitated patients is a well-known cause for ascending parotitis, but in the 109 patients only one (the patient with disseminated lymphosarcoma) fell into this category. This reflects improved oral, dental, and nursing care.

The parotid duct stricture which improved on dilatation justifies this approach. The second stricture had much induration around it, and on sialography it was tight with extravasation of the contrast medium in parotid tissue. The idea of a plastic operation on the parotid duct from the outer aspect of the cheek was not entertained because (1) the two branches of the facial nerve running along with the parotid duct could have been easily injured amid the fibrous tissue; (2) even though the procedure might have been technically feasible, the stricture might recur. Therefore superficial parotidectomy, with the main nerve trunks followed forward and preserved, was performed and produced effective permanent cure.

We feel that parotid follow-up should be protracted. No patient is discharged from the clinic: even symptomless patients are seen annually. Therefore we have been left with a small group of patients who are now symptom-free for more than one year: while a final diagnosis has yet to be made, our provisional diagnosis, that a calculus had passed, is probably correct.

In only two of the 109 patients was the aetiology of the chronic parotitis obscure. We would like to stress the fact that, if the evidence we have given for our diagnosis of ‘presumed calculus’ is accepted, then causes of recurrent parotid gland swelling were obscure in only 1-8%. Therefore, the much larger proportions of patients in other series assigned to such obscure causes as congenital abnormality of the parotid gland, stricture at the termination of the parotid duct, hypersensitivity to mumps virus, or allergy (Smith, 1953; Rose, 1954) are unlikely to be correct: many of these were probably caused by stones.

References